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Speaker Abstracts

Robin Sequence

OS 0.1

C. Breugem and K. Evans


1Amsterdam, The Netherlands, 2Seattle, USA, 3Utrecht, The Netherlands, 4Alberta, Canada, 5Rotterdam, The Netherlands, 6Paris, France, 7Tuebingen, Germany, 8Toronto, Canada, 9Boston, USA, 10New York, USA, 11Chapil Hill, USA, 12Amsterdam, The Netherlands, 13Utrecht, The Netherlands, 14Bauru, Brazil, 15Paris, France, 16Toronto, Canada

This preconference symposium on Robin Sequence is designed to continue the discussion generated at the two previous International Robin Sequence Conferences.

Evaluations among infants with Robin sequence varies substantially. Quantifying respiratory compromise in infants with Robin sequence is challenging, however is crucial to determine and compare treatment approaches and outcomes. If we do not have a way to objectively measure and compare treatment options, we will never be able to assess if our treatments are successful.

The main goal is to reach a consensus regarding objective evaluations of the airway among international experts involved in the care of infants with Robin sequence.

Robin Sequence Working group:

Corstiaan Breugem, Amsterdam, The Netherlands
Kelly Evans, Seattle, USA
Bob Logjes, Utrecht, The Netherlands
Joanna Maclean, Alberta, Canada
Koen Joosten, Rotterdam, The Netherlands
Veronique Abadie, Paris, France
Christian Poets, Tuebingen, Germany
Chris Forrest, Toronto, Canada
Cory Resnick, Boston, USA
Roberto Flores, New York, USA
Carlton Zdanski, Chapil Hill, USA
Frea Kruisinga, Amsterdam, The Netherlands
Maartje Haasnoot, Utrecht, The Netherlands
Ivy Kiemle Trindade Suedam, Bauru, Brazil
Brigitte Fauroux, Paris, France
Resha Amin, Toronto, Canada
Language development in children with 22q11.2 deletion syndrome
E Van Den Heuvel

Abstract content: 22q11.2 deletion syndrome (22q11DS) is a common copy number variant (CNV) with high risk for neurodevelopmental disorders (NDD). Language is a key predictor for developmental outcomes and a warning sign for families to note that their child might be at risk for a NDD. Evaluation of language ability in children with genetic syndromes supports the interactional exploration of the behavioural phenotype. Considerable research has been devoted to speech disorders and velopharyngeal insufficiency, rather less attention has been paid to the language abilities of children with 22q11DS. Some children with 22q11DS seem to acquire some near-normal language form and language content skills in line with their level of intellectual functioning, but recent studies indicate challenges in language comprehension and pragmatic language skills in school-aged children with 22q11DS. The inability to use contextual cues for effectively understanding, organising and expressing language differentiates children with 22q11DS from children with idiopathic intellectual disability (IID). We summarize results of our recent studies regarding language skills in children with 22q11DS and address implications for clinical practice and management. Finally, we indicate some opportunities for future research.
Speech outcomes at age 10 in the scandcleft studies: velopharyngeal function and consonant proficiency part 1


1Speech and Language Pathology Unit, Institute of Neuroscience and Physiology, Göteborg, Sweden, 2Cleft Palate and Craniofacial center, Helsinki University Central Hospital, Helsinki, Finland, 3Department of Speech and Language Disorders, Statped sorost, Oslo, Norway, 4Greater Manchester Cleft Unit, Royal Manchester Children's Hospital, Manchester, United Kingdom, 5Division of Speech and Language Pathology, Sahlgrenska University Hospital, Göteborg, Sweden, 6The Royal Hospital fo Sick Children, Belfast, United Kingdom, 7Department of Speech and Language Disorder, Statped sorost, Oslo, Norway, 8Cleft Palate and Craniofacial Center, Helsinki University Central Hospital, Helsinki, Finland, 9Copenhagen Cleft Palate Center, University Hospital of Copenhagen, Copenhagen, 10Cleft Palate Center, Cleft Palate Center, Aarhus, Denmark, 11Division of Speech and Language Pathology, Karolinska Institutet and Karolinska University Hospital, Stockholm, 12Division of Speech and Language Pathology, Linköping University, Linköping, 13Stockholm Craniofacial Team, Karolinska University Hospital, Stockholm, Sweden, 14Statped Vest, Bergen, Norway, 15Department of Nordic Studies and Linguistics, University of Copenhagen, Copenhagen, Denmark

Background: In the Scandcleft studies, 448 children born with non-syndromic UCLP are being followed from birth to adulthood to assess the influence of surgical treatment protocols on speech and dentofacial development. At five years of age, no significant differences were found in velopharyngeal competence (VPC) or incompetence (VPI) between arms in any trial (or centre). The frequency of VPC was disappointingly low ranging between 39-62% in arms. Articulation outcomes were also poor with an average Percent Consonants Correct (PCC) score around 80% in all trials (which is more than 2 SDs below that of typically developing peers). A significant difference was found between arms in trial 1, with the poorest outcome in children who received hard palate closure at 36 months. Burden of care in terms of number of secondary speech surgeries and speech visits differed. Thus, it is important to follow-up speech outcome at 10 years of age.

Aims: To study the overall speech proficiency in 10-year-old children born with unilateral cleft lip and palate and to compare speech outcomes, velopharyngeal function and consonant proficiency, between the four surgical methods. A further aim was to investigate the longitudinal speech outcomes between 5 and 10 years of age.

Methods: Three different surgical protocols for primary palatal repair were tested against a common procedure in the total cohort of 448 children born with complete UCLP. Video recordings of 394 children (139 girls, 255 boys) were available and perceptually analysed independently by SLTs blinded to the surgical protocol. Target sounds from a single word test were phonetically transcribed and the SLTs rated the velopharyngeal function based on continuous speech from the Bus story test. Longitudinal data were available for 349 children. Background data on secondary velopharyngeal surgery and number of speech therapy visits will be presented.

Results: Overall data on articulation proficiency will include PCC with and without /s/, as well as cleft speech characteristics and developmental speech characteristics. Results on velopharyngeal function will include an overall measure of velopharyngeal function based on single words (the VPC-sum), and from connected speech (the VPC-rate). We will also present outcome data for each of the surgical protocols included in the three trials. Finally, correlations will be presented between the speech variables at 5 and 10 years of age to provide longitudinal developmental results within each trial.

Summary/Conclusion: Results will be discussed in light of previously reported findings of 10-year-olds with UCLP and the previously reported 5 year findings of the participants.
Orthopedic manifestations in the 22q11.2 deletion syndrome
J. Homans

Abstract Content: The 22q11.2 Deletion syndrome (22q11.2DS) is a relatively common genetic disorder with broad phenotypic expression. The orthopedic manifestations are not as well described as some other, more vital, disorders. However, skeletal anomalies are common within 22q11.2DS and occur throughout the body: from the cervical spine to the toes. Nearly all 22q11.2DS patients have at least one cervical spine anomaly. These patients frequently suffer from increased segmental motion in the neck (56%), which may occur at multiple levels, in some cases this is accompanied with the development of neurological symptoms. Moreover, there are four studies that describe platybasia in 11.5-91.2% of the cases. In the case of neurologic signs and/or symptoms suggestive of spinal canal encroachment or impingement, advanced imaging such as (dynamic) MRI is recommended. Whether these findings should lead to active counseling and the advice to refrain from collision sports seems to be highly dependent on surgeons preference throughout the different centers. Nearly 50% develops a scoliosis and there is a 30 times increased risk of a club foot as compared to the general population. In a study by Ming et al., the largest study on extremity abnormalities within 22q11.2DS, six percent of the 22q11.2DS patients had abnormalities of the fingers. These abnormalities include polydactyly (4%), clinodactyly (1%), camptodactyly (1%) and rotation deformities (1%, usually the fifth finger). Moreover, in a study by Derbent et al. syndactyly was described. Last, the same deformities that can occur at the fingers, can occur at the toes (clinodactyly, polydactyly and syndactyly), as well as overfolded toes and hammer toes. These extremity abnormalities, such as polydactyly, can be a clue towards the diagnosis of 22q11.2DS.
Abstract Content: The 22q11.2 deletion syndrome (22q11DS) is associated with increased risk of congenital abnormalities across multiple organ systems. Associated phenotypic features include congenital heart defects and palatal abnormalities. The same deletion also influences development and function of the brain, such that individuals with 22q11DS are at increased risk for neurodevelopmental disorders (e.g. intellectual disability, language disorder, autism spectrum disorder, ADHD) and psychiatric disorders. Regarding the latter, the risk of psychotic disorders, in particular schizophrenia, is 25%, which represents the highest known risk for this illness conferred by a single genetic variant. In this presentation, I will review developmental and psychiatric presentations in individuals with 22q11DS, what is known about their association with other phenotypic expressions in 22q11DS and their genetic underpinnings.
Abstract Content: Regular cardiac follow-up is mandatory in adult patients with 22q11.2 deletion syndrome who have congenital cardiac abnormalities. Between 25%-75% of patients with 22q11.2 deletion syndrome have a congenital heart disease, depending on the age studied. These congenital heart diseases mostly consist of conotruncal defects, for example tetralogy of Fallot. Genetic testing for 22q11 deletions is advised for patients with conotruncal cardiac defects, for recognition and management of comorbidities and for counseling on the potential risk of recurrence in offspring. During follow-up they may need repeat cardiac interventions.
22q11 Age 0-12 Years

OS 3.3

High number of healthcare professionals involved in the care for children with 22q11.2 deletion syndrome at time of first referral to national reference center
M Houben1, L Heestermans1, M van Susante2, H Veye2, A Mink van der Molen3
1Pediatrics, 2Psychology, 3Plastic Surgery, University Medical Center Utrecht, Utrecht, Netherlands

Background: The 22q11.2 deletion syndrome (22q11DS) has a high prevalence (1:2,000–4,000), resulting in the birth of approximately 45-90 affected children per year in the Netherlands. The syndrome has a high phenotypic variability, with potential involvement of virtually all human organ systems. Intellectual disability and psychologic and psychiatric problems further increase the complexity and the morbidity of the disorder. Delayed diagnosis and a large physical and psychosocial burden have been associated with a high number of healthcare professionals (HCP) in North-American countries.

Aims: To estimate the number of HCPs that are involved in the care for children with 22q11DS in the Netherlands at time of first referral to the national reference center. To describe the spectrum of HCPs involved. To study the relation between age at referral and total number of HCPs involved.

Methods: Patients were reviewed for all newly referred patients at the Wilhelmina Children's Hospital (WKZ / UMCU, Utrecht, The Netherlands). Relevant demographic and HCP data were extracted. The IRB waived the necessity of informed consent, given the retrospective and descriptive nature of the study.

Results: In 2017, a total of 26 children with 22q11DS (15 boys, 11 girls) were newly referred, from 8 of the 12 Dutch provinces. The median age at time of first visit was 6 years (interquartile range (IQR) 3-11 years). The mean total number of HCPs involved at time of first visit was 4.1, mostly appointed in local regional hospitals and facilities (90%). The HCPs that were all involved in >10% of patients were a General practitioner (100%), Pediatrician (73%), Speech therapist (31%), Physiotherapist (23%), Pediatric cardiologist (19%), Dietician (15%), Pediatric pulmonologist (12%), Ophthalmologist (12%), Rehabilitation medicine (12%). The children without a general pediatrician (median 12 years; IQR 3-13) tended to be older than those with a general pediatrician (median 5 years, IQR 3-7), P= 0.08). The number of HCPs involved was negatively associated with age at referral (Spearman’s rho -0.46; P= 0.02).

Summary/Conclusion: Children with 22q11DS in the Netherlands are referred from large parts of the country to the national reference center, at a median age of 6 years. Those that do not have a general pediatrician tend to be older (12 years), suggesting that morbidities have become less complex at that age. The top-five of most frequently involved HCPs are General Practitioner (100%), Pediatrician (73%), Speech therapist (31%), Physiotherapist (19%) and Dietician (19%). The number of active HCPs is negatively associated with age at referral, suggesting inactivation of morbidities. More qualitative and transition research is needed to achieve a better understanding of disease activities and patient and family needs throughout childhood and adolescence.
Models of 22q team care in the united states
Emily Gallagher*, Jill Arganbright1, Daniela Schweitzer2, Donna McDonald-McGinn3, Courtney Hall4, Susan Hughes5, Kaylee Paulsgrove6, Alexis Johns7, Adriane Baylis8, Oksana Jackson9 1Otolaryngology, Children's Mercy Hospital, Kansas City, MO, 2Genetics, Children's Hospital of Los Angeles, Los Angeles, CA, 3Genetics, Children's Hospital of Philadelphia, Philadelphia, PA, 4Plastic Surgery, Nationwide Children's Hospital, Columbus, OH, 5Genetics, Children's Mercy Hospital, Kansas City, MO, 6Speech and Language Pathology, Seattle Children's Hospital, Seattle, WA, 7Psychology, Children's Hospital of Los Angeles, Los Angeles, CA, 8Speech and Language Pathology, Nationwide Children's Hospital, Columbus, OH, 9Plastic Surgery, Children's Hospital of Philadelphia, Philadelphia, PA, United States

Background: Numerous cleft/craniofacial teams in the United States provide care for patients with 22q11.2-related disorders. Care includes evaluation and management of feeding difficulties, immunodeficiency, developmental delays, behavior and mental health conditions, and speech and language disorders, including velopharyngeal dysfunction (VPD). Because of multisystem involvement and care needs that vary with age of the child, these patients require complex coordinated care from multidisciplinary teams. Unlike the well-established team care standards for cleft and craniofacial care, management models for patients with 22q vary widely.

Aims: The primary aim of this presentation is to review models of team structure and function across several large institutions in the United States. We will compare management approaches, differences in resources, and highlight challenges that these centers experience when managing the complex care of these patients.

Methods: Five cleft/craniofacial teams in the United States who provide care for large numbers of patients with 22q led a course at the American Cleft Palate-Craniofacial Association (ACPA) in 2019. The collaborative presentation included discussion of the structure and format of the teams providing care for patients with 22q, including specialist composition, clinic format, care coordination strategies, team conferencing, and approaches to patient education and outreach. Challenging clinical scenarios were also discussed, including management of VPD, perioperative management, evaluation and support of learning needs, evaluation of psychiatric disorders, and overall care coordination challenges.

Results: While some core team members providing care for patients with 22q are consistent across regions of the United States, there is variability among the largest teams. Availability of resources on the team and in the community often influences management decisions, particularly around evaluation and management of VPD, perioperative concerns, learning evaluations, developmental support, and management of behavioral and psychiatric disorders. Coordinating care for patients is challenging for many teams, and successful models will be highlighted.

Summary/Conclusion: The structure of teams providing care for patients with 22q varies across regions, although core team members are usually consistent. Teams face unique challenges, often related to geographic distribution of patients and available resources. As a result, management approaches vary in ways that may impact clinical outcomes of patients with 22q
Movement disorders in adults with 22q11.2 deletion syndrome

E Boot¹
¹'s Heeren Loo, Amersfoort, Netherlands

Abstract Content: 22q11.2 deletion syndrome (22q11.2DS) is the most common microdeletion syndrome in humans, occurring in approximately 1:3000 live births. Manifestations of 22q11.2DS involve multiple organ systems, both congenital and later onset, displaying considerable variation in the spectrum and severity of its expression between individuals. Due to improved pediatric survival rates, individuals with 22q11.2DS are an increasing population as the majority now lives into adulthood. Recent studies suggest that, in addition to early-onset.
Scandcleft Randomised Trial Update

OS 6.1
Scandcleft randomised trials: planning and management
G Semb

1Department of Plastic and Reconstructive Surgery, Oslo University Hospital, Oslo, Norway

Background: Longstanding uncertainty surrounds the selection of surgical protocols for the closure of unilateral cleft lip and palate, and randomised trials have only rarely been performed. Aims: This series will describe three randomised trials of primary surgery for children born with complete unilateral cleft lip and palate (UCLP). It presents the protocol developed for the trials in CONSORT format, and describes the management structure that was developed to achieve the long-term engagement and commitment required to complete the project.

Method: The study involved 4 years of planning and patient recruitment started in 1997. Ten established national or regional cleft centres participated. These were Aarhus, Copenhagen, Gothenburg, Helsinki, Linkping, Stockholm, Oslo, Bergen, Manchester and Belfast. Lip and soft palate closure at 3–4 months, and hard palate closure at 12 months served as a common method in each trial. Trial 1 compared this with hard palate closure at 36 months. Trial 2 compared it with lip closure at 3–4 months and hard and soft palate closure at 12 months. Trial 3 compared it with lip and hard palate closure at 3–4 months and soft palate closure at 12 months. The primary outcomes are speech and dentofacial development, with a series of perioperative and longer-term secondary outcomes. Multi-disciplinary follow-up at age 5 years have been completed and published. Ongoing orthodontic follow-up is done at 8 years (before any orthodontic treatment and alveolar bone grafting), at 10 years, then before and after orthodontic treatment in the permanent dentition and at 18-20 years. Speech follow-up is done at 10 years and at 18-20 years.

Results: Recruitment of 448 infants took place over a 9-year period. Only 1 patient failed to attend at the 5 years multi-disciplinary record taking. One UK centre discontinued the follow-up at age 5 years. Outcomes of velopharyngeal function and consonant production at age 10 years and occlusion and maxillary growth, naso-labial appearance and dental anomalies at 8 years will be discussed in the following presentations. A hundred and thirty individuals have now reached early adulthood (18-20 years). Standardised record collection continues and will be completed in 2025.

Conclusion: Longitudinal multidisciplinary, multi-national, multi-linguistic and multi-cultural collaboration has been met by a high level of enthusiasm supported by a flat, inclusive organisational structure. Efforts have been made to communicate with each and every one of the 85 Scandcleft members to encourage openness and ownership. The outcomes recorded and the numbers of analyses for each outcome and time point are described in this series of presentations.
**OS 6.2**

**Surgical results**

J. Rautio\(^1\), M. Andersen\(^2\), S. Bolund\(^3\), J. Hukki\(^4\), H. Vindenes\(^4\), P. Davenport\(^5\), K. Arctander\(^6\), O. Larson\(^7\), A. Berggren\(^8\), F. Åbyholm\(^9\), D. Whitby\(^6\), A. Leonard\(^10\), J. Lilja\(^11\), E. Neovius\(^7\), A. Elander\(^11\), A. Heliövaara\(^1\), P. Eyres\(^12\), G. Semb\(^13\)

\(^1\)Cleft Palate and Craniofacial Center, Helsinki University Hospital, Helsinki, Finland, \(^2\)Copenhagen Cleft Palate Centre, University Hospital of Copenhagen, Copenhagen, \(^3\)Cleft Palate and Craniofacial Center, Helsinki University Hospital, Helsinki, Denmark, \(^4\)Center for Cleft Lip and Palate, Bergen University Hospital, Bergen, Norway, \(^5\)Department of Plastic Surgery, Royal Manchester Children’s Hospital, Manchester, United Kingdom, \(^6\)Department of Plastic and Reconstructive Surgery, Rikshospitalet Oslo University Hospital, Oslo, Norway, \(^7\)Stockholm Craniofacial Team, Karolinska University Hospital, Stockholm, \(^8\)Department of Plastic Surgery, Linköping University Hospital, Linköping, Sweden, \(^9\)Department of Plastic and Reconstructive Surgery, Rikshospitalet Oslo University Hospital, Oslo, Norway, \(^10\)The Royal Hospital for Sick Children, The Royal Hospital for Sick Children, Belfast, United Kingdom, \(^11\)Department of Plastic Surgery, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden, \(^12\)Dental School, University of Manchester, Manchester, United Kingdom, \(^13\)Dental School, University of Manchester, Manchester, Norway

**Background:** Ten cleft centres in Denmark, Finland, Norway, Sweden, and the UK participated in a set of three randomised trials of primary surgery of unilateral cleft lip and palate. Three groups of centres (Trials 1, 2 and 3) tested a newly defined common technique for palatal repair Arm A, closing the soft palate at the same time as the lip at 3-4 months and the hard palate at 12 months, against their local protocols: Arm B, same as above but closing the hard palate at 36 months, Arm C closing the lip at 3-4 months and entire palate at 12 months. Arm D closing the lip and hard palate at 3-4 months and the soft palate at 12 months. Arm A was familiar to most of the surgeons in Trial 1 but not to the surgeons in the other Trials.

**Aims:** To evaluate surgical events and complications of the 448 (293 boys, 155 girls) patients with complete unilateral cleft lip and palate (UCLP) enrolled in the three trials.

**Methods:** Airway problems and rates of fistula and surgery for velopharyngeal incompetence (VPI) were assessed until the youngest patient of the study had reached the age of 9 years. Pearson Chi-square statistical analysis was used to compare the outcomes.

**Results:** In Trial 1 there were no significant differences in the above parameters between methods A and B. In Trials 2 and 3 there were more airway problems in Arm A than the traditional local protocols (Arms C or D). In Trial 2 fistula rates were higher than in Trial 1. In Trial 3 both fistula and VPI surgery rates were higher than in Arms A and B.

**Summary/Conclusion:** The results do not provide statistical evidence that one technique is better than others when the surgeons were performing the method they were most familiar with. Using the more unfamiliar technique A in trials 2 and 3 led to an increase in fistulas.
Scandcleft randomised control trial: a descriptive study of postoperative nursing care following first stage cleft repair and the issues faced in providing care

P Bannister¹, N Lindburg², K Jeppeson³, U Elfving-Little⁴, A Semmingsen⁵, A Paganini⁶, A Gustavsson⁷, E Slevin⁸, P Eyres⁹, G Semb¹⁰

¹dental, manchester university, Manchester, United Kingdom, ²Oslo University Hospital, Dental, Bergen, Norway, ³Cleft Palate Center, Cleft Palate Centre, Copenhagen, Denmark, ⁴Cleft Palate and Craniofacial Centre, University Hospital, Helsinki, Finland, ⁵Division of Surgery and Clinical Neuroscience, Hospital Rikshospitalet Oslo University, Oslo, Norway, ⁶Department of Plastic Surgery Sahlgrenska Academy Gothenburg, SE, University of Gothenburg, Gothenburg, ⁷Maxillofacial Unit, University Hospital, Linköping, Sweden, ⁸Cleft Unit, Royal Belfast Hospital for Sick Children, Belfast, ⁹Dental School, University of Manchester, ¹⁰University of Manchester, Dental School, Manchester, United Kingdom

Background: The surgical repair of a cleft lip and palate is one of the most common scheduled procedures carried out on young children under the age of one year. Little clinical evidence exists to guide nurses in providing optimal care following surgery.

Aims: The aim of the study was to describe the post-operative care after the first surgical procedure in the Scandcleft project and to compare how variation in both surgical and non-surgical factors affect this care.

Methods: A post-operative recovery form was developed and sent to the 10 trial sites in the Scandcleft project. Prospective and retrospective data was recorded on post-operative environment, conceptual age and weight, post-operative complications, medication history, oral feeding outcomes and length of hospital stay.

Results: 403 nursing forms from the first surgical procedure were returned for analysis. Variations in postoperative care was influenced by both local protocols, surgical technique and difficulties in providing care following the use of unfamiliar surgical techniques. Calculation of age at time of surgery was not uniform, calculated either from date of birth or expected date of delivery. This may have had some effect on the post-operative results.

Summary/Conclusion: Many of the differences identified in post-operative care were influenced by local protocols. As little clinical evidence exists to guide optimal management, there is little evidence-based practice to guide care when nurses are caring for infants following both familiar and unfamiliar surgical techniques. Post-operative recovery may play a significant role in the future selection of surgical protocols and future trials need to consider cross study site training to familiarize nurses, prior to any changes in surgical method.
Background: In the Scandcleft studies, 448 children born with non-syndromic UCLP are being followed from birth to adulthood to assess the influence of surgical treatment protocols on speech and dentofacial development. At five years of age, no significant differences were found in velopharyngeal competence (VPC) or incompetence (VPI) between arms in any trial (or centre). The frequency of VPC was disappointingly low ranging between 39-62% in arms. Articulation outcomes were also poor with an average Percent Consonants Correct (PCC) score around 80% in all trials (which is more than 2 SDs below that of typically developing peers). A significant difference was found between arms in trial 1, with the poorest outcome in children who received hard palate closure at 36 months. Burden of care in terms of number of secondary speech surgeries and speech visits differed. Thus, it is important to follow-up speech outcome at 10 years of age.

Aims: To study the overall speech proficiency in 10-year old children born with unilateral cleft lip and palate and to compare speech outcomes, velopharyngeal function and consonant proficiency, between the four surgical methods. A further aim was to investigate the longitudinal speech outcomes between 5 and 10 years of age.

Methods: Three different surgical protocols for primary palatal repair were tested against a common procedure in the total cohort of 448 children born with complete UCLP. Video recordings of 394 children (139 girls, 255 boys) were available and perceptually analysed independently by SLTs blinded to the surgical protocol. Target sounds from a single word test were phonetically transcribed and the SLTs rated the velopharyngeal function based on continuous speech from the Bus story test. Longitudinal data were available for 349 children. Background data on secondary velopharyngeal surgery and number of speech therapy visits will be presented.

Results: Overall data on articulation proficiency will include PCC with and without /s/, as well as cleft speech characteristics and developmental speech characteristics. Results on velopharyngeal function will include an overall measure of velopharyngeal function based on single words (the VPC-sum), and from connected speech (the VPC-rate). We will also present outcome data for each of the surgical protocols included in the three trials. Finally, correlations will be presented between the speech variables at 5 and 10 years of age to provide longitudinal developmental results within each trial.

Summary/Conclusion: Results will be discussed in lig
OS 6.5
Scandcleft randomised trials: speech outcomes at age 10 years: velopharyngeal function and consonant proficiency: part ii

E Willadsen1, A Lohmander2, S Alaluusua3, R Aukner4, M Boers5, M Bowden6, J Davies6, C Havstam7, C Hayden8, Ø Hide4, E Hölttä3, L Dahl Jørgensen5, M Kisling-Møller9, I Lundeborg Hammarström10, J Bogh Nielsen9, J Nyberg2, N Pedersen11, T Rasmussen11, H Søgaard Andersen7, C Persson12
1Department of Nordic Studies and Linguistics, University of Copenhagen, Copenhagen, Denmark, 2Division of Speech and Language Pathology, Karolinska Institutet and Karolinska University Hospital, Stockholm, Sweden, 3Cleft Palate and Craniofacial Center, Helsinki University Central Hospital, Helsinki, Finland, 4Department of Speech and Language Disorder, Statped Sørøst, Oslo, Norway, 5Copenhagen Cleft Palate Center, University Hospital of Copenhagen, Copenhagen, Denmark, 6Greater Manchester Cleft Unit, Royal Manchester Children's Hospital, Manchester, United Kingdom, 7Division of Speech and Language Pathology, Sahlgrenska University Hospital, Gothenburg, Sweden, 8, The Royal Hospital for Sick Children, Belfast, United Kingdom, 9Cleft Palate Center, Cleft Palate Center, Aarhus, Denmark, 10Division of Speech and Language Pathology, Linköping University, Linköping, Sweden, 11Statped Vest, Statped Vest, Bergen, Norway, 12Speech and Language Pathology Unit, Institute of Neuroscience and Physiology, Gothenburg, Sweden

Background: In the Scandcleft studies, 448 children born with non-syndromic UCLP are being followed from birth to adulthood to assess the influence of surgical treatment protocols on speech and dentofacial development. At five years of age, no significant differences were found in velopharyngeal competence (VPC) or incompetence (VPI) between arms in any trial (or centre). The frequency of VPC was disappointingly low ranging between 39-62% in arms. Articulation outcomes were also poor with an average Percent Consonants Correct (PCC) score around 80% in all trials (which is more than 2 SDs below that of typically developing peers). A significant difference was found between arms in trial 1, with the poorest outcome in children who received hard palate closure at 36 months. Burden of care in terms of number of secondary velopharyngeal surgeries and speech visits differed. Thus, it is important to follow-up speech outcome at 10 years of age.

Aims: To study the overall speech proficiency in 10-year-old children born with unilateral cleft lip and palate and to compare speech outcomes, velopharyngeal function and consonant proficiency, between the four surgical methods. A further aim was to investigate the longitudinal speech outcomes between 5 and 10 years of age.

Methods: Three different surgical protocols for primary palatal repair were tested against a common procedure in the total cohort of 448 children born with complete UCLP. Video recordings of 394 children (139 girls, 255 boys) were available and perceptually analysed independently by SLTs blinded to the surgical protocol. Target sounds from a single word test were phonetically transcribed and the SLTs rated the velopharyngeal function based on continuous speech from the Bus story test. Longitudinal data were available for 349 children. Background data on secondary velopharyngeal surgery and number of speech therapy visits will be presented.

Results: Overall data on articulation proficiency will include PCC with and without /s/, as well as cleft speech characteristics and developmental speech characteristics. Results on velopharyngeal function will include an overall measure of velopharyngeal function based on single words (the VPC-sum), and from connected speech (the VPC-rate). We will also present outcome data for each of the surgical protocols included in the three trials. Finally, correlations will be presented between the speech variables at 5 and 10 years of age to provide longitudinal developmental results within each trial.

Discussion: Results will be discussed in light of previously reported findings of 10-year-olds with UCLP and the previously reported 5 year findings of the participants.
OS 6.6
Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate. Dental arch relationships at 8 years.

A Heliövaara¹, P Skaare², A Küseler³, K Mølsted⁴, A Karsten⁵, A Marcusson⁶, E Brinck⁷, S Rizell⁷, P Sæle⁸, M Najar Chalien⁷, H Bellardie⁹, J Mooney⁹, P Eyres¹⁰, W Shaw¹¹, G Semb¹²
¹Department of Plastic Surgery, Cleft Palate and Craniofacial Center, Helsinki, Finland, ²Department of Plastic and Reconstructive Surgery, Oslo University Hospital, Rikshospitalet, Oslo, Norway, ³Cleft Palate Center, Aarhus, ⁴Copenhagen Cleft Palate Center, University Hospital of Copenhagen, Copenhagen, Denmark, ⁵Division of Orthodontics, Department of Dental Medicine, Stockholm Craniofacial Team, Karolinska Institutet, Stockholm, ⁶Dentofacial Orthopedics, Maxillofacial Unit, University Hospital, Linköping, ⁷Orthodontic clinic, University Clinics of Odontology Gothenburg, Gothenburg, Sweden, ⁸Oral Health Center of Excellence/Western Norway, Bergen, Norway, ⁹Greater Manchester Cleft Unit, Royal Manchester Children's Hospital, ¹⁰Division of Dentistry, ¹¹Dental School, University of Manchester, Manchester, United Kingdom, ¹²Department of Plastic and Reconstructive Surgery, Oslo University Hospital, Rikshospitalet and Statped Sørøst, Oslo, Norway

Background: Ten cleft centres in Denmark, Finland, Norway, Sweden, and the UK participated in a set of three randomized trials of primary surgery. Three groups of centres (Trials 1, 2, and 3) tested their traditional local protocols (Arms B, C, and D) against a newly defined common method (Arm A).

Aims: To evaluate dental arch relationships at age 8 years after four different protocols of primary surgery for unilateral complete cleft lip and palate (UCLP).

Methods: Study models of 411 patients (270 boys and 141 girls) at the mean age of 8.1 years (range 7.0-10.0) were available. Dental arch relationships were assessed using the Goslon index by a blinded panel of 11 orthodontists. Kappa statistics were calculated to assess reliability. The trials were tested statistically with t- and Chi-square tests.

Results: Comparisons within each trial showed no statistically significant differences in the mean 8-year index scores or their distributions between the common method and the local team protocol. The mean index scores were Trial 1: Arm A 3.03, Arm B 2.82, Trial 2: Arm A 2.78, Arm C 2.64 and Trial 3: Arm A 3.06, Arm D 3.08. The intra- and interrater reliabilities were acceptable.

Summary/Conclusion: The results of the three trials do not provide statistical evidence that one technique is better than the others.
Airway Management for Patients with Craniofacial Anomalies

OS 7.2
Perioperative adverse events in patients undergoing primary cleft palate repair: a prospective analysis.
O Jackson1, A Garcia-Marcinkiewicz2, K Peeples2, L Sequera Ramos2, P Hu2, M Basta3, J Fiadjoe2
1Plastic Surgery, 2Anesthesiology and Critical Care Medicine, The Children’s Hospital of Philadelphia, Philadelphia, 3Plastic Surgery, Brown University, Providence, United States

Background: Cleft palate repair is a common surgical procedure performed worldwide in the pediatric population. The reported incidence of airway-related complications in patients undergoing palatoplasty ranges from 5-30%, and can result in significant patient morbidity, parental distress, and increased health care costs.

Aims: This study aimed to identify risk factors for adverse perioperative events including escalation of care and readmission after cleft palate repair.

Methods: Prospective data were collected of consecutive patients less than 18 years of age who underwent primary palatoplasty at a single institution between January 2015 and January 2019. The study was approved and a waiver of consent was granted by the hospital Institutional Review Board. Perioperative Adverse Event (PAE) was defined as hypoxemia, laryngospasm or bronchospasm, airway obstruction, hypoventilation, accidental tracheal extubation, or reintubation, and vomiting. Secondary outcomes included unplanned intensive care unit admission, or urgent care or emergency department visits.

Results: Two hundred ninety-two patients with median age 11.5 months [25-75% interquartile range (IQR) 10, 17.5 months] were included. The majority of patients were American Society of Anesthesiology (ASA) class II or III (n = 181, 62.9%), (n = 76, 26.4%), respectively. Cleft distribution included submucous (n = 41), 14.1%, Veau 1 (n = 45) 15.5%, Veau 2 (n = 94), 32.3%, Veau 3 (n = 81), 27.8%, Veau 4 (n = 30), 10.3%. A primary Furlow palatoplasty was performed in (n = 272), 96.5%. Pierre Robin Sequence (n = 44), 15%, was the most common syndrome/anomaly. Fifty-one patients (17.5%) experienced at least one PAE, with 13 patients (4.5%) experiencing an event intraoperatively, and 38 patients (13%) at least one post-operative event. Twelve patients (4%) had two or more separate post-operative events. The common PAEs (separate events) were hypoxemia (40 events), obstruction (17 events), hypoventilation (9 events), laryngospasm (4 events), bronchospasm (4 events), vomiting (4 events), reintubation (3 events), and one patient experienced Acute Respiratory Distress Syndrome (ARDS). The majority of post-operative events occurred close to the time of surgery with 9 (23.1%) occurring while still in the operating room, 15 (38.5%) in the post-anesthesia care unit, and 10 (25.6%) on the inpatient floor within 12 hours of surgery. PAEs were associated with syndromic status OR 2.0, 95% CI (1.04, 3.8), p = 0.04; nasopharyngeal (NP) airway placement at the end of the case OR 3.8, 95% CI (1.6, 8.6), p = 0.0011; and prolonged emergence time with every minute increase in emergence time associated with 6% increase in the risk of PAE, OR 1.06, 95% CI (1.03, 1.09); p < 0.0001. Patients who received paralytic and had no reversal agent experienced increased risk of post-operative adverse events, OR 3.45, 95% CI (1.7, 6.9), p = 0.0003. Unplanned Intensive Care Unit (ICU) admissions occurred in 15 (5%) of patients. Patients with PAE had a greater median length of stay (LOS) than those with no PAE [Median LOS with PAE = 2, IQR (1, 2)], [Median LOS without PAE 1, IQR (1,2)]; p = 0.0001.

Summary/Conclusion: PAE incidence was 17.5% after palatoplasty, with most PAEs occurring in the immediate post-operative period. Patients with PAEs had a greater length of hospital stay.
Pierre Robin Sequence: Patient Care in 14 Francophone Centers

A De Buys Roessingh, O El Ezzi, G Herzog, J Leca, V Lesne, C Zbinden-Trichet, A Picard, M Mitrofanoff, C Chapuis-Vandenbogaerde, N Degardin, B Grollemund

AFFF, Paris, France

Background: Pierre Robin Sequence (PRS) is the triad microretrognathia, glossoptosis and cleft palate, with presence of respiratory and feeding problems at birth. Definition and patient care varies largely between centers.

Aims: The aim is to compare the definition, patient care and follow-up of 14 francophone centers.

Methods: The survey consisted of 30 questions sent by E-mail. One center was the reference center (Paris, Necker Hospital) and the others were considered competence centers. The questions concerned PRS definition, assessment and treatment of airway obstruction, method and monitoring, patient care, follow-up and surgical considerations for children born with a diagnosis of PRS.

Results: Fourteen centers answered the survey. Forty-two (42%) of these centers based their definition of PRS on anatomy alone; another 42%, on anatomy and respiratory problem. 22q/11 and Stickler were the most frequent syndromes found. For severe respiratory problems at birth, all kinds of conservative and surgical treatment were performed, such as glossopexia, tongue muscular section, mandibular traction and tracheotomy. No distraction was realized in the first months. Children were operated at six months in 28% of the centers, at nine months in 43%. A polygraphy was organized in 57% before surgery. A palatoplasty following the Langenbeck technique was used in 43%, other techniques in the other centers. Grommets were inserted systematically in 21% of the centers, from case to case in 64%. Ninety-two (92)% organized ENT controls by microscope. Palatal fistula was never seen in 30% of the centers, every two to three years in the others. Orthognatic surgery at the end of growth was done in 50% of the centers. In 30% of the centers there was no psychological follow-up. Seventy-nine (79)% offered early speech therapy, and 50% performed a pharyngoplasty. In 50% long-term alimentary difficulties were noted in PRS children. Eighty-five (85)% of the centers conducted no survey on the quality of life.

Summary/Conclusion: The definition of PRS is not always the same in the francophone community. Patient care may largely differ from center to center. Respiratory problems at birth are always challenging and all kinds of surgical acts may be considered. For the follow-up, all children are seen by a cleft team, without psychological support in 30% of the centers, but with regular speech therapy and controls. Documented surveys on their quality of life would broaden our knowledge in the long term.
Predicting perioperative respiratory events and successful extubation following mandibular distraction osteogenesis (MDO) for micrognathia: a retrospective cohort study

J Swanson¹, R Zhang¹, L Lin¹, I Hoppe¹, J Taylor¹, S Bartlett¹
¹Children’s Hospital of Philadelphia, Philadelphia, United States

Background: The frequency of respiratory events in the perioperative period, and optimal duration of intubation during early mandibular distraction osteogenesis (MDO) activation, are poorly understood.

Aims: This study assesses potential risk factors associated with perioperative respiratory events, particularly the need for reintubation, following MDO surgery.

Methods: A retrospective review was conducted for infants (less than 1 year of age) undergoing MDO for tongue-based airway obstruction between November 2010 and December 2017. Univariate and multivariate analysis of sentinel events and outcomes was performed.

Results: Ninety infants (median age 35 days) were included in the study (50% syndromic, 52% male). 27 subjects (30%) experienced a respiratory event requiring intervention prior to discharge, including 14 subjects who failed initial extubation. 33% of subjects extubated earlier than post-operative day 5 (POD5) failed extubation, compared to 9% of subjects extubated on POD5 or later (p=0.005). Respiratory events occurred more frequently when extubation was attempted at distraction lengths of 5mm or less (42%, 16/38) compared to >5mm (21%, 11/52; p=0.032.) Logistic regression modeling showed that syndromic status (odds ratio, OR 14.8) and secondary airway anomaly (OR 6.1) were significant predictors for respiratory events, while greater length of distraction at time of extubation was protective (OR 0.8) (p<0.05).

Summary/Conclusion: Post-operative intubation of at least 5 days, with associated mean distraction of 5mm appears to be associated with successful extubation trial following MDO surgery. Patients with congenital syndromes and secondary airway anomalies are more likely to experience perioperative respiratory events, and this group of patients merits increased attention at extubation.
OS 8.1
3d printing in cleft and craniofacial care: development of a self-supported nasopharyngeal airway
David Zopf
UMC Utrecht, Utrecht, Netherlands Speaker

Abstract Content:
Dr. Zopf has used 3D Printing for several applications in Cleft and Craniofacial care. This talk will summarize the technology, highlight some previous applications, and focus on the design, development, and implementation of a novel self-supported nasopharyngeal device to mitigate upper airway obstruction, such as that seen in Robin Sequence and other craniofacial anomalies. Do you have a conflict of interest to declare?: No Name the other party to the relationship : -
OS 8.6

Scandcleft randomised trials: occlusion in 8-year olds according to the modified huddart and bodenham index
A Karsten¹, A Marcusson², S Rizell³, M Najar Chalien³, A Heliövaara⁴, A Kuseler⁵, P Skaare⁶, E Brinck⁶, W Shaw⁷, H Bellardie⁷, K Mooney⁷, K Mølsted⁸, P Sæle⁹, P Eyres⁷, G Semb¹⁰
¹Orthodontics, Dental Medicine, Karolinska Institutet, Huddinge, Stockholm, ²Dentofacial Orthopedics, University Hospital, Linkoping, ³Odontology, Sahlgrenska Academy, Gothenburg, Sweden, ⁴Cleft Palate and Craniofacial Center, Plastic Surgery, Helsinki University Hospital, Helsinki, Finland, ⁵Cleft Palate Center, University of Aarhus, Aarhus, Denmark, ⁶Department of Plastic and Reconstructive Surgery, Oslo University Hospital Rikshospitalet, Oslo, Norway, ⁷Dental School, University of Manchester, Manchester, United Kingdom, ⁸Copenhagen Cleft Palate Center, University of Copenhagen, Copenhagen, Denmark, ⁹Oral Health Center of Expertise, University of Bergen, Bergen, ¹⁰Department of Orthodontics, University of Oslo, Oslo, Norway

Background: The Scandcleft project studies prospectively the long-term outcome after four different surgical protocols for palatal closure in patients born with unilateral cleft lip and palate (UCLP).

Aims: To evaluate the dental occlusion at age eight years of age with the Modified Huddart and Bodenham (MHB) index.

Methods: In an international multicenter study by ten cleft teams in five countries: Denmark, Finland, Sweden, Norway and UK, three different surgical procedures for primary palatal repair (Arms B, C and D) were tested against a common procedure (Arm A) in a total cohort of 448 children born with non-syndromic UCLP. At eight years of age 429 children remained in the study. Dental casts of 411 patients (270 boys, 141 girls), at a mean age of 8.1 years (range 7.0-10.0) were blindly assessed by four orthodontists with the MHB index. The main outcome measures were the anterior and posterior dental occlusion.

Results: The inter- and intra-examiner reliability was good to excellent (0.75-0.90; 0.73-0.97) respectively. The mean total scores varied from -7.09 (Trial 2C) to -10.13 (Trial 3D). The mean anterior scores varied from -1.75 (Trial 2C) to -3.18 (Trial IA). The mean posterior cleft-side scores varied from -4.32 (Trial IB) to -5.21 (Trial 3D) and the mean non-cleft-side scores varied from -0.88 (Trial 2C) to -2.40 (Trial 3A). No significant differences were found within the trials. Between the trials there was a significant difference between Trial 2 and 3 (Arm C/D) for the anterior score (p=0.025), between Trial 1 and 3 (Arm B/D) for the posterior cleft score (p=0.033) and between Trial 2 and 3 (Arm C/D) for the total score (p=0.002). Only the last remained significant after Bonferroni correction (p=0.004).

Summary/Conclusion: There was no evidence of a clinically significant difference in occlusion between the two surgical methods in each trial. All mean scores showed more negative values at eight years compared to previously reported values at five years.
OS 8.7
Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: maxillary growth at eight years of age.
A Küseler

Aims: The aim of this particular study was to assess differences in maxillary growth at the age of eight years according to the different surgical protocols for primary cleft surgery, as defined in the Scandcleft project.

Methods: A common surgical method (a) with soft palate closure at 3-4 months of age and hard palate closure at 12 month of age was tested in Trial 1 against similar surgery but with hard palate repair at 36 months (delayed hard palate closure) (1b). In Trial 2 the common method (2a) was tested against simultaneous closure of both hard and soft palate at 1 year (2c) and in Trial 3 the common method (3a) was tested against closure of hard palate together with lip closure at 3 months of age and soft palate closure at 1 year of age (3d). Participants were randomly allocated to one of two arms within their Trial. Total number of participating patients at 8 years of age was 428. Lateral cephalograms (n=408) were analyzed. The cephalometric angles SNA and ANB were chosen for assessing maxillary growth for this part of the presentation.

Results: Within each Trial (Trial 1a/1b, Trial 2a/2b and Trial 3a/3b) there was no difference in cephalometric values between the common and the local arm. There was no statistical significant differences in the SNA and ANB angles between the common arm in Trial 1a (mean SNA 77.8, mean ANB 2.6) and Trial 2a (mean SNA 79.8, mean ANB 3.6) and no difference between Trial 1a and Trial 3a but at statistical difference could be seen between Trial 2a and Trial 3a (mean SNA 76.9, mean ANB 1.7), however, the confidence interval was rather large. Intra- and interrater reliability were within acceptable range.

Summary/Conclusion: The timing and the surgical method is not of major importance concerning growth outcomes (SNA and ANB) in UCLP.
OS 8.8
Scandcleft randomised trials of primary surgery for unilateral cleft lip and palate: naso-labial appearance at 8 years. K Mølsted1, A Heliövaara2, E Brinck3, M Chalien4, L Riis1, A Küsele5, A Karsten6, A Marcusson7, S Rizell8, P Sæle9, H Bellardie10, J Mooney11, P Eyres12, W Shaw12, G Semb3

1Copenhagen Cleft Palate Center, University Hospital of Copenhagen, Hellerup, Denmark, 2Department of Plastic Surgery, Cleft Palate and Craniofacial Center, Helsinki, Finland, 3Department of Plastic and Reconstructive Surgery, Oslo University Hospital, Oslo, Norway, 4Orthodontic clinic, University Clinics of Odontology, Gothenburg, Sweden, 5Cleft Palate Center, Aarhus, Denmark, 6Division of Orthodontics, Department of Dental Medicine, Stockholm Craniofacial Team, Karolinska Institute, Stockholm, 7Dentofacial Orthopedics, Maxillofacial Unit, University Hospital, Linköping, 8Orthodontic clinic, University Clinics of Odontology Gothenburg, Gothenburg, Sweden, 9Oral Health Center of Excellence, Bergen, Norway, 10Greater Manchester Cleft Unit, Royal Manchester Children's Hospital, Manchester, United Kingdom, 11Orthodontic Department, The University of the Western Cape, Cape Town, South Africa, 12Dental School, University of Manchester, Manchester, United Kingdom

Background: Nine cleft centres in Denmark, Finland, Norway, Sweden, and the UK participated in this part of the project evaluating the long-term outcome after primary surgery of the lip and nose. The local protocol for closure of the lip continued to be used, because primary lip and nose operations were not part of the randomisation in the Scandcleft project.

Aims: To assess naso-labial appearance at 8 years of age.

Methods: Standardized photos taken according to a specific protocol developed for the Scandcleft project was taken. Only the naso-labial area was shown, the surrounding facial features were masked. A modification of the original method described by Asher-McDade et al was used. This rating method uses a 5-point ordinal scale, the method has been improved by adding reference photos used in the Americleft studies (Americleft Yardstick). 375 patients participated in this part of the project. The photos were blindly assessed by 5 orthodontists from the Scandcleft group.

Results: A majority of the centres used the Millard technique for closure of the lip and the McComb procedure for nose correction. There were statistically significant differences between the centres concerning upper lip form, nasal form and deviation and cleft side profile. The intra-and inter-rater reliability scores were acceptable.

Summary/Conclusion: The results showed statistically significant differences in naso-labial appearance between the centres in the Scandcleft group.
**OS 8.9**

**Scandcleft randomised trials: dental anomalies at 8 years**

S Rizell¹, H Bellardie², A Karsten³, J Mooney², P Sæle⁴, E Brinck⁵, A Heliövaara⁶, A Küseler⁷, K Mølsted⁸, A Marcusson⁹, M Najar Chalien¹⁰, P Skaare⁵, P Eyres², W Shaw², G Semb¹¹

¹Department of Orthodontics, University of Gothenburg, Gothenburg, Sweden, ²Dental School, University of Manchester, Manchester, United Kingdom, ³Division of Orthodontics, Division of Dental Medicine, Karolinska Institutet, Huddinge, Sweden, ⁴Oral Health Center of Expertise, University of Bergen, Bergen, ⁵Department of Plastic and Reconstructive Surgery, Oslo University Hospital Rikshospitalet, Oslo, Norway, ⁶Cleft Palate and Craniofacial Center, Plastic Surgery, Helsinki University Hospital, Helsinki, Finland, ⁷Cleft Palate Center, University of Aarhus, Aarhus, ⁸Copenhagen Cleft Palate Center, University of Copenhagen, Copenhagen, Denmark, ⁹Dentofacial Orthopedics, University Hospital Linköping, Linköping, ¹⁰1 Department of Orthodontics, University of Gothenburg, Gothenburg, Sweden, ¹¹Department of Orthodontics, University of Oslo, Oslo, Norway

**Background:** The Scandcleft trials is a multi-center prospective project for long-term outcome after four different surgical protocols. This material additionally brings about opportunity to study secondary outcomes in a well-controlled material.

**Aims:** To study dental anomalies for the total group of individuals in the Scandcleft trials.

**Methods:** Panoramic radiographs (n=426) were obtained at 8 years of age, together with occlusal films prior to secondary bonegrafting from 147 girls and 279 boys, born with unilateral cleft lip and palate. Assessment of dental anomalies as agenesis of permanent teeth supernumeraries position and anatomy of cleft lateral as well as ectopic eruption was performed independently by four different examiners.

**Results:** Dental agenesis was found in 52.7% and supernumerary teeth were found in 16.9% of the individuals. The cleft lateral was found to be peg shaped in 44.3% and ectopic eruption was found in 14.9% of the participants.

**Summary/Conclusion:** Individuals born with unilateral cleft lip and palate exhibit multiple dental anomalies at 8 years of age.
Scandcleft randomised trials: parents’ experience of support and reactions to their 5-year-old child’s cleft diagnosis

K Feragen¹, E Willadsen², C Havstam³, J Bogh Nielsen⁴, N Pedersen⁵, A Heliövaara⁶, P Eyres⁷, A Marcusson⁸, G Semb⁹
¹Centre for Rare Disorders, Oslo University Hospital, Oslo, Norway, ²Division of Speech and Language Pathology, Karolinska Institutet and Karolinska University Hospital, Stockholm, ³Division of Speech and Language Pathology, Sahlgrenska University Hospital, Gothenburg, Sweden, ⁴Cleft Palate Center, Cleft Palate Center, Aarhus, Denmark, ⁵Statped Vest, Bergen, Norway, ⁶Department of Plastic Surgery, Cleft Palate and Craniofacial Center, Helsinki, Finland, ⁷Division of Dentistry, University of Manchester, Manchester, United Kingdom, ⁸Dentofacial Orthopedics, University Hospital Linköping, Linköping, Sweden, ⁹Department of Orthodontics, University of Oslo, Oslo, Norway

Abstract Content:
Aims and objectives: Parental experiences and perceptions are important when evaluating the complexity of satisfactory treatment outcomes. Having a child with a cleft lip and palate may be emotionally distressing for parents and affect their future relationship with the child. The objective of this study was to examine parents’ social and emotional experiences related to their child’s cleft diagnosis.

Materials and methods: Three parallel group randomised clinical trials were undertaken as an international multicenter study by 10 cleft teams in five countries: Denmark, Finland, Sweden, Norway and UK. A total of 356 parents completed the Scandcleft Parent Questionnaire.

Results: The majority of parents experienced practical and emotional support from family, friends, and health professionals. Nevertheless, parents had to cope with other people’s reactions to the cleft, experiences that were described as ranging from hurtful to neutral and/or positive. Parents were mostly satisfied with their child’s treatment. Nonetheless, more than half of the parents reported specific worries related to their child’s future.

Discussion and summary: Parents have to cope with a number of challenges related to their own and the child’s emotional and psychological responses to the diagnosis, in addition to other people’s reactions and comments. Even if most parents experienced social, emotional and practical support from family, friends, and health professionals, some parents also reported emotionally challenging and hurtful social experiences. Vulnerable subgroups of parents should be identified by the treatment teams and offered appropriate support and help.
Scandcleft randomised trials: what next?
W Shaw¹, G Semb²
¹Division of Dentistry, University of Manchester, Manchester, United Kingdom, ²Division of Plastic Surgery, Oslo University Hospital, Oslo, Norway.

Background: Longstanding uncertainty surrounds the selection of surgical protocols for unilateral cleft lip and palate (UCLP), and randomised trials have only occasionally been performed. Aims: The Scandcleft Project consists of three trials initiated in 1997. The results at age 5 have been published in the series of reports that precede this manuscript, which aims to distil some of the issues that future trialists may need to address. Methods: The shared experience of this group of trials is reviewed with reference to the current literature on trial management and more specifically, trials of surgical technique. Results: The main discussion points relate to the challenges associated with research bureaucracy, learning curve, and individuality of skill, and ethics. Summary/Conclusion: Compliance with current regulatory requirements for clinical trials and recruitment rates for cleft sub-types represent substantial challenges. Mastery of new surgical techniques prior to trials raises important ethical issues, though recent reports in the wider surgical literature suggest that learning may be hastened with the assistance of anonymised peer review ratings of intraoperative performance. The three Scandcleft Trials succeeded in meeting the planned recruitment targets, and completed follow-up with remarkably high retention rates. The design of the study required the majority of participating surgeons to master a new technique and though overall outcomes were comparable with those in the literature, the associated learning curve impacted on the results of one of the trials.

Though ethical issues accompanying trials of timing and sequence are less dependent on operator experience, it is evident that trials requiring standardisation of new operative techniques and the associated learning curve require special consideration. In the first place, it is already clear that many established techniques can achieve good outcomes if performed well. So if new complex techniques constitute a major learning curve challenge and are difficult to master, they are unlikely to be of much value in a wider sphere. Indeed, just about every technique that could be tried, has been tried, and there are several protocols to choose from that yield good outcomes in the right hands.

If future trials were to involve new technical challenges, planning should include a peer review feedback process that would assist rapid mastery of new techniques and determine the point when an appropriate level of skill has been reached. The study of Birkmeyer et al. might be a suitable model, and it is certainly an approach that could become a routine feature of training and appraisal. Further work on the essence of surgical skill in cleft care and the transfer of skills could be a fruitful endeavour.

Rather than testing new variations of surgical protocols however, it is more likely that we can anticipate some breakthroughs in tissue engineering such as the long awaited scar free wound healing, novel tissue cultures for grafting, and so forth. These new developments may well need RCTs to validate their worth. And well structured trials are long overdue for other interventions associated with primary surgical outcomes, such as cost-effectiveness of naso-alveolar moulding, and presurgical orthopaedics.
3d printing in cleft and craniofacial care: development of a self-supported nasopharyngeal airway
David Zopf
UMC Utrecht, Utrecht, Netherlands Speaker

Abstract Content: Dr. Zopf has used 3D Printing for several applications in Cleft and Craniofacial care. This talk will summarize the technology, highlight some previous applications, and focus on the design, development, and implementation of a novel self-supported nasopharyngeal device to mitigate upper airway obstruction, such as that seen in Robin Sequence and other craniofacial anomalies.
Developing algorithms of care in robin management
H Mcclements¹, V beale¹, D phare¹, H robson¹, S van eeden¹
¹cleft unit, Manchester University NHS Foundation Trust, Manchester, United Kingdom

Background: In the past in our Network the majority of babies born with Robin sequence were transferred to the centralised specialist centre. An audit of parent’s views showed however that parents wanted to be cared for locally rather than a distance from their home and family. The need for guidance pathways to manage these patients safely in local units was therefore identified. With clinical nurse specialists in our network teams providing local support this was felt to be achievable. This presentation looks at the production and use of algorithms of care to guide both clinical nurse specialists and maternity staff in the safe care of the airway in these babies locally.

Aims: To develop algorithms for the safe local management of the airway in Robin patients.

Methods: A joint consultation of cleft surgeons, clinical nurse specialists, anaesthetists and otolaryngologists devised two algorithms of for the management of the airway in babies born with an isolated cleft palate and for Robin sequence babies needing an naso-pharyngeal airway(NPA).

Results: The majority of babies born with a cleft palate within our cleft network are cared for in local maternity units. Diagnosis of Robin sequence usually precipitated referral to the central surgical unit for further management. Institution of the algorithm resulted in local management and a shorter hospital stay for the majority of Robin patients.

Summary/Conclusion: With the new algorithms of care babies can be safely assessed by a clinical nurse specialist and a plan of care put in place for the local maternity unit, which might include the use of a nasopharyngeal airway. Frequent assessment by the clinical nurse specialist locally means that amendments to the plan of care as the baby’s condition changes can be made as set out in the algorithm of care, and if more specialist care is required the baby can be transferred to a specialist centre.
OS 9.3

Developing algorithms of care in robin management
H McClements¹, V Beale², D Phare², H Robson², S van Eeden¹
¹Cleft, Alder Hey Childrens Hospital, Liverpool, ²Cleft, Royal Manchester Childrens Hospital, Manchester, United Kingdom

Background: In the past in our Network the majority of babies born with Robin sequence were transferred to the centralised specialist centre. An audit of parent’s views showed however that parents wanted to be cared for locally rather than a distance from their home and family. The need for guidance pathways to manage these patients safely in local units was therefore identified. With clinical nurse specialists in our network teams providing local support this was felt to be achievable. This presentation looks at the production and use of algorithms of care to guide both clinical nurse specialists and maternity staff in the safe care of the airway in these babies locally.

Aims: To develop algorithms for the safe local management of the airway in Robin patients.

Methods: A joint consultation of cleft surgeons, clinical nurse specialists, anaesthetists and otolaryngologists devised two algorithms of for the management of the airway in babies born with an isolated cleft palate and for Robin sequence babies needing an naso-pharyngeal airway(NPA).

Results: The majority of babies born with a cleft palate within our cleft network are cared for in local maternity units. Diagnosis of Robin sequence usually precipitated referral to the central surgical unit for further management. Institution of the algorithm resulted in local management and a shorter hospital stay for the majority of Robin patients.

Summary/Conclusion: With the new algorithms of care babies can be safely assessed by a clinical nurse specialist and a plan of care put in place for the local maternity unit, which might include the use of a nasopharyngeal airway. Frequent assessment by the clinical nurse specialist locally means that amendments to the plan of care as the baby’s condition changes can be made as set out in the algorithm of care, and if more specialist care is required the baby can be transferred to a specialist centre.
**Prediction of recurrent osa after neonatal mandibular distraction**

A Kaye¹, K Shedd¹, S Jiang¹

¹Plastic Surgery, Children’s Mercy Kansas City, Kansas City, Missouri, United States

**Background:** Patients presenting with Pierre Robin Sequence and significant upper airway obstruction are at risk for short and long term medical complications. Clinical evidence of significant airway obstruction warrant assessment and intervention. Intervention options are numerous and more than one may be required for a given patient. The use of mandibular distraction for severe upper airway obstruction has the primary goal of lengthening the mandible and thereby relieving the tongue base obstruction associated with Pierre Robin. The secondary benefits may include avoiding tracheotomy, improved oral feeding, and decreased reflux. After successful distraction patients can be noted to have decreased work of breathing, fewer desaturations, fewer apneas, and improved feeding ability. Despite these clear clinical indications of airway improvements after distraction, however, patients may still have significant residual or recurrent OSA that may manifest as growth failure over time.

**Aims:** The aim of this study was as a pilot investigation of nutritional/feeding predictors of recurrent obstructive sleep apnea in patients with Pierre Robin sequence.

**Methods:** This study was performed as a retrospective look at patients with Pierre Robin born over a 3 year period and treated with distraction as infants. Timing of management, evaluations, feeding and growth curve data was assessed.

**Results:** Twenty-five patients were identified with Pierre Robin Sequence treated with distraction for airway obstruction. Twelve of the 25 had known or suspected syndromes. Six patients were diagnosed with recurrent/residual OSA, of which 5 underwent repeat distraction. Four of the 6 demonstrated faltered growth curves in advance of clinical recognition of recurrent OSA, suggesting a potential opportunity to improve efficiency of diagnosis and management of problems in this patient group.

**Summary/Conclusion:** Clinicians should have ongoing awareness of growth patterns even after successful initial therapy. It should be recognized that plateaus in the growth curve may indicate residual or recurrent symptomatic obstructive sleep apnea. Growth failure is a frequently overlooked symptom of sleep apnea irrespective of Pierre Robin and providers should not simply assume growth delays are purely intrinsic to an underlying syndrome.
Postnatal and perioperative management of upper airway obstruction with pre-epiglottic baton plate in children with pierre robin syndrome

G Schmidt¹, J Ervens², C Matuschek³, T Bartzela⁴
¹Oral and Maxillofacial Surgery, Germany, ²Oral and Maxillofacial Surgery, Charité Universitätsmedizin Berlin, corporate member of Freie Universität Berlin, Humboldt Universität zu Berlin und Berlin Institute of Health, ³Institute for surgical Prothetik and Epithetik, Klinikum Westend, ⁴Orthodontics, Dentofacial Orthopedics and Pedodontics, Charité - Universitätsmedizin Berlin, CC3 Center for Dental and Craniofacial Sciences, Berlin, Germany

Background: Pierre Robin Sequence (PRS) is associated with retrognathy, glossoptosis and upper airway obstruction (UAO) which can be life threatening. Treatment alternatives are ranging from non-invasive methods like positioning to invasive actions like lip-tongue adhesion. Even after the neonatal period, these patients remain for anesthesiologists as the group with great intubation risk and postoperative respiratory distress. Nevertheless, until now, there is no consensus for the treatment management of these children.

Aims: The aim of this study was to report our experience on the treatment management of UAO with pre-epiglottic baton plate (PEBP) in children with PRS.

Methods: Data were extracted from 128 patients with isolated PRS with cleft palate that were treated with PEBP between 2006-2018. Informed consent was obtained. The principles of Declaration of Helsinki with regard to research in human subjects were followed. Clinical data included postnatal frequency and grade of desaturation, signs of respiratory distress, mode and effect of positioning, duration of hospital stay, mode and duration of food intake and weight gain. Perioperatively, intubation difficulties and postoperative respiratory complications were classified using the Cormack-Lehane scoring system and furthermore were compared to 128 patients with isolated cleft palate.

Treatment protocol from the year 2006 to 2014: PEBP was in situ for 3 months, then intravelar veloplasty and 6 months later, hard palate repair were performed.

Treatment protocol since 2015: One-stage surgical procedure for velar repair and hard palate closure and the wearing time of the PEBP was extending from 3 to 6 months.

Results: Postnatal results:
98 patients presented clinical signs of increased breathing work and oxygen desaturation immediately after birth. 23 patients required high flow, and four patients needed intubation after birth. After insertion of PEBP, neither oxygen desaturation nor retractions were observed and the patients could be released from the hospital after 2 – 4 weeks. Four intubated patients were successfully weaned from the respirator. Three patients needed a gastric tube for feeding.

In 31 patients, positioning restored sufficiently breathing so that they could be discharged without plate. However, nine patients developed breathing problems, in two patients aspiration occurred, 15 had feeding problems and consecutively failure to thrive, and nine had to be rehospitalized in the course. Finally, only in 16 patients the positioning alone proved to be a sufficient long-term measure.

Perioperative results:
During intubation for cleft palate closure 37,5 % of the patients had III – IV according to Cormack-Lehane classification [1]. Since the protocol adaptation in 2015, the patients with III – IV Cormack-Lehane classification has been declined to only 17,9 %, indicating a statistical significant difference (p ≤ 0.05) in the degree of intubational difficulty. All other parameters, concerning postoperative respiratory distress didn’t differ significantly between the two treatment protocols.

Summary/Conclusion: Treatment with PEBP in patients with PRS is a non-invasive approach, which not only eliminates the postnatal UAO but also minimizes the perioperative complications of these patients.

Reference
Perceptual, acoustical and tomographical responses to experimentally induced hypernasality and nasal air emission by temporary velum paralysis: a pilot study

N Loomans¹, Y Maryn², A Zarowski²
¹cleft team GZA hospitals, department of cranio-maxillofacial surgery, European Institute for ORL-HNS, ²European institute of ORL-HNS, GZA hospitals Antwerp, Antwerpen, Belgium

Background: Velopharyngeal inadequacy (VPI) can result in one or more of the following speech sound disruptions: hypernasality, nasal air emission, weak or omitted consonants, erroneous articulatory compensations and even dysphonia. VPI is a major problem in cleft lip and palate patients, and the ability of good speech is the most difficult topic in the multidisciplinary cleft treatment. Even after palatal surgery in cleft patients VPI remains a concern which resulting in a continuous search for the optimal surgical protocol.

Aims: To better understand the physiology of excessively nasal sound, the present pilot study investigated the correlation between (a) conebeam computed tomography (CBCT)-based velopharyngeal orifice volume and areameasurements and (b) auditory-perceptual ratings of hypernasality, articulation disorders and nasal emission. To control for craniofacial morphology and to simulate VPI, neuromuscular junctions of the levator palatine muscle and the palatopharygeus muscle were temporarily paralyzed.

Methods: This study is based on 5 female participants. They all received 4 intramuscular injections with articaïne hydrochloride 40 mg/ml and epinephrine 0,01 mg/ml with a total volume of 4 ml. Audio recordings of sustained [i] and the oronasal text as well as CBCT during sustained [i] were made before and after this anaesthesia, with normally functioning velum as control condition and with temporarily paralyzed velum as experimental condition. CBCT measurements were performed in an axial plane tangential to the top of dens axis and the cervical border of thv central incisor. Velopharyngeal orifice volume and area were calculated.

Results: Temporary velar paralysis clearly raised hypernasality and nasal emission ratings in three of the five participants. Spearman correlation coefficients (rs) pointed towards positive association between velopharyngeal orifice volume and area and these ratings.

Summary/Conclusion: To better comprehend VPI-based excessive nasal airstream during speech while controlling for craniofacial morphology, this preliminary study intended to investigate the feasibility of temporary paralysis of the velum. As shown in both CBCT as in the perceptual ratings, temporary velar paralysis was clearly accomplished in three of the five participants.
Meet the Master

MTM 1.1

Meet the master presentation: buccal flaps in palate repair, the development of the buccal flap philosophy

R. Mann

1Pediatric Plastic Surgery, Helen DeVos Childrens Hospital, Grand Rapids, Michigan, United States

Abstract Content: This presentation will illustrate how a very useful reconstructive tool, the buccinator myomucosal flap, became the key to a new philosophic approach that encompasses all phases of care for the patient born with a cleft lip and or palate. The new philosophy is named the Buccal Flap Approach, reflecting the importance of addressing the embryologic deficiencies inherent to the cleft malformations, and encompassing modern reconstructive principles. The Buccal Flap Philosophy has a goal of achieving a complete reconstruction of the cleft while recognizing inherent complexities of tissue changes that our patients exhibit during normal growth. This presentation will be the introduction to the Buccal Flap Approach and will establish some of the thought processes that went into the development of the numerous techniques based on the philosophy. This introduction will be helpful for those delegates that wish to attend the presentation on the Practical Applications of the Buccal Flap Philosophy, which will be a video based teaching session with the goal to help participants get comfortable using the Buccal Flap Approach for primary cleft palate repair and VPD as well if time allows. The Friday session on Speech Enhancing Surgery will further illustrate more of the practical uses of the Buccal Flap approach to treat VPD
MTM 1.2
Cleft orthodontic treatment of maxillary hypoplasia: when is early sagittal correction indicated and when is late maxillary advancement with the alt-ramec protocol indicated
M Meazzini

Introduction: A thorough literature review on long term results of maxillary protraction in CLP patients are the basis for the current limited indications to early treatment. On the other hand, long term results of late protraction will be shown.

Objective: The objective of this retrospective longitudinal study was to evaluate short and long-term results of the application of the Liou Alt-RAMEC (Alternate Rapid Maxillary Expansion and Constriction) technique, a late orthopedic maxillary protraction technique, with intraoral anchorage, in cleft patients. Materials and Methods: 37 Unilateral Cleft Lip and Palate (UCLP) patients were consecutively treated with the Alt-Ramec technique. The average age of the patients was 12.3 years (10.7-14.2 years) before protraction and 18.3 years (17.4-24.1 years) at long term follow-up. A sample of non-treated UCLP patients was used as a control group. It was matched for sex, skeletal class III and age (12.1 years). The control sample had records at the end of growth (18.9 years).

Results: The sagittal advancement of A-point, after the application of the technique, was 5.8±2.7 mm. Some mandibular dentoalveolar and positional adaptation was noted. The position of the maxilla was stable in the long term. On the other hand, the UCLP control group showed hardly any growth at the maxillary level during the long term follow up period.

Conclusion: Our results showed that the Alt-RAMEC technique, performed at the correct time, with a double hinged expander, followed by class III spring or elastic traction, 24hours/day, allows for satisfactory maxillary protraction, with stable long term results.
Future Perspectives

OS 10.4
Assessment of canonical babbling status in infants with cp: agreement between parents and speech therapists results of 485 infants in the rct, timing of primary surgery for cleft palate (tops)
E Willadsen¹, R Cooper², B Conroy³, C Persson⁴, C Gamble²
¹Nordic Studies and Linguistics, University of Copenhagen, Copenhagen, Denmark, ²Clinical Trials Research Centre, ³Clinical Trials Research Centre, University of Liverpool, Liverpool, United Kingdom, ⁴Institute of Neuroscience and Physiology, Speech and Language Pathology Unit, University of Gothenburg and Sahlgrenska University Hospital, Gothenburg, Sweden

Background: Canonical babbling is an important prelinguistic measure. If not achieved by 10 months of age, further investigation is needed. Previous studies have suggested that parents of typically developing children can reliably report canonical babbling status, but this has not been studied in children with CP. Also, little attention has been given to speech and language therapists’ (SLTs) ability to make this judgment in real time. The latter is important for clinical practice. If parents and SLTs agree, parent classification may prove useful within the clinical setting. To explore the agreement between parents and SLTs, 12 months old infants had canonical babbling classified as present or absent by the following three assessor types: 1) three TOPS SLTs (research setting), 2) a Site Therapist (clinical visit), 3) Parent/carer.

Aims: First, we examined how many children were assessed as ‘canonical’ by the three TOPS SLTs as well as their inter assessor agreement. Secondly, we compared these results to the classification of canonical obtained from the clinical visit (Site Therapist) and from the parent/carer, as well as the inter assessment agreement across the three assessment types.

Methods: Fourty-five minutes long video recordings of play interaction between infant and parent/carer were obtained from the 12 months site visit. Recordings were assessed independently by three TOPS SLTs blinded to the infants’ randomisation at a TOPS assessment meeting. Recordings were split into two halves (22 min), SLTs listened to the entire recording without pausing, no notes were taken. Afterwards, the SLTs classified canonical babbling as present or absent. Present was ascribed if at least one part of the recording was classified as such.

At the 12 month clinical visit, the Site Therapist recorded her/his classification of canonical status on the TOPS case report form (CRF) after the clinical visit including video recording, based on her/his overall perception of the infant’s vocalisations. Afterwards, she/he asked the parents: “what sounds is your baby able to produce regularly?” The parents’ classification of canonical babbling was recorded on the CRF.

Results: The three TOPS SLTs classified 86.2% of the infants as canonical and 13.8% as non-canonical, and they agreed in their classification in 85.2% of infants. The Site Therapists classified 79.6% of the infants as ‘canonical’, 15.0% as non-canonical and were unsure of 5.4%. The parents classified 91.1% of infants as ‘canonical’ and 8.9% as non-canonical. The TOPS SLTs and Site Therapists made the same classification with 77.2% being ‘canonical’ and 9.3% ‘not canonical’. The TOPS SLTs and Parent/carer made the same classification with 82.3% being ‘canonical’ and 4.3% ‘not canonical’. The Site Therapists and Parent/carer classified the children the same with 78.0% being ‘canonical’ and 6.5% ‘not canonical’. Across all assessor types, the infants were classified the same with 75.8% being ‘canonical’ and 4.1% ‘not canonical’.

Summary/Conclusion: All assessor types showed similar classifications. Parents reported a lower frequency of non-canonical than both groups of SLT assessors.

*The study is co-authored by the TOPS SLT group.
**Substantial reductions in secondary cleft surgery following centralisation of cleft services in the united kingdom**

T Sitzman¹, M Temkit², M Britto³, D Sell⁴, A Wills⁵, J Sandy⁶, A Ness⁷

¹Plastic Surgery, ²Clinical Research, Phoenix Children’s Hospital, Phoenix, ³James M. Anderson Center for Health Systems Excellence, Cincinnati Children’s Hospital Medical Center, Cincinnati, United States, ⁴North Thames Regional Cleft Service, Speech and Language Therapy Department and Centre for Outcomes and Experience Research in Children’s Health, Illness and Disability (ORCHID), Great Ormond Street Hospital NHS Foundation Trust, London, ⁵National Institute for Health Research (NIHR) Biomedical Research Centre, University Hospitals Bristol NHS Foundation Trust and the University of Bristol, ⁶Bristol Dental School, University of Bristol, ⁷National Institute for Health Research (NIHR) Biomedical Research Centre, University Hospitals Bristol NHS Foundation Trust and University of Bristol, Bristol, United Kingdom

**Background:** Centralisation of cleft services in the United Kingdom (UK) was a multi-faceted process that included consolidation of care at high-volume multidisciplinary centers, implementation of formal practice guidelines, establishing minimum training standards and competencies for providers, and creation of a system for continuous monitoring of treatment outcomes. These changes in cleft care provision have been associated with improvements in speech and aesthetic outcomes, but little is known about the effects of centralisation on the use of secondary cleft surgery.

**Aims:** 1) To compare the cumulative incidence of secondary cleft surgeries before and after centralisation of cleft services in the UK
2) To compare the proportion of children achieving good clinical outcomes without secondary surgery before and after centralisation of cleft services in the UK

**Methods:** Two cross-sectional studies of 5-year-old children with non-syndromic unilateral cleft lip and palate were completed: the first study included children treated in the UK prior to centralisation (born 1989-1991) and the second study included children treated in the UK after centralisation (born 2005-2007). Investigators collected information on subject’s receipt of secondary lip, palate, and/or nasal surgery(s). Investigators also evaluated subject’s facial appearance (using frontal photographs and a validated 5-point rating scale) and speech hypernasality (using audio-video recordings and the CAPS-A Audit tool). The cumulative incidence of secondary surgery from birth until evaluation at age five was compared pre- and post-centralisation with Fisher’s exact test. Risk ratios (RR) were estimated using log-binomial multivariable regression models that adjusted for gender and age at evaluation. Parental consent was obtained for all participants.

**Results:** Information on receipt of secondary surgery and clinical outcomes was available for 192 children treated prior to centralisation and 262 treated after centralisation. The incidence of secondary lip surgery was 28.9% pre-centralisation and 5.3% post-centralisation (RR 0.19; 95% CI, 0.11-0.32; p<0.0001). The incidence of secondary palate surgery, which included fistula repair, palate re-repair, and pharyngoplasty, was 53.9% pre-centralisation and 29.0% post-centralisation (RR 0.54; 95% CI, 0.43-0.68; p<0.0001). The incidence of secondary nasal surgery was 15.1% pre-centralisation and 1.9% post-centralisation (RR 0.13, 95% CI, 0.05-0.32; p<0.0001). After adjusting for child’s gender and age at evaluation, the relative risk of receiving any secondary surgery post-centralisation was 0.65 (95% CI, 0.50-0.85; p=0.001), compared to the risk pre-centralisation. The risk of achieving a good clinical outcome, defined as both a nasolabial appearance rated as good or excellent on frontal photo and no consistent hypernasality on speech evaluation, without the use of any secondary surgery, was 3.29-fold higher post-centralisation (95% CI, 1.78-6.07; p<0.001).

**Summary/Conclusion:** Centralisation of cleft services in the UK was associated with a reduction in the use of secondary surgery. Centralisation was also associated with an increase in the proportion of children achieving good clinical outcomes without secondary surgery, suggesting that changes in care delivery implemented during centralisation improved outcomes of primary lip and palate repairs. Overall, centralisation appears to have both reduced the use of secondary surgery and improved clinical outcomes for children with cleft lip and palate.
**OS 10.6**

**Educational impact and development of a novel cleft palate surgical simulator: results of initial testing demonstrate acceptability and improvement in surgical trainees’ knowledge and confidence**

Rebecca Nicholas*1, Rute Fiadeiro2, Alexandra Burke-Smith1, Duncan Atherton1, Norma Timoney1, Kezia Echlin1,3.

1 Evelina London Children’s Hospital, London. 2 Brunel University, 3 Birmingham Children’s Hospital, Birmingham.

**Background:** Gaining hands-on experience in cleft surgery can be difficult due to limited access within the infant oral cavity and the delicate tissues of the velum. Even minor errors may lead to complications with serious consequences for the patient. Within the UK, a reduction in the number of hours spent in training and the centralisation of care has further limited access to cleft surgery with many trainees struggling to fulfil the curriculum requirements, while globally an estimated 250,000 infants are born with cleft lip and/or palate in low resource countries each year and it is estimated that a significant proportion are not repaired. This is, in part, due to a lack of local cleft surgeons. Training in cleft surgery is time and resource-heavy and so tools which can accelerate the learning curve are of significant interest. Simulation allows surgeons-in-training to gain experience in a low-risk, low-stress environment. Evidence suggests that skills acquired through simulation translate into the operating theatre, resulting in better in-vivo performance. Simulators for cleft palate surgery do exist but are either over-simplified or prohibitively expensive.

**Aims:** To develop a high-fidelity yet cost-effective simulator for cleft palate repair. Initial testing to determine the educational value for surgeons-in-training.

**Methods:** The simulator was developed jointly by a surgeon and product design student. Accurate skeletal elements were obtained through high-resolution scanning of a pathologic specimen. This was initially 3D printed and subsequently moulded in plastic. Modelling and surface-scanning were used to design the soft tissue components, which are formed from layers of silicone or bio-plastic through sequential moulding. The reusable parts can be manufactured locally, further reducing costs. The simulator is adjustable, allowing increasing levels of difficulty. 26 UK specialty trainees attended a 1 hour workshop and performed a vomerine mucosal flap and intra-velar veloplasty under instruction. Pre and post-simulation questionnaires were administered in order to assess knowledge of cleft palate repair and surgical confidence.

**Results:** The simulator had high acceptability among postgraduate trainees: 89% agreed or strongly agreed that the workshop was a valuable learning experience and 92% agreed that it had helped them understand and learn the procedure. Trainees had little baseline experience of cleft palate surgery with 32% having never observed a cleft palate repair. Only 2 participants had performed any part of a palatoplasty under supervision. Following the workshop, there was an increase in knowledge of cleft palate repair by 21% and increase in surgical confidence by 18%.

**Summary/Conclusion:** We have demonstrated not only the feasibility and acceptability of a new and cost-effective cleft palate simulator, but have also shown that it can be used as a powerful educational tool that is effective in improving cleft palate surgical knowledge and confidence across all grades surgical trainee. The simulator is currently undergoing further validation and has a patent pending.
Simulation-based skills and knowledge acquisition by plastic surgery residents: a prospective randomized blinded trial

R Kantar1, A Alfonso1, E Ramly1, M Gonchar1, S Maliha1, O Cohen1, J Diaz-Siso1, B Eisemann1, P Saadeh1, R Flores1

1Hansjörg Wyss Department of Plastic Surgery, New York University Langone Health, New York, United States

Background: Simulation is a standard component of residency training in many surgical subspecialties, yet its impact on knowledge and skills acquisition in plastic surgery training remains poorly defined.

Aims: Evaluate the potential benefits of simulation-based learning on residency training through a prospective, randomized, blinded trial.

Methods: Thirteen plastic surgery residents were randomized to a digital simulator or textbook demonstrating unilateral cleft lip (UCL) repair. Performance on the following tasks before (pre-intervention) and after (post-intervention) studying were evaluated: knowledge of surgical steps, lip markings on a three-dimensional (3D) stone model, and lip repair using a hands-on/high-fidelity 3D haptic model. Participant procedural confidence and satisfaction with each educational tool were also evaluated. Two expert reviewers blindly graded markings and surgical performance. Intra-class correlation coefficients (ICC) were calculated. Wilcoxon signed-rank and Mann-Whitney U tests were used.

Results: Interrater reliability was strong for pre-intervention and post-intervention grading of markings (ICC=0.97; p<0.001 and ICC=0.96; p<0.001) and surgical performance (ICC=0.76; p=0.01 and ICC=0.85; p=0.001). Compared to pre-intervention, post-intervention marking performance (8.0±2.5 vs. 2.9±3.1; p=0.03), procedural confidence (24.0±7.0 vs. 14.7±2.3; p=0.03), knowledge (40.3±4.4 vs. 33.5±3.7; p=0.03), and performance (20.3±3.6 vs. 15.3±3.1; p=0.04) significantly improved in the digital simulation group, but not in the textbook group. All participants were more satisfied with the digital simulator as an educational tool (27.7±2.5 vs. 14.4±4.4; p<0.001).

Summary/Conclusion: We present level I evidence suggesting that digital cognitive simulators lead to significant improvement in surgical markings, as well as procedural confidence, knowledge and performance.
Experiences of turkish parents taking care of their children through nam treatment: a qualitative analysis
S. Zeytinoglu-Saydam

Abstract Content: For the purposes of this qualitative study, the researcher and her trained assistant interviewed 20 parents (10 mothers and 10 fathers) of 12 children born with cleft lip palate and receiving NAM (naseo-alveolar molding) treatment at Yeditepe University’s Faculty of Dentistry, Department of Orthodontics. Throughout the process of NAM treatment, parents were interviewed 4 times; first, when they came for their first appointment to start the NAM treatment, second, a month after they started the NAM treatment, third, before their child’s lip surgery and fourth a month after the lip surgery. Some parents were not able to complete all the interviews; overall, 56 interviews took place. During this process, parents discussed their experiences during the NAM treatment as well as their experiences during the diagnosis, birth and first surgery with regards to challenges and coping mechanisms, their reactions, impact of this process on their couple relationships and social lives.
Maximising patient quality of life lies at the heart of craniofacial care and is the responsibility of the entire multidisciplinary team. Yet, consensus regarding the key psychosocial aspects of craniofacial care, and how and when to assess them has been difficult to achieve. As a result, prior research has utilised a variety of different measures at different time points, rendering findings difficult to compare across studies. Meaningful conclusions about how individuals and families adjust to the challenges associated with craniofacial conditions have been difficult to draw, and current evidence for psychosocial intervention is weak. Based on extensive research and clinical expertise, this talk will provide an overview of the common psychosocial challenges reported by individuals born with craniofacial conditions and their families. Key time points at which these challenges are likely to occur will be identified, and outcome measures for use in this population will be recommended. Finally, a tiered approach to the assessment of psychosocial adjustment will be proposed, applicable to all countries irrespective of resource level. Information regarding how to access outcome measures and all related materials free of charge will be provided.
OS 11.3
Clapa whole of life survey for adults born with a cleft and living in the uk
K Ardouin¹, N Stock², C Cunniffe¹
¹Cleft Lip and Palate Association, London, ²University of the West of England, Bristol, United Kingdom

Background: The Cleft Lip and Palate Association (CLAPA) Adult Services Project, based in the UK, in conjunction with the Centre for Appearance Research based in Bristol, UK have undertaken the world’s most comprehensive survey of those over the age of 16 born with a cleft, focusing on medical journey, emotional wellbeing, discrimination, family, friendships and romantic relationships, genetics, education, employment and dietary habits. The study was designed with the view of using its findings to develop new strategies to support adults who were born with a cleft.

Aims: To understand the unique experiences and challenges faced in adulthood by people who were born with a cleft lip and/or palate. To explore the residual medical issues that having a cleft can pose in adulthood. To investigate the social experiences of adults who were born with a cleft (e.g. work experiences, bullying and discrimination, dating and intimacy etc.) to determine if these are influenced by cleft.

Methods: The survey was available in both online and paper format from August – October 2018 and contained 12 sections consisting of a variety of qualitative and quantitative questions (240 questions in total). A number of questions incorporated standardised measures, whilst others were bespoke. Participants had to be 16 or over, living in the UK at the time of the survey, or residing outside of the UK for a period of 12 months or less. The survey was advertised online using social media, email newsletters, posters and a BBC Radio interview. The study received ethical approval from the University of the West of England and met all requirements in the Declaration of Helsinki.

Results: The survey attracted 259 respondents, ranging in age from 18-84. The survey has revealed a number of concerns that often persist into adulthood including fistulae, speech concerns, difficulties with dentition, hearing and breathing. Additionally the survey revealed that there were a number of emotional adjustment and psychological concerns. These included areas such as self-esteem, dating and intimacy and decision making regarding having children.

Summary/Conclusion: This presentation will offer a summary of the survey’s most pertinent findings and an overview of the analysis expected to be completed during year two of the project. The presentation will discuss the findings relevance to the wider European and international cleft community. Where relevant, data gathered from the CLAPA Adults Engagement Roadshow, a tour of 12 UK cities in Summer 2018 where adults born with a cleft came together to share their experiences and discuss aspects of the survey will be included.
OS 11.4
Facilitating patient led care for adults seeking cleft related input in scotland: what do they want and need?
J Hare1, R Crawford1
1National Cleft Surgical Service for Scotland, NHS Scotland, Glasgow, United Kingdom

Background: In Scotland, following a pathway of fairly standardised treatment in childhood, individuals over the age of 15 vary both in the issues (if any) that their cleft causes them, and their motivations/expectations for further (elective) treatment regarding cleft. Previously, any ‘adult’ patient looking (or encouraged) to discuss further treatment attended a multidisciplinary clinic with a variety of clinicians working in cleft, allowing each to contribute to the treatment plan as appropriate. However, some patients reported feeling intimidated by this setting, and/or leaving with plans to address issues they did not personally find problematic (i.e. clinician led care). Additionally, having all potentially relevant clinicians present where not all were needed was an inefficient use of resource Consequently, the Cleft Clinical Psychology Service developed an initial assessment appointment for individuals aged 15 or over who had requested or been referred to discuss any new (elective) cleft related care. The aim of this appointment was to determine the issues patients would like support for (i.e. facilitate patient led care), so they could be referred to the relevant clinician(s) within the team.

Aims: The study aimed to answer these questions:
- What concerns do adults returning to cleft care seek input for?
- What are the psychological needs (if any) of adults considering further cleft care?

Methods: Participants: 34 individuals aged 15 or over with a cleft lip and/or palate, living in Scotland, who were awaiting an appointment to discuss current issues and/or additional care related to cleft. The age range was 15-67 years (median 24 years), and 21 (62%) were female.
Materials: Measures including standardised scales related to cleft and wellbeing were completed, as well as categorical information on interest in cleft team specialties, and present issues.
Design: This was a non-experimental design.
Procedure: All patients aged 15 or over awaiting new input from the cleft team between January-December 2018 attended a 45-60 minute appointment with a team clinical psychologist. Patients completed questionnaires before discussing these and their reasons for return and expectations of/motivations for further cleft related care. An assessment report/referral summary was then sent to relevant members of the cleft team.

Results: Concerns prompting treatment: 22.73% of patients wanted input for functional issues only (i.e. discomfort, speech, breathing), whilst 36.36% wanted support for aesthetic issues only and 40.91% wanted support for both aesthetic and functional issues.
Surgeons were the clinicians that patients most often said they wanted input from prior to discussion (90.00% of patients), followed by Restorative Dentists (56.67%), Orthodontists (46.67%), Speech Therapists (26.67%), Clinical Psychologists (26.67%), Geneticists (26.67%) and Audiologists (6.67%).
Psychological needs: 35.71% of patients were offered an appointment with (or already seeing) Clinical Psychology to discuss psychological issues related to cleft, with the commonest issue being appearance/speech related social anxiety. A significant proportion of patients demonstrated clinically significant levels of anxiety (44.83%) and/or depression (27.59%).

Summary/Conclusion: - Adults seek cleft related input for a variety of issues
- Adults most often sought surgical input, but were not always familiar with other specialties.
- Adults seeking cleft related care may have psychological needs in addition to physical issues
Relational development in infants with cleft lip and palate and impact of the malformation on parents. Results of a prospective multicentric french study

P. GAVELLE¹, B GROLLEMUND², C PEREZ MARTINEZ³, A GUEDENEY⁴, J MULLAERT⁵, T ALFAIATE⁵

¹Maxillo-facial surgery, Hôpital Necker Enfants malades, Paris, ²Dentofacial orthopedic Department, Hospices civils, Strasbourg, France, ³Perinatal and infant psychopathology, AM, Cambridge, United States, ⁴Child psychiatry Department, ⁵Biostatistics, Epidemiology, and Clinical Research Department , Hôpital Bichat Claude Bernard, Paris, France

Background: Previous studies have focused on several topics about the impact of cleft lip and palate on the infant relationship and on babies. These findings support the fact that having a child with a cleft lip and palate may adversely affect the parent relationship and the further child development. No consensus were found in literature about the impact of a late or neonatal repair on mothers, and on the influence of the severity of the cleft on the mother infant relationship.

Aims: This study is the first prospective study on a french cohort, observing babies development, mother infant relationship but also the impact on mothers and fathers, depending on the moment of the diagnosis (antenatal or at birth), type of cleft and neonatal or late repair.

Methods: Four inclusion centers have been selected, according to the time-lapse before the first surgery (2 neonatal repair centers and 2 late repair centers). The main hypothesis is that the longer the time-lapse before the first surgery, the more likely are the psychological perceptions of the parents to affect the harmonious development of the child. Parents and children are seen twice, when the child is 4 months (T0) and when he is 1 year old (T1). At these 2 times, an observation of both relationship and baby's relational withdrawal is filmed and an ADBB scale is filled by a psychologist. Self administered questionnaires measuring factors liable to affect child-parent relationship are issued to the mothers and fathers (Edinburgh Postpartum Depression Scale, Impact On Family Scale and Parental Stress Inventory). For all analysis, multiple variables will be taken into account: neonatal or late surgery (≥2.5 months), antenatal or at birth diagnosis, and the type of cleft.

Results: 156 patients were included.
- Babies with clefts don't show more withdrawal behavior than other babies in general population. No linkable variables like type of cleft, time lapse or diagnosis period. No link between infant's relational withdrawal and mother's depression.
- Both parents are more stressed (PSI) and mothers are more depressed (EPDS) in the later surgery group.
- Higher impact of the cleft lip and palate compared to cleft lip: Mothers are more depressed (EPDS). However, parents are more impacted in their family (IOFS) when the infant has a cleft lip only and a neonatal surgery.
- Higher post partum depression scores for both parents (EPDS) compared to general population.

Summary/Conclusion: This study can show us the parental psychological distress after the birth of a baby with a cleft, even if there is an antenatal diagnosis, and especially when the surgery is late (after 2.5 months) and the cleft associating lip and palate. These results could influence the way we organize the screening of babies and both parents to provide adjusted psychological care, respond to their real needs and improve further infant development.
OS 11.6
Psychological screening for UK cleft orthognathic patients: pre and post surgery outcomes
K Le Marechal¹, L Grey¹, R Cassidy¹
¹South Thames Cleft Service, Evelina London Children’s Hospital (Guy’s and St Thomas’ NHS Foundation Trust), London, United Kingdom

Background: Psychological aspects of orthognathic surgery have been shown to be an important factor in decision making and satisfaction post-surgery (Girod et al., 2015; Oliveira et al., 2016). Cadogan and Bennun (2010) explored the experiences of cleft patients before, during, and after orthognathic surgery. The reported that, pre-surgery, patients felt that their appearance was negatively impacting their lives. Those who decided to go ahead with orthognathic surgery were highly motivated to make changes to improve their quality of life and did indeed find that their social confidence improved post-surgery.

Cheung et al (2006) considered the early psychological changes of cleft versus non-cleft patients undergoing orthognathic surgery. They measured patient’s anxiety and distress and subjective well-being and found cleft patients displayed lower levels of social anxiety and distress than non-cleft patients – possibly because cleft patients are better prepared psychologically.

The UK Cleft Clinical Psychology Clinical Excellence Network (CEN) protocol for the psychosocial screening of patients therefore recommends pre and post orthognathic psychological assessment as part of their MDT care. The assessment includes measures of psychological well-being, orthognathic quality of life and feelings about appearance, speech and hearing.

Aims: The aim of the paper was to examine data collected from UK cleft centres in order to consider the psychological outcomes for cleft patients who go undergo orthognathic surgery.

Methods: All UK cleft centres were invited to contribute data regarding the pre/post assessments for orthognathic patients. The assessments were comprised of the Hospital Anxiety and Depression Scale (HADS), Orthognathic Quality of Life Questionnaire and Cleft Hearing Appearance and Speech Questionnaire. Demographic data and the timings of assessments relative to the surgery were also collected. Data for 385 patients was received from four cleft centres. The authors are awaiting data from one further cleft centre.

Results: 325 patients had at least one pre surgery assessment and 83 had at least one post. 84% of patients scored in the normal/mild range for anxiety at the pre orthognathic stage and 95% in the normal/mild range for depression. No significant change was found in depression or anxiety post-surgery. Initial analysis showed significant improvements in satisfaction with facial appearance, satisfaction with overall appearance and orthognathic quality of life (p< .001).

Summary/Conclusion: In the UK, our protocol is that all cleft patients considering orthognathic surgery meet with a Clinical Psychologist during the treatment process. Patients are well-supported psychologically throughout the process and, whilst they are not generally found to be experiencing significant anxiety or depression before, during or after the process, patients report increased satisfaction with facial appearance and overall appearance as well as significant improvements in orthognathic quality of life. The importance of psychological support to protect positive wellbeing is considered crucial.
Operation is coming: how to act

SIEMANN1

1Medical Psychology, Radboudumc, Nijmegen, Netherlands

Background: Children with cleft undergo multiple surgeries. The jaw-closure around the age of 10 years is the first operation they consciously experience. This awareness can create feelings of fear among some of these children. We have taken a look at whether assistance of a psychologist in the preparation for an operation can be helpful.

Aims: Reducing the level of anxiety through psychological assistance/counseling. And also getting more involvement from parents.

Methods: Before the operation (while visiting the cleft team) patients are asked about the level of anxiety. They are being referred to a psychologist. The psychologist first contacts the parents. And together with the parents determines what the question is. One or more appointments are scheduled. Acceptance and Commitment Therapy (ACT) is used during the sessions with the psychologist.

Results: In a questionnaire taken after the procedure, parents indicated that they appreciated the preparation/psychological support. The anxiety level from children and parents dropped considerably because both children and parents had a good understanding of what was going to happen. Deployment of ACT also seems to be helpful.

Summary/Conclusion: For a cleft operation psychological preparation and psychological support can reduce the anxiety level for both children and parents.
"Strange and beautiful": parents’ perceptions of their child’s visible difference and the impact of appearance-altering surgery
K Billaud Feragen¹, A Myhre¹, N Stock²
¹Oslo University Hospital, Centre for Rare Disorders, Oslo, Norway, ²Centre for Appearance Research, University of the West of England, Bristol, United Kingdom

Background: While appearance concerns are now considered normative within the general population, those born with a visible difference vary not only from the societal ideal, but also from the norm. Previous research has investigated parents’ experiences of having a child born with a visible difference, the potential impact of the visible difference on parental attachment, and on parents’ satisfaction with the outcomes of their child’s aesthetic treatment.

Aims: The aims of the present study were to explore parents’ perceptions of their child’s visible difference, the impact of appearance-altering surgery from the parents’ perspective, and how parents felt about having appearance-related conversations with their child.

Methods: Individual semi-structured interviews were conducted with 44 parents of children born with a range of rare craniofacial conditions. Interviews were transcribed verbatim and subjected to thematic analysis.

Results: Some parents reported conflicting feelings toward their child’s visible difference, often stating that they accepted their child’s appearance while also acknowledging that their child looked different or strange. Many parents worried about how their child would cope when they became more aware of their difference, and were unsure how to talk to their child about appearance-related concerns. While some parents perceived appearance-altering surgery to be a positive step toward normalising their child’s appearance, others found it difficult to adjust to these changes.

Summary/Conclusion: Parents can experience difficult emotions in response to their child’s visible difference, which may be further complicated by their child undergoing appearance-altering treatment. There is a need for parental guidance to be developed in order to help parents adjust to their child’s different and changing appearance, and to facilitate positive conversations around appearance both within and outside the family home.
Using telehealth to empower parents of pre-schoolers with CLP: providing high quality information addressing school- and psychosocial aspects in life.

M Ditlfsen\textsuperscript{1}, J Ottesen\textsuperscript{1}
\textsuperscript{1}Department of Speech and Language, Statped, Oslo, Norway

**Background:** The Cleft Lip and Palate (CLP) Oslo-team aims to deliver a complete service for its users. This consequently involves a holistic and interdisciplinary coordinated healthcare service, ranging from speech therapeutic and psychological interventions, to different orthodontic and medical procedures throughout life. As psychologists, we are concerned about the psychosocial well-being of the population. Our clinical experience and the feedback from parents indicate that transitioning from kindergarten to primary school can be especially demanding. Besides becoming increasingly self-aware around the age of 6, being brought out of a well-known, safe and relatively small environment with many adults to a new, unfamiliar and bigger environment with new classmates and fewer adults also tend to represent a challenge. As Norway is a country that spreads over a large geographical area, and the CLP team being regionally divided into two teams, reaching out to the total population also represents a challenge. In lack of ideal interventions before age 10, we have developed a digital parental educational program, using telehealth technology.

**Aims:** By offering an educational program for parents, we aim to empower parents by increasing their competence and awareness on their child’s needs transitioning from kindergarten to school. Naturally, the impact we are looking for, is that the children will develop in a conscious and well reflected environment, and thereby reduce the possible psychosocial impact of being born with CLP. Another aim for us is to evaluate the impact of a follow-up online session where 5 of the participants are invited as a group to ask the team’s psychologists questions and reflect on topics addressed in the program. If successful, the educational program will be included as part of the standardised psychological treatment plan for children born with CLP in Norway.

**Methods:** Participants were selected by sending a text message to all of the parents with children aged 5-6 (pre-schoolers) born with CLP in the area of the Oslo team, with a short intro about the project. They were asked to reply with their personal email address if interested. The participants that replied received more details about the program as well as a questionnaire about their expectations and thoughts about their children starting school. After completing the program, they all received another questionnaire evaluating the impact of the program. We also wanted to evaluate the impact of a follow-up online group session (video conference) carried out with five of the participants after they completed the education program. The five participants invited to the video conference were selected randomly within the group of parents with children with CLP only, excluding parents of children with additional difficulties. After the meeting they received another questionnaire evaluating the impact of the group session.

**Results:** There is an ongoing data collection. The results will be shared, and implications discussed at the conference.

**Summary/Conclusion:** In this project, we have developed and administrated an online educational program for parents with children born with CLP. The aim of the project is to increase the parents’ competence and awareness about their children starting school.

At the conference, we will present the content of the educational program. The method is evaluated, and implications and further research plans will be discussed.
"Exposed and vulnerable": parent reports of their child’s experience of multidisciplinary craniofacial consultations

A Myhre¹, K Feragen¹, N Stock²

¹Oslo University Hospital, Centre of rare disorders, Oslo, Norway, ²Centre of Appearance Research, University of the West of England, Bristol, United Kingdom

Background: Complex multidisciplinary treatment and long-term follow-up is normally required for children born with a craniofacial condition (CFA). Few studies have investigated the child’s treatment experiences.

Aims: The objective of this study was to explore children’s experiences of multidisciplinary team (MDT) consultations from the perspective of their parents.

Methods: Thirty-eight parents of children with a CFA were interviewed in person or over the telephone. Interviews were transcribed verbatim, and explored using thematic analysis.

Results: Background factors influencing the child’s experience of the consultation included age, developmental stage, personality, and prior treatment experiences. Participants tried to prepare their child for meeting the MDT, but did not fully understand what to expect themselves. During consultations, participants were acutely focused on their child’s emotional state, making it difficult to balance their desire to protect the child from potentially negative experiences, and the need to engage in a constructive dialogue with health professionals. Participants believed that health professionals’ conduct could considerably influence the child’s wellbeing and subsequent treatment decisions. Finally, participants highlighted the need to debrief their child to help them adjust positively.

Summary/Conclusion: The ultimate goal of craniofacial care is to help children develop into confident adults who are able to cope with the challenges associated with their condition. MDTs play a vital role in creating a safe and supportive environment in which children feel genuinely informed and involved in key aspects of their care.
3D technology benefits for orthodontic preparation to early secondary alveolar bone graft for CLP patients
A Majourau-Bouriez¹, M Lê-Dacheux¹, A Picard¹
¹MaxilloFacial and Plastic Surgery, Hopital Necker Enfants Malades, Paris France, Paris, France

**Background:** Multidisciplinary CLP teams who are performing early secondary alveolar bone graft at age 4 to 5 should optimize patients cooperation toward orthodontic treatment.
One of the most common and efficient way to expand the maxilla in CLP patients prior alveolar bone graft is to use a quad'helix appliance.
The traditional way to place a quad'helix requires a number of appointments and cooperation from the patient which can become difficult to manage for many orthodontists and patients and their family.
Nowadays 3D Technology offers an alternative to simplify treatment, reduce the number of appointments and decrease treatment burden

**Aims:** The goal of this presentation will be to show and describe the benefits of using 3D Technology in Orthodontics for young CLP patients prior early secondary alveolar bone graft

**Methods:** Clinical and lab steps will be described from recording useful accurate data of the CLP patient to 3D print custom made appliances for each of our patient with extensive possibilities

**Results:** The use of 3D technology for our young CLP patients from recording initial data to 3D printed bands and quad'helix has considerably simplified orthodontic treatment care prior secondary alveolar bone graft. It is not only a benefit for the orthodontist but also for the surgeon and the patient and his family.

**Summary/Conclusion:** Nowadays, 3D technology in Orthodontics for our CLP patients offers a cost effective way which can be very efficiently used in order to obtain high performance appliances and to reduce the burden of treatment
3D analysis of maxillary arches in cleft lip and palate patient after using nasoalveolar molding device
A Manosudprasit¹, W Chantadilok ¹
¹Orthodontics, Khon Kaen University, khon kaen, Thailand

Background: Presurgical nasoalveolar molding (PNAM) is the device accepted as an option for correcting of nose and lip shape and also alveolar ridge segment position before primary lip repair, especially for patients with large cleft and significant misalignment of the dentoalveolar arches.

Aims: To evaluate the 3D changes of maxillary arches after using khon kaen university presurgical nasoalveolar molding device (KKU-PNAM) in complete unilateral cleft lip and palate (UCLP) patients.

Methods: The samples composed of maxillary arch models of eight infants with UCLP who treated with Khon Kaen University PNAM (KKU-PNAM). Dental casts were digitized using a 3D scanner, they were evaluated at initial visit (T1) and a visit before cheiloplasty (T2). For statistical analysis, Wilcoxon Signed-Rank test was used to compare the difference between treatment periods.

Results: The result showed that measurement at T1 was significantly decreased compared to T2 in the width dimension (P<0.05), while vertical dimension, length dimension, and ratios between posterior and anterior cleft width were significantly increased (P<0.05).

Summary/Conclusion: The KKU-PNAM is effective in improving maxillary arch shape which could achieve a better result in alveolar cleft repaired.
Facial asymmetry changes in patients with oculo-auriculo-vertebral spectrum using 3d morphometry

P Polackova¹, V Moslerova², M Kotova¹

¹Department of Orthodontics and Cleft Anomalies, 3rd Faculty of Medicine, Charles University and Kralovske Vinohrady University Hospital, ²Department of Biology and Medical Genetics, 2nd Faculty of Medicine, Charles University and Motol University Hospital, Prague, Czech Republic

**Background:** Oculo-auriculo-vertebral spectrum (OAVS) is a congenital anomaly. Typical unilaterally affected structures are ear, mandible and temporomandibular joint, facial skeleton, nerves and surrounding soft tissue.

**Aims:** The aim of this study was to objectify and evaluate changes in individual asymmetry in OAVS patients using 3D facial scans in two different ages.

**Methods:** The facial scans were obtained in 6 patients (age from 6 to 15; 5♂, 1♀) with OAVS using 3dMD FaceSystem, in all individuals twice: at the time t1 and t2 (ø interval 14,5 months). Using the construction of dense correspondence mapping by CPD-DCA method (coherent point drift - dense correspondence analysis) between facial meshes, model registration were performed. An ideally symmetric mesh was created as an average of the original and the mirror mesh. By subtracting the points of the original image from the symmetrical image, individual asymmetry were visualized by heat maps. All the analysis and subsequent visualizations were performed in the Morhome3cs software.

**Results:** Comparison of the individual asymmetry in patients at the time t1 and t2 showed only slight differences, on average 0.5-1 mm. These changes are within the meaning of both, increasing and also decreasing deviations from perfect symmetry. No correlation between the change of asymmetry during the observed period and the age of the patient were found. There were also no correlation between the severity of the defect and the progress of asymmetry.

**Summary/Conclusion:** Evaluation of facial asymmetry changes using 3D facial morphometry can provide important information in serious rare diseases such as OAVS. Based on the results of this pilot study seems that facial growth in patients with OAVS is relatively steady and there is no significant changes in facial asymmetry. These findings could help clinicians in indication of conservative treatment during ongoing growth. To confirm this hypothesis, detailed longitudinal evaluation of facial development is necessary.
New proposal for three-dimensional measurement of results in pre-surgical functional maxillary orthopedic treatment for cleft lip and palate patients

O Pelliccioni Monroy¹, T Pannaci³⁴, A Pares Acosta⁵⁶
¹Biomechanics Group of Simón Bolívar University, ²Departamento de Mecánica, Universidad Simón Bolívar - VE, Caracas, Venezuela, Bolivarian Republic Of, ³Maxillofacial Department, Vall d’Hebron Hospital, Barcelona, Spain, ⁴Dentist, Operation Smile, Norfolk-Virginia, United States, ⁵Technical sales and consulting, Ineltk Nord GmbH, Hamburg, Germany, ⁶Electrical Engineering, Universidad Simón Bolívar - Ve, Caracas, Venezuela, Bolivarian Republic Of

Background: A dental cast model reflects the current state of a patient's upper maxilary, which is why it represents valuable information about the morphology of the cleft for a given moment. Always when the patient attends its functional maxillary orthopedics treatment (FMO) check, the cast models are taken. The evolution in time of the cleft palate after the intervention by FMO is observed with all of the models. There are some control procedures that allow the characterization of the malformation. A protocol that includes anatomical points for characterization of the dental arch and the nasal area in patients with cleft lip and palate are developed by Adus & Pruzansky (1967). On the other hand, the specific anatomical points characteristic of the cleft and the dental arch are described by the protocol of Mazaheri et al (1971). A methodology to reduce the error associated with the positioning of anatomical points linked to cleft palate are described by Seckel et al (1995) and Brief et al (2006). Finally, the position of these by using medical images were studied by Bayerlein et al (2006).

Aims: The cleft lip and palate involves an abnormality in the facial skeletal structure that affects the three planes: sagittal, transverse and axial or coronal. Most of the current procedures that report this changes in the oral cavity are limited to the transverse plane. A new non-invasive methodology to report and quantify the modeling and external remodeling of the oral cavity is required.

Methods: Firstly, the cast models of the cleft patient and its respective intraoral plate adapted for each moment of the FMO treatment are digitalized in 3D geometries. Based on the procedure published by Mazaheri et al (1971), reference markers are placed on the surface of the model. In the present work, an adaptation of these procedure to three-dimensional place considering changes in the orthonormal planes was proposed. During FMO treatment, all cast models were measured. These novel changes on the known version of Mazaheri in two dimensions, expand in detail the report of changes in the oral cavity by allowing: the spatial calculation of cleft palate segments' displacement and the level of asymmetry present, volumetric and external topological growth. All the results were synchronized with each moment of intervention with the FMO treatment, and stimulated areas of contact between the palate and the intraoral plate.

Results: The volumetric digital reconstruction of the cleft palate presents great advantages with respect to the traditional report in two dimensions: the exposed methodology allows studying the closure of the alveolar and palatal gap after the application of FMO treatment. The asymmetry can be quantified and will not depend on the perception of the doctor; lineal and rotational displacements on each cartesian axis can be calculated. Another advantage of this work is that the specific areas of the palate that are undergoing major changes can be recognized on the digital model. It is not possible with the Mazaheri technique in the plane.

Summary/Conclusion: The study presented a non-invasive digital methodology that allows a more detailed study and program of pre-surgical FMO treatment through three-dimensional scanning of the palate and the intraoral plate.
Maxillofacial anthropometric measurement on cleft patients: validity and reproducibility of the three dimensional imaging system
S Othman¹, L Saffai², W Wan Hassan¹
¹Department of Paediatric Dentistry and Orthodontics, University of Malaya, ²Oral Health Program, Ministry of Health, Kuala Lumpur, Malaysia

Background: Cleft lip and palate is the most common cranio-facial abnormality in the world. Various methods had been used to qualify pre and post treatment craniofacial variation and evaluation. Stereophotogrammetry is currently the gold standard for these purposes due to its many advantages.

Aims: The aim of this prospective experimental study is to validate the accuracy and reproducibility of linear measurements of three-dimensional (3D) images and to compare the measurements with direct anthropometry method on cleft lip and palate patients.

Methods: Twenty-six linear facial measurements were derived from twenty-five standardized surface landmarks obtained from thirty-six cleft patients. These linear measurements were taken manually with calipers and compared it with the digitally calculated distance on the 3D images captured using VECTRA M5-360 Imaging System (Canfield Scientific Inc, USA) with pre-marked landmarks. Another pair of 26 linear measurements were computed on the 3D images 2 weeks apart for intra and inter observer agreements. Statistical analysis was conducted using intraclass correlation coefficient index and paired t-test.

Results: The intra-observer and inter-observer reliability showed good to acceptable agreement. There were no statistically significant differences for the linear measurements taken by direct measurement and the digitally calculated distance on the 3D images except for eye fissure height, nose width and upper vermilion thickness (p<0.001). The mean differences were 1.30 mm, 1.91 mm, 1.35 mm and 1.02 mm respectively, thus clinically they were not significant (>2mm). Five measurements for inter-observer reproducibility possessed clinically significance mean differences of merely slightly more than 2 mm. They were upper face height (2.13 mm ± 3.77), lower face height (-2.31 mm ± 4.46), right eye fissure length (2.23 mm ± 2.89), nose width (2.62 mm ± 2.93) and pronasale to left alar base (2.47 mm ± 2.75).

Summary/Conclusion: The 3D imaging system is both accurate and reproducible in obtaining data for cleft patients; hence it has potential to be applied clinically and is suitable for clinical application. However, extra attention must be taken when measuring the areas around the eyes, nose and the upper lips. In addition, training of the operator is strictly advisable.
Bone Grafting Part One

OS 13.1
Early alveolar bone grafting at 5-6 years
B. Grollemund

The objective of residual alveolar cleft bone graft is to restore maxillary segment union and stability, to allow tooth eruption, to close alveolar fistula when it exists, and to provide support both to the lip and to the nose. The protocol described manages cancellous iliac bone graft before 6 years old in deciduous dentition. Unlike the usual protocols, which are done before the eruption of the canines, this one is realised before the eruption of lateral incisor when present or before the eruption of central incisor when agenesis occurs. As for the others protocols, a maxillary arch expansion is necessary before grafting. This expansion restores a normal deciduous intercanine space in order to allow good canine function on the cleft side with symmetrical occlusion. After the bone graft, the anterior expansion is continued to stimulate the interincisive suture and increase the width, to allow a good alignment of the rotated incisor teeth. Why this choice? The goal is to restore all the functions concerned by the repair of the lip, the nose, the maxilla, the dentition and the palate. That means to restore properly, mastication in good occlusion, deglutition, speech and nasal breathing and an essential function of the face, which is communication. It needs to look good at rest with a good aptitude for normal movement and a symmetrical expression, which are necessary for a good social life and self-esteem. There’s a necessity to give to the patient and the family the benefit of a good social life very early, before school age. Normal function results in normal growth. Furthermore, it’s of the utmost importance to do all the surgeries before reading learning process.
Alveolar bone grafting in cleft patients: timing and technique
F. Bierenbroodspot

Patients with a unilateral or bilateral cleft lip, alveolar arch (and palate) will at some point undergo alveolar bone grafting of the cleft. It provides a continuous and stable maxillary arch and thereby supports the base of the ala nasi on the cleft side. Persistent oro-nasal communications are closed at this stage. Alveolar cleft closure facilitates canine and/or lateral incisor eruption into the alveolar cleft. Timing and technique of this procedure are important. Optimal timing and technique of the alveolar bone grafting procedure were the two important clinical questions to be answered in the chapter on this subject in the (Dutch) Guideline on Treatment of Patients with Clefts of the Lip and Palate. The following recommendations concerning timing and technique were made:

Recommendations on timing of bone grafting:
1. Preferably close the alveolar cleft by means of an early secondary alveolar bone grafting procedure.
2. Base the timing for the alveolar bone grafting procedure on the position and the stage of root formation (½ - 2/3) of the canine on the cleft side. The presence of a lateral incisor and the moment of its eruption can bring the timing forward. For certain indications, the position of the central incisor can be of importance.
3. Decide on the timing of the alveolar bone grafting procedure in close consultation with the treating orthodontist.
4. Consider tertiary alveolar bone grafting only in cases where no (secondary) alveolar bone grafting procedure was performed or when insufficient bone is present in the alveolar cleft for - for example - an implant at a later age.

Recommendations on technique of bone grafting:
1. Reconstruct the alveolar cleft using bone harvested from the iliac crest or the chin (if needed supplemented by a bone substitute).
2. If a larger volume is required, use bone from the iliac crest, or opt for bone from the chin supplemented by a bone substitute. Based on the literature, it is not possible to make a recommendation for the choice of the bone substitute to be used.
3. The use of only a bone substitute for the reconstruction of an alveolar cleft, in other words without an autologous bone transplant, should only be performed in a research context and is as yet not suitable for use in general practice. The considerations behind the above recommendations of the Guideline will be discussed. Besides that the procedure of early secondary bone grafting will be presented.
**OS 13.4**

**Assessment of palatal fistulas after orthodontic expansion**

M Aksu¹, F Yaz¹, S Gunes¹

¹Orthodontics, Hacettepe University, Ankara, Turkey

**Background:** Palatal fistula is a patency between the oral and nasal cavities. It is a known complication of cleft palate repair. Its incidence ranges between 12 and 45%. Fistula may be present anywhere along the primary or secondary palate. They are a result of inadequate dissection of the flaps, closure under tension, post-operative bleeding, hematoma formation between the oral and nasal layers, and infection. As a late complication, orthodontic expansion is considered to be a risk factor in developing palatal fistula. Although it has been assumed, no study has been conducted.

**Aims:** The purpose of this study was to determine the occurrence of cleft palatal fistula in a series of nonsyndromic cleft lip and palate children treated by maxillary expansion at the authors' institution. This prospective analysis of 39 patients includes 20 bilateral (BCLP) and 19 unilateral (UCLP) cleft lip and palate cases, whose mean age was 9.7 years for BCLP and 9.2 for UCLP at the time of maxillary expansion. All the patients were operated by the same surgeon and none of the patients had secondary alveolar bone grafting before expansion.

**Methods:** Before the start of treatment all the patients were examined regarding the fistulas and the data was recorded according to the Pittsburgh palatal fistula classification system. The structure of the hard and soft palate and alveolar ridge were double-checked by two authors until coming to an agreement. Intraoral photographs and dental casts were examined if necessary. The BCLP group had bonded rapid maxillary expander because of the symmetrical maxillary constriction while UCLP group had quad helix expander due to the asymmetrical arch constriction. The expansion was ended until 2-3 mm overexpansion was gained. The expanders were debonded in order to examine the patients according to the fistulas. All differences were recorded and the expanders were rebonded in order to keep in retention.

**Results:** For 20 CLP patients, 10 patients (%50) had palatal fistulas and 5 of them (%25) had type V fistula before expansion. The remaining 5 had type IV, types IV-V, types IV-VII and types V-VI-VII. After expansion 14 patients had fistulas, which type V was dominating (8 patients, %40). The remaining 6 patients had types IV-V (3 patients, %15), types IV-VII (1 patient %5), types V-VI-VII (1 patient %5) and types IV-V-VII (1 patient %5).

For UCLP patients; of 19 patients, 9 did not have any fistulas before expansion, and these 9 patients did not develop any type of fistulas during expansion (%47.4). Of 12 patients, 5 patients had types V-VII (%26.3), 4 patients had type VII (%21.1), and 1 patient had type V (%5.3). After expansion the combination of type V-VII fistulas increase to 7 patients (%36.8), while in 1 patient types V-VI-VII occurred (%5.3).

Regardless of the fistula type, %50 fistulas were increased dimensionally in BCLP and %32 in UCLP.

**Summary/Conclusion:** BCLP patients were more prone to developing fistulas by expansion even they had not have the fistulas at the start of treatment in this study. In UCLP patients the new developing fistulas were relatively low, but not seem to be unavoidable. The UCLP patients who had not have any fistulas at the start of treatment, did not develop any fistulas, keeping in safe zone.
Orthodontic pre grafting of large alveolar bony and soft tissue gaps: orthodontic-surgical cooperation reduce the burden of care

C Incorvati¹, M Meazzini², L Autelitano²

¹ORTHODONTICS, SMILE HOUSE S. Paolo Hospital University of Milano, ²SMILE HOUSE, S. Paolo Hospital University Of Milano, Milano, Italy

Background: Patients with cleft lip and palate require a team approach for their treatment, comprised of several specialists. This multidisciplinary care starts from birth and continues into adulthood, and coordination amongst specialists is a major contributor to success in cleft treatment. Closure of wide alveolar clefts with large soft tissue gaps and reconstruction of the dentoalveolar defect are challenging for the surgeon because of the difficulty in achieving complete soft tissue closure by using local attached gingiva. In the more severe cases lingual grafts are needed. Some authors successfully used interdental segmental distraction, which nevertheless requires extra surgical burden.

Aims: The objective of this study was to assess the effectiveness of tooth borne devices utilized to advance the lesser segments with a complete approximation of the soft tissues of the alveolar stumps. This allowed traditional simultaneous soft tissue closure and bone grafting avoiding the need for supplementary surgery.

Methods: A Case series of 11 unilateral and bilateral complete cleft lip and palate patients, who presented prior to bone grafting with large soft tissue and bony alveolar defects was prospectively selected. Selection criteria were bony gaps larger than 10mm or less than 5mm but associated to locally deficient or scarred soft tissues. An orthodontic protraction was applied by means of an RPE device combined with protraction face mask in younger patients or with the Alt-Ramec technique in patients older than 8-9 years. Traditional x rays were available in 8 patients and a CBCT in 4 patients at T0 to assess the bony cleft defect. Soft tissue deficiency was photometrically quantified and given a qualitative score. Records were available a T1 (end of the orthodontic distraction) and after bone grafting.

Results: Clinical and findings showed a significant reduction of the cleft area, in particular the approximation of the cleft segments when large soft tissue defect were present.

Summary/Conclusion: The present data seems to indicate that, in patients affected by large clefts, orthodontic distraction could be an efficient method to reduce the cleft defect, resulting in a favorable area for graft placement, minimizing the risk of post grafting fistulas or reducing the need for supplementary invasive surgical procedures and favoring long term stability of the graft survival.
Sagittal and vertical skeletal relationships of complete uclp patients with early secondary gingivo – alveolo – plasty
M Drevenšek¹, A Plut², A Eberlinc³, M Kočar³
¹Division of Stomatology, University Medical Center Ljubljana, ²Division of Stomatology, University Medical Center Ljubljana, ³Division of Surgery, University Medical Center Ljubljana, Ljubljana, Slovenia

Background: The early secondary gingivo alveoloplasty (ESGAP) at the age of 30 months together with hard palate repair was introduced in Ljubljana cleft centre in 2004. Before that ESGAP was not performed in UCLP patients.

Aims: The aim of this study was to evaluate the growth and development of craniofacial complex at the age of 10 years in complete UCLP patients where ESGAP was carried out and to compare them with complete UCLP patients where ESGAP was not performed.

Methods: 13 consecutive complete UCLP patients without ESGAP (mean age 9.77±0.84 years) and 16 consecutive patients with complete UCLP where ESGAP was performed (mean age 9.96±2.13 years) were included into the study. The analysis of sagittal and vertical relationships was performed on lateral cephalograms. The data was analysed using unpaired Student’s t-test.

Results: The ANB values was significantly decreased in ESGAP group compared to non ESGAP group (1.5°±3.3° in ESGAP; 4.4°±22° in non ESGAP; p=0.009). The Witts value was less in ESGAP group but the difference in comparison to non ESGAP group was not significant (1.1 mm±2.0 in ESGAP; -0.6 mm±2.8 in ESGAP group; p=0.06). In analysis of vertical relationships of maxilla and mandible there was a significant difference in the facial axis values (85.5°±2.1° in ESGAP; 88.4°±1.8° in non ESGAP; p=0.01). Other parameters showed less vertical parameters in ESGAP group, but the differences were not significant.

Summary/Conclusion: According to the results there was less growth of maxilla in sagittal plane observed in ESGAP group at the age of 10 years. The growth in vertical plane did not differ significantly in ESGAP group compared to non ESGAP group.
Secondary alveolar bone grafting performed in different age groups: a comparative analysis of subsequent craniofacial development
A Brudnicki, E Sawicka, P Fudalej

Maxillo-facial Surgery Unit at the Department of Surgery of Children and Adolescents, Department of Surgery of Children and Adolescents, INSTITUTE OF MOTHER AND CHILD IN WARSAW, Warsaw, Poland, Department of Orthodontics and Dentofacial Orthopedics, School of Dental Medicine, School of Dental Medicine, University of Bern, Bern, Switzerland

Background: Currently, bone grafting is recognized as the integral part of contemporary surgical protocols when treating cleft defects of the alveolus but its optimal timing is still debated. A recent trend to preform bone grafting in younger and younger age urges evaluation of the potential negative influence of early bone grafting on craniofacial development, since the surgical techniques of well documented primary bone grafting – popular in the last century, and early secondary bone grafting are different and their results cannot be compared unambiguously in regard to timing of these procedures.

Aims: To evaluate the influence of secondary alveolar bone graft timing on sagittal maxillary development of patients suffering from unilateral cleft lip and palate (UCLP), as a single variable, in relation to the non-grafted analogous group of patients with UCLP and population norm.

Methods: Main outcome measures were medical documentation and cephalometric assessment around 10 years of age. The material consisted of the non-syndromic patients born with complete UCLP who were consecutively operated on using single-stage primary cleft repair at one center in a time span of 10 years. The study group, which included 128 patients, was divided into four age-related groups established according to patients’ age at bone grafting: <3, 3-5, 6-8, 8>. Additionally two groups were established for comparisons: a group of 39 patients with UCLP who had not had bone grafting but otherwise treated according to the same method of primary UCLP repair and a control group of 56 non-cleft children.

Results: The following characteristics of the study group were established: mean age at the primary cleft repair was 8.1 months (SD 1.4), age at cephalometric evaluation was 10.0 years (SD 0.8) while age at bone grafting varied from 1.4 to 11.5 years. The comparison between the cleft groups revealed a lack of growth differences in the vertical dimension and the tendency to more pronounced anterior maxillary development inhibition in earlier bone grafting groups. The correlations between timing of bone grafting and cephalometric measurement values calculated using Pearson’s correlation coefficients were at very low level. The correlations of A-N Perp distance or ANB angle did not even reached the statistical significance whereas the most relevant measurement appeared to be Co-A distance (r = 0.254; p = 0.001977). The comparisons of the age subgroups by ANOVA and Tukey’s HSD test and the age subgroups with the non-cleft group and the non-grafted group using t-test showed that maxillary restriction became significant when bone grafting was performed before three years of age, therefore bone grafting before that age should be avoided.

Summary/Conclusion: The study indicated the relationship between bone graft timing and sagittal maxillary development inhibition, existing however, on a verge of statistical significance. The obtained results imply that in case of a highly effective method of primary cleft repair the potentially negative influence of early bone grafting in clinical practice loses its visibility. This is in contrast to what was previously assumed, based on the studies describing devastating effect of primary bone grafting.
OS 13.8
A review of 30 years of alveolar bone grafting in the mixed dentition using a standardized protocol in western australia.
D Gillett1, P KERBIN2, W Nicholls3, M WALTERS4
1Paediatric Plastic Surgery, Perth Children's Hospital, Perth, Australia, 2Faculty of Dentistry, Université de Nantes, Nantes, France, 3Dentistry, 4Plastic Surgery, Perth Children's Hospital, Perth, Australia

Background: The Cleft Lip and Palate Clinic was established in Perth, Australia in 1955. We developed an alveolar bone graft protocol in 1982 and have followed it since with minimal changes.

Aims: Outcomes for the western Australian Alveolar Bone Grafting (ABG) protocol, established in 1982, are reported and compared to previously published outcomes from our unit and other published reports.

Methods: A descriptive, retrospective cohort study of ABG outcomes at a tertiary referral cleft centre. Records of all ABGs between 2002 and 2014 were reviewed (223 grafts). Three-year post-operative periapical radiographs were evaluated using the Bergland, Kindelan and Standard Way to Assess Graft (SWAG) scores by an external rater. Incomplete records, a syndromic diagnosis or primary surgery performed elsewhere resulted in 123 grafts being excluded leaving 100 grafts for assessment. The distribution of scores was compared to our previous studies and international reports. We also tested for any impact on the outcome of grafts based on: cleft type, laterality, timing for incisor or canine eruption and level of surgeon experience.

Results: 95.6% of grafts were considered “successful” by Bergland scores and 96% by Kindelan scores. 89% of grafts were “very good” based on SWAG score. No significant differences could be detected in outcomes based on timing, cleft type or laterality. Surgeon experience was found to have a significant impact (p

Summary/Conclusion: The Western Australian ABG protocol has consistently achieved very high success rate (96%) for over 30 years despite multiple staff changes. Surgeon training and experience were significant in achieving these outcomes.
Evaluation of dental arch relationships in 5-year-old children with unilateral cleft lip and palate after neonatal cleft lip repair

M Rousi¹², A Brysova¹, J Vokurkova²³, O Koskova²³
¹Clinic of Dentistry, St. Anne's Faculty Hospital, Pekarska 53, 656 91, Brno, Czech Republic, ²Faculty of Medicine, Masaryk University, Brno, Czech Republic, ³Department of Pediatric Plastic Surgery - Pediatric Surgery, Orthopedics and Traumatology, University Hospital Brno, Cernopolni 9, 613 00, Brno, Czech Republic

Background: Clefts are one of the most common craniofacial anomalies. Treatment of children with cleft starts soon after birth and continues to adulthood. The first step of treatment of unilateral cleft lip and palate (UCLP) is the surgery of the lip and later of the palate which seem to be important for the future growth of the maxilla.

Aims: The purpose of this study was to evaluate the dental arch relationships of 5-year-old children with nonsyndromic unilateral cleft lip and palate (UCLP) operated in the department of Pediatric Plastic Surgery, University Hospital Brno. For the assessment, 5-year-old index according to Attack was used. The results were compared with data published from other cleft centers.

Methods: Dental casts of 46 patients with UCLP born between 2009 and 2013 were assessed at the age of five years. All patients were operated by one surgeon and with the same protocol (neonatal cleft lip repair and one-stage palatal closure from 7-12 months of age). None of the patients had any active orthodontic treatment. Outcomes of dental arch relationships were judged and categorised by applying the 5-year-old index. Evaluation of the index was done by three experienced orthodontic specialists of the Clinic of Dentistry, St. Anne's Faculty Hospital Brno twice at different times.

Results: The mean age of the patients was 5.3 years old. 35 patients were boys(72.9%). Unilateral cleft lip and palate affected left side in 70% of the children in the study. The mean score of 5-year-old index was 2.42. Standard deviation was 1.04 and median value 2.0. Of all the patients, 57% were rated as belonging to group 1 or 2 (good result), 24% to group 3 (fair result) and 19% to group 4 or 5 (poor result). The interrater agreement was very good, represented by kappa value of 0.873. These results were compared with other data published in databases such as Pubmed and Web of Science.

Summary/Conclusion: The 5-year-old index is a reliable assessment of the primary outcome in unilateral cleft lip and palate patients. The results from our study showed that neonatal cleft lip repair and one-stage palatal repair can lead to a satisfying maxillary growth and interdental relationships, and can be compared with the results of the centres where the surgical protocol was different.
Compaction force appliance on alveolar cleft bone graft
C Dissaux¹, D Wagner², D George³, Y Remond³
¹Maxillofacial and plastic surgery, ²Orthodontic department, Strasbourg University Hospital, ³ICube Laboratory, University of Strasbourg, CNRS, Strasbourg, France

Background: Alveolar cleft bone grafting is now widely accepted as one step of cleft surgical treatment. The peculiarity of this graft stands in the particular geometry of the alveolar cleft: bone is placed in between two cortical surfaces. Several factors could influence alveolar bone graft results such as age of the child at operation, dental environment, origin of bone graft, vascularization density, technical precisions such as compaction force at the time of the bone graft placement, growth factors adjunction.

Aims: The objective of this research project is to use modeling based on in vitro and experimental animal models to dispense with experimentation on children. The model aims to first get a better comprehension of graft integration and second to simulate graft results according to the influence of different parameters.

Methods: First step of this project includes in vitro and in vivo analysis to establish the effect of mechanical compaction on bone graft. Graft features (Young modulus, cellular vitality and density) are observed relying on magnitude of the applied force (0 to 50N). Cancellous human femoral and iliac bones are used. Micro-CT scanning and CFUs culture are performed.

Results: Micro-CT scan gives a precise characterization of the structure of the graft depending of the applied compaction forces 0, 5, 20 and 50N. The number of CFUs (Colony Forming Units) was higher when a compaction force is applied. This number is even more important at 50N than at 20N, however a flattening of the CFUs number curve is observed. These data are integrated into a biomechanical model of the human cleft bone graft.

Summary/Conclusion: This biomechanical model tends to better understand the bone remodeling influenced by compaction forces in the alveolar cleft particular environment, with the ultimate objective to guide the surgical procedure.
Craniosynostosis Surgery Part One

**OS 14.3**
The impact of surgically treated isolated craniosynostoses on dental age and morphology in panoramic radiographs
S Leinonen¹, J Leikola², A Heliövaara²
¹Department of Orthodontics, University of Helsinki, Faculty of Medicine, ²Cleft Palate and Craniofacial Center, Department of Plastic Surgery, Helsinki University Hospital, Helsinki, Finland

**Background:** Craniosynostoses are defined as a premature fusion of one or more of the calvarial sutures with reported incidence rate of 1:2000-2500. Most often craniosynostoses are isolated and non-syndromic, but they do appear in conjunction with different syndromes. Little is known about the dentition of the children with isolated craniosynostoses.

**Aims:** The aim of this retrospective cross-sectional study is to evaluate the dental ages, dental morphology, and mandibular condylar morphology from dental panoramic radiographs of children with surgically operated isolated non-syndromic craniosynostoses.

**Methods:** The study population consists of patients treated in the Cleft Palate and Craniofacial Centre in Helsinki University Hospital and it is divided into three groups: patients with sagittal synostosis (n=97, out of which 19 were girls), coronal synostosis (n=23, 15 girls) and metopic synostosis (n=18, 2 girls). In each of the three groups, the dental panoramic radiographs were taken when the patients were on average 8.19, 8.21, and 8.34 years old, respectively. The surgical methods used to treat the mentioned synostoses included cranial vault expansion, strip-craniectomy, pi-plasty, and fronto-orbital advancement. For dental age assessment we used the method developed by Demirjian et al. (1973), and as a control group we used dental age tables developed for the Finnish population by Nyström et al. (2004). Existing radiologists' statements of the radiographs, if available, were used to assess dental morphology and condylar morphology.

**Results:** The dental ages in different groups were on average 0.36, 0.51, and 0.49 years ahead of the Finnish normative values, respectively. In the sagittal synostosis group, 10 (10%) children had missing permanent teeth. The lower left second premolar on was missing in eight patients, and the lower right second premolar was missing in five patients. One patient had a missing upper right lateral incisor. Taurodontic teeth were found in two patients. In the coronal synostosis group, one patient lacked the upper left lateral incisor, and in the metopic synostosis group, one patient lacked the upper right second molar. Interestingly, in the metopic group, two patients (11%) had a geminated tooth (an upper- and a lower lateral incisor). No condylar changes were found in any of the groups.

**Summary/Conclusion:** Dental characteristics in isolated, non-syndromic craniosynostoses is a field not comprehensively studied. More research is needed to lighten the burden of care in craniosynostosis patients through better treatment planning.
Intracranial pressure patterns in children with craniosynostosis utilizing optical coherence tomography

J Swanson11, L Lin1, I Hoppe1, S Bartlett1, S Lang1, G Heuer1, W Xu2, G Liu1, T Aleman2, G Ying2, J Taylor1
1Children’s Hospital of Philadelphia, 2University of Pennsylvania, Philadelphia, United States

Background: Better understanding the incidence and patterns of elevated intracranial pressure (ICP) in patients with craniosynostosis may facilitate more timely intervention to alter neurocognitive outcomes. Spectral-domain optical coherence tomography (OCT) of the retina can non-invasively diagnose elevated ICP, and has demonstrated high sensitivity and specificity among patients with craniosynostosis.

Aims: This study sought to utilize OCT to characterize patterns of elevated ICP among patients with craniosynostosis.

Methods: Quantitative retinal parameters were prospectively assessed in both eyes of patients with craniosynostosis using spectral-domain OCT. Based on retinal OCT thresholds associated with elevated ICP (>15mmHg), subjects were assigned an OCT diagnosis of elevated or non-elevated ICP which was analyzed relative to clinical characteristics and craniosynostosis patterns.

Results: 80 subjects (aged 0.2-18 years) with craniosynostosis were enrolled; among these, 67 (84%) were nonsyndromic. OCT evaluation was performed at initial vault expansion in 56 (70%) patients. Among this subset, 27 (48%) patients had peri-papillary changes suggestive of elevated ICP, reflecting a 44% incidence in nonsyndromic and 83% in syndromic patients. The median age at initial vault expansion was higher among those with elevated ICP (11.1 months) than those without (7.8 months; p=0.04). Multi-suture synostosis was associated with changes consistent with elevated ICP in 9 (75%) patients compared to 18 (41%) with single suture synostosis (p=0.05).

Summary/Conclusion: OCT of the retina produces a potentially a sensitive indicator of ICP in craniosynostosis patients. Elevated ICP may be associated with number of involved sutures and older patient presentation, and refining appropriate “cut-offs” will be important as the technology becomes more widespread.
In search of a single standardised system for reporting complications in craniofacial surgery: a comparison of three different classifications

M Söfteland¹, A Paganini¹, S Fischer¹, D Kölby¹, E Hansson¹, J O’Hara², G Maltese¹, P Tarnow¹, L Kölby¹
¹Plastic surgery, Sahlgrenska University, Gothenburg, Sweden, ²Plastic surgery, Great Ormond Street, London, United Kingdom

Background: Comparing complication rates between craniofacial centres is difficult due to the lack of criteria regarding what adverse events should be defined as complications and how they should be classified in craniofacial surgery (CF). Several well-designed classifications exist in the surgical literature to evaluate complications; however, there has been no gold standard established in CF surgery across units.

Aims: The aim of this study was to determine the effectiveness of three different complication classifications in craniofacial surgery and perform a comparative analysis.

Methods: Between 2006 and 2015, a total of 1023 consecutive craniofacial procedures in 641 patients were identified. A systematic review of peri and postoperative complications was carried out using three different classifications (Clavien Dindo, Leeds and Oxford) for comparison.

Results: The Clavien Dindo captured 74 complications in 74 procedures (7.2%) whereas the Leeds and Oxford scales captured 163 complications in 134 procedures (13.1%) and 85 complications in 83 procedures (8.1%), respectively.

Summary/Conclusion: In summary, Clavien-Dindo classification appears unsuitable for paediatric CF surgery, whereas the Leeds classification captures both perioperative and postoperative complications, systematically. The Oxford classification highlights relevant complications within CF surgery and provides a broad overview suitable for auditing purposes. Therefore, depending on how one wishes to tabulate complications, and for what purpose, both the Leeds and the Oxford classification are applicable for CF surgery.
OS 14.6
Evaluating the utility of routine computed tomography scans after cranial vault reconstruction in children with craniosynostosis
C Ahammout1, F Perez2, C Birgfeld3, C Heike4
1Erasmus University, Rotterdam, Netherlands, 2Radiology, 3Plastic surgery, 4Pediatrics, University of Washington, Seattle, United States

Background: Post-operative computed tomography (CT) scans are often obtained in patients with craniosynostosis after cranial vault reconstruction to evaluate surgical results and assess for complications such as an intracranial hemorrhage. However, the benefits of these scans must be weighed against the risks of the cumulative ionizing radiation exposure in these children.

Aims: To evaluate the frequency with which clinically relevant findings are identified on routine post-operative computed tomography scans in patients with craniosynostosis after cranial vault reconstruction.

Methods: We conducted a retrospective study of all post-operative CT scan reports for all patients with craniosynostosis who underwent cranial vault reconstruction surgery between November 2009 and September 2018 at our large tertiary pediatric hospital. We categorized common CT findings and the reporting radiologists’ assessment of whether the post-operative CT findings were typical, atypical or indeterminate. All indeterminate and atypical findings were reviewed by a pediatric neuroradiologist. We reviewed medical charts to determine clinical outcomes of patients for whom an abnormal post-operative CT finding was confirmed.

Results: Post-operative reports for 548 surgeries in 506 participants, 39 of whom had more than one surgery, were included in the study. Most of the surgeries were performed in children under one year of age (n=429, 78%), and most of patients were male (n=326, 64%) and had had single suture craniosynostosis (> 90%). Most participants had an isolated form of craniosynostosis (n=416, 82%). Among those with single suture synostosis, sagittal (n=228, 45%), metopic (n=88, 17%) and coronal (n=59, 12%) were the most common sutures involved. Ninety patients had multiple congenital anomalies (n=33, 7%) or syndromic (n=57, 11%) forms of craniosynostosis. The posterior vault modified PI was the most common surgery performed (n=256, 47%), followed by frontal orbital advancement (n=213, 39%) and endoscopic strip craniectomy (n=41, 8%). Atypical CT findings were described in 9% (n=52) of the CT reports. Our pediatric neuroradiologist confirmed abnormal findings in 69% of these reports (n=36/52). Atypical findings included: extra-axial hemorrhages (n=21, 58%), bone abnormalities (n=5, 14%), brain injuries (n=4, 11%), vascular injuries (n=3, 8%), and increased ventricular sizes/hydrocephalus (n=2, 6%). Most of the children for whom an abnormal finding was identified on CT (n=19, 53%) did not require an intervention. However, 39% (n=14) of the cases underwent additional observation and three (8%) cases required further surgical intervention (n=1) or anti-thrombolytic management (n=2). The CT reports that led to an intervention described dural venous sinus injury, incomplete osteotomy, or a subdural extra-axial hemorrhage.

Summary/Conclusion: Although it is common to obtain a postoperative CT scan after the cranial vault reconstruction, our data showed low rates of abnormal findings. The CT scans demonstrated an abnormal post-operative in 6.6% of cases, which led to an intervention in only 3 cases. The decision of obtaining a CT-scan should be considered by balancing the risks of radiation exposure and costs with the potential benefits of identifying clinically relevant post-operative findings.
Is early repair really necessary: comparative evaluation of the effect of palate repair timing on speech results with objective parameters

M Kara¹, M Çalış², I Kara², Ö İncebay², M Kulak Kayıkçı², R Günaydın³, F Özgür¹
¹Plastic, Reconstructive and Aesthetic Surgery, Hacettepe University Faculty of Medicine, ²Speech and Language Therapy, Hacettepe University, ³Ear, Nose and Throat, Hacettepe University Faculty of Medicine, Ankara, Turkey

Background: Optimal timing of cleft palate repair has been one of the major topics of discussion among the cleft literature. There are many studies questioning the right time for cleft palate repair from the speech, postoperative complication and maxillary growth perspective whereas there are limited studies evaluating the results using objective parameters.

Aims: The main of this study was to evaluate the effect of timing of cleft palate repair on speech results, postoperative complication profile and necessity for secondary procedures using objective evaluation tools.

Methods: 90 patients divided into three groups according to their age of repair. Patients in group I (n = 31) operated before 12 months of age, group II (n = 30) operated between 12-18 months, and group III (n = 29) operated after 18 months of age. The demographic and perioperative data of the patients included in the study such as gender, cleft type, repair technique, fistula ratio, additional surgical intervention due to velopharyngeal insufficiency were recorded. Speech evaluation was performed with objective parameters. Velopharyngeal closure was evaluated anatomically by nasopharyngoscopy, and the nasalance values were recorded and evaluated objectively by nasometer.

Results: Of the 90 patients 46 were boys (51.1%) and 44 were girls (48.90). The highest nasalance scores were recorded in group 3 in all syllables and counting scores. In the statistical analysis of the nasal scores, there was no statistically significant difference between group 1 and group 2. However, there was a statistically significant difference observed between these two groups and group 3. When the function of the velopharyngeal unit was evaluated, the absence of velopharyngeal closure defining the non-functional velopharyngeal unit was found to be lowest (12.9%) in group 1, 16.7% in group 2 and the highest (48.3%) in group 3. There was no statistically significant difference between group 1 and group 2 as a result of intergroup analysis of closure rate variable, whereas it was seen that there was a statistically significant relationship between these two groups and group 3 in the "absence of velopharyngeal closure" variable (p=0.022). When the rates of postoperative fistula development and secondary surgical intervention were examined according to the groups, it was found the lowest in group 1 and the highest in group 3.

Summary/Conclusion: In respect to the objective evaluation using nasometric and nasopharyngoscopic evaluation in means of operation timing, the most unfavorable results were observed in the late cleft palate repair group. It appears that optimal timing of cleft palate repair is an important determinant in obtaining optimal speech results. From this perspective running further prospective studies using objective evaluation tools to confirm the postoperative results would clarify an important issue in cleft practice.
Cephalometric and ct evaluation of syndromic craniosynostosis after le fort iii halo distraction: a long term follow up

F Mazzoleni

1san gerardo hospital, Monza, Italy

**Background:** Midface distraction osteogenesis in craniofacial synostosis has largely been described. However, very few cephalometric and computed tomography (CT) long-term follow-up studies are present in current literature.

**Aims:** Evaluation of possibility in advancement of the midface through Le Fort III and RED and of post-surgical stability.

**Methods:** A total of 45 consecutive patients affected by craniofacialsynostosis subjected to Le Fort III and rigid external distraction (RED) were examined. All patients had preoperative cephalometric records, immediately postoperative and 6e12 months postoperative.

**Results:** Excellent post-surgical stability was recorded. Short- and long-term CT data demonstrated excellent ossification at the osteotomy sites post distraction osteogenesis. In the growing patients, surface resorption in the zygomatic-temporal and in the subspinal area (p < 0.05) was observed in the long-term follow-up, as well as a mild increment of the corrected exorbitism (p <0.05), as only appositional and no sutural growth occurs post Le Fort III, whereby orbital volume does not increase after surgery.

**Summary/Conclusion:** Significant advancement of the midface can be achieved and maintained through Le Fort III and RED. In the long term, in growing patients, in general a class III malocclusion does not re-occur, but physiological remodelling processes at the maxillary-zygomatic level, not coupled with sutural growth, tend to mildly re-express the original midfacial phenotype.
Photometric study of cranio-facial symmetry in hemicoronal single suture synostosis treated with surgical fronto-orbital remodeling

F Mazzoleni

1san gerardo hospital, Monza, Italy

Background: Unilateral coronal synostosis is the third most common type of single suture craniosynostosis. Although there is no scientific evidence on the effects of cranioplasty on neurocognitive development, a good and lasting aesthetic result can improve patients quality of life.

Aims: Evaluation of frontal vault symmetry and progressive facial symmetrization in a cohort of patients with hemicoronal single suture synostosis treated with a standardized cranioplasty and rigid fixation

Methods: Fifty-nine patients with hemicoronal synostosis operated between 1999 and 2018 were reviewed retrospectively. Pre, immediately postoperative and yearly photographs were taken with the same head position and projection. A photogrammetric method was applied to quantify the pre and postoperative contour changes. The anterior skull hemispheres were traced, divided into two equal parts and the areas were compared. Angular measurements obtained by the intersection of the interpupillary line and the glabella perpendicular vertical line were calculated.

Results: The average advancement on the affected side was 18 mm. The angular measurements documented the frontal symmetry and the progressive improvement of facial symmetry

Summary/Conclusion: Cranioplasty with rigid fixation achieved a good and stable correction of anterior plagiocephaly and a subsequent symmetrical facial growth.
Practical Applications of the Buccal Flap Philosophy

OS 15.1
Practical applications of the buccal flap philosophy
R Mann

Abstract Content: This presentation will be a video, photograph, and diagram based teaching session designed to help delegates get comfortable using the Buccal Flap Philosophy based techniques when reconstructing the primary cleft palate. The course will cover how to raise a Buccal Flap and when and how it can be used. Other flap options will be described. Individual cases, will illustrate how to develop the initial reconstructive treatment plan that is integrated with long term planning to maximize each individuals natural growth potential. The delegates will be able to see how this approach can achieve both successful speech and normal facial appearance. There will be ample time for discussion and questions. Dr Mann will share his experiences with both success and failures. What post op issues you will encounter and how to manage them will be outlined. For more on complications seen, you may wish to attend the presentation on the Double Opposing Z Plasty +/- Buccal Flap Repair Of Cleft Palate: A 29 Year Review Of The Complications, And Presenting A Fistula Grading Scale, shortly following this presentation. An introduction to using the Buccal Flap Philosophy to treat VPD will presented as time permits focusing on how to reestablish normal speech resonance while maintaining normal velar closure. Friday’s session on Speech Enhancing Surgery will continue the practical use of the Buccal Flap Approach for the Treatment of VPD, specifically focusing on what to do when your palate lengthening is not enough.
Early active presurgical orthopedics and gingivoperiosteoplasty protocols opposed to a protocol giving the priority to the nose. The current dilemma over cleft treatment

OS 15.2
Early active presurgical orthopedics protocols opposed to a protocol giving the priority to the nose. The current dilemma over cleft treatment
J Talmant¹
¹Clinique Jules Verne, Nantes, France

Abstract Content: “Boy, Plastic surgery is not easy...” said Harold Gillies. He added: “When planing the restauration, function should be the first consideration.” Such concern in cleft treatment should lead to save the space of the missing piece of the lateral incisive field which supports the piriform orifice, the nasal valve and the incisive arch. Therefore, to start repair of a cleft new born with early active presurgical orthopedics to settle the contact of both gingiva, whatever the procedure might be, is a mistake. It means that this space disappears with the bony fusion of the alveolar arch, leaving severe consequences. During the same period we have explored a totally opposite hypothesis, giving the priority to the establishment of nasal ventilation that involves not only primary nasal cartilage repositioning with large dissection and prevention of retraction, but altogether the reconstruction of the missing piece. Nasal ventilation only allows correct resting oral cavity postures necessary for a good and balance facial growth. Of course, our 21 year follow up has confirmed that cleft treatment is not simple: we have had to improve late deformities. The lesson drawn from this experience is that secondary repositioning of the normal structures with the same technique and precaution than in primary surgery is the solution. The procedures developed for this cleft nasal surgery have great potential for any complex reconstructive nasal surgery. Now it is time to take a stand on a rational cleft treatment.
My life with Treacher Collins syndrome and its connection to my research in craniofacial morphology

OS 15.3
My life with treacher collins syndrome and its connection to my research in craniofacial morphology

F Smith

Department of Craniofacial Biology, University of Colorado Denver Anschutz Medical Campus, Aurora, United States

My unique life experience, with Treacher Collins syndrome, has given me the opportunity to engage in scientific research in craniofacial embryology and anomalies as well as public outreach for craniofacial differences awareness. My research, in the UK, USA, and Canada, has focused on the genetics and morphology of embryonic craniofacial development. In my earliest craniofacial genetics research at King's College London, I have traced the expression of a novel gene in mouse embryos, and examined regulation of craniofacial myogenesis in chick embryos. For my PhD dissertation, I examined the role of early embryonic hypoxia in craniofacial malformations in chick embryos. My subsequent research has focused on craniofacial 3-D imaging and morphometrics, upper airway volumetrics in Treacher Collins syndrome, and genomic analysis of craniofacial gene expression among embryonic mouse tissues. Outside the laboratory, I have been involved in a global campaign of public outreach for awareness and acceptance of craniofacial differences and the need for continuing research, through public speaking and lecturing on my background, expertise, and research.
Abstract Content: The author has been using Furlow palatoplasty in various cleft conditions since 1988. The technique has been used in:

1. Primary cleft palate repair
   The technique can be applied in different types of cleft palate. Double opposing Z-plasty is used for soft palate repair. Relaxing incision or raising the mucoperiosteal flap can be added in hard palate closure. The overall incidence of secondary surgery is 4% in author’s experience.

2. Velopharyngeal insufficiency after primary repair
   The author started to use Furlow for the management of velopharyngeal insufficiency from 1988. The technique can achieve a successful rate around 80% despite patient selection. The indication for patient selection is based on nasendoscopic examination with closing ratio better than 0.7.

3. Submucous cleft palate
   The patient selection criteria for submucous cleft palate is the same, i.e., closing ratio better than 0.7. Eighty percentage of the patients can get improvement.

4. Furlow with pharyngeal flap
   Combined Furlow palatoplasty with pharyngeal flap is used for patients with closing ratio less than 0.7. The width of the flap can be narrowed to reduce the risk of obstructive sleep apnea.

5. Revision of previous pharyngeal flap with obstructive sleep apnea
   For patients with previous pharyngeal flap and obstructive sleep apnea, the pharyngeal flap is divided for enlarging airway and a Furlow palatoplasty is performed on soft palate for better velopharyngeal closure. It can achieve a better result than flap division alone or enlarging of the portals.
The double opposing z plasty +/- buccal flap repair of cleft palate: a 29 year review of the complications, and presenting a new fistula grading scale

R Mann

1Pediatric Plastic Surgery, Helen DeVos Childrens Hospital, Grand Rapids, Michigan, United States

Background: The Double Opposing Z-plasty +/- Buccal Flap Approach (DOZP +/- BFA) is a useful alternative to traditional palate repairs. Identical excellent speech outcomes are demonstrated regardless of the patients cleft width or classification. Palate surgery complications vary dramatically in their complexity. The complex, nightmare fistulas are difficult to treat, requiring multiple operations and extensive distant flaps. Nightmare fistulas can have significant negative impact on speech. Smaller nuisance type fistulas are easy to fix and have little impact on speech. The widely varying impact between fistulas types makes it very difficult to make sense of reported fistula rates.

Aims: The purpose of this presentation is present the long term complication results of the (DOZP +/- BFA). Outline the types of fistulas occurred and how they were managed. To present a new usable way to describe fistulas that may help caregivers have a better understanding of the impact fistulas can have and to make practical sense of what a complication rate really means for the patient.

Methods: Retrospective review of 505 patients over 29 years. All patients treated with the (DOZP +/- BFA). Outcomes included cleft width, Veau class. Fistula data included incidence, etiology, size and location. Review of all treatments required to treat the fistula. To assess the long term impact, all patients received speech assessments, including nasal resonance scores and a review of any secondary speech surgeries.

Results: Overall complications 30/505 pts. Fistula rate = 5.5 %. Fistulas > 2mm = only 2.5%. Non syndromic, Non Pierre Robin, Fistula rate = 4.8%, fistulas > 2mm = only 1.8%. The overall patient impact: Success closing the fistula, how many treatments are required for closure? Only one patient required two repairs to achieve stable closure, and zero patients requiring addition outside flaps for closure. All fistulas were closed with local tissue rearrangement. More secondary speech surgeries required for patients with fistulas than for patients without fistulas. However the final speech resonance scores for patients with fistulas were similar to the scores achieved in patients without fistulas

Summary/Conclusion: The (DOZP +/- BFA) is a safe approach with few complications. The types of fistulas seen over 29 years, indicates that the majority of the fistula seen are of the small nuisance variety and are easily managed with low impact on the patients. The fact that zero nightmare fistulas occurred shows the patients are at low risk of having to go through the dangers of multiple surgeries and the fistulas have limited effect on long term speech outcomes. Reporting the incidence of the nuisance type and the nightmare type fistulas is extremely helpful to better differentiate which surgical procedures in fact have the best overall outcome.
Outcomes of two versus one stage for unilateral cleft lip and palate repair
R Pereira¹, D do Vale², E Melo², N Siqueira², N Alonso³
¹CADEFI, IMIP, Recife- PE, ²CADEFI, IMIP, Recife-Pe, ³Craniofacial Surgery Department, FMUSP, São Paulo, Brazil

Background: Adequate maxillofacial growth is one of the main goals in unilateral cleft lip and palate (UCLP) treatment. Few randomized studies were published comparing the maxillomandibular relationship at 5 years of age after two treatment protocols: one with single stage palatoplasty and another with late hard palate closure (LHPC).

Aims: We hypothesized that the LHPC protocol provides better dentofacial growth

Methods: A randomized controlled trial was performed, with surgeries performed by a single surgeon and blind evaluation to evaluate maxillomandibular relationships in both groups. The intervention group (GI) underwent LHPC between 3 and 4 years; the control group (CG) underwent complete palatoplasty between 9 and 15 months. We evaluated the prevalence rates of oronasal fistula. The dental arch relation was evaluated through blind panels using the Five Year's Old Index (FYOI) index. The Kappa concordance test was performed to evaluate the panel's inter- and intra-rater reliability. Tests were statistically tested with Student's t-test and chi-square test

Results: Sixty-four patients were the study sample. The incidence of oronasal fistulas was 9.4% (GI) and 6.7% (CG). Sixty-two models with mean age of 55 months were available for analysis. Good reliability was found intra and inter-evaluators (0.73-0.93 and 0.60-0.94, respectively). Mean index scores ranging from 2.04 (GI) to 2.76 (GC) were significantly different (p = 0.007). Significant differences between groups (p = 0.006) were found in scores 1 and 2 in the GI (74%) and GC (52%) groups. When the distributions were compared by the median, a difference (p = 0.024) was found in the score, 1 between the GI (31.2%) and GC (3.3%) groups

Summary/Conclusion: Although the LHPC protocol presented better outcomes related to dentofacial growth, with a statistically significant difference, other protocols should be tested and the findings verified by other researchers.
**OS 16.5**

**A cadaveric anatomical study of the buccal fat pad flap and implications for cleft surgery**

K Echlin¹,², H Whitehouse³, M Schwaiger¹, R Nicholas³, N Fallico¹, D Atherton¹

¹South Thames Cleft Service, Guys and St Thomas' Hospital, London, ²Birmingham Childrens Hospital, Birmingham, ³Dept of Plastic Surgery, Guys and St Thomas' Hospital, London, United Kingdom

**Background:** The buccal fat pad flap is a useful tool for the cleft surgeon both in fistula repair and in primary surgery. Most of the literature has tended to concentrate on its role as a part of aesthetic surgery and as a tool for dento alveolar surgeons however. We wanted to examine the anatomical implications for cleft surgeons in particular.

**Aims:** We aimed to perform a series of cadaveric anatomical dissections on fresh frozen specimens. A standard raise of the buccal fat pad flap would be performed as if to augment a standard cleft palate surgery, and to assess its potential reach in terms of assessing its suitability for fistula closure. We also wished to make an assessment of its donor site implications by examining how much of the fat pad was utilised in the surgery.

**Methods:** 30 buccal fat pad flaps were raised from fresh frozen cadavers. Measurements were taken to assess the arc of rotation from the base of the pedicle behind the alveolus, to its maximum reach, with no tension, across the midline; posteriorly towards the uvula and anteriorly towards the incisors. Flaps were then transected at their base, measured and weighed. Finally, an external facial dissection was carried out, removing the external tissue of the face and cheek to assess the donor site. The remaining buccal fat pad was dissected free, excised and also weighed to determine the proportion of fat pad used in the flap and the amount of tissue retained in the face/cheek.

**Results:** All 30 buccal fat pad flaps reached across the midline without tension and with an average reach beyond the midline of 12mm. 29/30 flaps reached posteriorly to the uvula. All flaps would have covered a small to moderate fistula in the Pittsburgh classification Zone III and posterior zone IV (posterior hard palate). 73% of flaps would reach a mid zone IV palate fistula. 30 % of flaps would cover an anterior zone IV fistula. When comparing the flap weight to the weight of the residual fat pad; on average 36% of the entire fat pad had been used in the surgical procedure. A series of consistent retaining ligaments were identified that appear to limit complete extraction of the fat pad through the intra oral approach which should hopefully protect the surgeon and patient from over resection.

**Summary/Conclusion:** An arc of rotation has been described to show the potential reach of the buccal fat pad which has implications for cleft surgeons. A typical buccal fat pad procedure will use 36% of the mass of the buccal pad, with most of the fat pad remaining in the cheek/face.
Is it possible to predict if the primary palatoplasty can be the only surgical procedure for the good speech outcome in the treatment of submucous cleft palate?

B Offert, E Radkowska, W Piwowar

Maxillo-facial Surgery Unit in the Department of Surgery of Children and Adolescents, Institute of Mother and Child, Warsaw, Poland

**Background:** A range of surgical treatments have been proposed and used for patients with submucous cleft palate (SMCP). It is being discussed whether pharyngeal flap or pharyngoplasty operation should be performed as a primary procedure, as primary palatoplasty seems to be a more physiologic solution. In our clinic pharyngeal flap is a secondary procedure in the treatment of SMCP in the cases when primary palatoplasty is not efficient for good speech outcome.

**Aims:** The aim of this study was to find if there is a significant predictor of speech outcomes following primary palatoplasty surgery for SMCP in patients aged 3 or older. We searched for the factors that can indicate need for choosing pharyngeal flap operation to be performed in our clinic as a primary procedure. Our goal is to reduce the number of operations in order to achieve a correction of velopharyngeal insufficiency and speech improvement.

**Methods:** Out of 600 patients with isolated cleft palate operated on in our centre between 12/2014 and 01/2018 (3 years and 2 months), 75 were diagnosed with SMCP. A consistent group of 30 patients with SMCP was studied to determine specific risk factors that can be predictors of outcome following primary palatoplasty surgery. The age of these patients ranged between 3 to 9 years old. All confirmed to have hypernasal speech before operation. All cases were treated with the same method of palatoplasty performed by 2 experienced surgeons. Factors studied included: type of palatal cleft (classic or occult SMCP), feeding difficulties in the neonatal period, chronic middle ear disease, operative age (the age at the time of operation), adenoidectomy before diagnose of SMCP and 22q11.2 Deletion Syndrome.

**Results:** After palatoplasty 8 patients (27%) persisted with hypernasality postoperatively and required secondary pharyngeal flap operation for correction of residual velopharyngeal insufficiency. 22 patients (73%) improved to normal nasal resonance.

25% of patients (3 out of 12) with the occult SMCP and 28% of patients (5 out of 18) with the classic SMCP persisted with hypernasality postoperatively. 38% of patients (6 out of 16) with feeding difficulties and 33% of patients (5 out of 15) with chronic middle ear disease persisted with hypernasality postoperatively. 27% of patients (6 out of 22) ranged from 3 to 6 years old and 25% of patients (2 out of 8) ranged from 6 to 9 years old persisted with hypernasality postoperatively. 50% of patients (3 out of 6) after adenoidectomy and 50% of patients (2 out of 4) with 22q11.2 Deletion Syndrome persisted with hypernasality postoperatively.

**Summary/Conclusion:** So far we have been unable to determine preoperative characteristics which may help us predict the outcome after palatoplasty. The factors, that correlated most strongly with remaining speech disorders after primary palatoplasty, were adenoidectomy before diagnose of SMCP and 22q11.2 Deletion Syndrome (50% of patients with specific factor). Operative age of patients was not an important predictor of the outcome in our group of patients older than 3 years. We need more studies on a larger number of patients to confirm our results.
Comparative evaluation between two palatoplasty techniques in patients with unilateral cleft lip and palate

K Gigov

Plastic and Craniofacial Department, University Hospital `Sv. Georgy`, Medical University - Plovdiv, Bulgaria

Background: Cleft lip usually has a greater aesthetic and social impact, but cleft palate has the greatest functional impacts. There are several palatoplasty techniques described in the literature, the choice based on several criteria: type of fissure, fissure extension, preference and technical skill of the surgeon. The goals of palatoplasty are to closing the oral and nasal communication, to have a good speech outcome, less fistulas, better results in the maxillary growth.

Aims: The aim of this study was to make a comparative evaluation between two palatoplasty techniques - Veau-Wardill-Kilner and a modified palatoplasty (without lateral releasing incisions) - in regards of the number of fistulas, speech outcome, recovery period, dental arch relationships with 5 years old index and GOSLON Yardstick in Bulgarian children with Unilateral cleft lip and palate (UCLP).

Methods: One hundred patients with UCLP between 4-15 years of age were divided in two groups - Group I 80 patients treated by a modified Veau-Wardill-Kilner technique and Group II - 20 patients without a releasing lateral incisions technique. In both groups a transversal section of the nasal layer was performed. All of them were treated by a team of two surgeons. Both groups had a cleft of the palate limited to 10 mm in the larger part of the cleft and can be equally treated by the two of this techniques of palatoplasty.

Results: In the first group we registered 22 oronasal fistulas (almost 30%) and 5 fistula from the second group which is (25%). Speech was assessed perceptually by the group of four independent experts for the presence of hypernasality, compensatory articulations, audible nasal air emissions, articulation disorders, compensatory articulation and speech intelligibility. No statistically significant differences were found between the two groups. The overall speech intelligibility was good in 79% of all cases. We compare two groups of patients at the same age range. The evaluation of dental arch relationships for the first group, by the 5-years old index has an average score of 3.20. The second group has a score of 2.66. We also evaluate the patients over 10 years by GOSLON Yardstick index and their average score is 3.6. The recovery period in the second group of patients is better.

Summary/Conclusion: The conclusion of this study between the two techniques is: no differences in the speech outcome, the number of fistulas, easy and significantly better recovery period in the second group of patients. The evaluation of dental arch relationships by the 5-years-old index for the second group of cases have a slightly better results. The index evaluates the anteroposterior relationship of maxillary/mandibular dental arches but does not evaluate the collapse of maxillary segments.
**Background:** To facilitate the best approach during cleft palate surgery, children are positioned with hyperextension of the neck according Rose position. Extensive head extension may induce intraoperative cerebral ischaemia if collateral flow is insufficient. To evaluate and monitor the effect of cerebral blood flow on cerebral tissue oxygenation, near-infrared spectroscopy has proved to be a valuable method.

**Aims:** The aim of this study was to evaluate and quantify whether Rose position affects the cerebral tissue oxygenation in children during cleft palate surgery.

**Methods:** This prospective study included children (ASA 1 and 2) under the age of 3 years old who underwent cleft palate repair at the Wilhelmina Children’s Hospital, in the Netherlands. Data were collected for date of birth, cleft type, date of cleft repair and physiological parameters (MAP, saturation, heart rate, expiratory CO2 and O2, temperature and cerebral blood oxygenation) during surgery. The cerebral blood oxygenation was measured with NIRS.

**Results:** A total of 34 children were included for cleft palate surgery during this study. The majority of the population was male (61.8%, n= 21). The mixed model analyses showed a significant drop at time of Rose position of -4.25 (69-74 95% CI; p < 0.001) and -4.39 (69- 74 95% CI; p <0.001). Postoperatively, none of the children displayed any neurological disturbance.

**Summary/Conclusion:** This study suggests that Rose position in children leads to a significant decrease in cerebral oxygenation. Severe cerebral desaturation events during surgery were uncommon and do not seem to be of clinical relevance in ASA 1 and 2 children.
May radical intravelar veloplasty lead to denervation of the levator muscle?
M Krimmel¹, B Hirt², S Reinert¹
¹Department of Oral and Maxillofacial Surgery, ²Department of Anatomy, University Hospital Tübingen, Tübingen, Germany

Background: Intravelar veloplasty was first described by Kriens. Recently the technique was further refined by Sommerlad. The term “radical intravelar veloplasty” was chosen for this approach. The goal of radical intravelar veloplasty is to dissect the levator muscle of the soft palate far laterally in order to obtain a muscle sling that is positioned as far posterior as possible. Thus an optimal muscle function is sought. However, muscle function depends also on intact nerve supply to the palatal muscles.

Aims: The question that arises is therefore, whether such a radical preparation of the muscle may lead to a denervation of the levator palati muscle?

Methods: We dissected the soft palate of 6 formalin conserved cadaver specimens at the Insitute of Anatomy and identified the motor nerve to the levator palate muscle.

Results: The motor nerve, that innervates the levator palati muscle, comes in a bundle of 2 – 3 fibers from the area of the sphonopalatine fissure/ musculotubary canal. It runs parallel to the muscle and then enters the muscle in steps and fan like. The nerve fibers enter the muscle at its posterior border on the oral and palatal side. A connection to the glosspharyngeal nerve could not be established. On the anterior and lateral aspect of the muscle, we could not identify any relevant nerve fibers.

Summary/Conclusion: In the literature the origin of the motor nerve to the levator palati muscle is still unclear and under debate. Our results support the concept that in radical intravelar veloplasty the dissection of the levator palati muscle may proceed far laterally. As long as the posterior border of the muscle is not exposed, a denervation of the muscle is probably not a problem. These findings are in accordance with our own clinical findings.
Micro-structured β-tcp for alveolar cleft reconstruction: a two-center study
Nard G Janssen1, Ruud Schreurs2, Adrianus P de Ruiter1, Hans Christian Sylvester-Jensen* 3, Grethe Blindheim4, Gert J. Meijer2, Ronald Koole1, Hallvard Vindenes5
1Department of Oral and Maxillofacial Surgery, Utrecht University Medical Center, Utrecht, 2Department of Oral and Maxillofacial Surgery, Radboud University Medical Center, Nijmegen, Netherlands, 3Cleft Lip and palate team Bergen, Haukeland University Hospital, 4Radiology, Tannhelsetjenestens kompetansesenter Vest, 5Plastic Surgery, Haukeland University Hospital, Bergen, Norway

Background:
The current standard of care in alveolar cleft repair is timing the procedure in the mixed dentition stage and making use of autologous bone to restore the maxillary defect.

Aims: Using a synthetic bone substitute bypasses the risk of donor site morbidity and reduces the operation time.

Methods: Twenty patients were enrolled prospectively in this study, divided between two centres. Continuity of the alveolar process, recurrence of oronasal fistulas, and eruption of teeth into the repaired cleft were evaluated at 1 year postoperative. Also, cone beam computed tomography scans were analyzed using a volume-based semi-automatic segmentation protocol.

Results: No adverse events were reported. The mean residual bone volume in the repaired cleft at 1 year postoperative was 65%. There was no recurrence of oronasal fistula. Furthermore, 90% of the teeth adjacent to the cleft erupted spontaneously and all patients showed a continuous alveolar process.

Summary/Conclusion: Secondary alveolar grafting using microporous β-TCP can safely be used in the clinical situation. Residual calcified tissue, canine eruption, and complication rates at the recipient site are comparable to those with autologous grafts.
Bone Grafting Part Two

OS 17.2
Replacing the autologous bone graft in cleft lip and palate surgery
Nard G Janssen, MD, DDS, PhD. Department of Oral and Maxillofacial Surgery, Utrecht University Medical Center, the Netherlands

In this presentation, more than ten years of research on replacing the autologous bone graft for alveolar cleft repair procedures is discussed. Nowadays, the cleft lip and palate team of the Wilhelmina Children’s Hospital Utrecht successfully makes use of instructive calcium phosphate scaffolds in order to bypass donor site morbidity after autologous bone grafting procedures. Furthermore, future improvement of bone grafting with calcium phosphate based scaffolds is revealed.
3d bioprinting cartilage for craniofacial reconstruction

A Ujam1, O Gardner1, E Gomez1, N Agabalyan1, E Zucchelli1, N Bulstrode2, P Ferretti1

1Stem cells and Regenerative Medicine, University College London, Great Ormond Street Institute of Child Health., 2Plastic and Reconstructive Surgery, Great Ormond Street Hospital for Children, London, United Kingdom

Background: Patients with cleft lip or craniofacial abnormalities can present with a primary or secondary nasal deformity that often requires difficult septorhinoplasty using costochondral grafts. Patients with microtia will also undergo similar challenging surgery for reconstruction. Common nasal problems requiring cartilage augmentation include an underprojected nasal tip, short columellar, flat nasal bridge and deviated septum. Although rib provides excellent quality and volume of cartilage for septorhinoplasty and microtia reconstruction, it is far from ideal due to significant morbidity at the chest donor site including post-operative pain, risk of infection, risk of pneumothorax and scarring. Alternatives to rib grafting such as silicone or porous polyethylene implants, are associated with high risk of infection and extrusion.

Aims: This work focuses on bioengineering cartilage tissue using autologous human adipose derived stem cells and auricular chondrocytes to generate clinically useful cartilage for nasal and ear reconstruction. Engineered grafts will be fabricated using three dimensional bioprinting technology with cell seeded bioink and a resorbable polycaprolactone (PCL) scaffold to increase mechanical strength. We aim to tissue engineer patient specific and custom-made mature cartilage with similar biomechanical properties to native nasal and auricular cartilage. Ultimately this will supersede current costochondral graft procedures and eliminate the associated morbidity.

Methods: Lipoaspirate and cartilage samples were obtained from paediatric patients undergoing elective surgery under ethical approval. Cells were then isolated and expanded using established protocols. Cells were seeded in a range of bioinks and printed using a pneumatic extrusion-based 3D bioprinter. Simultaneous printing of cell seeded bioink onto a PCL scaffold was achieved using a dual head bioprinting system. Bioprinted constructs were then chondrogenically differentiated and assessed for cartilage formation using staining for common cartilage markers including type I, II, X collagen as well as elastin. Cell survival following 3D bioprinting was also assessed with live/dead assays. Mechanical testing of bioprinted constructs to determine tensile strength was measured using a nanoindenter device.

Results: Cell survival following 3D bioprinting remains high within printed constructs. We demonstrate that both cells lines are suitable for 3D printing and that a high expression of collagen II is achieved in prints as detected by 2-photon fluorescence microscopy and histological staining. This indicates early cartilage formation and maturation of the extracellular matrix (ECM).

We have optimised protocols for simultaneous bioink/PCL 3D printing to allow reproducible custom shaped grafts. Bioink printing protocols were developed to allow homogenous mixing of cells with bioink and to achieve reproducible print patterns.

Summary/Conclusion: Tissue engineering and regenerative medicine aims to replace current surgical techniques that rely on costochondral harvesting and therefore eliminate their associated morbidity. We demonstrate that 3D bioprinting technology utilising autologous human stem cells generate cartilage-like tissue. Future research will focus on enhancing maturation of constructs in order to achieve cartilage that is clinically useful. This forms the focus of our future work.
Development of an osteogenic construct derived from stem cells for treatment of primary cleft palate.

M Perez-Dosal¹, M Velasquillo², J SANCHEZ BETANCOURT³
¹Experimental Research, NATIONAL PEDIATRIC INSTITUTE, ²Biotechonology, Rehabilitation National Institute, ³PORCINE PRODUCTION, AUTONOMA NATIONAL UNIVERSITY, MEXICO CITY, Mexico

Background: The possibility of generating a construct osteogenic created by tissue engineering techniques would allow that cleft palate patients to avoid the complications of the donor area.

Aims: 1. Standardize the technique to produce an osteogenic construct derived from stem cells and 2. Evaluate its effectiveness to achieve bone regeneration in maxillary defects in a growing porcine model.

Methods: It is an experimental design. The project will run from January 2013 to December 2018 in 3 stages: I-Standardization of culture technique, II-Application of an animal model osteogenic construct and III-Surgical treatment of a pilot group of alveolar-cleft patients. This is the report of the first and second stage.

The following were performed: 1. Periosteal biopsy, 2. Isolation of stem cells. 3. Cultivation and cell expansion, 4. Osteogenic differentiation and 6) typing culture. For the second step: In this third stage the osteogenic implant generated by techniques of Tissue Engineering was implanted in a growing experimental model, evaluating its capacity for integration, degree of resorption, quality of bone tissue generated (histological, biochemical, molecular and morphological properties), as well as the safety and adverse effects found in its clinical application. Statistical analysis was performed using descriptive statistics and non parametric tests.

Results: We included 10 pigs: the cultures showed cell viability to 100% and one count of 2.5x10⁶ (±1.14 x10⁶). The isolated cells showed stem cell phenotype (CD73, CD90 and CD34 and CD166 positive and CD45 negative). After being seeded on the scaffold (hydroxyapatite / xanthan / chitosan) showed bone matrix composed of collagen I and X (immunohistochemistry), the matrix is mineralized (von Kossa positive) and expressed alkaline phosphatase, RUNX2 and osteopontin (RT-PCR). All these features are compatible with osteogenic differentiated lines.

For the second step we implanted the osteogenic construct, evaluating its capacity for integration, degree of resorption, quality of bone tissue generated (histological, biochemical, molecular and morphological properties), as well as the safety and adverse effects found in its clinical application. We will present the biomolecular characterization (PCR, immunohistochemical test: cells expressing RUNX2 (osteoblasts), osteopontin (osteoclasts), they produce an extracellular matrix composed of Collagen I and X and alkaline phosphatase, ), histological assays (osteoblasts, osteoid and calcium deposits) and organic compounds and micro elements.

Summary/Conclusion: The osteogenic implant derived from cells adult mesenchymal mother has been shown to be effective in achieving bone regeneration in a standardized porcine experimental model of maxillary bone defects.
Alveolar bone graft: evaluation of the surgical outcomes using cone-beam computed tomography
E Fabbri¹, L Pannuto², L Cercenelli³, B Bortolan³, E Marcelli³, P Morselli¹
¹Bologna University - Sant'Orsola-Malpighi Hospital, ²Sant'Orsola-Malpighi Hospital, ³Bologna University, Bologna, Italy

Background: Alveolar bone graft is the standard of care for alveolar cleft treatment. A quantitative assessment of the result, in term of real bone integration, was difficult using bi-dimensional imaging methods. The introduction of the cone-beam computed tomography (CBCT), with a 3D reconstruction of the defect, allowed an effective evaluation of the surgical outcome.

Aims: We analyse the cleft patients with CBCT before orthodontic treatment, immediately before the surgery and 4-6 months after surgery. Thanks to a process of segmentation and 3D modelling, it is possible to obtain a 3D reconstruction of the anatomical defect that has to be restored and the exact volume of bone missing that we will need during the surgery. Finally, the bone graft reabsorption is evaluated through the comparison between the pre-operative and the post-operative CBCT.

Methods: In the last 2 years, 16 non-syndromic patients with alveolar cleft were treated with alveolar bone graft procedure. All the patients were studied with OPT and CBCT before the orthodontic treatment, pre-operatively and 4-6 months after surgery. With software of segmentation (Dicom to Print) and 3D modelling (MeshMixer) the anatomical defect was reconstructed and the volume of bone needed to be grafted was established. Two different volumes were, therefore, calculated for each patients:

1. The “functional volume”, corresponding to the quantity of bone needed to recreate the alveolar process;
2. The “morphologic volume” corresponding to the bone needed in order to restore the anterior nasal aperture.

A container with the shape of a spoon was created for both the functional and the morphologic volume. The virtual 3D models of the alveolar cleft and of the measuring spoons were then converted in a .stl-file and printed. A second CBCT was performed between 4 and 6 months after surgery and processed with the same software. Then, a comparison of the two 3D models allowed a measurement of the quantity of neo alveolar bone and even its distribution into the space of the cleft.

Results: 88% of the patients had a satisfactory result with a correct recreation of the alveolar process. In all of these patients, the volume of bone that was calculated with the post-op CBCT was higher than the volume defined as “functional” and the ratio between the “obtained volume” and the “functional volume” was 114%. In 4 patients, the volume obtained was similar to the “morphologic” one with a ratio greater than 90%. Only 2 patients had poor results because of a post-operative infection. No other complications were reported.

Summary/Conclusion: The introduction of the CBCT allowed several advantages. We can recreate a 3D model of the anatomy of the bone defect. In this way the surgeon can harvest a more suitable pocket for the bone graft. We developed a method to produce a measuring spoon corresponding to the volume of the bone needed for the graft, in order to facilitate the surgeon and minimize the trauma on the donor site. Comparing the CBCT results, it is also possible to measure the exact volume of new bone present in the alveolar cleft and make an evaluation of the reabsorption.
Tibial periosteal graft for cleft lip and palate repair. 25-year follow-up and controversies

M Zama¹, O Rajabtork Zadeh¹, D Buccii¹, S Vagnoni¹, M Rizzo¹
¹Plastic and Maxillofacial Surgery Dept, Cleft and Craniofacial Malformation Center, Bambino Gesù Children Hospital, Rome, Italy

Background: Hard palate repair has been more controversial than cleft lip repair. Despite the considerable progress in the treatment of children with cleft lip and palate, there is no agreement. The main discussion is about maxillary growth. Different authors have been discussing for many years the possibility to delay hard palate repair in order to minimize the effect on maxillary growth. Up to 1995 the cleft center of Bambino Gesù Hospital has performed primary repair with periosteal tibial graft according to a modified Stricker technique.

Aims: The aim is to objectively evaluate the long-term outcome in adults treated for Unilateral or Bilateral Cleft Lip and Palate (UCLP or BCLP) using periosteal tibial graft in primary repair during the period 1992-1995. We investigated and analyzed long-term outcomes on maxillary growth leg length discrepancy, residual oronasal fistula occurrence and aesthetical results.

Methods: 108 patients were identified. Exclusion criteria were: syndromic disorders, incomplete medical records, and refusal of the patient. 52 non-syndromic patients, affected by cleft lip and palate and treated by periosteal graft, were included in the study. These patients underwent: evaluation of plastic surgeons, dentists and speech therapist, lateral and antero-posterior radiographs for evaluation of the maxillary and mandibular growth, lower extremity radiograph. In order to obtain facial growth evaluation, the radiographs were analyzed by the same doctor using Dolphin Imaging Software.

Results: In the current study we analyzed 25 females and 27 males. Median age at follow-up was 25 years old. We observed 13 BCLP (25%), 39 UCLP (75%). They were operated at a median age of 8 months. During clinical exam, performed by dentist and by plastic surgeon, no patient showed an oral-nasal communication. In the past, 3 patients out of 52 reported fistulae in clinical history. Of these, 2 patients resolved spontaneously; only 1 patient underwent surgery. 49 patients accepted to be evaluated for leg discrepancy through the X-ray which highlighted leg discrepancy in 6 patients (12.2%). Cephalometric parameters were tested for normality; on the basis of our results, we cannot exclude that these variables were normally distributed. Overall, we found mean value of maxillary depth of 86°±4.5°. Patients completed a satisfaction tests about the overall physical aspect, 42 subjects (85.71%) claimed themselves satisfied, 4 subjects (8.16%) considered themselves highly satisfied, 3 patients (6.12%) were not satisfied.

Summary/Conclusion: The percentage of maxillary retrusion obtained is the same if compared to other techniques. Tibial periosteal graft reduces extraordinarily the risk of fistula and the share of bone graft reabsorbed during secondary bone graft. About the effect on the tibia, our study did not observe negative impact on leg growth after 25 years. Subjects are overall satisfied with the morpho-functional outcomes.
Comparaison between alveolar bone graft with only cancellous bone, and alveolar bone graft with cancellous bone combined with nasal cavity floor’s reconstruction using a cortical block.

V Thomas¹, P tramini², M bigorre⁴, G captier⁶
¹Centre de soins dentaires, CHU de Montpellier, ²EA2415, laboratoire d'aide à la médecine personnalisée, montpellier university, ³Centre de soins dentaires, CHU de Montpellier, ⁴Service de chirurgie plastique pédiatrique, CHU montpellier, ⁵Service de chirurgie plastique pédiatrique, CHU Montpellier, ⁶EA2415, laboratoire d'aide à la médecine personnalisée, Université de montpellier, montpellier, France

Background: Alveolar bone graft is one of the main step of the surgical care of the patients who are suffering from cleft lip and palate. They are usually performed before the lateral incisor eruption, at about 5 years old and using cancellous bone taken from the iliac crest.

Aims: The aim of the study is to show if adding a cortical graft to the cancellous causes more complications, which may be source of partial or total graft loss in the 6 months following the observation.

Methods: This is an observational, retrospective and monocentric study.
To be included, patients had to suffer from a clef lip and palate or alveolar cleft and had to have an alveolar bone graft with gingivo-periostoplasty in the pediatric plastic surgery unit of the Montpellier hospital from June 1999 to Mai 2016. They were divided into two groups : the first with patients who had a cortical and cancellous bone graft and the second with patients who had a cancellous graft only.
Both groups were followed during 6 months after the operation to look for the occurence of complications which lead to a graft loss. Bilateral cleft are seen as 2 graft events.
Percentages and proportions were calculated for the categorial variables, and chi-square tests were performed in bivariate analysis between the no failure/partial failure/total failure of the bone graft and the potential influencing factors. A stepwise multiple ordinal logistic regression was used in a multilevel mixed model to analyse the relationships between the surgery outcome and the independant variables, particularly the presence of “cortical bone. The random effect was the subject level. All analysis have been performed with the statistical software Stata 14.2 and the significance level was set at 0.05.

Results: 302 patients were included, which means 369 grafting event. In the cortical group, 13 failures were found in the cortical group among the 182 patients, 9 in the cancellous group with 187 patients. No significant difference was found between the two groups.

Summary/Conclusion: Alveolar bone graft is a reliable technic with a 5% partial failure rate and a 1% total failure rate. Add a cortical graft to rebuild the nasal cavity floor doesn’t bring more complications. It will be interesting to follow these patients for a longer period to see if this cortical graft bring better results.
Ten years follow-up of ours first alveolar bone graft in cleft lip and palate with bone morphogenetic protein (rhBMP-2)
R Carvalho¹, T Okada Ozawa², C Resende Leal¹
¹Cirurgia Bucomaxilofacial, ²Ortodontia, Hospital de Reabilitação de Anomalias Craniofaciais - Universidade de São Paulo, Bauru - SP, Brazil

**Background:** This is the third part of a previous study (a long term follow-up) of alveolar bone grafting in patients with unilateral cleft lip and palate using recombinant human bone morphogenetic protein (rhBMP-2).

**Aims:** This study intended to assess bone formation in alveolar clefts with rhBMP-2 in a collagen sponge carrier after ten years follow-up and eventual side effects and/or complications during this period.

**Methods:** Twelve patients with complete unilateral cleft lip and palate had undergone secondary alveolar bone grafting procedures, using rhBMP-2 in an absorbable collagen sponge. They were evaluated ten years later by a questionnaire and periapical radiographs from cleft area.

**Results:** Interdental septum height was evaluated according to Chelsea scale and the images revealed 100% success index as well as at one and three years post operative. None side effect and/or complications were referred by patients and their parents.

**Summary/Conclusion:** Bone formation from rhBMP-2 in alveolar bone grafts seems to be as stable as the one we observe from iliac cancellous bone.
Preliminary results of a bone substitute graft (glassbone®) in alveoloplasty
M Gatibelza1, A ARNAUD1, D BREZULIER2, O AZZIS1
1pediatric Surgery, 2dental department, Rennes Hospital, Rennes, France

Background: Alveolar bone graft is a major step in the care of complete cleft lip and palate. In order to decrease the morbidity of the autologous iliac bone procurement, currently performed, some authors studied bone substitutes with promising results.

Aims: We aimed to evaluate the effectiveness and morbidity of a synthetic and bioactive bone graft substitute (Glassbone®, Noraker) for secondary alveoloplasty.

Methods: All consecutive patients who underwent alveoplasty in our department using this bone substitute were prospectively included from July 2015 to December 2018. Informed consent was obtained.

Results: 57 patients were included, presenting with 46 unilateral and 11 bilateral clefts. Median age at surgery was 9 years [5-17]. Day case surgery was possible for 54 patients (95%), the three others were discharged the day after. Paracetamol and ibuprofen were required during 48 hours postoperative. Antibiotic prophylaxis by Amoxicilline and Acid clavulanic was prescribed for one week. Patients had 7-days school and 1-month sport exemptions. Short term follow-up showed a good tolerance in 54 patients (95%). Three patients with gingivitis were noted including one with loss of the substitute. At 6 months postoperative, all patients underwent a CT-scan; 44 (77%) showed a satisfactory filling of the alveolus cleft with a good integration of the substitute. At one year postoperative, 33 patients underwent a CT-Scan; 27 (82%) showed good maxillar fusion with satisfactory alveolar volume, 3 (9%) an incomplete bone fusion and 3 (9%) a persistant alveolar cleft. 25(78%) had complete Glassbone® resorption.

Summary/Conclusion: Glassbone® graft in alveoloplasty is a simple, reliable and reproducible technique. It represents a cost effective alternative with a low morbidity rate. Long term studies are needed to confirm these results and promote this new technique as a real alternative to iliac bone graft.
Lip/Nose

OS 18.1
Secondary cleft lip nose surgery. The normal structures are present. The secret is to respect them.

J Talmant¹
¹Clinique Jules Verne, Nantes, France

Abstract Content: Since 21 years we follow the same protocol in unilateral and bilateral cleft which aims to establish nasal ventilation through the first operation. The deformities of the alar cartilage and of the Nasalis-Myrtiform muscle are intermingled. Their accurate simultaneous repositioning restores a normal nose. Another key point is to save the space of the missing lateral incisive field which is the bony support of the nasal valve. At 6 months of age, without any presurgical orthopedics, - repositioning of naso labial muscles and nasal cartilages after wide subperiosteal and subperichondrial dissections, - efficient control of the healing process with closure of dead space and 4 months nasal stenting settle nasal ventilation and good esthetics. In secondary surgery, these rigorous principles improve the result as in tertiary surgery where any cartilage grafts should be removed and the original structures repositioned. Numerous cases are shown. This concept is opposite to the cosmetic trend of external approach and rigid framework leaving thick and narrow nostrils. Nevertheless, we should not focus only on the nose, for inter labial contact at rest is pre requisite for nasal ventilation which is the major function with regard to facial growth. To restore labial competence early enough during mixed dentition is indispensable to amazing late orthopedics and orthodontics success we had not dared hope for. The procedures developed for this cleft nose have great potential for any complex nasal reconstructive surgery.
Using facial averageness to measure the impact of secondary surgeries in cleft lip & palate
A Chadha¹, P Georg¹, P Haers¹
¹South Thames Cleft Service, St. Thomas' Hospital, London, United Kingdom

Background: The role of secondary surgery in cleft lip and palate is to approximate facial features of the patient to those of a normal face, however defined. One such definition is the mean features of the racial group to which the patient belongs. Each secondary surgical procedure, in a prescribed sequence, should result in an approximation towards this mean. Furthermore, it is useful to be able to predict the incremental benefit of each secondary procedure relative to the final result.

Aims: This study was granted permission as a Service Evaluation within South Thames Cleft Service. Its aims were to:
1. Characterise the sequence of secondary surgeries prescribed for CLP patients undertaken by one surgeon
2. Demonstrate the final result relative to the status pre-secondary surgery
3. Demonstrate the incremental benefit of each secondary surgery and to highlight those procedures that disproportionately accounted for the results of secondary surgery

Methods: This was a retrospective cohort study analysing routinely collected 3D photographs taken before and after each secondary surgical procedure. For each photograph, a detailed anthropometric analysis was undertaken and summed by way of a singular Facial Averageness Index (FAI) calculated from facial proportions. FAI results were then distributed graphically to calculate both the incremental benefit of each procedure as well as the overall benefit of secondary surgery relative to the operative baseline.

Results: 53/241 patients were identified as having 3D photographs of sufficient quality for analysis taken at time points consistent with study aims. A total of 122 photographs were analysed, across the spectrum of cleft deformities, using 35 anthropometric landmarks on each to generate 65 proportion indices. Summation of these indices resulted in the “Facial Averageness Index”. There was a strong correlation (r = 0.71, p<0.01) between pre-secondary surgery baseline facial appearance and final facial appearance in terms of approximation to the facial average. The greatest proportionate improvement was achieved through orthognathic surgery.

Summary/Conclusion: These results suggest a role for Facial Averageness Index as a tool to evaluate the results of secondary cleft surgeries using 3D photography. In our study, this tool indicates that orthognathic surgery is the greatest agent of approximation to the average face in the cleft sample analysed. Such a tool can be used to assess burden of surgical care and related cost-effectiveness between different cleft populations.
Development of the submental nasal appearance scale for the assessment of repaired unilateral complete cleft lip: a pilot study

R Tan¹, K Isaac¹, I Ganske ᵁ, D Mosmuller², H De Vet³, J Don Griot², J Mulliken¹
¹Plastic and Oral Surgery, Boston Children's Hospital, Boston, MA, United States, ²Plastic, Reconstructive and Hand Surgery, ³Department of Epidemiology and Biostatistics and the Amsterdam Public Health research institute, Amsterdam UMC, location VUmc, Amsterdam, Netherlands

Background: The goal of unilateral cleft lip repair is to create nasolabial symmetry and to correct the deformed nasal structures. Traditionally, appearance of the lip and nose is assessed by scoring frontal and lateral two-dimensional photographs. Given the importance of the nasal appearance, a new tool focused on the submental view would enable complete three-dimensional evaluation when used in combination with lateral/frontal validated scales. Important nasal structures, such as the columella and ala, are optimally visualized in this view and could be important when comparing surgical techniques or surgeons and cleft teams. As expert opinion is still the standard when grading operative outcome of cleft lip repair, this study aimed to identify nasal characteristics that are predictive for a cleft surgeon’s standard.

Aims: To develop the “Submental Nasal Appearance Scale” (SNAS), which is an easy-to-use objectified tool to represent a cleft surgeon’s standard for assessment of the nasal appearance from the submental perspective.

Methods: Eighty-five photographs of patients with unilateral complete cleft lip and palate were selected and cropped, displaying the submental view. Sixty-one photographs were used to develop 5 sets of reference photographs. Three cleft surgeons graded 24 photographs with these sets and subjectively graded the overall nasal appearance as well. Internal agreement for both methods was calculated, as well as correlation between them. The SNAS was created, by only using the combination of sets that showed the highest reliability and correlation.

Results: The intra-rater and inter-rater reliability was 0.84 and 0.79, respectively, for the SNAS and 0.76 and 0.62, respectively, for the overall appearance assessment. The correlation was 0.74 between the methods.

Summary/Conclusion: The SNAS is a reliable tool that reflects a cleft surgeon’s standard and could be used independently or in combination with existing rating scales using the frontal and/or lateral view, for assessment after cleft lip repair.
Validation of the unilateral cleft lip severity index for surgeons and laypersons

A Campbell¹, C Restrepo², G Deshpande³, S Bernstein⁴, C Tredway Goldin⁵, L Wendby¹, B Schönmeyer⁶
¹Operation Smile, Virginia Beach, ²Operation Smile, Virginia Beach, United States, ³MGM Dental College and Hospital, Navi Mumbai, India, ⁴Emory University, Atlanta, ⁵University of Colorado, Aurora, United States, ⁶Department of Hand and Plastic Surgery, Umeå University Hospital, Umeå, Sweden

**Background:** Severity of the unilateral cleft lip deformity (UCL) is postulated to play a key role in postoperative complications, aesthetic result, and need for secondary surgery. There is currently no validated and widely accepted classification scheme of initial cleft severity.

**Aims:** The purpose of this study was to validate the Unilateral Cleft Lip Severity Index as a reliable tool for evaluating presurgical UCL deformity by both surgeons and laypersons.

**Methods:** Twenty-five participants (10 surgeons and 15 laypeople) evaluated 25 sets of randomly selected presurgical standardized photographs of UCL patients. Each participant rated patients on a scale of 1–4 using the Unilateral Cleft Lip Severity Index. Grade 1 was defined as incomplete cleft lip involving less than 50% of the lip height. Grade 2 was defined as incomplete cleft lip involving more than 50% of the lip height. Grade 3 was defined as complete cleft lip with a nostril width ratio (cleft side nostril width/noncleft side nostril width) less than 2. Grade 4 was defined as complete cleft lip with a nostril width ratio greater than 2. Interrater reliability for surgeons, laypersons, and all participants was determined using the intraclass correlation coefficient. Histograms and regression analysis were performed to compare average ratings between groups.

**Results:** Interrater reliability for all groups was classified as "very good" determined by intraclass correlation coefficients of 0.837 (laymen), 0.885 (surgeons), and 0.848 (all participants). These results indicate that there was a high degree of agreement across all 3 groups and that both surgeons and laypersons can reliably rate cleft severity using the Unilateral Cleft Lip Severity Index.

**Summary/Conclusion:** This study validates the use of the Unilateral Cleft Lip Severity Index by both surgeons and laypersons as a reliable tool for evaluating the degree of presurgical severity of patients with UCL. This index can thus serve as a reproducible and reliable grading system for UCL deformity and to categorize patients for future outcomes studies.
Background: Patients with cleft lip and palate (CLP) can exhibit a thick and everted lower lip. The purpose of this study was to measure lower lip thickness and eversion in patients with CLP and maxillary hypoplasia.

Aims: The specific aims were to (1) compare lower lip thickness/eversion in patients with CLP to non-cleft controls with maxillary hypoplasia, (2) determine differences between patients with unilateral (UCLP) and bilateral (BCLP) CLP and (3) document lower lip changes that occur with Le Fort I advancement.

Methods: Retrospective case-control study of patients with available pre- and postoperative CT scans and 2D lateral photographs that underwent Le Fort I advancement between 2009-2017. Lateral cephalograms were generated in natural head position from CT scans and 11 cephalometric landmarks were identified. Soft tissue thickness of the lips and distance between lips were measured parallel to a true horizontal line, and height of the upper lip was measured perpendicular to the true horizontal. Lateral photographs with a marked lower lip vermilion border were superimposed with lateral cephalograms. Angle between most superior point of the lower lip and vermilion border was measured. To test intra-rater reliability 33% of all measurements were performed twice.

Results: The study included 32 patients with CLP (17 females: 15 males; mean age 17.6 ± 1.9 years) and 33 non-cleft controls (21 females: 12 males; mean age 18.7 ± 2.5 years): there was no difference in sample characteristics between the 2 groups. An independent t-test showed that patients with CLP have a significantly thicker lower lip (p = .019) and increased outward rotation of the vermilion border (p = .003) compared to non-cleft controls. The lower lip was significantly thicker in patients with BCLP than in those with UCLP (p = .035). Comparison of pre- and postoperative measurements with a paired t-test found the lower lip thickness and rotation did not change after maxillary advancement. Intra-rater reliability was good (ICC>0.9) for all measurements.

Summary/Conclusion: Patients with CLP and maxillary hypoplasia have a thicker and more everted lower lip than non-cleft controls. Patients with BCLP have a significantly thicker lower lip than patients with UCLP. Maxillary advancement does not improve the lower lip deformity.
**OS 18.7**

**Aesthetic and psychosocial impacts of primary rhinoplasty in the dento-facial appearance of children with unilateral cleft lip and palate.**

C Dissaux¹, V DIOP², J Talmant³, B Morand⁴, I Kauffmann⁵, C Bruant-Rodier¹, B Grollemund²

¹Maxillofacial and plastic surgery, Cleft competence center, ²Orthodontic department, Strasbourg University Hospital, Strasbourg, ³Plastic surgery, cleft competence center, Clinique Jules Verne, Nantes, ⁴Maxillofacial surgery, Cleft competence center, Grenoble University Hospital, Grenoble, ⁵Pediatric surgery department, cleft competence center, Strasbourg University Hospital, Strasbourg, France

**Background:** Primary rhinoplasty at the same time as lip closure has become more and more popular these last years and stands now as a major step of the primary surgical treatment of cleft lip and palate.

**Aims:** The primary aim of this study was to demonstrate whether primary rhinoplasty shows aesthetic and psychosocial advantages for children with a complete unilateral cleft lip and palate (CLP).

The second aim was to determine the satisfaction levels concerning the dentofacial appearance.

**Methods:** Two cohorts of patients who had or not undergone primary rhinoplasty at 6 months of age were evaluated at preadolescence. Three French cleft centers, of which only one performs primary rhinoplasty, provided 97 frontal and low-angle facial photographs of 9 to 12 year-old children (mean age: 11.5). Ten operated noses (A) and ten non-operated noses (B) were randomly selected and assessed by a jury of 305 people made up of 136 health professionals, 57 lay persons and 56 families of CLP patients (56 children and 56 children). Moreover children and their parents filled in a questionnaire about the satisfaction of the children dentofacial appearance and the psychosocial impact of the cleft. Bayesian statistic model was used.

**Results:** The aesthetic perception test realized on 20 noses revealed that primary-operated noses were statistically significantly more attractive than non-operated ones.

The questionnaire filled in by the patients’ families revealed that the children did not rate statistically differently their social relationships and their specific nose teasing if they had primary rhinoplasty or not. Parents however expressed very different views. The parents considered the nasal appearance of the operated children as statistically more attractive and those whose kids had undergone nose surgery evaluated the psychosocial experience of their children as significantly improved.

Overall, in both groups, satisfaction levels of dentofacial appearance and psychosocial comfort were good (over 80%).

**Summary/Conclusion:** General and families evaluation showed a positive impact of primary rhinoplasty on facial appearance and social life, even though if children themselves, before puberty, were not aware of it yet.
Common techniques of lip enhancement in secondary cleft surgery and their influence on the labial appearance

M Schwaiger1, K Echlin2, S Edmondson3, A Paddle4, D Atherton2
1South Thames Cleft Service, Department of OMFS, Medical University of Graz, Austria, 2Guy's and St. Thomas' Hospital, Department of OMFS, Medical University of Graz, Austria, 3South Thames Cleft Service, Guy's and St. Thomas' Hospital, 4Guys and St. Thomas' Hospital, London, United Kingdom

Background: In secondary cleft lip deformities, volume deficiency in the upper lip vermilion is commonly encountered. Typical characteristics include asymmetry, an imbalance between upper lower lip volume and poor upper lip projection. Aesthetic standards are known to fluctuate over time and differ between cultures and ethnicities, yet, it has been established that full lips are perceived as aesthetically more pleasing and attractive than thin lips. Different surgical techniques are available to address volumetric deficiencies, according to extent and localisation. However, data examining their influence on the labial appearance is limited.

Aims: The purpose of our study was to subjectively and objectively assess postoperative outcomes of common surgical techniques for lip volume enhancement in cleft patients regarding lip fullness, upper vermilion height and lip projection.

Methods: Standardised pre- and postoperative photographs of 54 adult patients, who underwent secondary lip volume enhancement surgery, were assessed according to the parameters lip fullness, upper lip projection and upper vermilion height. Subjective and objective assessment measures were used to assess outcome. 7 examiners (3 cleft surgeons and 4 lay persons) independently evaluated anonymised pre- and postoperative front and side views. The examiners were blinded regarding the patients' procedure and the status of the photograph. The Allergan lip fullness score and a 7-point Likert scale were used for the subjective evaluation. Objective analysis was performed using Adobe Photoshop. Landmarks as recently described by Thomas et al. were used.

Results: 42 patients (17 BCLPs, 25 UCLPs) were included in the final analysis. 15 lip revisions, 4 Abbe flaps, 12 dermal grafts, 6 Permalip implants, 2 autologous fat graftings and 3 V-Y-plasties were performed. All procedures were performed by a single surgeon. In BCLPs, the Abbe flap showed most significant improvement in terms of the assessed parameters. In UCLPs, Permalip implants improved upper lip fullness and lip projection most distinctly. The lip height was shown to change most considerably in dermal grafts. Statistically significant differences between Abbe flaps, dermal grafts and silicone implants, when compared to lip revisions were found. Overall, Abbe flaps showed most significant improvement.

Summary/Conclusion: In secondary lip deformities, Abbe flaps, dermal grafts and Permalip implants have been shown to reliably address volume deficiency in the upper lip vermilion and simultaneously contribute to a more attractive labial appearance.
The healing with minimally visible scar - our thirteen years of experience with neonates cleft- lip-surgery
J Borsky¹, M Jurovčík², L Jaklová³, J Velemínská³
¹Motol University Hospital in Prague, 2nd Faculty f Medicin, Charles University in Prague, Prague 6, ²Motol University Hospital in Prague, 2nd Faculty f Medicin, Charles University in Prague, Prague 5, ³Anthropology and Human Genetics, Faculty of Science, Charles University, Prague 2, Czech Republic

Background: We present the results of our thirteen years of experience with neonates cleft-lip-surgery with new-born patients up to 8 days old.
Aims: Cleft lip surgery is usually performed at the age of 3 months or later. Nursing babies with visible facial disfigurement for three months can adversely affect the psychological wellbeing of these patients' families. By early surgical intervention we achieved not only good anatomical correction but we significantly improved the quality of life of the whole family.
Methods: We operated on 644 patients cleft lip or cleft lip and palate at the age of 1 to 8 days after birth: 532 neonates with unilateral cleft lip and 112 with bilateral cleft lip. One surgeon from December 2005 to December 2017 performed all operations. A neonatologist was responsible for preoperative care in the Well Baby nursery and post-operatively in the NICU. Before surgery we performed 3D scans of palate shaped casting using laser scanner as well as facial scanner 3D. The ENT specialist who evaluated torus tubarius initiated the first phase. Following this, the eardrum was examined and if present, middle ear fluid was sucked out. We believe this is the earliest detection and treatment of OME. New-borns left hospital between the 3rd and 4th postoperative day. All patients wore supportive silicon nostril retainers for 2-3 months.
Results: We have experienced only 5 complications resulting from this surgery. At the time of patients' discharge the wounds were usually healed. We presume that the aesthetic results are superior to patients operated in 3 months and later. Comparison of 3D scans of palate and face between study group and controls revealed no significant difference in maxilla and face growth.
Summary/Conclusion: If performed under high quality anesthesiological conditions, neonatal, and ENT care settings, and of course with experienced hands, the early correction of cleft lip is a safe method for neonates and gives very good aesthetic results.
**OS 18.10**

**Benefits of tongue reduction surgery (trs) for macroglossia in beckwith wiedemann syndrome (bws)**

C Shipster

1Speech and Language Therapy Department, Great Ormond Street Hospital for Children NHS Foundation Trust London, London, United Kingdom

**Background:** TRS is recommended for children who have macroglossia associated with BWS to overcome or reduce the secondary negative functional effects of the macroglossia on speech, drooling, oral motor skills and feeding. However, few studies describe the exact nature of these functional effects and the degree of improvement that is achieved post surgically. Furthermore, only two small retrospective studies to date, report on the functional and psychosocial profiles both pre and post operatively within the same cohort of children (Shipster. et al 2006; 2012).

**Aims:** In contrast, this prospective study carried out at the UK multidisciplinary national service for this patient group, describe systematically the pre-operative functional and psychosocial effects of the macroglossia and the changes that occur post-operatively. This study also discusses the optimal age to perform the surgery. This is the largest cohort studied to date.

**Methods:** Between April 2012 and March 2017, a consecutive series of 87 participants underwent the same TRS technique. This was a primary procedure for 83 participants and a secondary procedure for 4 participants. Age at surgery ranged between 1 and 17 years. 85 % had surgery below 3 years of age. All were evaluated pre-operatively and 3-6 months post-operatively using the same battery of clinical measures. Data was also collected on any co-morbid factors which could affect the functional outcomes.

**Results:** Anterior articulatory errors and oral stage feeding difficulties were present in the majority pre-operatively and were eliminated by TRS. Excessive drooling was present in 80% pre-operatively and significantly reduced or eliminated post-operatively. Positive psychosocial changes and satisfaction with the functional changes following surgery was reported in all cases. Speech impairment and feeding difficulties unrelated to the macroglossia also occurred in the cohort.

**Summary/Conclusion:** Macroglossia causes distinct articulatory and feeding difficulties; increased drooling and psychosocial difficulties due to the enlarged tongue and the functional difficulties that arise as a direct consequence of the macroglossia. TRS has a positive impact on these difficulties with consistently good outcomes for children with BWS.
Craniosynostosis Surgery Part Two

OS 19.1
Spring assisted surgery for craniosynostosis - 20 years experience
L Kölby

Abstract Content: Spring assisted surgery (SAS) was introduced Gothenburg in 1997 and has developed to a simple, safe and reproducible surgical technique, especially for sagittal synostosis. The obvious advantages include less invasiveness, less blood loss, less postoperative pain and shorter hospital stay compare to more complex cranioplasties. SAS is preferably used in children below 6 months of age; after that the skull becomes too hard for the springs to be able to exert their action. To date we have performed X SAS for sagittal synostosis and when the result is compared to that after the more extensive pi-plasty, the cephalic index (CI) and intracranial volume (ICV) compares favorably. SAS can be used in several other variants of surgery for craniosynostosis. In multi sutural synostosis springs can act as a simple first step to correct complex deformities. Springs can also be used as a part of cranioplasty for brachycephaly. The forehead remodeling is combined with springs over open lambdoid sutures or eventually over posterior osteotomies to gain extra ICV. The combination of forehead remodeling and posterior springs have been proven to result in normal ICV in patients operated for brachycephaly. Posterior skull expansion is often performed with osteodistractors. A simple way to achieve excellent distraction is to put springs across an osteotomy that releases the occipital part of the skull. In recent years two cases of unicoronal synostosis have been operated with a simple coronal osteotomy combined with springs. The effect on facial scoliosis has been remarkably good and the forehead is also well corrected. The springs have a few, at present, unavoidable drawbacks. i. They need to be removed at a second minor operation. This is a quick procedure but requires full anesthesia. ii. The springs also have a certain degree of unpredictability. If not adequately placed, the distraction by the springs may be asymmetric. Future development of the spring technique could include development of an absorbable material to avoid the second operation.
OS 19.2
Thirty years later – what has craniofacial distraction osteogenesis surgery replaced?
R Hopper

Abstract Content:
Background: Since introduced thirty years ago, the role of craniofacial distraction osteogenesis has been debated and challenged. The purpose of this presentation is to critically assess the current evidence in the literature, identifying what traditional procedures craniofacial distraction has replaced, and which it has not.
Methods: This continuing education presentation is a review of comparative studies and expert opinion on the current state of craniofacial distraction compared to traditional surgeries. Through this critical evaluation, a participant will be able to identify when distraction techniques are appropriate, when traditional techniques are more favorable, and the potential evolution of distraction osteogenesis in the future.
Results: The existing literature demonstrates that mandible distraction can achieve large stable movements, but has not overcome the traditional challenges of high occlusal plane angle, condylar resorption, or neurosensory changes. Sagittal split osteotomy remains the standard to achieve final occlusion at maturity. Alveolar distraction is a powerful and evolving technique. Its higher complication rate and technical difficulty has prevented it from replacing traditional grafting for implant placement, but it shows promise in severe defect cases. Subcranial and monobloc distraction have both replaced traditional advancement due to improved safety profiles. More long-term outcome studies of comparable populations treated with monobloc verses a staged approach will help us tailor the appropriate treatment for syndromic patients. Posterior cranial distraction has not been proven to be safer, but has replaced traditional posterior remodeling due to its ability to achieve large cranial volume increases with favorable intracranial changes. The potential of anterior cranial distraction continues to be explored, but long-term comparisons of shape and function will be required before it can compete with the standard of fronto-orbital advancement.
Conclusions: Certain craniofacial distraction techniques have replaced traditional advancement and fixation, whereas others require further evolution before this may be possible. The first three decades of distraction has been one of exploration, whereas the road ahead will be one of structured evaluation and comparison of long-term outcomes.
A comparison of the clinical application of autologous fat vs. Allograft adipose matrix grafting (aam) for craniofacial and cleft soft tissue reconstruction in the pediatric patient.

K Hopkins

Background: Autologous fat (AF) grafting has been successful in soft tissue augmentation and contouring primarily in the adult population. It is also a viable tool in restoring soft tissue in the pediatric patient for congenital and traumatic defects but there are inherent issues in this population such as the accessibility and amount of available autologous fat. Recent FDA approval of allograft adipose matrix (AAM) provides a viable alternative tool for correcting soft tissue congenital and traumatic defects in the pediatric patient.

Aims: This paper recounts a comparison of our experience using both autologous fat and AAM in the pediatric population.

Methods: Autologous Fat: 34 patients (21 female, 13 male) ages 2-18 years (average age 10.6) presented with 23 congenital defects (20 cleft lip/palate, 1 hemifacial microsomia, 1 hemangioma, 1 frontonasal dysplasia) and 11 traumatic defects (6 MVA, 4 burns, 1 dog bite) with follow up 7 weeks to 26 months. The cleft lip and palate defects included palatal fistula with severe scarring and fibrosis (7); velopharyngeal insufficiency (5); asymmetric soft tissue volume (8) and scarred,contracted nasal tip (2). Three congenital defects were hemifacial microsomia (1), malar deformation - involuted hemangioma (1), and frontonasal dysplasia (1). Traumatic deformities involved face (4), upper extremity (5), trunk (1), and lower extremity (3). Three patients had multiple deformities (3). Seven patients underwent serial AFT. Fat was harvested using the Coleman technique and a 12-hole Khouri cannula, The volume of fat ranged from 0.5 ml to 10 ml per site. Allograft adipose matrix(AAM): 27 patients (9 female, 18 male); ages 1-16 years (average age 10.3) presented with 23 congenital defects (17 cleft lip/palate, 6 craniofacial soft tissue deficits and 4 traumatic defects (3 craniofacial and 1 trunk with follow up time ranging from 2 weeks to 18 months. The cleft lip and palate defects include palatal fistula with severe scarring and fibrosis (1); velopharyngeal insufficiency (9); a asymmetric soft tissue volume (7) Congenital defects were hemifacial microsomia (1), hemifacial atrophy (1), and frontonasal dysplasia (1). The traumatic deformities involved the face (3), trunk (1). Five patients underwent serial AAM grafting. Dehydrated allograft adipose matrix was reconstituted with sterile 0.9% saline. The volume of AAM transferred ranged from 0.9 ml to 15 ml per site.

Results: Direct observation and serial photographs demonstrate increased soft tissue volume and enhanced tissue quality as well as improvement in all patients with VPI following both autologous fat and AAM grafting.

Summary/Conclusion: Autologous fat may be safely harvested and effectively grafted in pediatric patients. Care must be taken to safely harvest fat in these children so as to not create a contour deformity or perforation. Allograft adipose matrix may be safely used and effectively grafted to correct contour deformities in children. Thus far, AAM grafting has been used for the same indications as autologous fat grafting with improvement in contour and soft tissue defects similar to autologous fat. Very early studies suggest allogenic fat has similar results to autologous fat. There is less potential morbidity, shorter operative time, ease of use and potential cost savings to consider. Further studies are warranted.
OS 19.5

The use of throat packs in pediatric cleft lip/palate surgery.
B Smarius¹, C Guillaume¹, G Jonker², A Mink van der Molen¹, C Breugem¹
¹Department of Pediatric Plastic Surgery, ²Department of Anesthesia, UMC Utrecht, Utrecht, Netherlands

Background: Throat packs are commonly used to prevent ingestion or aspiration of blood and other debris during cleft lip/palate surgery. However, dislodgement or (partial) retainment after extubation could have serious consequences. The aim of the present study was to investigate the effect of omitting pharyngeal packing during cleft lip/palate surgery on the incidence of early postoperative complications in children.

Aims: The aim of the present study was to investigate the effect of omitting pharyngeal packing during cleft lip/palate surgery on the incidence of early postoperative complications in children.

Methods: A retrospective review was performed on all children who underwent cleft lip/palate surgery at the Wilhelmina Children’s Hospital. This study compared the period January 2010 through December 2012 when pharyngeal packing was applied according to local protocol (group A) with the period January 2013 till December 2015 when pharyngeal packing was no longer applied after removal from the protocol (group B). Data were collected for sex, age at operation, cleft lip/palate type, type of repair, lateral incisions, length of hospital stay, and complications in the first six weeks after surgery. Early complications included: wound dehiscence, postoperative bleeding, infection, fever, upper respiratory tract infection (URTI) and lower respiratory tract infection (LRTI).

Results: This study included 489 cleft lip/palate operations (group A: n=246, group B: n=243). A total of 39 (15.9%) early complications were recorded in group A and a total of 40 (16.5%) in group B. There were no significant differences (P=0.902) in complications between the two groups, however there was a significant difference (P<0.001) in length of hospital stay between the two groups (group A: 3.6 days vs group B: 3.2 days).

Summary/Conclusion: Omitting routine placement of throat packs in cleft lip/palate surgery was not associated with an increased early postoperative complication rate when compared to routine placement of throat packs. The traditional, routine placement of a throat pack during cleft lip/palate surgery can be questioned.
Relevance of positional cranial deformity and therapeutic need evaluated by laymen
S Kluba¹, S Hoepfl¹, K Haas-Lude², M Krimmel¹, S Reinert¹
¹Department of Oral and Maxillofacial Surgery, University Hospital Tuebingen, Tuebingen, ²Department of Paediatric Neurology, University Children's Hospital Tübingen, Tübingen, Germany

Background: The relevance of positional cranial deformity is still controversially discussed. While many specialists support therapeutic interventions with a helmet for severe cases, some are convinced that the fears are exaggerated and helmets superfluous.

Aims: We therefore asked unaffected, non-expert people to evaluate positional cranial deformities and the necessity for therapeutic interventions.

Methods: 395 laymen were interviewed. Standardized photos of 10 children with different degrees of positional deformity (CVAI 3.5% - 12.5%, CI 85.0% - 100%) were presented to them in a randomized order. Values from the literature (CI: 85%, CVAI: 3.5%) served as normal reference. The people were asked to evaluate, if the children looked abnormal to them and if they should be treated. The deformity was regarded as obviously abnormal when 50% of the respondents perceived the head as conspicuous. The cut-off value for a therapeutic recommendation was set at 25%.

Results: All heads above the reference values were perceived as abnormal by the great majority of laymen independently of the severity of deformity (plagiocephaly: 93.9% - 95.7%, brachycephaly: 87.3% - 93.2%, combination: 77%). In all abnormal infant heads laymen saw the need to treat (plagiocephaly: 51.1% - 72.9%, brachycephaly: 59.5% - 62.0%, combination: 37.2%). The lower border of confidence intervals exceeded the cut-off value of 25% in all cases.

Summary/Conclusion: Our results confirmed the existing normal values. As already very mild deformities were perceived as conspicuous and worth to treat by the laymen, the problem is clinically relevant and should not be underestimated. A social stigmatization cannot be excluded when untreated. Therefore adequate treatment (e.g. physio-, osteopathy, helmet or a combination) should be offered regularly.
Evaluation of smile esthetics in treated cleft lip and palate patients – a photographic study
A Husain¹, A kuipers-jagtman², A gafoor³
¹Department of orthodontics, Yenepoya Dental college, Mangalore, Karnataka, India, ²Faculty of Dentistry, Universitas Indonesia, jakarta, Indonesia

Background: Smile is one of the most important expressions contributing to facial attractiveness. It influences a person’s perceived attractiveness and is the cornerstone of social interaction.

Aims: This study compares the components of a smile, between pre treatment and post treatment subjects with unilateral cleft lip and palate and comparing the post treatment results with normal subjects.

Methods: 40 frontal smiling pictures of subjects with unilateral cleft lip and palate before and after orthodontic treatment as test group and 40 frontal smiling pictures of non-cleft subjects with apparently symmetrical faces with full complement of teeth who haven’t undergone orthodontic treatment were selected as control group. The face was divided into Facial, Nasal, Labial, Naso-Labial & Dento-gingival components based on the AKTHER-ANNE Smile Diagram. MAKHTER facial analysis software was used for the facial analysis. A ‘1cm x 1cm’ square sticker was put on the forehead for standardization of the photographs. Paired t test was used to compare smile parameters between pre and post orthodontic treatment in the cleft group and unpaired t test was used to compare post treatment results with the non-cleft group group. Statistical significance was set at p < 0.05.

Results: Lower anterior facial height, Nasal concha – endocanthon horizontal height, Crista philtri width, Commissure height, Commissure width, Buccal corridor, Posterior maxillary dentition visibility, Upper incisal display and Lower incisal display showed a statistically significant improvement after orthodontic treatment in subjects with unilateral cleft lip and palate when compared to pre treatment values. The post treatment results didn’t show significant differences when compared to non-cleft subjects.

Summary/Conclusion: Orthodontic treatment in patients with unilateral cleft lip and palate improves the smile significantly especially on dento-gingival components. So orthodontic treatment in conjunction with suitable surgical procedures will help to achieve a face without cleft stigmata in patients with unilateral cleft lip and palate.
**Background:** Autologous adipose tissue transplantation is a broadly used procedure in reconstructive and aesthetic surgery; however, a universally accepted processing method has not been reached. The “LULL-PGM System” is based on a closed washing circuit. The harvested tissue is purified by Ringer solutions with a gentle lull motion of the syringe without expensive devices.

**Aims:** Our purpose was to compare three processing techniques: decantation, centrifugation and washing through the LULL-PGM system. Secondly we analyse some clinical application in patients with different degrees of malformations.

**Methods:** Liposuction was performed in the lower abdomen of 19 patients; three 30 ml specimens were collected from each patient. Each sample was randomly processed in the operating room through one of the compared methods; 10 ml sample of purified tissue from each specimen were sent to the laboratory. Investigations were focused on hematopoietic cellular lines, adipocytes, stromal vascular fraction (SVF) and adipose-derived mesenchymal stem cells (AMSCs).

**Results:** Centrifuged and Lull samples, containing less red blood cells than decanted specimens, have proved to be better purified (p < 0.05). SVF concentration was significantly higher in Lull samples compared to decanted ones (p < 0.05), while centrifuged samples presented intermediate value. The colony-forming units (CFUs) evaluation showed a significantly higher number of AMSCs for centrifuged and washed samples (p < 0.05).

Osteoblast and adipocyte commitment of SVF cells obtained from all the procedures confirmed that the subpopulation of AMSCs presents features of stemness toward mesenchymal linages.

**Summary/Conclusion:** The Lull PGM system seemed to be the most suitable processing technique, as it achieved the best compromise between adipocytes integrity, purification from contaminants and concentration of SVF and AMSCs. Given the satisfying results obtained in “in vitro” evaluations, the Lull has been progressively implemented into our clinical practice. Positive results and patient satisfaction have been obtained and no side effects or disadvantages have been observed. In patients with velopharyngeal insufficiency we obtained almost complete resolution in mild to moderate cases. In patients with facial asymmetry or deficiency of the middle third of the face, a good morphological restoration was achieved.
Development of a method for rating nasolabial appearance in children with bilateral cleft lip and palate
T Bartzela1, G Semb2, T Okada-Ozawa3, S Rizell4, A Kuijpers-Jagtman5
1Orthodontics, Dentofacial Orthopedics and Pedodontics, Charité - Universitätsmedizin Berlin, CC3, Berlin, Germany, 2Plastic and Reconstructive Surgery, University Hospital of Oslo and Statped Sørøst, Oslo, Norway, 3Dental Department in the Hospital for Research and Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru, Brazil, 4 Section of Jaw Orthopedics, University Clinics of Odontology, Gothenburg, Sweden, 5Faculty of Dentistry, University of Indonesia, Jakarta, Indonesia

Background: Nasolabial appearance is one of the outcomes in patients with cleft lip and palate (CLP) that have the greatest importance for patients and parents (1). A major goal for cleft surgeons is to get a good aesthetic appearance of the cleft lip and distorted nose. A method and yardstick for rating dental arch relationships in children with bilateral CLP (BCLP) has been described (2) but no method to assess the nasolabial aesthetics has been defined.

Aims: To develop a method for rating the nasolabial appearance in children with BCLP, at 6, 9 and 12 years of age.

Methods: Children with complete BCLP at age 6, 9 and 12 years from 4 different centers (Nijmegen, Oslo, Gothenburg and Bauru) were included in the study. Informed consent was obtained. The principles of Declaration of Helsinki with regard to research in human subjects were followed. A panel of judges, consisting of 11 orthodontists, with experience in the treatment and outcome of cleft patient. The judges were shown: the frontal, profile and worm’s eye facial views of children with BCLP. The facial features (hair and eyes) were eliminated. The upper lip and the nose were judged separately. Three different features were assessed in the upper lip: red, white, length. Three features were assessed for the nose: shape, tip, columella. A 5-point scale was used, rating nasolabial appearance from excellent (1) to very poor (5). A sample consisting of 50 photographs for each age group were discussed and analyzed by the panel of judges until they agreed on the pictures that would become part of each yardstick. To assess the validity, 30 randomly selected pictures were rated by the judges twice. Weighted Kappa statistics were performed to evaluate intra-rater and inter-rater reliability.

Results: This yardstick has fair to good level of reproducibility (kappa= 0.21-0.8). The interrater weighted kappa scores range for the 6-year old patients for the lip: 0.468-0.681, for the nose: 0.383-0.748, for the 9-years old for the lip: 0.380-0.660 and for the nose: 0.338-0.738 and for the 12-year old group, for the lip: 0.267-0.680 and for the nose: 0.248-0.624. During the test runs, consensus was reached on the final use of the yardstick for three parameters for the lip and three for the nose. The guidelines of the five categories for each yardstick will be presented.

Summary/Conclusion: The nasolabial appearance yardstick for BCLP patients has good level of reproducibility for the youngest age group and for most of the parameters. The red of the lip and the tip of the nose have the best reliability in all age groups.

References:
Nasolabial appearance in 5-year-old patients with repaired complete unilateral cleft lip and palate – comparison of two different techniques of lip repair

S Lux1, M Mayr1, G Danner1, C Koenig1, A Gaggl1, P Schachner1

1Clinic for Oral and Maxillofacial Surgery, Private Medical University Salzburg, Salzburg, Austria

Background: For lip repair in patients with unilateral cleft lip and palate (UCLP) many different techniques are described. Aim of all these techniques is a reconstruction of the lip and nose especially in functional as well as in aesthetic aspects. At the age of approximately five to six years it should be decided if any correction of the upper lip or nose should be performed because of changing the social environment with entering school.

For assessment of the nasolabial appearance in patients with cleft lip and palate the Asher-McDade Aesthetic Index is the most common method.

Aims: Aim of the study was to assess aesthetic appearance in five year old patients with unilateral cleft lip and palate and to compare the results of lip repair according to Pfeifer’s “wave-line” technique with the results according to Randall’s technique. Nasolabial appearance was assessed qualitatively using the Asher-McDade Aesthetic Index in a blind retrospective analysis.

Methods: Frontal and lateral photographs of 53 patients were cropped according to the CARS Cleft Aesthetic Rating Scale. Twenty-seven patients received lip repair according to Pfeifer’s wave-line procedure (group I) and twenty-six patients according to Randall’s technique (group II). All patients underwent only primary surgery (of lip and palate) without any correction of the nose or upper lip. Three components of the nasolabial area (in the frontal view: 1. nasal form and deviation/symmetry of the nose (nasal frontal, NF), 2. shape of the vermilion border (vermilion border, VB); and in the side profile view: 3. nasal profile including upper lip (nasal lip profile, NLP)) were rated separately using a five-point ordinal scale.

Five professionals highly experienced in UCLP treatment (three surgeons, one speech and language therapist, one orthodontist) scored the cropped photographs.

Results: The average rating of the VB, NF and NLP in the Randall-group (group II) was 1.99 (±0.78), 2.44 (±0.59) and 2.29 (±0.57). In the Pfeife-group (group I) the same parameters were evaluated with an average of 2.48 (± 0.95), 2.54 (±0.57) and 2.52 (±0.56). There was a significant difference between both groups at the comparison of the upper lip/vermilion border: -0.51 (95% confident interval: -0.98; -0.059; p=0.03). No significant difference was found at the rating of the nose (nasal profile, nasal frontal view) p>0.05. Intraclass correlation of the first rating was 0.681 (95% confidence interval: 0.573; 0.778). Intraclass correlation was calculated for each individual rater. The values ranged from 0.837 (95% confidence interval: 0.71; 0.907) to 0.670 (95% confidence interval: 0.051; 0.867) with an average of 0.75.

Summary/Conclusion: This retrospective investigation showed that the patients with lip repair according to Randall’s technique had a better aesthetic outcome of the upper lip and shape of the vermilion border than the patients with lip repair according to Pfeifer’s “wave-line” technique. The rating of the nose showed no differences. Further evaluations during growth (at the age of 10 years and later) have to be done.
Background: The main aim of the COST Action “European Cleft and Craniofacial Initiative for Equality in Care” (www.ecce.nu) is to ensure that children born with orofacial clefts and other craniofacial conditions receive optimum multidisciplinary care enabling them to grow up like any other child and attain equal status within their societies. Estimates indicate that there are over one million individuals with clefts in Europe - a significant figure, especially when one considers that not only the patients but also their families are affected in terms of psychosocial adjustment and having to endure the burden of a long treatment pathway. One aspect of the Action is to generate awareness the impact of the long term outcomes together with how social determinants influence provision of care.

Aim: Europe currently lacks a harmonised approach to evaluate access to care, the current provision of care, the impacts on key areas of the affected families and society at large, especially in relation to inequalities and social determinant factors. At the same time, about 23% of our general population is at risk for poverty or social exclusion together with that studies in Europe show that around half of all patients cannot understand basic health care information that is presented to them by healthcare professionals. It is paramount that healthcare professionals have understood how these factors influence the provision of care and how individuals with a cleft are affected.

Method: A review of the current status in Europe concerning social determinants, education and mental health for the general population and the population with a cleft.

Result: For individuals with a cleft, research shows that their educational achievement can be negatively affected and that they have a significant risk for experience for mental health problems in comparison to the general population.

Summary/Conclusion: The resources in our European health care system are quite varied, and the demand for resources is increasing rapidly. Healthcare professionals need to be aware of how social determinants affect individuals and at the same time recognise that screening for educational outcomes and mental health for individuals with a cleft should be incorporated in their provision of care.
Neurodevelopment in children with isolated oral cleft: findings from structural and functional imaging studies
A Conrad

Abstract Content: This presentation will discuss the past 20 years of neuroimaging research that has been conducted across the age-span on patients with isolated oral clefts. Structural and functional imaging has provided new insights in brain development and potential connections to academic, cognitive, and behavioral outcomes. This body of literature will be discussed in relation to the neural migration theory and the next step of bringing together neuroimaging and genetic disciplines. Participants will learn about different imaging methods, structural development in patients with isolated oral clefts, how structural findings correlate to functioning, and emerging work on neurocircuitry.
Os 20.3
Eurolinkcat -establishing a linked european cohort of children with congenital anomalies- cleft lip: the parent's voice
A J Neville¹, J Clemensen², A Pierin³, J Morris⁴
¹IMER Registry, University of Ferrara and Azienda Ospedaliero Universitario di Ferrara, EUROCAT Registry Leader, Ferrara, Italy, ²Odense University Hospital and Head of Research at Centre for Innovative Medical Technology, Hans Christian Andersen Hospital for Children and Adolescents, Odens, Denmark, ³Institute of Clinical Physiology National Research Council, Registry of Congenital Defects, Pisa, Italy, ⁴St George's, University of London, Population Health Research Institute, London, United Kingdom

Background: EUROlinkCAT is establishing a linked European Cohort of Children with Congenital Anomalies involving 14 countries.
Aims: To investigate the health and educational outcomes of children with congenital anomalies and facilitate a reciprocal relationship between families, health care professionals and researchers.
Methods: Cleft lip (CL) was chosen for a focus group to address parent's experiences. Eleven Italian parents explored themes such as the discovery of the condition, the birth, daily life, support and information and their ideas for research questions to be answered regarding CL.
Results: Many parents expressed that the focus group was the first opportunity to talk about their child and share experiences. A clear gap exists between parents and clinicians in communication and information. The parents wished to receive more information on what their child could potentially achieve rather than on how limited the child is likely to be. Parents commented that they loved their babies face and that surgery and change was difficult psychologically. Parents used the internet but abandoned it finding horror stories, conflicting information or not the needed information. However, following the focus group, the parents spontaneously took the initiative to create a Whatsapp group that they now use for sharing experiences, pictures and invitations. A representative of the parents also presented their experiences at a regional scientific conference on Cleft Lip. CONCLUSION: Parents did have different views from clinicians concerning the information they wished to receive about their children’s potential futures and therefore it is important for researchers to involve parents in such focus groups when planning their research. Although parents of children with anomalies can feel isolated contact with other parents needed to be in person before it could continue through the use of social media.
Summary/Conclusion: Parents did have different views from clinicians concerning the information they wished to receive about their children’s potential futures and therefore it is important for researchers to involve parents in such focus groups when planning their research. Although parents of children with anomalies can feel isolated contact with other parents needed to be in person before it could continue through the use of social media.
Background: Neuropsychological studies on the CLP population and statements of the Consensus Conference 2011-assessed that two or more general anesthesia postpartum and prior to age four are considered as a risk factor associated with the development of learning disabilities.

Aims: The study aims to test the automation of the reading process on a group of adolescents and young adults born with isolated cleft lip and palate (CLP) and exposed to two general anesthetics prior to age four. The speed and accuracy of the results of the mentioned group were compared with the ones of an equal number of peers born without CLP and not exposed to the considered risk factor.

Methods: The CLP sample consists of a consecutive group of 32 young adults, 26 UCLP and 6 BCLP, with a mean age of 21.11, who came to the CLP center of the Santi Paolo e Carlo Hospital of Milan for a follow-up at the end of the craniofacial growth. The control group consists of 32 participants, recruited and observed in high school, university or working professionals, with a mean age of 20.8. The assessment of the reading ability has been carried out with the MT 16-19 test (Cornoldi, Candela, 2015) using word reading list, non word reading list and text reading.

Results: The CLP group showed a significant lower performance in comparison to the control group: speed is worse in word reading, and text reading tests, while accuracy is worse in the non word reading and text reading test. The 22% of CLP tested obtained values in line with Learning Disabilities (LD), compared with 6% of non CLP. Furthermore, 19% of CLP has obtained at least 2 values between 5° and 15° (borderline group), while in the control group was 6%. In addition the CLP group scored significantly lower than controls on some reading list tests. As expected, the CLP sample demonstrated a significant difference in the speed and accuracy parameters based on the school attended by the group. To this day, a neuropsychological assessment has been conducted on 6 of the 7 adults belonging to the CLP group that achieved values in line with learning disabilities. Five patients were certified with LD giving a prevalence of 16% in the CLP group in comparison to the 5% of the mean that we have in the Italian population while one has been certified with a low cognitive level and one decided not to participate in this follow-up.

Summary/Conclusion: The study’s results indicate that CLP have more difficulties in reading tasks than the non cleft population. The present project, begun as a screening for the over 16 yrs CLP population, suggests a need to consider children with isolated CLP outcomes as a population at risk for Learning Disabilities.
Playing, sharing and learning: a two days group intervention as a valuable treatment approach for children born with CLP?

M Ditlefsen¹, J Ottesen¹, T Særvold¹, H Tveit¹, R Kjerstad¹
¹Department of Speech and Language, Statped, Oslo, Norway

Background: As part of the routine examination of children born with cleft-, lip-, and palate (CLP), the Oslo team performs a psychological semi-structured interview at age 10. By assessing the child’s experiences of their environment, satisfaction with appearance, insight in their CLP, and emotional insight, we get a broader picture of the child’s psychosocial development and adjustment living with CLP. The screening tools Child Experience Questionnaire, Satisfaction With Appearance, Strength and Difficulty Questionnaire are administrated. The majority of children born with CLP show signs of normal psychosocial development. Some children, though, report negative CLP-related experiences and challenges coping with others’ reactions to visible scars or audible differences. We find that they often have a negative CLP-related self-perception, and lack adaptive strategies to deal with the reactions of their peers. Some children also report feelings of alienation and have never met others born with CLP.

Aims: To create a safe environment where children born with CLP can meet and share experiences about growing up with a noticeable difference. Facilitate the development of a positive self-perception. Increase the cognitive, emotional and behavioral flexibility by dealing with own and other people’s reactions to the cleft.

Methods: Each group consists of 5 to 7 children. Ideally, there is an equal number of boys and girls in every group. The children are selected based on the screening performed at the routine check-up at 10 years of age. When selecting participants we are looking for expressed tendencies of negative self evaluation or difficulties coping with others’ reaction. The program has parallel sessions for children and parents. Sessions with the children consist of different psychological founded games and education: role-play to practice social skills, psychoeducation on feelings and emotions and strategies and techniques to deal with worries. The theoretical framework for these sessions are mainly based on mentalization and metacognitive therapy. The parents have parallel sessions where they are invited to share experiences. A few weeks after the group intervention, each family receives a “mystery box” containing an individual letter where the two days are summarized, a golden envelope containing simple words of praise from the other children, and an USB stick containing a video about what they as a group have experienced and learned made by the CLP-team. They also receive an evaluation form that we encourage them to fill out and return to us.

Results: The response has been overwhelmingly positive from the participating children and parents. A recurring finding seems to be that both the parents and children in particular enjoyed meeting others in a similar situation. Also, the children reported that they feel more confident in how to respond to CLP-related attention.

Summary/Conclusion: The evaluations suggest that this type of group intervention is valuable for the children as well as for the parents. It leaves us with a hope that the group interventions might have a positive effect on the children’s coping and social interaction skills. Also, having emotional experiences with other children in similar situations might reduce the feeling of alienation. At the conference it will be discussed to what extent such a group intervention is a valuable treatment approach considering the above-mentioned findings, the cost benefit aspect and other related aspects of such a group intervention.
OS 20.6

Strengths and difficulties in children with cleft, lip and palate
M Klajic¹, M Linde², H Mark¹
¹Department of Plastic Surgery, Sahlgrenska University Hospital/Sahlgrenska Academy/Gothenburg University,
²Department of Psychology, Gothenburg University, Gothenburg, Sweden

Background: Previous studies have shown that children with cleft, lip and palate (CLP) often have other developmental problems such as delayed language development, learning difficulties, and attention problems. However, research in the psychological field is still limited.

Aims: The aim of the study was to assess psychological well-being and psychosocial skills in children treated for CLP.

Methods: Eighty-two children treated for CLP, 30 five-year olds and 52 ten-year olds, participated in the study during a clinical visit. Strengths and Difficulties Questionnaire (SDQ) was used to assess emotional problems, conduct problems, hyperactivity, peer problems and prosocial skills. Proxy-reports were used for the five-year olds, and for the ten-year olds both self- and proxy reports were registered.

Results: There were no differences between girls and boys. Two significant differences were found between self- and proxy-reports were the children reported higher points of total problems (P<.05) and a lower prosocial scale (P<.01). The total mean of emotional problems, conduct problems, hyperactivity and peer problems were within normal range, for both five-year olds proxy-report and ten-year olds self- and proxy-report. The strengths as measured by the prosocial scale was also within normal range for all groups.

Summary/Conclusion: Children with CLP do not seem to have a higher frequency of general psychological problems. We also conclude that it is of great importance to use both self- and proxy-reports, when possible, since we found them to be not consistent.
Attention-related functioning of children with non-syndromic craniosynostosis
M Kljajic¹, G Maltese¹, P Tarnow¹, P Sand², L Kölby¹
¹Department of Plastic Surgery, ²Department of Psychiatry and Neurochemistry, Institute of Neuroscience and Physiology, Sahlgrenska University Hospital/Sahlgrenska Academy/Gothenburg University, Gothenburg, Sweden

Background: Cognitive- and attention-related problems have been claimed to be common in patients with craniosynostosis. Since craniosynostosis is a rare condition, many studies face challenges with selection bias, small samples and wide age-ranges.

Aims: The aim of this study was to assess attention-related functioning of school children previously treated for non-syndromic craniosynostosis.

Methods: To reduce age range and selection bias, children that had previously undergone surgery for craniosynostosis and now had reached 8-16 years and lived within 200 km from the craniofacial center were invited. The Conner Continuous Performance Test 3rd Edition™ (Conner CPT 3™) was used for measurement of attention-related functioning.

Results: Sixty-five children (response rate 73.6 percent), aged 8-16 years, performed the test. The attrition analysis revealed no differences between responding and non-responding group regarding background variables. Minor differences between the sagittal synostosis (SS) and metopic synostosis (MS) groups regarding age and working memory were adjusted for.

Only one difference was found between the SS and MS groups; the SS-group performed significantly better at Hit Reaction Time (HRT) (P<0.05). When compared to the normative sample, the SS-group had significantly worse response style, detectability, omission, commissions, perseverations, HRT-SD, HRT isi Change (P<0.01) and Variability (P<0.05). The MS-group also had significantly worse detectability, commissions, perseverations, HRT-SD, Variability (P<0.01) and HRT isi Change (P<0.05) compared to the normative sample.

Summary/Conclusion: A sample of children previously operated for non-syndromic craniosynostosis, with narrow age range and small selection bias, underwent a test specific for attention. It revealed several minor short-comings compared to the normative sample both in the sagittal- and metopic synostosis groups. In general, the deviations were small.
The relationship of self-concept, parenting practices and mental health with academic achievement in children with cleft lip and/or palate

D Lopez Garcia¹, A Espinoza Leal²
¹Psychologist, Instituto Nacional de Pediatría, Ciudad de México, ²Psychologist, Clinica LPH Shriners, Tijuana, Mexico

Background: In Mexico, the cleft lip and/or palate (CLP) is usually approached on a priority basis from the medical side; first due to the urgency of a surgical intervention to correct the physical injury presented at birth and second due to the high incidence existing, leaving aside the concern to know the psychological impact, emotional and learning, so it is difficult to find research to address the associated problems that may arise in the development of the child with CLP.

Aims: Within the present research "The relation of the self-concept, the practices of parenting and the mental health with the academic achievement in children with cleft lip and/or palate" were analyzed the psychosocial variables such as: self-concept, practices of Parenting and mental health present in the development of the child with cleft lip and/or palate and its relation with the academic achievement, thus trying to answer the question ¿What is the relationship between self-concept, parenting practices and mental health with academic achievement in children with cleft lip and/or palate from 8 to 12 years of age?

Methods: A sample of 100 parents (86 mothers and 14 fathers) and 100 children (61 boys and 39 girls) were assessed, of the states of Baja California, Mexico City, state of Mexico, Oaxaca and Guanajuato, which have surgically corrected cleft lip and/or palate, with ages between 8 and 12 years studying elementary school level at the moment. To which they were applied the self-concept scale piers-Harris 2, inventory of practices of parenting, brief questionnaire of screening and diagnosis (CBTD), raven and scales of comprehension verbal (WISC-IV). In accordance with the Declaration of Helsinki and promulgated by the World Medical Association (WMA), the present investigation was submitted for approval by the ethics committee of the corresponding health institutions and the informed consent of the parents was obtained and of informed assent for the children.

Results: Correlation of academic achievement with self-concept, parenting practices and mental health. A multiple linear regression analysis was generated, to know the relation between the self-concept, the practices of parenting and the mental health with the academic achievement, adding also the two mediator variables (IQ and language) and thus determine if there is a significant relation. The results showed that, of the variables mentioned, only the mental health variable showed a significant negative correlation (r= -.322 p=.001) with the academic achievement variable, which indicates an association between mental health and achievement Child's academic. Since the correlation is negative, it is shown that the higher mental health problems present the lower the academic achievement obtained.

Summary/Conclusion: In this research, 63.5% of the sample reports normal self-concept levels and only 8% a low self-concept, related to their intellectual level and academic achievement, the 0% of the evaluated population does not present difficulties in the achievement academic, in relation to mental health, if there is the risk of presenting problems of internalization and/or problems of externalization (1-3).

It is considered important from the present research, to develop other studies in Mexico that allow to identify, to confirm and to characterize more precisely the difficulties present in the development of the children with CLP that supports in the integral management and multidisciplinary of this population.
Parental and health care professional views on psychosocial and educational outcomes in patients with cleft lip and/or cleft palate

M Stiernman¹, K Österlind¹, H Svensson¹, N Rumsey², M Becker¹, M Persson³

¹Department of Plastic and Reconstructive Surgery in Malmö, Sweden, Clinical Sciences in Malmö, Malmö, Sweden, ²Department of Health and Social Sciences, University of the West of England, Bristol, United Kingdom, ³Department of Health and Society, Högskolan Kristianstad, Kristianstad, Sweden

Background: Earlier research has investigated psychosocial and educational issues in populations of patients with cleft identifying several areas of concern (REF SHARIF). Health care professionals working with patients with visible differences have reported that 30-60% of their patients experience psychosocial challenges (Clarke and Cooper 2001, Rumsey, Clarke et al. 2004).

Aims: The aim of this study was to investigate current beliefs and knowledge about psychosocial and educational issues in parents and health care professionals (HCP) of children with Cleft Lip and/or Palate (CL/P).

Methods: Parents were interviewed concerning the view of society on people with visible differences, their child’s social life, educational progress and information regarding psychosocial care. Interviews with HCPs concerned characteristics related with educational achievement, behavior, and social relationships. Sixteen parents of children 9 to 13 years of age with CL/P and 10 HCPs and were interviewed. Data from interviews was analysed with thematic analysis.

Results: Eight children were reported to have emotional issues related to their cleft. Twelve parents however, did not perceive that their child was treated differently in society. HCPs expressed concerns regarding for example coping with being different, low self-esteem, shyness, disadvantage on first impression and acceptance of themselves. A majority of the HCPs did not think patient cognition, behavioural or physical development was specifically affected. It was noted by some HCPs that there was a lack of time to address such issues in their clinic, that they didn’t want to stigmatize the patients by asking about their psychosocial health or suggesting treatment and neither did they feel that this was ‘their job’.

Summary/Conclusion: The results revealed that parent experience and views were diverse - from no specific problems related to the cleft, to both emotional and educational issues. The beliefs and level of knowledge in HCPs also varied. Few HCPs appeared to be aware of the potential of counseling or psychological treatment for patients with cleft to improve psychological wellbeing, to alleviate the negative impacts of shyness or to improve levels of self-confidence (N. Rumsey and Harcourt 2014, Jenkinson, Williamson et al. 2015, Norman and Moss 2015). This lack of awareness could in turn lead to under-utilization of existing pathways of referral for psychological treatment. It may also reflect an unwillingness to stigmatize patients by suggesting they should be referred for further treatment. All HCPs, however, wished for more information and training regarding psychosocial issues.

Major themes are in line with earlier research and shed light on issues regarding the variability present in the subjective views of individual participants, and the importance of including a focus on the psychosocial health and educational progress of patients as an integral part of care (ACPA 2018).
Prevalence of reading difficulties in 9-10 year old children with cleft palate in Sweden

J. Weinfeld1, C. Persson2, J. Åsberg2
1Department of Otorhinolaryngology, Sahlgrenska University Hospital, Gothenburg, Sweden, 2Department of Neuroscience and Physiology, University of Gothenburg, Gothenburg, Sweden

Background: Previous research into the reading and writing skills of children with cleft palate have documented a higher prevalence of reading and writing difficulties in this population compared to the general population. Research shows a difference in risk of having reading and writing difficulties according to cleft type, where males with isolated cleft palate have a higher prevalence of reading difficulties. The higher prevalence of reading difficulties have also been connected to comorbidity. No population based study have previously been conducted examining the prevalence of reading difficulties in the cleft group. In Sweden the cleft-care is centrally organized which means that recruitment for studies are not as sensitive to sample-bias and are geographically and socially diverse. Earlier studies have utilized designs with clinical testing of subjects or retrospective clinical data designs, whereas the current study utilizes a parent-report of reading difficulties.

Aims: The aim of the current study was to investigate the prevalence of reading difficulties in children with cleft palate at ages 9 and 10 in Sweden. Additional aims were to investigate the prevalence of reading and writing difficulties according to cleft type and in comorbidity with other conditions. Correlations of reading difficulties with gender, adoption history and parental education were also conducted.

Methods: Parents of children born with cleft palate 2008 and 2009 and treated at four out of six regional cleft centres in Sweden were asked to participate in the study. A total of 246 potential families were identified. No specific exclusion criteria were adopted. A parent report concerning reading and writing skills together with a separate questionnaire including background questions and consent form was posted to potential participants from each respective center. 139 families (57%) responded and returned the parent report, background questionnaire and consent form. The parent report consisted of seven questions regarding their child’s reading and writing skills. The parent report has previously been validated in two Swedish studies and significantly correlated with clinical reading and writing tests. The definition of clinical reading difficulties was set to ≥8 points on the parental report and represented the 10th percentile.

Results: Of the 139 subjects 88 boys; 51 girls. Cleft type distribution (n=137): 58 Unilateral CLP; 38 bilateral CLP and 41 Isolated CP. Chronbach’s alpha indicated excellent internal consistency (α = .91). Great variability on the total score of the parent report was observed with max and min scores represented (0-14 p). M: 3.54, SD: 4.21. Utilizing the clinical high cut off (≥8) the results revealed that 21% (95 CI: 15-30%) prevalence of reading difficulties in the cohort. This is clearly greater than in the general population. In order to identify important correlates to reading difficulties, a series of Chi-square tests were conducted. Parental education, adoption history, and gender showed no significant associations with reading difficulties (p > .14). Isolated cleft palate was associated with reading difficulties with a prevalence of 40%. Reported comorbidities in the total group was also marginally associated with reading difficulties (p = .084).

Summary/Conclusion: The results corresponds to earlier research with a higher prevalence of reading difficulties in the group compared to the general population. Higher prevalence in the isolated cleft group could also be confirmed.
**Esthetical Outcome (Standards)**

**OS 21.1**

*Why it is difficult to create a validated cleft aesthetic outcome measure.*

B Richard

**Abstract Content:** Asher McDade is commonly used for assessing facial aesthetic outcomes in cleft children. However, it has weak validity compared with the facial growth and speech outcome methods. National cleft teams and Randomised Controlled trials are using outcomes to compare surgical techniques, protocols, timings, surgeons and units. The Cleft Collective UK repeated the 1998, CSAG study of 5-year olds with UCLP, born 5-7 years after the establishment of a National cleft service, which showed improvements in cleft care in regard to facial growth and speech but minimal change in facial aesthetic results. Several teams have tried to improve the validity by using yardsticks of suggested outcomes for the scorers to use, (Dutch Cleft & Americleft) and others hope that a 3-D or 4-D system will produce an outcome measure that is clinically useful for comparing results. The Current Asher Mc-Dade system has been deconstructed to try and analyse the reasons for the weak Kappas and we now know that the raters themselves are a cause of bias in choosing from a Likert scale of 1-5, (excellent, good, fair, poor and very poor), when looking at 2-D images. Cropping the image, trapezoid or circle, and reversing all right sided to clefts to look like left sided ones removes some unconscious observer bias. Using computer programmes to enable scoring means less human error occurs. The lip appears to dominate the opinion of the rater if they can see the lip whilst scoring the nose. A few humans rate almost randomly, and 10% can ‘never see’ an excellent and a similar percentage can ‘never see’ a poor result. Increasing the numbers of raters does not improve reliability as these bias’s remain. Training raters, or screening out biased raters, has not been tested, but might improve reliability. Cleft professionals, lay people and patients are similar in scoring abilities. Crowd sourcing gains large numbers of raters and the result is as valid as professionals. SymNose, is a computer program that provides a semi-objective measure of lip/nose symmetry, created by tracing the lower border of the nose and upper lip on a digital track pad and software to analyse the symmetry. Using results from this technique, removes the human bias of right versus left, and excellent versus very poor ‘blindness’. Further use of computer graphics technology to automate the process of symmetry analysis is being developed such that 2-D photos can be scanned and an outcome delivered with no human opinion involved!
The cleft aesthetic rating scale.
Measuring aesthetic outcome after unilateral cleft lip repair.
P Don Griot

Abstract Content: The goal of unilateral cleft lip repair is to create nasolabial symmetry and to correct the deformed nasal structures. In order to assess and compare the outcome of cleft lip and palate surgery it is essential to have a reliable scoring system. However, for the assessment of aesthetic outcome of cleft-related facial deformities, a widely accepted, reliable scoring system is not available. One of the most frequently used scoring systems in literature is the system proposed by Asher-McDade, which uses frontal and lateral views. However, this scoring system is complicated and time-consuming which results in a significant burden when assessing a large number of photographs. In this presentation the development of the Cleft Aesthetic Rating Scale is described, its advantages, lessons learned and possible pitfalls. The importance to reach for an international consensus on an esthetical assessment tool is stressed and the use of a multiphase Delphi method approach to achieve this is, will be discussed.
An instrument to provide specific, actionable evaluations of nasolabial aesthetics after unilateral cleft lip repair

T Sitzman, A Allori, S Beals, M Bezuhly, D Fisher, J Marcus, D Matic, T Samson, R Tse

Duke University, Durham, Barrow Cleft and Craniofacial Center, Phoenix, United States, Dalhousie University, Halifax, The Hospital for Sick Children, Toronto, University of Western Ontario, London, Canada, Penn State Hershey Medical Center, Hershey, Seattle Children's Hospital, Seattle, United States

Background: Existing methods for evaluating nasolabial aesthetics after unilateral cleft lip (UCL) repair do not identify the specific components of a surgeon’s technique that consistently achieve excellent results or the areas where a surgeon can improve.

Aims: To develop an instrument for evaluating nasolabial aesthetics after UCL repair that provides specific, actionable information to guide development of each surgeon’s approach to UCL repair. Methods: A panel of nine surgeons met regularly over a three-year period to develop a component-based instrument. Development began with defining the instrument’s purpose and then proceeded through conceptualizing the individual objectives of UCL repair, developing questions to elicit evaluation of each objective, testing of an initial instrument, revising the instrument including addition of reference images, testing of a revised instrument, and production of a final scale. Intra- and inter-rater agreement was evaluated with the kappa statistic. The panel then used the scale to conduct an audit of each surgeon’s UCL repairs.

Results: An instrument was developed that assesses eleven distinct objectives of UCL repair: nasal tip symmetry, equal nostril circumference, symmetric nostril shape, correction of columnella angulation, alar base symmetry, balancing of Cupid’s bow, level vermillion-cutaneous junction, level vermilion-mucosal junction, level free border of the lip, satisfactory scar form, and optimal scar healing. Success in achieving these objectives is assessed for individual patients using cropped frontal and worm’s eye photographs to answer eleven questions. Intra-rater agreement for the questions ranged from moderate to very good (median kappa=0.71, range 0.55-0.81). Inter-rater agreement ranged from fair to good (median kappa=0.46, range 0.27-0.72). Six surgeons used the scale to audit their UCL repairs as part of a continuing medical education course. All surgeons found the experience an excellent educational activity and very effective in evaluating their UCL repair outcomes; all surgeons identified specific components of their repair they would change.

Summary/Conclusion: A new instrument for evaluating nasolabial aesthetics is proposed that evaluates attainment of individual objectives of UCL repair. Surgeons who conducted a self-assessment using this component-specific approach rated the experience positively and identified specific, actionable opportunities for improvement.
Cleft Surgery in Developing Countries

OS 22.2

Comprehensive management of cleft deformities in India. What is needed in the future?

K Bonanthaya

Cleft lip and palate deformities occur, 1 in about 800 live births in India. This translates to about 35,000 new cases being added in the country every year. Although there is Public and Private health sector involvement in the care of these patients, currently it is the NGO driven efforts that deliver most of the care provided to these patients. Not only has this been able to reach most of the nook and corners of the country, but also has started to focus on Comprehensive care, in the recent past. However this is by no means universal, or comprehensive in all instances. There are a number of issues that need to be addressed if this has to become a reality i.e., Universal and Comprehensive care for all patients. Obviously top amongst the priorities will be financial resources. A much larger amount than being spent now, needs to be available and has to be found. Lack of adequately trained carers, particularly in the disciplines of Speech Pathology and Orthodontics is a huge stumbling block in this effort. Logistical problems which leads to patient dropout needs to be tackled with the help of efficient and economical technologies. Clinical research pooling the vast amount of data, leading to a better understanding of outcomes in these circumstances is of great importance. This also is essential to modify current protocols which then should result in a significant reduction in the burden of care while providing optimum outcomes. About 35000 new cases of clefts are being added in the country every year. Currently it is the NGO driven efforts that delivers most of the care provided to these patients. Not only has this been able to reach most of the nook and corners of the country, but also has started to focus on Comprehensive care, in the recent past. However this is by no means universal, or comprehensive. There are a number of issues that need to be addressed if this has to become a reality. Obviously top amongst the priorities will be financial resources. Lack of adequately trained carers, in the disciplines of Speech Pathology and Orthodontics is a huge stumbling block in this effort. Logistical problems which leads to patient dropout needs to be tackled with the help of technology. Clinical research pooling the vast amount of data, leading to a better understanding of outcomes is of great importance. This also is essential to modify current protocols which then should result in a significant reduction in the burden of care.
Sustainable cleft care through education: simulation-based comprehensive workshops
U Hamdan

Abstract Content:
Background/Purpose: Cleft lip and/or palate (CLP) affect nearly one in 500-700 births, and lead to increased morbidity and mortality if untreated. Nevertheless, significant global disparities in access to care still exist. The relatively basic infrastructure required to surgically correct these deformities and large unmet disease burden, have resulted in a significant number of foundation-based cleft care clinical initiatives in developing countries. In addition to clinical activities, these foundations have the unique opportunity to contribute to sustainable cleft care through educational initiatives.

Methods/Description: Global Smile Foundation (GSF) is a non-profit organization dedicated to providing free comprehensive multidisciplinary cleft care to individuals born with CLP around the world. GSF providers and practitioners have been providing comprehensive clinical care to patients with CLP for over 3 decades. The GSF model of care consists of surgical mission trips to developing countries with an unmet cleft care need, with the ultimate goal of establishing local cleft care centers and training local surgeons and other cleft care providers for long-term sustainability.

Results: Relying on our commitment to provide free comprehensive cleft care to individuals with CLP in developing countries and train the next generation of cleft surgeons in these countries, we have organized the first simulation-based comprehensive cleft care workshop in the Middle-East and North Africa (MENA) region. The workshop included close to 100 participants from over 20 countries. All participants reported high satisfaction with all aspects of the workshop. All participants reported they would recommend it to colleagues (100.0%) and participate again (100.0%). Based on the success of the workshop, we have organized a second international simulation-based comprehensive cleft care workshop including world authorities in cleft care, to be held in Lima, Peru: October 16-18, 2019.

Conclusions: Multidisciplinary simulation-based cleft care workshops are well received by cleft practitioners in developing countries, can serve as a platform for intellectual exchange, and are only possible through strong collaborations. Stakeholders invested in providing comprehensive cleft care should translate these successes from the regional to the global arena in order to contribute to sustainable cleft care through education.
Predictors of complication following cleft lip and palate surgery in a low-resource setting: a prospective outcomes study in Nicaragua

J Swanson¹, M Swanson¹, A Auslander², T Morales³, A Siu³, W Magee III⁴, R Ayala⁵
¹Operation Smile Nicaragua, Managua, Nicaragua, ²University of Southern California, Los Angeles, United States, ³Operation Smile, Managua, Nicaragua, ⁴Children’s Hospital Los Angeles, Los Angeles, ⁵Operation Smile, Virginia Beach, United States

Background: Higher rates of postoperative complication following cleft lip or palate repair have been documented in low resource settings, but their causes remain unclear.

Aims: This study sought to delineate patient, surgeon, and care environment factors in cleft complications in a low-income country.

Methods: Candidate patients presenting for cleft lip or palate repair or revision in a Nicaragua comprehensive cleft care program were enrolled and followed prospectively. Patient anthropometric, nutritional, environmental and peri- and postoperative care factors were collected. Post-operative evaluation occurred at standard 1-week and 2-month postoperative intervals. Complication was defined as fistula, dehiscence and/or infection.

Results: Among 408 patients enrolled, 380 (93%) underwent surgery, of which 208 (51%) underwent lip repair (124) or revision (84), and 178 (48%) underwent palate repair (96) or revision (82). 322 (85%) were evaluated 1 week and 166 (44%) 2 months postoperatively. 50 (16%) complications were identified, including: 25 (8%) fistulas, 24 (8%) dehiscences, 17 (5%) infections. Mid-upper arm circumference (MUAC) ≤12.5cm was associated with dehiscence after primary lip repair (OR=28, p=0.02). Leukocytosis ≥11,500 on pre-operative evaluation was associated with dehiscence (OR=2.51, p=0.04) or palate revision fistula (OR=64, p<0.001). Surgeons who performed fewer than 25 previous-year palate repairs had higher likelihood of palate complications, (OR=3.03, p=0.01) although there was no difference in complication rate with years of surgeon experience or duration of surgery.

Summary/Conclusion: Multiple patient, surgeon, and perioperative factors are associated with higher rates of complication in a lower-resource setting, and are potentially modifiable to reduce complications following cleft surgery.
**OS 22.5**

**Narrative ability and verbal working memory in internationally adopted children with uclp**

A Larsson¹, I Johansson¹, L Sandström¹, C Persson¹

¹Speech and Language Pathology Unit, Department of Health and Rehabilitation, Institute of Neuroscience and Physiology, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden

**Background:** During 2007-2010 European CLP centres had a large increase of patients that were internationally adopted (IA). Many children had open palates at arrival and a majority of them were adopted at age 2 years and older. Previous studies have found severe speech impairments, mainly with velopharyngeal impairment but also with restricted speech production. A few studies have described the risk for language impairment as a suspected consequence of these early speech impairments. Narrative ability (NA) and verbal working memory (VWM) are closely intertwined and are considered to be important areas of core language functions. Assessing narrative ability is also reported to be an ecologically valid measure to capture overall language function and communicative ability in all children. However, this is not fully studied yet and there is a lack of studies on language abilities in this group of children.

**Aims:** To investigate narrative ability and verbal working memory in a group of IA children with unilateral cleft lip and palate (IA+UCLP) and compare with a group of IA children without cleft lip and palate (IA).

**Methods:** Two groups of participants were recruited. Twenty-seven IA children with UCLP were recruited from two CLP centres and via national adoption agencies in Sweden. These children were all adopted from China at ages of 0:10 - 6:1 years and had a mean duration of exposure (MDE) to Swedish of 6:9 years (min-max 2:4 – 7:5) at time of assessment. A comparison group of 29 IA children without CLP was recruited via national adoption agencies. They were adopted from countries in Africa, Asia and Europe at ages between 0:7-4:7 years and had a MDE to Swedish of 7:0 years (min-max 2:3 – 7:10) at time of assessment.

Language assessment was performed at a mean age of 7:7 and 7:9 years for each group respectively. The Bus Story test (Information score, Sentence length and Subordinate clauses) was used to capture narrative ability. The subtest Number repetition from the CELF-IV was used to assess VWM.

**Results:** No significant differences were found between groups regarding NA. However comparing the results from both groups to the Swedish norms, showed that their results were equivalent to children at 5:0-6:1 years of age on all three scores. Regarding VWM, no significant differences were found between groups. However, only ≈15% of children in both groups scored at expected levels. In both groups many children (IA+UCLP 41% and IA 38%) performed -2 SD below the mean for their age groups.

**Summary/Conclusion:** The present study aimed at investigating narrative ability and VWM in IA children with UCLP and compare with IA children without CLP. No statistically significant differences between groups were found. However, compared to Swedish test norms, a majority of children in both groups scored low and at levels of much younger children. IA children with UCLP are in need of broad assessments of both speech production and language abilities. Assessing narratives and VWM should be included in those assessments. Results from the present study suggests that IA children are in great risk of having language impairments.
The impact of short-term reconstructive surgical missions: a systematic review
T Hendriks¹, M Botman¹, C Rahmee², J Ket³, M Mullender¹, B Gerretsen³, E Nuwas⁴, K Marck⁵, H Winters¹
¹Department of Plastic, Reconstructive and Hand Surgery, ²VU medisch centrum, ³KIT royal tropical institute, Amsterdam, Netherlands, ⁴Surgery department and director, Haydom Hospital, Haydom, Tanzania, United Republic of, ⁵Department of Plastic Surgery, Medisch Centrum Leeuwarden, Leeuwarden, Netherlands

Background: Short-term missions providing patients in low-income countries with reconstructive surgery are often criticised because evidence of their value is lacking.
Aims: This study aims to assess the effectiveness of short-term reconstructive surgical missions in low- and middle-income countries.
Methods: A systematic review was conducted according to PRISMA guidelines. We searched five medical databases from inception up to 2 July 2018. Original studies of short-term reconstructive surgical missions were included, which reported on patient safety measurements, health gains of individual patients and sustainability. Data were combined to generate overall outcomes, including overall complication rates. PROSPERO registration: CRD42018099285.
Results: Of 1662 identified studies, 41 met full inclusion criteria, which included 48,546 patients. The overall study quality according to Oxford CEBM and GRADE was low. Ten studies reported a minimum of six months follow-up, showing a follow-up rate of 56.0% and a complication rate of 22.3%. Twelve studies that did not report on duration or follow-up rate, reported a complication rate of 1.2%. Fifteen out of 20 studies (75%) that reported on follow-up, also reported on sustainable characteristics
Summary/Conclusion: Evidence on the patient outcomes of reconstructive surgical missions is scarce and of limited quality. Higher complication rates were reported in studies which explicitly mentioned the duration and rate of follow-up. Studies with a low follow-up quality, might be underreporting complication rates and overestimating the positive impact of missions. This review indicates that missions should develop towards sustainable partnerships. These partnerships should provide quality aftercare, perform outcome research and build the surgical capacity of local healthcare systems.
An integrated approach has been used in repairing the unilateral cleft lip nasal deformity by the author since late 90’s. The approach includes: (1) presurgical nasoalveolar molding, (2) modifications in surgical techniques and (3) postoperative care and maintenance. The presurgical nasoalveolar molding can improve the symmetry in skeletal base as well as the alar shape. A modified device using the spring mechanism can reduce the numbers of clinical visits for nasoalveolar molding thus greatly reduce the burden of care for the parents. The key steps in surgical techniques are: (1) several mucosal flaps for nasal floor reconstruction, correction of mucosal deficiency in piriform area, (2) mobilization of alar base for alar base repositioning, (3) muscle release and reconstruction, (4) anchoring of advancement flap to nasal septum to maintain the position of alar base, (5) semi-open rhinoplasty with reverse U incision on the cleft side and rim incision on the non-cleft side, (6) atraumatic dissection to release the fibrofatty tissue from lower lateral cartilages, (7) advancement and fixation of the cleft side lower lateral cartilage to the non-cleft side lower lateral cartilage and to the skin in an over-corrected position, (8) primary septal cartilage graft, and (9) definition of the ala-facial groove with alar transfixion sutures. A set of asymmetric silicone nasal stents are used for maintenance of the over-corrected nasal shape. These changes have resulted in a more consistent and better results thus greatly reduce the need for secondary revisions in children in preschool age or even adolescences.
**OS 23.2**  
**Nasal resistance in cleft lip and palate patients: a computational fluid dynamics study.**  
L Pimenta¹, E Lages², J Kimbell³, A Drake³  
¹Dental Ecology, Craniofacial Center - UNC at Chapel Hill, Chapel Hill, United States, ²Orthodontics, Federal University of Minas Gerais, Belo Horizonte, Brazil, ³Department of Otolaryngology/Head and Neck Surgery, School of Medicine - UNC, Chapel Hill, United States  

**Background:** Cleft lip and palate (CLP) causes morphological and geometric deformities in the facial, nasal and oral structures. Such alterations may change the nasal airflow and increase nasal resistance. The recent application of computational fluid dynamic (CFD) modeling in medicine and specifically in otolaryngology has opened new pathways to evaluate physiologic changes within the human airway. It can be used to characterize and compare the effect of upper airway geometry on nasal resistance in both CLP and non CLP subjects. Geometric variation of the nasal cavity in CLP individuals, noting the airflow pattern which is constructed based on realistic nasal cavity models, has not been previously studied.

**Aims:** The purpose of this study was to compare nasal resistance in unilateral and bilateral CLP patients with controls using CFD models.

**Methods:** De-identified CT scans from patients with CLP who underwent CBCT imaging were obtained at a regional craniofacial center (n=20: 13 UCLP and 7 BCLP). The control group (n=15) comprised of patients who had received CT imaging for conditions unrelated to the upper airways. All DICOM (Digital Imaging and Communications in Medicine) files from CBCT images were imported using Mimics 19.0 software and 3D reconstructions were made. The nasal airway was properly captured in the 3D models, including accurate shapes of the inferior, middle, and superior turbinates and nasopharynx. The airflow allocation was assessed for each 3D model using CFD model through Fluent 14.5. Analysis of the CFD simulations was performed, measuring the flow rate, pressure and resistance in the choanae plan. Data were tested for their normality and analyzed by Mann-Whitney Test. The level of significance was p<0.05.

**Results:** The mean volumetric flow rate in choanae plan for CLP group was 187.24 Q and control group was 222.55 Q (p=0.000). The mean pressure in choanae plan for CLP group was 30.65 Pa and the control group was 21.04 Pa (p=0.729). The mean nasal resistance in choanae plan for CLP group was 0.17 and the control group was 0.10 (p=0.353).

**Summary/Conclusion:** This is one of the first studies to compare nasal airflow in CFD models in CLP and normal individuals. Since anatomic changes in CLP alter nasal airflow, a computational fluid dynamics (CFD) study assessing air dynamics was crucial, demonstrating differences in flow rate. Nasal resistance was not different when comparing groups. Correlation of airflow alteration, pressure and nasal resistance measurements in the context of subjective symptoms would allow for a deeper appreciation of the alterations in the pediatric CLP.
Background: 3-dimensional midfacial hypoplasia and asymmetry in cleft patients are common and are frequently connected to impairment of the aesthetic facial appearance. Restriction of maxillary growth and consecutive skeletal class III malocclusion, as well as lacking paranasal and malar prominence may lead to a concave facial profile. The lack of paranasal and subnasal hard-tissue support may further contribute to the typical nasal appearance in cleft patients, such as impaired nasal tip projection and displacement of the alar base. A wide range surgical approaches to correct and augment relevant facial regions, including orthognathic surgery, cleft rhinoplasty and autologous tissue transfer, are available. Alloplastic facial implants have been established as an adjunct to orthognathic surgery and a viable alternative to autologous tissue augmentation in various circumstances. However, in cleft patients, the application of facial implants has rarely been reported.

Aims: This retrospective study aimed to evaluate the use of Medpor implants for midfacial contouring in cleft patients.

Methods: A retrospective analysis regarding the use of Medpor implants in cleft patients was conducted. Male and female patients with orofacial cleft deformities having been treated with at least one Medpor implant to correct midfacial hypoplasia and specific asymmetry were included. Patients with insertion of other solid alloplastic implants (Goretex, Silicone) were excluded. The surgical technique used, implant distribution and combination of implants, surgical pathways, combination of implant insertion with other cleft-related surgical procedures and age at implant insertion were evaluated.

Results: 51 patients with orofacial clefts were assessed with regard to defined parameters. A range of Paranasal, Malar and Nasal Dorsum Medpor implants have been used (122 implants). UCLPs represented the most common indication for Midfacial implant insertion, followed by BCLPs. Bilateral implant insertion was done as a general rule with few exceptions. Insertion of implants was frequently combined with other cleft-related surgical procedures. Most commonly implant placement was combined with primary or revision rhinoplasties and corrective procedures of the upper lip. Different surgical pathways with regard to Medpor implant insertion were identified. 1. Implants were applied as a temporary measure in adolescent cleft patients, who were psychologically affected by their hypoplastic facial appearance, to bridge the time to definitive correcting/orthognathic procedures. 2. Implants were applied in adult patients who did not choose to undergo orthognathic surgery, or who were not affected by skeletal malocclusion. 3. After orthognathic surgery, midfacial augmentation was implemented to specifically address residual volume deficiency, particularly in the malar region. The complication rate amounted to 4.9% (6/122 implants).

Summary/Conclusion: Based on our findings, Medpor implants are reliable and long-term stable materials to successfully augment paranasal, subnasal and malar areas as well as a solid nasal dorsum material with few complications in cleft patients.
Correction of caudal septum in secondary deformity of unilateral cleft lip and nose at preschool age population

J Kwon¹, K Koh¹

¹Department of Plastic and Reconstructive surgery, Asan Medical Center, Seoul, Korea, Republic Of

Background: Regarding the treatment of unilateral cleft lip and nose, the medialization of the columella is one of the key factors for success. To achieve optimal results, correction of skeletal structures, most importantly the nasal septum, should be performed along with soft tissue rearrangement. It is universally acknowledged that repositioning the caudal part of the dislocated septum during the initial cheiloplasty is beneficial for unilateral cleft lip and nose patients.

Aims: The purpose of this article is to evaluate the efficacy of septal repositioning during the secondary nasal correction in the preschool age population, with regards to nasal asymmetry.

Methods: 130 consecutive patients who underwent secondary nasal correction for secondary deformity of unilateral cleft lip and nose between the ages of 4.5 and 6.5 years old at Asan Medical Center from October 2009 to November 2017 were analyzed. Both primary correction and secondary correction were performed by a single surgeon, KS Koh. The study population was divided into two groups according to the two different operative methods, used within the study period. Group 1 consisted of 65 patients who underwent the septal repositioning procedure during the secondary nasal correction, the method used from August 2015 on words. For septal repositioning, the perichondrium of the caudal septum was dissected through the hemitransfixion incision at the non-cleft side. After relocation of the caudal septum to the cleft side, the septum was fixed to the anterior nasal spine using a 5-0 polydioxanone suture. Group 2 consisted of 65 patients operated on without septal repositioning. Using two dimensional photography, which were taken preoperatively and one year postoperatively, alar width (the distance from the midpoint of the columella base to the lateral alae in anterior posterior view), deviation of columella (the angle between the columella and the vertical axis to the line between the medial canthus), nostril width, nostril height, and columella height were analyzed. The independent t-test was used in statistical analysis to compare measurements between groups.

Results: The mean follow up period was 420 days. The average columella angle deviation decreased from 6.3(3.7) degrees to 1.2(1.1) degrees at group 1 and from 6.4(4.9) degrees to 4.7(3.7) degrees at group 2. The difference between groups was statistically significant.(p<0.05) The average nostril height ratio (cleft side/non-cleft side) improved by 16.7(14.2)% at group 1 and 12.1(10.9)% at group 2, which was also statistically significant.(p=0.041) Regarding the alar width, nostril width and columella height, there was no statistical significant difference.

Summary/Conclusion: This preliminary report details initial results on the efficacy of septal repositioning in the unilateral cleft lip patient requiring secondary nasal correction at preschool age. At one year follow up, septal repositioning in the preschool age population provides an effective method to medialize the columella. To ensure safe nasal growth and maintenance of postoperative outcome, these patients will additionally need long-term follow up.
OS 23.5

Long term follow-up in refinements of secondary cleft lip and nose reconstructions treated with simultaneous use of cartilage, dermal, fat and bone grafts.


1Plastic Surgery, ST. Joseph’s University Med. Ctr., 2Plastic Surgery, St. Jospeh’s, 3Plastic Surgery, St. Joseph’s, Paterson, United States, 4Plastic Surgery, Ospedali di Piacenza, Piacenza, 5Plastic Surgery, Ospedali Perugia, Perugia, Italy, 6Plastic Surgery, St. Joseph’s Univ., 7Plastic Surgery, St. Joseph’s Hospital, Paterson, United States

Background: Secondary cleft lip and nose correction is often challenging because of the long-standing and severe associated deformities. Hypoplasia of the maxilla and the pyriform rim, “slanting” and deformity of the affected hypoplastic lower lateral cartilage, anomalous insertion of the surrounding muscles and severe soft tissues deficiencies are the major stigmata to be corrected, if possible, in a single surgical intervention.

Aims: Aim of our study is to present our anatomically-oriented approach with the simultaneous use of multiple grafts in order to improve outcomes, reducing the reoperation rate.

Methods: The technique has been used in 17 patients from 2013 to 2015. Mean age was 16 and male to female ratio was 9:8. Informed consent was obtained before surgery. We repaired the cleft nose first and then the lip to avoid the “locking down effect” of the repaired lip. Dermal grafts (from the bone graft incision), fat grafts, cartilage grafts (chonchal, costal and/or septal) and bone grafts were all used in one stage to correct the deficiency of the pyriform rim, the affected alar cartilage, the upper lip and the surrounding soft tissues. The grafts were inserted in separate layer to facilitate graft take. Antibiotics were given in the perioperative period. During follow-up visits symmetry, volume and projection were measured with quantitative anthropometric assessments.

Results: Mean follow up was 4 years. No complications occurred. All patient showed and maintained adequate symmetry and appropriate volume augmentation. Nose projection and alar cartilage position appeared to be reinstated. Bone augmentation at the piriform rim appeared to give a solid scaffold to support the affected ala position and the tip support. Augmented soft tissues with fat and dermal grafts were stable over time. Lip length was appropriate in all cases and did not require further revisions. According to our long-term quantitative anthropometric data graft resorption was minimal.

Summary/Conclusion: Adequate use of multiple grafts in secondary cleft lip and nose surgery can improve outcomes and reduce reoperation rate. Performing the cleft-rhinoplasty first and giving support to the deficient alar base prevents recurrence of the nose anomaly and allows adequate lengthening of the reconstructed lip. Augmenting the upper lip with grafts also gives reliable tissue to adequate lengthening of the deficient soft tissues. Our long-term anthropometric data seem to confirm the adequacy of our surgical approach.
**OS 23.6**

**Nasal flap extended. A new technique**

**E Alvarez¹, D Alvarez¹**

¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

**Background:** The treatment of a palatine fistula is a challenge even for an experienced surgeon, the lack of adequate tissue around the hole makes many attempts to rebuild it become flawed and determine a larger defect than the original. Local hinge flaps have been applied widely with good results and sometimes with failure. The extended nasal flap taking advantage of the basic principles of reconstructive plastic surgery referring to the revascularization of the edges of the flap and thus obtaining additional tissue has incorporated it into this technique.

**Aims:**

When the edges of the palatal tissue and the nasal mucosa heal by second intention after an infection, dehiscence or necrosis of the previous surgery that determined a fistula, the same cicatricial edges will allow the reappearance of arterial flow between the two mucous membranes, nasal and palatal. restore circulation, as it was known in the principles of tissue transfer as described by Gilles and Filatov. In this way palatal tissue is added to a nasal pedicle for purposes of adequate repair of the nasal mucosal plane.

**Methods:** Six patients between 7 and 32 years were selected with multiple attempts to repair fistula that failed and on average their sizes ranged from 1 x 1.5 cm to 5 x 3 cm. After they or their representatives voluntarily and informed agreed to undergo the procedure, they underwent surgery under general anesthesia where they first proceeded to determine the amount of palatal mucosa necessary to close the defect, equidistant to the midpoint of the fistula, after an infiltration of lidocaine with epinephrine at 1 per 100,000 and 7 minutes of waiting for latency, pedicle flaps were elevated to pedicle level. the nasal mucosa, and continuous submucosal dissection on the nasal side until reaching the lateral walls of the nose below the inferior turbinate, which achieved an adequate repair and free of tension of the nasal floor with the extended nasal flap. After this first repair phase, the oral side of the fistula was covered with mucosal palatal flaps, buccinator flaps or reverse flow lingual flaps as we have reported in another paper.

**Results:** Of the six cases taken for the report of this work, the resolution of the integral fistula was achieved in 5 cases and partial resolution in one of them due to dehiscence evidenced 5 days after their primary surgery, the same case evolved with healing by a second intention. on the nasal plane adequately repaired.

**Summary/Conclusion:** The ideal goal for the repair of a cleft palate is to redo in two mucosal planes, both nasal and oral, the same objective seeks the repair of a fistula. Undoubtedly the repair in a single palatal mucosal plane in any of the two intentions has a high risk of failure. Getting adequate tissue for the repair of the nasal side of a primary cleft palate surgery or for a recurrent fistula is the key to success, it has been determined in some studies that adequate repair of the nasal plane provides 80% safety in surgery.
**Background:** The repair of nasal deformities in cleft patients remains most challenging. The complexity of nasal deformities is caused by the inborn deformity and by prior surgical corrections. Nasal deformities in unilateral and bilateral CLP patients are typical.

**Aims:** To rate functional and aesthetic outcome of cleft septorhinoplasty.

**Methods:** From 2002 to 2017 more than 450 cleft nose repairs were performed by a single surgeon, 340 complete septorhinoplasties in the age of skeletal maturity, more than 110 corrections in childhood and adolescence due to severe functional or aesthetic problems including columella lengthening procedures. For final correction open approach is the standard procedure. The septum was addressed through an interdomal approach. Septal cartilage is harvested as necessary, spreader grafts are often used to preserve septal straightening, the severely deformed septum can be completely removed with ex vivo creation and replacement of an L-strut. The dorsum is addressed by rasping or osteotomy. We changed the regime from internal to external osteotomies. Diced cartilage was used in 75% of the cases. If needed radix augmentation is achieved with diced cartilage in fascia. The correction of the nasal tip includes creating a symmetrical platform using existing lower lateral cartilages, septal extension grafts and augmenting with tip grafts. For outcome rating a morphometric analyses and a rating by a neutral jury of 90 medical professionals and non-professionals was performed.

**Results:** All patients with compromised nasal ventilation reported about improvement of nasal ventilation after surgery. In a follow up investigation 88 % of the patients were satisfied with the aesthetic outcome. A neutral jury rated the preoperative situation as aesthetic good in 9%, in 91 % as aesthetic bad, the postoperative result was rated as good in 85 % and in 15 % as not good or bad. Morphometric measurements demonstrated a significant improvement in all parameters, however remaining asymmetries were still seen depending on the degree of deformity.

**Summary/Conclusion:** Complete analysis of the nasal functional and aesthetic deformity as well as analysis of the facial pathology are the key to for successful management of the cleft nasal deformity. Initial closed rhinoplasty together with lip repair is helpful in reducing the nasal asymmetry in childhood. The results of final cleft septorhinoplasty using a standard algorithm with use of septal or rib cartilage are good. The patient should be told that a second corrective procedure might be indicated.
Background: The treatment of patients affected by unilateral cleft lip and palate (UCLP) is based on a multistage procedure of surgical and nonsurgical treatments in accordance with the different types of deformity. The septal deformity of UCLP is well described and involves convexity of the cartilaginous and bony septum towards the cleft-side, preventing the passage of the air, and deviation of the caudal and anterior margin toward the noncleft-side. Enlargement of the inferior turbinate on the noncleft-side has been reported in these patients. Inferior turbinate hypertrophy is related to increased airflow through the unobstructed nostril, which triggers a reactive hypertrophy in the turbinate mucosa. The management of the nasal deformity in patients with UCLP has traditionally focused on the appearance of the nose, but, in these patients, even the restoration of the normal nasal physiology should be a major objective.

Aims: The authors introduced a primary rhinoseptoplasty during the cheiloplasty since 2008. The aim of the present study was to assess, not only the nasal deformity of the septum and nasal shape symmetry, but even the inferior turbinate size and the degree of nasal airway patency, in patients who have undergone primary correction of the nasal septum during lip repair, compared to patients operated on without primary septal surgery.

Methods: The authors compared two groups: group A, which underwent septoplasty during cleft lip repair, and group B, which did not. A physical examination with relief of different anthropometric measurement was used to assess the nasal shape symmetry. Moreover the Nasal Obstructive Symptom Evaluation (NOSE) scale and nasal endoscopy were used to evaluate the nasal obstruction.

Results: After the anthropometric evaluation of the two groups, the authors observed better symmetry regarding nasal shape, correct growth of the nose, and a strong reduction of the nasal deformity in the patients who underwent primary JJ septum deformity correction. In reference to nasal physiology, the untreated group reported severe symptoms across all NOSE scale dimensions more frequently than children who have undergone primary rhinoseptoplasty; furthermore the turbinate size decreased significantly compared to pre-operative data and even between the two groups there is a statistically significant difference.

Summary/Conclusion: The authors can assume that the septum can be repositioned during the primary surgery, without causing growth anomaly, improving the morphology and the physiology of the nose.
OS 24.1
Traditional conservative is now called aggressive treatment
C Van Loveren

Abstract Content: Over the recent years there have been important paradigm shifts in cariology especially related to caries in the primary dentition. Three of them and their consequences will be discussed in this presentation: 1. the process of caries dentine is driven by the dental plaque on top of the mineral and not by dental plaque in the mineral, 2. by consequence new rules for excavation are less aggressive than they used to be, 3. caries can be controlled by the patient self. Based on these 3 paradigms there are treatment alternatives for the traditional restauration as in order of decreasing aggressiveness atraumatic restorative treatment, Hall-technique treatment and the non-restorative caries treatment (NRCT or NRCC). These variety of treatments options gives the practitioner ample possibilities to preserve the teeth till natural exfoliation. Another key for this success is that our patient education shifts from knowledge transfer to breaking obstacles for and with the patient for good oral hygiene behaviour because all cavities are failed prevention (G. Koch)
**OS 24.4**

**Cone beam computed tomography analysis of dentoalveolar morphology in patients with uni and bilateral cleft lip and palate**

H Muhtar¹, E Esenlik²

¹Orthodontics, Private practice, Gaziantep, ²Orthodontics, Akdeniz University, ANTALYA, Turkey

**Background:** Alveolar bone loss, root resorption, fenestration and gingival recession may occur during tooth movements especially in patients with cleft lip and palate. Therefore assessing the alveolar characteristics in detail may be helpful for avoiding these side effects.

**Aims:** The aim of this retrospective study was to compare dentoalveolar morphology of the patients with uni and bilateral cleft lip and palate (UCLP, BCLP) and patients with Skeletal Class I, II and III malocclusions by using Cone Beam Computerized Tomography (CBCT).

**Methods:** Patients having CBCTs which were taken before orthodontic treatment were evaluated. A total of 97 patients aged 9-28 years were included to the study. CBCT images of these patients were divided into 5 groups based on their anomalies. There were 16 patients with UCLP, 14 patients with BCLP, 29 patients with skeletal Class I (ANB°: 0-4°), 18 patients with Class II (ANB°>4°) and 20 patients with Class III (ANB°< 0°) in study. Dentoalveolar measurements were acquired from reoriented CBCT images. Buccal/Labial and palatal alveolar bone thicknesses of all permanent teeth at alveolar crest, 3.6 mm apical levels and apex level. Labial and lingual alveolar heights were also measured and compared among the groups. Kruskall Wallis test was used for comparisons.

**Results:** There were no differences between UCLP and other noncleft groups in terms of buccal and palatal alveolar bone thicknesses of maxillary molar and premolar teeth. However buccal alveolar thickness in premolar teeth was significantly higher in BCLP group than other groups. The labial alveolar bone thicknesses of the maxillary left central tooth at the apex level and at 6 mm apical level of the palatal side of the right central tooth in UCLP group were found to be significantly lower than those of skeletal Class I, II and III groups (noncleft groups) (p<0.05). However labial side of the thicknesses of the canine and lateral teeth in UCLP group were similar to non cleft groups. UCLP and BCLP groups exhibited similar labial and palatal alveolar thicknesses in maxillary central, lateral and canine teeth. Labial and lingual alveolar heights were found to be lesser in Class III group than all other groups.

**Summary/Conclusion:** Sagittal and transversal movements of maxillary central teeth in patients with UCLP and BCLP should be carefully monitored during orthodontic tooth movement because of less alveolar bone support than non cleft patients.
Recognition and management of dental caries in cleft children undergoing alveolar bone graft – the impact of a consultant paediatric dentist appointment.

R Heer¹, V Beale²
¹Royal Manchester Children’s Hospital, OMFs, ²OMFS, Clinical Director North West England, Isle of Man and North Wales Cleft Lip and Palate Network, Manchester, United Kingdom

Background: Studies have shown that school aged children with a cleft lip and/or palate (CLP) have a higher incidence of dental caries. Good oral hygiene and dental health results in better outcomes for these patients. In recognition of this the North West, North Wales, Isle of Man Cleft Lip and Palate Network appointed a consultant paediatric dentist in October 2016.

Aims: To compare the recorded incidence of dental disease and its management for children with a CLP who underwent an alveolar bone graft (ABG) at the Royal Manchester Children’s Hospital (RMCH) before and after the appointment of a consultant paediatric dentist (CPD).

Methods: Retrospective case note review identified the documented caries incidence and treatment needs of patients attending the ABG clinic prior to surgery for 2 groups of patients: Group 1 (No CPD): November 15 - October 16; Group 2 (CPD) – November 16 – October 17. Subsequent dental treatment and presence of dental caries at the 6 month post ABG review were recorded.

Results: Group 2 patients who had been assessed by the CPD before ABG had significantly higher reported caries and treatment requirements than the Group 1 patients. Group 1 patients had a higher reported incidence of untreated decay at 6 months post ABG.

Summary/Conclusion: The appointment of a CPD has had a positive impact on the dental health of cleft patients undergoing ABG. Deficiencies in paediatric dental provision can result in missed diagnosis of dental disease and a lost opportunity for appropriate management alongside ABG.
Dental development in patients with unilateral cleft lip and palate: a retrospective study
J. Van Dyck¹, M. Cadenas¹, G. Willems¹, A. Verdonck¹
¹Orthodontics, KULeuven, Leuven, Belgium

Background: Recent research suggests that the presence of unilateral cleft lip and palate (UCLP) causes delay in dental age and tooth development.

Aims: This retrospective study aims to evaluate this hypothesis. Factors that may be related to the amount of delay; such as teeth proximity to the cleft, age and gender are also investigated.

Methods: Panoramic radiographs of 189 non-syndromic patients with UCLP were collected, aged 6 to 20 years. The OPGs of the cleft group were compared to a control group, matched for age and gender (n=189). A total of 378 OPGs were staged according to the validated methods developed by Demirjian and Willems. All present permanent teeth were investigated in this study. In order to evaluate the difference between UCLP and non-UCLP patients, a custom-made developmental score (DS) was used.

Results: At all ages, the dental age is significantly lower in the UCLP group, however, not at all ages this difference is statistically significant. The DS is significantly lower in the UCLP group compared to the control group. In the cleft group, the teeth in the upper jaw are more delayed compared to the teeth in the lower jaw.

Summary/Conclusion: The presence of UCLP needs to be taken into account for treatment planning in orthodontics. The delay in dental development in ULCP patients implies a delay in start of orthodontic treatment. Moreover, chronological age can be underestimated in UCLP patients when using age estimation methods based on permanent tooth development. It is important for forensic dentists to take this into account.
**Prenatal Genetics, Counselling**

**OS 25.3**

*Environmental factors causing cleft lip and palate in Spain. A preliminary retrospective study*

F Galíotto-Barba¹, M Viñas¹, C Martínez-Álvarez¹, M Cortez-Lede², M Rodríguez-González², I Moral³, E Delso³, B González-Meli⁴, F Lobo⁴, J López-Cedrún⁵, D Neagu⁵, J Garatea⁶, A Garatea⁶, B Berenguer⁶, C Lorca⁷, E Martí⁷, M Delgado⁴, J Gutiérrez⁷, C Hernández⁷, E Martínez-Sanz¹

¹Universidad Complutense de Madrid, Madrid, ²Servicio Murciano de Salud, Murcia, ³Servicio Aragonés de Salud, Zaragoza, ⁴Servicio Madrileño de Salud, Madrid, ⁵Servicio Gallego de Salud, La Coruña, ⁶Servicio Navarro de Salud, Pamplona, ⁷Salud de Castilla y León, Burgos, Spain

**Background:** Cleft lip and palate (CLP) and cleft palate only (CPO) affects 1.7/1000 new-born babies. Its aetiology includes genetic, mechanical and environmental factors. Amongst these last, contact with agents such as tobacco smoke, alcohol, organic solvents or drugs, together with vitamin deficit or bereavement in the antenatal period have been reported to constitute risk factors.

**Aims:** The aim of this work was to investigate the presence of environmental factors reported to cause CLP/CPO in a Spanish population of patients.

**Methods:** Under informed consent, a questionnaire on demographic information, family history of malformations, maternal and paternal status and diseases, contact with toxic products and drugs or exposure to tobacco smoke, alcohol and other environmental factors during the first three months of pregnancy was passed to the mothers of 70 patients of CLP or CPO aged 18-30 years. These patients were recruited from 8 Spanish hospitals belonging to 6 different Spanish autonomies. The questionnaire was an adapted translation of the questionnaire from the NIEHS National Institute of Environmental Health Sciences (https://www.niehs.nih.gov/research/atniehs/labs/epi/studies/ncl/index.cfm) used for similar studies in other countries.

**Results:** Preliminary results on the answers obtained from the analysed Spanish population of patients indicate that unilateral CLP is more frequent than bilateral CLP or CPO, with higher prevalence in women (60%). Most surveyed mothers were supplemented with folic acid and iron. Almost 20% of the mothers had a history of miscarriages previous or after birth of the patient, with an even higher percentage of family cases of CLP or CPO. A substantial number of mothers had contact with organic solvents, microwave devices, laser printers or photocopying machines at a shorter distance than two metres. About 35% of the mothers suffered physical or psychological stress, and about half of the surveyed mothers had active or passive exposure to tobacco smoke.

**Summary/Conclusion:** This preliminary study suggests the importance of specific environmental factors in the aetiology of CLP and CPO in the Spanish population, highlighting the relevance of planning advocacy campaigns on the causal factors of this disease that permit to reduce their frequency.

**Funding:** This work has been funded by Banco Santander-Universidad Complutense Madrid (PR41/17-21009).
OS 25.4

The ethics of pregnancy termination in non-syndromic cleft lip and/or palate: an investigation of extent and feasibility of the concept of reproductive autonomy

K Hens1,2, G Hens3

1Department of Philosophy, University of Antwerp, Antwerp, 2Department of Philosophy, 3Neurosciences, KU Leuven, Leuven, Belgium

Background: Pregnancy termination after ultrasound detection of an isolated cleft lip with or without cleft palate is a textbook bioethical dilemma. For some, it is the ultimate, and immoral, example for the search for a perfect child. For others, the desire not to continue with a pregnancy of a fetus with cleft palate or cleft lip falls well within the scope of reproductive autonomy. Still, gynaecologists and cleft care providers involved in prenatal counseling as well as ethical committee members face difficult ethical dilemmas when confronted with actual requests from couples.

Aims: Surprisingly, in comparison with discussions related to the detection of genetic abnormalities in fetuses and even in preimplantation embryos, relatively little ethics literature has been devoted to this. With this talk, we want to fill this gap. Using the European legislations regarding pregnancy termination as a starting point, and with reference to our own ethical and clinical practice, we will demonstrate that the issue of pregnancy termination after the detection of cleft lip with or without cleft palate raises some deeper ethical questions regarding autonomous choices that are as of yet unresolved in ethical discussions surrounding prenatal decisions. Moreover, future technological developments may complicate the issues even more. Still, despite of this uncertainty, we propose some guidelines that can be used in training documents for gynecologists who are faced with a diagnosis of cleft lip with or without cleft palate in the second half of a pregnancy.

Methods: In order to understand the ethical dilemma regarding pregnancy terminating we start by presenting two fictional cases that are adaptations of actual cases, and we will engage with the audience regarding their ethical intuitions about these dilemmas. We briefly discuss and compare European legislations regarding termination and pregnancy. We then discuss two prima facie ethical principals: non-maleficence and reproductive autonomy and the principle of ‘non-directive counseling’. We investigate whether and how an application of the local legislations or general principle could shed some light on the ethical dilemmas raised by the cases.

Results: Using the cases, ethical dilemmas and the intuitions of the audience, we demonstrate that a mere application of the legal or ethical principles will not straightforwardly yield general answers to dilemmas regarding isolated clefts and pregnancy termination.

Summary/Conclusion: The issue of pregnancy termination for cleft lip +/- palate points to some deeper questions regarding rights to choose, responsibility towards future children and rights and duties of clinicians. It is probably impossible to come up with a one-size-fits-all recommendation that gives a straightforward answer to such dilemmas. Nevertheless, we will point out that there is room for the development of some guidelines that will aid and streamline ethical decisionmaking in this context and that will ensure true informed decisionmaking. We will conclude by presenting some of these guidelines.
Antenatal diagnosis of cleft lip in northern ireland
S Martin¹, K McGarry², M McBride², E Slevin², C Hill²
¹Cleft Department, Royal Belfast Hospital for Sick Children, ²Cleft unit, RBHSC, Belfast, United Kingdom

Background: The diagnosis of cleft lip and/or palate (CLP) can be a difficult time for expectant parents. The benefit of antenatal diagnosis allows the parents time to adjust to the diagnosis and prepare for the arrival. It ensures the cleft team can spend time with them and counsel them regarding potential difficulties their baby may have in the early weeks of life as well as providing a support network. NHS guidelines target is 75% of babies born with a cleft lip should be diagnoses on the antenatal ultrasound.

Aims: To review the current standard achieved in Northern Ireland with regards to antenatal cleft lip and cleft lip and palate diagnosis.

Methods: The regional cleft lip and palate database was reviewed over a 5 year period (2014-2018). All babies born with CLP and the rates of antenatal ultrasound diagnosis was reviewed including the demographics of the different cleft types.

Results: Over the 5 year period 168 babies were born in the regional unit with CLP. Those with involvement of the lip (cleft lip only or cleft lip and palate) made up 55% of the cohort (92/168). Overall the rate of antenatal detection over the 5 years was 79% (73/92). The annual rates were; 2014: 75%, 2015: 74%, 2016: 89%, 2017: 77%, 2018: 86%

Summary/Conclusion: Over the past 5 years the antenatal diagnosis rate has remained above the NHS guideline target. Striving towards 100% in the future will require involvement from our radiography and obstetric colleagues. This will ensure parents are counselled antenatally and have time to adjust to the diagnosis and prepare themselves for the birth.
Public awareness of cleft lip and palate in turkey
I Kara1, A Bastug Dumbak1, M Kulak Kayikci1
1Speech and Language Therapy, Hacettepe University, Ankara, Turkey

Background: Isolated cleft palate (ICP), cleft lip and palate (CLP) isolated cleft lip (ICL) are common congenital deformities resulting from incomplete fetal development of face. Multidisciplinary team care is the best treatment approach for such deformities. As there are limited number of professionals specifically interested in CLP in Turkey, it’s a growing community. No information about Turkish sample of general public awareness and knowledge of CLP is available in the literature. It is thought that health professionals will need the general public’s awareness levels of CLP to spread the knowledge. This study is designed to contribute to this need.

Aims: The primary outcome of this study will be the public’s awareness and knowledge level of various aspects of CLP and its care in Turkey.

Methods: This is a cross-sectional descriptive study that was included 1000 participants whom was reached by snowball sampling technique. It was aimed to reach participants from various cities of Turkey. All participants provided written informed consent and the research was approved by the Hacettepe University Human Research Ethics Committee. The questionnaire elicited information with regard to the demographic characteristics of the participants and consisted of 13 questions in total which are closed questions. Questions were formed to estimate the participants’ general knowledge and awareness about CLP.

Results: One thousand (M/F:361/639) participants were included in this study. The mean age of the respondents was 36.4 (SD=13) years (min.-max:18-77). Majority of participants (68.4%) had heard or met a person with CLP. 43.1% of the participants indicated that CLP is a birth defect. 92.1% of the participants answered that CLP is treatable. 90.4% of the respondents were of the opinion that the IQ of persons with CLP is equal to that of peers without CLP. 78.6% of the respondents were of the opinion that the academic performance of persons with CLP is equal to that of peers without CLP. Almost one-third of the respondents (32.7%) answered they would consult a pediatrician in case they had a child with CLP themselves. Almost two-thirds of the participants answered person with CLP may face problems including hearing, speech, dental, cosmetic and nutrition. 76.9% of the respondents reported that they would like to learn more about CLP.

Summary/Conclusion: Although most participants were to some extent familiar about CLP, their overall knowledge appeared to be limited. Cleft lip and palate self-help groups and family support groups may play an important role to construct public awareness by information campaigns through continuous education. In addition, since almost 1/3 of the respondents indicated that they would consult with the pediatricians in case they had a child with CLP themselves, pediatricians play a crucial role in guiding the parents.
Meet the Master

MTM 2.4
Robin sequence – management with tuebingen palatal plate
S Reiner¹, B Koos², C Poets³
¹Dpt. of Oral and Craniomaxillofacial Surgery, ²Dpt. of Orthodontics, University Hospital Tuebingen, Tuebingen, ³Dpt. of Neonatology, University Hospital Tuebingen, Tuebingen, Germany

Robin Sequence (PRS) is characterized by microgenia, mandibular retrognathia, glossoptosis and possibly cleft palate. Consecutively, children suffer from upper airway obstruction resulting in intermittent hypoxia, disturbed sleep and failure to thrive. Current treatment options for children with Robin sequence range from prone positioning, use of a nasopharyngeal tube, glossoptomy via tongue lip adhesion, mandibular distraction to tracheostomy. An effective, non-invasive treatment protocol developed in our center includes implementation of an intraoral orthodontic appliance with velar extension (the Tbingen Palatal Plate, TPP) that shifts the base of the tongue forward, thereby widening the upper airway. We assumed that children treated with the TPP are less affected by intermittent hypoxia and thereby impaired neurodevelopment. In a randomized clinical trial using a crossover design, we compared the effect of the TPP with that of a conventional appliance without a velar extension and showed significantly improved upper airway obstruction with the TPP and no effect with a conventional appliance (Buchenau W et al, J Pediatr 2007). Additional retrospective studies concerning cognitive development, speech development and surgical management of palatal clefts in children initially treated with the TPP were also conducted. Children initially treated with the TPP and isolated RS showed a physical and neurologic development within test specific reference values. We did not observe significant neurologic or cognitive developmental delay in the PRS group. At a mean age of 6.1 years (8.1 months) all children showed an almost normal and age appropriate speech function. There was no significant difference between RS and non-RS children concerning speech outcomes (Drescher F et al., Acta Paediatr 2008, Brosch S, HNO 2006). Palate repair in more than 180 children could successfully be performed without significant morbidity at an age of 11 months. Discussion and Conclusion Our results support the hypothesis that RS children can be treated successfully with the TPP, showing normal physical and neurologic development. More invasive interventions like tracheostomy, mandibular distraction or tongue lip adhesion can be avoided. These results were similar in children with isolated and syndromic RS (e.g., craniosynostosis).
An integrated approach has been used in repairing the unilateral complete cleft lip/nasal deformity by the author since late 90’s.

The approach includes: (1) presurgical nasoalveolar molding, (2) modifications in surgical techniques and (3) postoperative care and maintenance. The presurgical nasoalveolar molding can improve the symmetry in skeletal base as well as the alar shape. A modified device using the spring mechanism can reduce the numbers of clinical visits for nasoalveolar molding thus greatly reduce the burden of care for the parents.

The key steps in surgical techniques are: (1) Mohler’s rotation incision, (2) mucosal flaps for nasal floor reconstruction, correction of mucosal deficiency in piriform area, (3) eliminate the perialar incision on advancement flap, limiting scars around the alar base and nostril floor, (4) mobilization of alar base, (5) nasal floor reconstruction with complete mucosal closure, (6) muscle release and reconstruction to simulate the philtral column, (7) anchoring of advancement flap to nasal septum for centralizing the Cupid’s bow, (8) correction of central vermilion deficiency with triangular vermilion flap from lateral lip, (9) semi-open rhinoplasty with reverse U incision on the cleft side and rim incision on the non-cleft side, (10) atraumatic dissection to release the fibrofatty tissue from lower lateral cartilages, (11) advancement and fixation of the cleft side lower lateral cartilage to the noncleft side lower lateral cartilage and to the skin in an over-corrected position, (12) primary septal cartilage graft, and (13) definition of the ala-facial groove with alar transfixion sutures.

The postoperative care consists of scar care with micropore tapes and silicone sheets, maintenance of the over-corrected nasal shape with a set of asymmetric nasal stents.

These changes have resulted in a more consistent and better results thus greatly reduce the need for secondary revisions in children in preschool age or even adolescences.
VPI Surgery

OS 26.1
Speech enhancing surgery: the buccal flap approach
Robert Mann

Abstract Content: This session will review how the Buccal Flap philosophy can be used to treat VPD. The presentation will discuss what the next step is when a Double Opposing Buccal Flap Palate Lengthening Procedure is not effective by itself. The session will introduce the Functional Palate Suspension Procedure, specifically designed to reposition functioning velar muscles so they can effectively reach the proper closure position on the retro pharynx. Re-palatoplasty options that are possible within the Buccal Flap Approach will also be described.
Cleft type and the correlation to speech outcome and secondary speech improving surgery in a longitudinal perspective.
Å Jonasson¹, M Blom Johansson², M Hakelius¹, D Nowinski¹
¹Dept. of Plastic surgery, Uppsala university hospital, ²Dept. of Neuroscience, Uppsala university, Uppsala, Sweden

Background: Several studies have compared how different cleft types and cleft width affect speech outcome and the need for secondary speech improving surgery during pre-school age. Most of the studies point in the same direction; there is a positive correlation between cleft severity and the incidence of velopharyngeal incompetence and cleft specific speech characteristics. It has also been suggested that an increased cleft width is associated with higher prevalence of velopharyngeal incompetence. However, there is a lack of data on the effect of cleft types and cleft width on speech outcome and the need for secondary surgery in the long term. 

Aims: To examine the possible correlation between cleft type with speech outcome and the need for secondary surgery in a longitudinal and long-term perspective.

Methods: 270 patients born with unilateral cleft lip and palate, bilateral cleft lip and palate and cleft palate only were included in the study. All patients were treated from birth to their sixteen-year follow-up at the Uppsala university hospital. Data on speech and cleft surgery were retrospectively extracted from medical records. The speech had been routinely evaluated at three, five, ten and sixteen years of age by a cleft team speech and language pathologists. These evaluations were organized into velopharyngeal competence, the presence of glottal articulation and intelligibility.

Results: The results show that there are no statistically significant differences between the different cleft types regarding velopharyngeal competence or glottal articulation at any age. There were no statistically significant differences between the different cleft types at three, five or sixteen years of age regarding intelligibility. However, at ten years of age, intelligibility was significantly worse in the group with bilateral cleft lip and palate. There is no difference between the groups regarding the need for secondary surgery.

Summary/Conclusion: In the long-term perspective, cleft type does not affect velopharyngeal function, presence of glottal articulation or the need for speech improving surgery. At the age ten the intelligibility was worse within the group with bilateral cleft lip and palate which indicates that they have poorer articulation skills than the two other groups.
A study on dynamic changes of velopharyngeal function after posterior pharyngeal flap surgery—a prospective study based on long-term follow-up with nasopharyngoscopy

W Zhang¹, S Tang¹, S Xie¹
¹Plastic Surgery & Cleft lip and palate Treatment, The Second Affiliated Hospital of Shantou University Medical College, Shantou, China

Background: Although many surgical procedures have been applied to the treatment of cleft palate, the changes of velopharyngeal function have not been revealed. Therefore, confusion is appearing about the arrangement of time of follow-up, speech therapy and re-operation.

Aims: In this study, a long-term follow-up study was carried out on patients with cleft palate after posterior pharyngeal flap surgery to investigate the dynamic changes of velopharyngeal function and its influence factors.

Methods: A total of 40 patients with cleft palate, older than 8 years old, who have undergone posterior pharyngeal flap surgery were included in the study. All patients were randomly divided into two groups, the ST group (Speech therapy were carried out from the first month after the operation) and the non-ST group (without speech therapy). All patients were followed up once a month, for subjective assessment and nasopharyngoscopy. Survival analysis was performed on velopharyngeal function and related factors were searched. Subsequently, the recovery process of velopharyngeal insufficiency after posterior pharyngeal flap surgery was simulated by software.

Results: A total of 33 patients were included for more than 8 times of follow-up, with 293 copies of nasopharyngoscopy examination. The median survival time to achieve the VPC of ST group was about 2 months, while the median survival time to achieve the VPC of non-ST group was about 5 months. The recovery time of velopharyngeal function was related to the movement of the lateral pharyngeal wall.

Summary/Conclusion: The recovery of velopharyngeal function after posterior pharyngeal flap surgery is a dynamic process and has its influence factors. Speech therapy has an obviously positive on the recovery of VP function, which is mainly reflected in the acceleration of lateral wall movement time.
**OS 26.4**

The role of preoperative cervical vascular imaging in patients with non cleft related vpd and velocardiofacial syndrome

N Fallico¹, N Timoney¹, D Atherton¹

¹South Thames Cleft Service, Guv's and St Thomas' Hospital, London, United Kingdom

**Background:** In velocardiofacial syndrome (VCFS) patients, medial displacement of the internal carotid arteries (ICAs) may increase the risk of vascular injury during the surgical correction of velopharyngeal dysfunction (VPD). Some surgeons advocate the use of vascular imaging studies prior to surgery. Nevertheless, the role of preoperative imaging is still controversial.

**Aims:** This study aimed to review the current practice of UK cleft units and also examine our own practice in the South Thames Cleft Service of children with VCFS undergoing speech surgery over the previous 5 years. Finally, a literature review would be performed to attempt to describe current best practice across the world.

**Methods:** A questionnaire was sent to the UK cleft surgeons to enquire about the management and use of preoperative vascular imaging in patients with non cleft related VPD and VCFS.

A retrospective study was also conducted of the unit's 5-year series of patients with VPD and VCFS.

A literature search of the PubMed database was performed according to PRISMA guidelines. Keywords for VCFS, VPD, medial dislocation of ICA, and preoperative vascular imaging were used to screen articles for final review.

**Results:** Twenty-nine completed questionnaires were returned (response rate 85%).

Most UK surgeons (75.8%) do not regularly order preoperative vascular imaging for patients with VCFS but some would consider it if a posterior pharyngeal wall pulsation was visible on clinical or endoscopic examination.

With respect to the surgical management of VPD, there was lack of consensus on the preferred technique: Palate exploration/re-repair (34.5%); Hynes (24.1%); Orticochoea (27.6%); Pharyngeal flap (10.3%); posterior wall augment (3.5%). Just over a third of the surgeons (34.5%) reported having had to modify or abandon the surgery as a result of posterior pharyngeal wall anomalies discovered intra-operatively. Finally, none of the surgeons had any postoperative morbidity (i.e. bleeding).

In the South Thames Cleft Service, between 2013 and 2018 a total of 36 patients affected by VCFS have been assessed for VPD. An MRA was performed for 19 patients. Medial deviation of the ICAs was identified in 6 patients. In these patients surgical planning was as follows: 2 patients underwent an Orthicochea pharyngoplasty; 2 patients did not proceed to surgical correction of VPD; 1 patient underwent a midline flap and 1 patient moved abroad before surgery was scheduled.

Nine studies were included in the literature review. A total number of subjects included in all studies were 304. Vascular imaging (MRA) was performed on a total of 201 patients; 111 (55.2%) of them was found to have medialization of the ICA.

Six papers concluded preoperative imaging of the major neck arteries is recommended for all patients with VCFS who are to undergo pharyngeal surgery; two articles questioned its cost-effectiveness and one maintained it is not essential.

**Summary/Conclusion:** The results of the survey showed most UK surgeons do not routinely perform vascular imaging but when posterior pharyngeal wall anomalies were discovered intra-operatively, surgery had to be abandoned or modified.

With reference to the surgical techniques, wide disparity has been observed among UK centers.

Most recent literature recognizes the value of preoperative cervical imaging in VCFS patients but acknowledges there may be cost issues.

Our retrospective study of our own data showed that in selected cases, the use of MRA influenced the choice of speech surgery.
Furlow re-palatoplasty for patients with unilateral cleft lip and palate (UCLP), a 16-year case series
A Saarikko¹, V Ahti¹, S Alaluusua¹, J Rautio¹
¹Department of Plastic Surgery, Helsinki Cleft Center, Helsinki, Finland

**Background:** Development of normal speech is the primary goal of palatoplasty in cleft patients. However, velopharyngeal insufficiency (VPI) is a common problem after cleft palate repair and it is often related to poor levator muscle function and palatal shortening. During the last 16 years, Furlow re-palatoplasty has been the most commonly used operation for patients with VPI symptoms in our cleft unit.

**Aims:** The purpose of this study was to assess the efficacy of Furlow re-palatoplasty in the treatment of VPI after primary surgery of unilateral cleft lip and palate (UCLP).

**Methods:** The study was a retrospective analysis of 69 consecutive non-syndromic Caucasian patients with a UCLP, born between 1997-2013, with VPI that required Furlow re-palatoplasty. Pre- and postoperative evaluation of velopharyngeal function was performed using auditory perceptual assessment (APA) and nasometry. Preoperative nasoendoscopy for velopharyngeal closure was performed, when needed, for preoperative planning.

**Results:** Average age at the time of Furlow re-palatoplasty was 6.0 years (range 3.1 - 14.5 years). Significant improvement of APA and nasalance scores was achieved in 87% of patients. Before Furlow re-palatoplasty velopharyngeal function was incompetent in 93% (64/69) of patients and borderline competent in 7% (5/69) of patients. After Furlow re-palatoplasty velopharyngeal function was competent in 59% (41/69) of patients, borderline competent in 28% (19/69) of patients and incompetent (did not improve) in 13% (9/69) of patients. Second VPI operation, pharyngeal flap (3 patients), Re-Furlow (2 patients) or palatal re-repair (1 patient), was performed for 6 patients. Postoperative complications included postoperative bleeding in two patients (3%) and mild temporary airway obstruction in one patient (1%).

**Summary/Conclusion:** Furlow re-palatoplasty is considered a safe and effective treatment option for VPI in patients with previously repaired UCLP with success rate of 87%.
Children without clefts referred for hypernasality. Experience from a norwegian cleft team.
Å Sivertsen¹, N Pedersen², H Skuladottir¹, C Kubon¹, H Vindenes¹
¹Dept Plastic Surgery, Haukeland University Hospital, ²Statped Vest, Bergen, Norway

Background: Most cleft teams have patients without known cleft palate referred to evaluate and treat hypernasality. In our experience it is a challenge to evaluate the velopharyngeal function in this group of patients and difficult to select the right patient for surgical treatment.

Aims: To achieve better understanding of hypernasality in patients without cleft palate.

Methods: We present patients referred to our plastic surgery department with the diagnosis hypernasality in a period of four years. This cohort of 15 children had multidisciplinary follow-ups in our cleft team till they were 16 years old. We describe the characteristics of the patients, our medical investigations and the long-term speech results for patients with and without surgical treatment.

Results: The children were between 4 and 11 years at the time of referral. At the age of sixteen, 8 patients had other complex medical diagnosis. 13 patients had been operated with pharyngeal flap and 9 of these were evaluated with good speech results.

Summary/Conclusion: This review of our clinical data contributes to better understanding of a heterogenic group of patients, and is a step forward to establish better routines for medical investigation and treatment of patients with hypernasality in our department.
Comparison of speech outcome after pharyngoplasty in 22q11.2 deletion syndrome: cranial based pharyngeal flap versus the modified Honig procedure.
A De La Mar¹, A Mink van der Molen²
¹Plastic Surgery, University of Utrecht, ²Plastic Surgery, UMC, Utrecht, Netherlands

Background: Most frequent human micro deletion syndrome (Saitta et al 2004), affecting around 1 in 2000-4000 newborns. 50-79% of 22q11.2 is diagnosed with velopharyngeal dysfunction (VPI). Functional outcome of VPI surgery has been reported to be worse in patients with 22q11DS compared to patients without the syndrome.

Aims: To compare the outcome in understandability, resonance, and complications between the Cranial Based Pharyngeal flap (CBPF) or the Modified Honig Procedures (MHP) in VPI patients with the 22q11.2 deletion syndrome (22qDS).

Methods: Indication in Velopharyngeal insufficiency (VPI) 22q11.2 patients for either (CBPF) or (MHP) was determined according to a standardized flowchart. All patients receiving either CBPF or a MHP from 2009 to 2016 were identified in databases of 2 Dutch centers. For a total, 37 patients undergoing pharyngoplasty were included. PBPF group contained 18 cases (48,6%), the MHP group contained 19 cases (51,4%). 8 patients were identified having an adynamic velum. Speech outcome was categorized in progression of understandability, resonance, detection of residual leakage in nasoscopy imaging and complication risk, all evaluated within 2 years after surgery. Complications were categorized into OSA, bleeding and the need for further surgery.

Results: Progression of understandability within two years after surgery was measured in 66,7% of the cranial based pharyngeal flap group and in 82,4% of the Modified Honig group (RR 1,889; 95% CI, 0,540-6,604 P= .306). In 20,0% of the cranial based pharyngeal flap group normal resonance was achieved, whereas the Modified Honig group achieved normal resonance in 26,3% (RR 1,760; 95% CI, 0,215-2,682 P= .666). According to the accomplished nasoscopy images in 33,3% of the cranial based pharyngeal flap group residual leakage was seen. In the Modified Honig group residual leakage was visible in 64,3% of the accomplished nasoscopy's (RR 1,867; 95% CI, 0,805-4,328 P=.147). No post operative bleeding occurred in the cranial based pharyngeal flap group, while one post operative bleeding appeared in the Modified Honig group (8,3%). In 60,0% of the patients suffering from an adynamic treated with the cranial based pharyngeal flap (n=5) there was progression of understandability, only in 40% normal resonance was received, only in 20% there was residual leakage and in none of the 5 patients bleeding occurred. None of the 2 patients treated with the Modified Honig due to an adynamic velum achieved progress in speech, there was no normal resonance accomplished, in 50,0% there was residual leakage and in neither bleeding occurred.

Summary/Conclusion: Although the cranial based pharyngeal flap procedure group included more patients with an adynamic velum, overall, the cranial based pharyngeal flap procedure achieved comparable results in understandability, resonance, residual leakage and post operative bleeding, to the Modified Honig procedure. Albeit the population was extremely low, the results seem to indicate the cranial based pharyngeal flap performs better in patients suffering from an adynamic velum. Achieving comparable results in patients expected to have a worse outcome shows the cranial based pharyngeal flap is a considerable procedure in challenging cases of 22q11.2 VPI patients with adynamic velum function.
OS 26.8
Speech of patients with unilateral complete cleft lip and palate – comparison of three different surgical protocols for primary repair
V Ahti1, S Alaluusua1, J Leikola1, J Rautio1, J Hukki1, A Saarikko1
1Cleft and craniofacial centre, Plastic surgery, Helsinki University Hospital, Helsinki, Finland

Background: The surgical treatment of unilateral cleft lip and palate (UCLP) varies among cleft treatment centers worldwide. Between 1997 and 2015, three different surgical protocols have been used in our cleft unit for primary repair of unilateral cleft lip and palate (UCLP). During the ScandCleft randomized controlled prospective trial closing the entire palate at 12 months (Protocol 1) was compared with closing the soft palate and lip at 4 months and the hard palate at 12 months (Protocol 2). The most commonly used protocol after 2008 in our cleft unit comprises closure of the lip and hard palate with a vomer flap at 4 months and the soft palate at 10 months (Protocol 3).

Aims: The purpose of this study was to compare subsequent velopharyngeal competence after three different surgical protocols for primary palatal repair.

Methods: The study was a retrospective analysis of 174 non-syndromatic Caucasian patients with a UCLP. Protocol 3 was retrospectively compared with Protocols 1 and 2 within the ScandCleft study. The study contained information of patients treated with Protocol 1 (50 patients) and Protocol 2 (43 patients) within the ScandCleft study between 1997 and 2006 and patients treated with Protocol 3 (81 patients) between 2004 and 2015. The medical history of all patients was obtained from the medical files. Patients’ speech outcome was evaluated by a speech pathologist at the age of three and five years.

Results: At 3 years of age, normal velopharyngeal competence was found in 58% of patients in Protocol 1, 47% of patients in Protocol 2, and 57% of patients in Protocol 3. At 5 years of age, the corresponding figures were 58%, 58%, and 64%. Two patients (4%) in Protocol 1, nine patients (21%) in Protocol 2, and 12 patients (19%) in Protocol 3 had palatal reoperations before the age of 5 years. Fistula rates were 4% in Protocol 1, 19% in Protocol 2 and 4% in Protocol 3.

Summary/Conclusion: No significant differences emerged in velopharyngeal competence at age 3 years between the most common local methods for primary palatal repair (Protocols 1 and 3). Surgery for velopharyngeal insufficiency (VPI) was performed earlier in patient group 3, explaining the difference in the VPI rate at the 5-year time-point. In Protocol 2, which was new for our cleft surgeons, there were significantly more reoperations needed due to fistulas.
**OS 26.9**

**Palatal lengthening using buccinator myomucosal flaps with furlow z-plasty technique for primary cleft palate repair**

M Khodir¹, H Moussa², E Helal³, T Aly¹, M Aboulhassan⁴

¹Oral Maxillofacial, Faculty of Dentistry- Alexandria University, ²Orthodontics, Faculty of Dentistry - Alexandria University, Alexandria, ³Medical Student, ⁴Plastic Surgery, Faculty of Medicine -Cairo University, Cairo, Egypt

**Background:** Cleft palate is a congenital deformity caused by abnormal facial development during intra-uterine life. Worldwide, the prevalence of cleft lip/palate is about 1:1000 live births. The levator vela palatini muscles are malpositioned sagittally, running postero-anteriorly and inserted onto the posterior edge of the hard palate in cleft palate patients. This configuration prevents the muscle from exerting its upward, backward, and lateral pull.

Various modalities have been described with major refinements over the past 30 years. Gradually, cleft surgeons began to appreciate the importance of dissection and retro-positioning of the levator muscle on improving speech outcomes. Disturbed speech resonance from velopharyngeal insufficiency is one of the major problems associated with cleft palate repair, with about 30 percent of the patients suffering from abnormal resonance resulting from anatomical abnormalities second surgery is commonly needed with this treatment modality to treat this attained insufficiency.

Furlow was the first to describe the palatoplasty technique, in which the levator muscle is dissected and freely released from its abnormal position and retro-positioned in a Z-plasty lengthening technique without dissection on the hard palate. However, among the problems that have been raised concerning the Furlow palatoplasty is the limitation of the procedure in wide clefts. Furthermore, the possibility of higher fistula rate associated with the technique.

These considerations have led to modified palatal lengthening by buccinator myomucosal flaps.

**Aims:** To assess the significance of the use of buccal myomucosal flap combined with Furlow double z palatoplasty on lengthening of the palate in primary cleft palate

**Methods:** Forty patients of non syndromic unilateral complete cleft palate were done using classical furlow z-plasty operation without suturing the right sided anteriorly based mucosal flap to the edge of the hard palate. This space was further filled with a left posteriorly based buccinator myomucosal flap. Infiltration of the buccinator flap with adrenalin was done then the flap was outlined inferior to the parotid duct. Its position on the buccal mucosa is 2 mm anterior to the base of the retromolar trigone and extends anteriorly to within 6 to 8 mm of the oral commissure. The base width will be determined by the width between the upper and lower alveolus. The buccal flap was then elevated in steps. The flap was then rotated such that its mucosa faces orally and sutured into the L-shaped defect as far forward into the hard palate defect. donor areas will be then loosely closed using 5/0 vicryl sutures.

Palatal length was measured pre and postoperative from the edge of the hard palate till the base of the uvula using a curved paper ruler.

**Results:** Among 40 patients with an age range of 9-18 months, the palatal lengthening was of a mean 29.65 (±4.72 ) compared to preoperative mean of 21.65 (±4.25 ).Paired t-test was used showing significant difference of pre and postoperative (P<0.001).we had 2 cases of fistula and one case of partial dehsicence.

**Summary/Conclusion:** Buccinator myomucosal flaps are effective and safe for palatal lengthening in primary cleft palate repair. they provide length and further more attachment by soft pliable tissue to the hard palate making it more mobile . however long term follow up is needed to validate this effect on speech.
OS 26.10
Speech outcomes following orticochea pharyngoplasty in patients with history of cleft palate and non-cleft velopharyngeal dysfunction
A Birch1, L Ferguson1, J Boorman1
1South Thames Cleft Service, Guys and St Thomas NHS Foundation Trust, London, United Kingdom

Background: The existing literature concerning speech outcome following Orticochea pharyngoplasty in cleft and non-cleft populations is limited. Previous studies indicate non-standardized reporting of speech which impede comparison of pre-operative and post-operative speech outcomes. This study aims to contribute to the current knowledge base.

Aims: To assess the speech outcomes following Orticochea pharyngoplasty in a group of patients with cleft palate and non-cleft velopharyngeal dysfunction (VPD).

Methods: 43 patients underwent Orticochea pharyngoplasty by a single surgeon in a regional UK cleft centre between 2004 and 2012. 23 patients presented with cleft diagnoses; 5 with unilateral cleft lip and palate, 1 with bilateral cleft lip and palate, 13 with cleft palate only and 6 with submucous cleft palate. 20 patients presented with non-cleft diagnoses; 8 of whom had a diagnosis of 22q11 deletion syndrome, 3 presented with other syndromes (NF1, Worcester-Drought and Turners Syndrome) and the remaining 9 patients had a non-syndromic diagnosis.

Pre and post-operative (range 6-12 months) speech samples were assessed blindly using the Cleft Audit Protocol for Speech-Augmented (CAPS-A) by a Specialist Cleft Speech and Language Therapist external to the team. Speech was rated on the following parameters: hypernasality, hyponasality, audible nasal emission, nasal turbulence and passive cleft speech characteristics. 37% of speech samples were assessed blindly by a second Specialist Cleft Speech and Language Therapist to calculate inter-rater reliability across each of the speech parameters. Intra-rater reliability for each speech parameter was also calculated using 37% of the sample.

Results: Speech results were examined as a whole group and in 2 separate groups (cleft group / non cleft group). Level of significance was set at p<0.05.

Whole group: A statistically significant difference in pre and post-operative scores for hypernasality (p<0.001) nasal emission (p<0.005) and passive cleft speech characteristics (p<0.005) were reported for the whole group. Nasal turbulence scores pre and post operatively were not found to be significantly different (p>0.05) Of the patients who had reported hypernasality preoperatively, 81% showed an improvement following the procedure. 100% of patients with nasal emission preoperatively showed an improvement post-operatively. 5 patients developed hyponasality following the procedure which was found to be statistically significant (p>0.05.)

Cleft Group: A statistically significant difference in pre and post-operative scores for hypernasality (p<0.001) nasal emission (p<0.005) and passive cleft speech characteristics (p<0.05) were reported for the cleft group. Nasal turbulence and hyponasality scores were not found to be statistically different (p>0.05)

Non cleft group: A statistically significant difference in pre and post-operative scores for hypernasality were noted (p<0.001). No statistical significance was noted for hyponasality, nasal emission, nasal turbulence and passive speech characteristics. (p>0.05). Inter-rater reliability scores ranged from moderate – good agreement (Kappa of 0.42 - 0.66). Intra-rater reliability scores ranged from moderate to very good (Kappa of 0.53 – 1.0)

Summary/Conclusion: Orticochea pharyngoplasty is a successful surgical procedure in treating velopharyngeal dysfunction in both the cleft and non-cleft population.
Could pharyngeal fat injection with palatal lengthening be a first-line treatment of velopharyngeal insufficiency?

G Diallo-Hornez1, K Khonsari2, J Mercier1, S Balandier1, B Isido3, G Rousteau1, J Talmant4, J Perrin1, P Corre1, H Bertin1

1Centre Hospitalier Universitaire de Nantes, Clinique de Chirurgie Maxillofaciale et de Stomatologie, Nantes, 2Hôpital Necker Enfants-Malades, Assistance Publique - Hôpitaux de Paris, Service de Chirurgie Maxillofaciale et Plastique, Paris, 3Centre Hospitalier Universitaire de Nantes, Service de Génétique clinique, 4Clinique Jules Verne, Chirurgie Plastique, Reconstrucctrice et Esthétique, Nantes, France

Background: Velopharyngeal insufficiency (VPI) affects the communication of patients concerned and their quality of life. Its main cause is the presence of a cleft palate, either syndromic or not. When speech therapy is not sufficient to correct the phonatory consequences of VPI, several surgical techniques are available to try to improve intelligibility: palatal lengthening (PL), velopharyngoplasty (VPP) and more recently, pharyngeal fat injection (PFI). VPP with pharyngeal flap is generally considered as the gold standard in the treatment of moderate to severe VPI, while PL and PFI, alone or in combination, are reserved for mild VPIs.

Aims: VPP with pharyngeal flap is generally considered as the gold standard in the treatment of moderate to severe VPI, while PL and PFI, alone or in combination, are reserved for mild VPIs. VPP is an effective but invasive surgical procedure leading to significant postoperative pain and the risk of obstructive sleep apnea (OSA) syndrome. PFI, with or without PL, is a less invasive and a well-tolerated procedure, but its efficacy in moderate to severe VPI has not yet been compared to that of VPP. The main objective of this study was to compare the phonatory results of patients with VPI, treated either with PFI, associated or not with PL, or with VPP with an inferiorly based pharyngeal flap.

Methods: This retrospective study was comprised of seventy patients with VPI that required surgery. Thirty-five patients were treated with VPP with an inferiorly based pharyngeal flap and 35 patients with PFI more or less associated with PL. All patients were evaluated pre-operatively and 6 months postoperatively by the same speech and language therapist at the University Hospital of Nantes according to the following criteria: Borel-Maisonny score, intelligibility, nasality and nasal air emission.

Results: There was no significant difference between the patients in the two groups according to age at treatment, sex, the presence of a syndrome, the presence of a cleft, or preoperative Borel-Maisonny scores. There was no postoperative complication in any of the patients. The Borel-Maisonny score and all the main speech parameters - intelligibility, nasality, nasal air emission - were improved in both groups, with no significant difference between them. The delay of resumption of oral intake and duration of hospitalization were significantly reduced in the PFI PL group compared to the VPP group.

Summary/Conclusion: PFI, whether associated or not with PL, produced phonatory results comparable to those of the VPP with an inferiorly based pharyngeal flap in the treatment of moderate to severe VPI. PFI PL was responsible for lower postoperative morbidity, resulting in a significant reduction in the delay of resumption of oral intake and duration of hospitalization. A prospective study is now needed to confirm these findings and to define, when appropriate, PFI as a first-line treatment for not only mild but moderate to severe VPI.
Subphenotyping and classification of orofacial clefts: a revised classification
Peter Mossey

Abstract Content: Nonsyndromic orofacial clefting (OFC) describes a range of phenotypes that represent the most common craniofacial birth defects in humans, with an overall birth prevalence of 1:700 live births. A range of different methods is used for recording and classifying OFC subphenotypes, one of which is the International Classification of Diseases (ICD) system. However, there is a general perception that research is being hampered by a lack of sensitivity and specificity in grouping those with OFC into subphenotypes, with potential heterogeneity and confounding in epidemiologic, genetic, and genotype-phenotype correlation studies. Besides the ICD system, there still remains a range of current OFC classifications and it is important to acknowledge their strengths as well as their limitations. The LAHSHAL classification is described in the context of a potentially useful tool for OFC that could complement the ICD-10/ICD-11 Beta coding systems to become a simply understood, universally accepted, clinically friendly, and research-sensitive instrument. Empowering registries, clinicians, and researchers to use a common classification system would have significant implications for OFC research across the world at a time when accurate subphenotyping is crucial and health care research is becoming increasingly tailored toward the individual.
Understanding the role of enhancer cis-mutations on human craniofacial variation
J. Mohammed

Abstract Content: It recently became clear that most genetic variants associated with intra-species morphological differences or inter-species normal range trait variation map to the non-coding parts of the genome. A substantial fraction of such variants is thought to reside within a class of cis-regulatory elements called enhancers, which are particularly fertile targets for mediating morphological variation. With respect to craniofacial development, we are broadly interested in using comparative genomics of primate species, results from craniofacial Genome-Wide Association Studies (GWAS), and epigenomic and transcriptomic profiling in a relevant cell type to systematically and quantitatively annotate divergence of enhancers that mediate craniofacial variation. Cranial Neural Crest Cells (CNCCs) are a relevant cell type and a direct in vitro model for elucidating both fundamental and evolving mechanisms underlying morphological facial variation because CNCC derivatives become the embryonic face. Recently, we performed comparative epigenetic profiling from human and chimpanzee CNCCs to identify and study cis-regulatory element variation from these species. From orthologous enhancers, we identified cis-mutations within transcription factor motifs that contribute to enhancer activity, discovered a novel transcription factor binding site specific to CNCC enhancers, which we named the ‘coordinator’ motif, uncovered broad clusters of species-biased enhancers near genes associated with human facial variation, and linked enhancer sequence divergence with quantitative expression differences in important neural crest regulators. At present, we are exploring the wealth of human genetic variants implicated in normal range facial variation from emerging craniofacial GWAS. Genomic manipulation of single nucleotide polymorphisms (SNPs) within candidate enhancers using CRISPR/Cas9 has narrowed the identification of causative SNPs for a limited number of facial features. Importantly, however, such experimental manipulations are beginning to provide insights into the mechanistic basis of human facial variation.
Value Based Healthcare and Measuring QoL

OS 28.2
What and when to measure in cleft care: my experiences in the uk
Scott Deacon

CRANE is a national register of children affected by cleft lip and/or palate covering England, Wales and Northern Ireland and shortly to include Scotland. https://www.crane-database.org.uk/ We have over 19000 children registered since 2000 and have collected clinical related outcomes at 5 years and some at 10 years for these children. This includes the facial growth, speech and the level of dental disease. We also have linked at the patient level to NHS records from Hospital Episode Statistics held by NHS Digital and the National Pupil Database, which has educational achievement and attendance records in state schools up to 16 years of age with the Department of Education. We are also approved to link to the National Hearing Screening Program. This is a unique dataset with health and nonhealth outcomes offering population level analysis on children affecting by a cleft. We offer our view on the potential benefits of linked data in population level analysis and the emphasis on 5 year data outcome data currently in the UK.
**OS 28.3**

**Evaluation of oral health outcomes in the various cleft types at different ages with the ichom cleft set.**

L Kind¹, L Kragt², I Apon², E Wolvius², S Versnel³

¹Cleft team, ²Department of Oral and Maxillofacial Surgery, Special Dental Care and Orthodontics, ³Department of Plastic and Reconstructive Surgery, Erasmus medical center, Rotterdam, Netherlands

**Background:** The care for cleft patients is complex due to the variety of functional disabilities and aesthetic impairment which differ depending on type of cleft. Previous research showed that cleft patients are considered to have a higher caries risk and a reduced oral-health related quality of life than non-cleft patients. Therefore, in the ICHOM cleft lip and palate set various data are collected considering oral health by a combination of patient-reported outcome measures (PROMs) and clinical observations.

**Aims:** The aim of our study is to gain more insight in the oral health perception of patients with different cleft types during the cleft treatment trajectory with the use of the dental and oral health related PROMs included in the ICHOM standard set for cleft lip and palate patients at the ages of 8, 12 and 22 years.

**Methods:** We performed a retrospective study of the CLEFT-Q Dental, Cleft-Q Jaw and the Child Oral Health Impact Profile – Oral Symptoms Subscale (COHIPOSS), completed between November 2015 and January 2019. The questionnaires were filled in before visiting the cleft team by patients, with the assistance of the parents when needed. Differences between age-groups and cleft phenotypes were evaluated with one-way ANOVA's including post-hoc analysis.

**Results:** A total of 334 participants have filled in the specific questionnaires depending to their cleft type and age. We found significant lower scores in the CLEFT-Q Dental scale between CLAP and CL(p = 0.036), between CLAP and CP (p = 0.07) as well as between CLAP and CLA phenotypes(p = 0.015). Furthermore, in the CLEFT-Q Dental scale patients reported significantly lower outcome scores at the ages of 8 and 12 than at 22 years (p < 0.001). No significant differences were found between 8 and 12 years of age. In the COHIPOSS and CLEFT-Q Jaw scores, no significant differences were found between age groups and between type of clefts.

**Summary/Conclusion:** Based on the results, children with CLAP perceive the aesthetic oral health status significantly worse than children with other types of clefts. Dental treatment results in overall improvement of outcome scores on the dental scale of Cleft Q at the age of 22 years. However, the perceptions of the jaw health status and the oral health status appear similar between all types of cleft, and ages in this population. The use of PROMs in cleft care enables the health care professional to approach patients more individually, namely based on their personal health perception.
Patient-reported outcome measures included in the ichom cleft standard set: examining their psychometric performance

Apon1, A Klassen2, E Wolvius1, S Versnel3

1Department of Oral and Maxillofacial Surgery, Erasmus Medical Center, Rotterdam, Netherlands, 2Department of Pediatrics, McMaster University, Hamilton, Canada, 3Department of Plastic and Reconstructive Surgery, Erasmus Medical Center, Rotterdam, Netherlands

Background: Cleft lip and/or palate is one of the most prevalent congenital craniofacial anomalies in the Netherlands affecting approximately one in 500 livebirths. This condition, treated by multidisciplinary teams, can be complex and directly impact appearance and how patients function and feel. Services and treatment protocols for the management of patients with cleft lip and palate vary substantially between and within countries. The Cleft Lip and Palate group of the International Consortium of Health Outcome Measures (ICHOM) seeks to address variation in practice with the introduction of a standard set of outcome measures and has recognized the importance of including the patient’s voice by incorporating the CLEFT-Q, Child Oral Health Impact Profile (COHIP) and NOSE scales. The Erasmus Medical Center Rotterdam is one of the first centers globally to adopt the complete ICHOM cleft set. The set was fully implemented as routine care by the end of 2015. Given the importance placed on patient-reported outcome measurements (PROMs) and the upcoming widespread adoption of the ICHOM Cleft Lip and Palate set, it is essential to evaluate the accuracy and reliability of these instruments to be certain they provide meaningful measurements.

Aims: The aim of our study was to examine the performance of the PROMs included in the ICHOM Cleft Lip and Palate set using the modern psychometric approach of Rasch Measurement Theory (RMT).

Methods: Patient characteristics and outcome data of 334 cleft patients of 8, 12 and 22 years were retrospectively extracted from the patient’s medical file. Data for the CLEFT-Q subscales (Face, Jaws, Teeth, Eating and Drinking, Speech, Social Function, School Function, Psychologic Function, Speech-related Distress), the COHIP and the NOSE scale completed between November 2015 and January 2019 were analyzed according to the RMT. This analysis is based on the probability that individuals will answer a set of items in a certain manner and can provide insight in the strengths and limitations of a scale.

Results: RMT analysis provided evidence that the CLEFT-Q scales worked together conceptually and statistically since almost all items had good fit and thresholds between response options were ordered. Reliability was high, with person separation index values ranging from 0.83 to 0.90 for all scales except speech distress, which was 0.66. The psychometric performance of the COHIP subscale and NOSE scale showed disordered item thresholds and low reliability with person separation index values of 0.34 and 0.35 respectively. The evaluation of the PROM scales revealed the absence of some cleft-specific outcome measures, such as the patient’s perspective on nose or lip appearance, which might be important in the treatment of cleft patients.

Summary/Conclusion: Some of the PROMs in the ICHOM cleft set evidenced poor psychometric properties based on data from a single centre. Importantly, not all cleft-specific treatments are evaluated with the included PROMs. Given the importance placed on the patient perspective in ICHOM standard sets, we recommend a reconsideration of the cleft set to include additional cleft-specific PROMs and to ensure that all components of the set provide meaningful measurements.
OS 28.5

Computerised adaptive testing accurately predicts cleft-q scores by selecting fewer, more patient-focused questions.

C Harrison¹², D Geerards¹³⁴, M Ottenhof¹³⁴, A Klassen⁵, K Wong Riff⁶, M Swan², A Pusic¹³, C Sidey-Gibbons¹³
¹Patient-Reported Outcomes, Value & Experience (PROVE) Center, Brigham and Women's Hospital, Boston, United States,
²Department of Plastic and Reconstructive Surgery, Oxford University Hospitals, NHS Foundation Trust, Oxford, United Kingdom,
³Department of Surgery, Harvard Medical School, Boston, United States,
⁴Department of Plastic and Reconstructive Surgery, Catharina Hospital, Eindhoven, Netherlands,
⁵Department of Pediatrics, McMaster University, Hamilton,
⁶Department of Plastic and Reconstructive Surgery, Hospital for Sick Children, Toronto, Canada

Background: The International Consortium for Health Outcome Measures (ICHOM) have recently agreed upon a core outcome set for the comprehensive appraisal of cleft care, which puts a greater emphasis on patient reported outcome measures, and in particular the CLEFT-Q. The CLEFT-Q comprises 12 scales with a total of 110 items, aimed to be answered by children as young as eight years old. Computerised adaptive testing (CAT) is a technique that can reduce the number of items in a patient reported outcome measure by selecting only the most relevant questions to administer, based on an individual's prior responses.

Aims: In this study we aimed to use CAT to reduce the number of items needed to predict results for each CLEFT-Q scale.

Methods: We used an open-source CAT simulation package to run item responses over each of the full-length scales and its CAT counterpart at varying degrees of precision, estimated by standard error (SE). The mean number of items needed to achieve a given SE was recorded for each scale's CAT, and the correlations between results from the full-length scales and those predicted by the CAT versions were calculated.

Results: Using CAT for each of the 12 CLEFT-Q scales we reduced the number of questions that participants needed to answer from 110 to a mean of 43.1 (range 34-60, SE <0.55) while maintaining a 97% correlation between scores obtained with CAT and full-length scales.

Summary/Conclusion: The response burden of questionnaires is of particular concern in the paediatric population, and the development of a CAT for the CLEFT-Q is an exciting advancement. The CAT version of the CLEFT-Q is likely to play a fundamental role in the uptake of the CLEFT-Q into clinical practice given the high degree of accuracy achieved with substantially fewer items.
Scores of the cleft hearing, appearance and speech questionnaire (chasq) in swedish participants with and without cleft

M Stiernman¹, H Svensson¹, M Persson², M Becker¹
¹Department och plastic and reconstructive surgery in Malmö, Sweden, Clinical Sciences in Malmö, Malmö, ²Department of Public Health, Högskolan Kristianstad, Kristianstad, Sweden


Aims: The primary aim of this study was to investigate if there were any differences in scores of the CHASQ between patients with CLP and a control population. The second aim was to compare cleft and control population scores in this study with an earlier established British norm population.

Methods: Data from 51 cleft patients were collected at routine visits to the cleft centre. Age ranged from 7 to 19, mean age was 12. Fifty-six participants were recruited to the control population. Age ranged from 9 to 20 years, mean age was 13. CHASQ consists of nine items regarding features typically affected by a cleft (factor 1 loading items) and six items regarding features not typically affected by a cleft (factor 2 loading items). Therefore, CHASQ produces two scores (sum of factor 1 loading items and sum of factor 2 loading items). In this study the sum of these two scores will be presented separately.

Results: There was no statistically significant difference in sum of factor 1 loading items (p=0.207) between the cleft and the control populations calculated with Mann-Whitney U test. There was a statistically significant difference in Factor 2 loading items (p=0.025) between the cleft and the control populations calculated with Mann-Whitney U test. Participants in the cleft population scored higher points (were more satisfied) than the control population. In total four (8%) participants with cleft scored less than 50 points on the total sum of factor 1 loading items and four (10%) participants with cleft scored less than 44 points on factor 2 loading items. Scores below these levels indicate lower satisfaction than expected according to the earlier established British norms of CHASQ. Also, eight (14%) participants in the control population scored less than 44 points on factor 2 loading items.

Summary/Conclusion: These results may indicate that children and young people with cleft in this study are generally as satisfied with their appearance, hearing and speech as children and young people without cleft. These findings are in line with some earlier research (Berger and Dalton 2009, Crerand et al. 2017). Since there was not a significant difference in the sum of factor 1 loading items between the cleft and control population, it is an indication that also participants in the control population are affected by low satisfaction with appearance, hearing and speech. The percentage of the cleft population in this study defined as less satisfied than expected according to their CHASQ scores are roughly in line with the British norm population. This is an indication that the cut-offs of the questionnaire work similarly in both the British and the Swedish cleft population.
Patient reported outcomes in the adult cleft lip and palate patient

H Gjengedal¹, K Sylvester-Jensen¹, A Littler Moi², H Vindenes³
¹Department of clinical dentistry, Medical Faculty, University of Bergen, ²Department of health and social sciences, Western University of Applied Sciences, ³Department of Plastic Surgery and Burn Centre, Haukeland University Hospital, Bergen, Norway

Background: This is a quality assurance project carried out by the Bergen Cleft team in Norway. Patients born between 1973 and 1979 were treated by the same surgeon at 3 months and 2 years at Haukeland University Hospital, University of Bergen, Norway. The surgical procedures were carried out by the same protocol. At 2 years the Pushback procedure was applied for palate closure.

Aims: To assess patient reported outcomes of cleft lip and palate patients at age 40.

Methods: Thirty patients with ULCP, consecutively treated from birth at HUS, were invited to a consultation at the age of 40. Twenty-one accepted, x men and y women and were examined and interviewed on the same day by the ULCP-team. They also completed the OHIP-14, OAS-N and Rosenberg questionnaires. The OHIP-14 assesses oral health related quality of life (OHRQoL), OAS-N is a questionnaire regarding satisfaction with facial aesthetics and the Rosenberg is a self-esteem questionnaire.

Results: The patients reported a mean OHIP-14 of 8.3 and this indicates that the ULCP patients report good OHRQoL. They also reported good satisfaction with facial aesthetics, mean OAS-N score was 46.6. The adjusted Rosenberg score was 33.1 which indicate good self-esteem. There was no correlation between Rosenberg and OAS-N, but strong correlation between OHIP-14 and OAS-N (p = 0.021) and between OHIP-14 and Rosenberg (p = 0.004).

Summary/Conclusion: ULCP patients at 40 years, treated by the same protocol since birth and by the same surgeon, report good OHRQoL, self-esteem and satisfaction with facial aesthetics.
Further psychometric validation of the cleft-q: ability to detect differences in outcome for four cleft-specific surgeries

C Harrison¹, C Rae², E Tsangaris³, K Wong Riff⁴, M Swan¹, T Goodacre⁵, S Cano⁶, A Klassen⁷
¹Department of Plastic Surgery, John Radcliffe Hospital, Oxford, United Kingdom, ²Department of Health Research Methods, Evidence and Impact, McMaster University, Hamilton, Canada, ³Patient Reported Outcomes Values and Experience Center, Brigham and Women's Hospital, Boston, United States, ⁴Department of Plastic Surgery, Hospital for Sick Children, Toronto, Canada, ⁵Nuffield Department of Surgery, Oxford University, Oxford, ⁶Modus Outcomes, Letchworth Garden City, United Kingdom, ⁷Department of Pediatrics, McMaster University, Hamilton, Canada

Background: The CLEFT-Q is a patient-reported outcome measure with validity in the field of cleft lip and/or palate. A significant indicator of this instrument’s power relates to its ability to distinguish between the impact of specific aspects of clefting in those who need treatment, compared with those who either have received treatment, or are not thought to need treatment.

Aims: This study compares relevant sub-scale scores of the CLEFT-Q for those requiring four specific surgical treatments against those who either had been treated or did not require surgery.

Methods: CLEFT-Q scores and clinical information regarding past and future need for rhinoplasty, jaw surgery, lip revision and speech surgery were obtained from the CLEFT-Q field-test. Eight one-way analysis of variance (ANOVA) models compared mean scores of relevant CLEFT-Q subscales between those who needed surgery, those who had completed surgery, and those who never needed surgery. The analyses only included patients treated in high income countries. In the rhinoplasty and lip revision models, patients without a cleft lip were excluded. In the jaw surgery and speech surgery models, patients without a cleft palate or alveolus were excluded.

Results: Participants who needed surgery reported significantly lower (poorer) scores than those who had completed surgery in each of the eight relevant CLEFT-Q scales (p<0.01 in each model).

Summary/Conclusion: The ability of the CLEFT-Q to report significantly lower scores for those with conditions deemed amenable to surgical correction lends further psychometric validity to its use in clinical practice.
**Background:** The Swedish healthcare quality registries aim to ensure a good quality of treatment for the patient, and to allow for comparison and open reporting of results, in order to promote quality control, research and innovation. The design of the Swedish cleft lip and palate (CLP) registry, results on coverage ratio and number of occasions with cleft related surgery before 5 years of age are presented.

**Aims:** The aim is to present a national quality register and the way to achieve valid and reliable data.

**Methods:** All six Swedish CLP centres are involved in the registry. Baseline data is recorded at first visit. Data on surgical treatment is recorded continuously. Treatment outcome regarding dentofacial development and speech are recorded at follow-ups at 5, 10, 16 and 19 years of age. A check against the central registry was performed to assess coverage ratio and reporting degree of cleft related surgery for children born 2009 to 2012. The number of occasions with cleft related surgery before 5 years of age was compared between centres.

**Results:** The average coverage ratio was 91.3% (range 87.4 – 96.8), and the proportion of reported cleft related surgeries was above 89% at all centres. The number of occasions with cleft related surgery before 5 years of age differed significantly between centres.

**Summary/Conclusion:** The coverage ratio was considered acceptable. Different surgical programs may explain the differences between centres regarding occasions of surgery before 5 years of age, and the final results can only be evaluated when the children have grown up. The coverage needs to be high, the included variables limited and checked for reliability, and the professionals must calibrate themselves regularly in order to achieve valid and reliable data.
Orthodontics

OS 29.1
Peculiar aspects of orthopaedic and orthodontic treatment in bilateral cleft patients
M Meazzini

Patients with cleft lip and palate require a multidisciplinary care, which starts from birth and continues into adulthood. Various difficult aspects of bilateral cleft treatment are greatly improved by a close coordination between orthodontist and surgeon. Some peculiar aspects will be discussed and as much evidence as possible will be given to support each protocol. 1) Naso-alveolar molding in BCLP. Performed with passive plates, with the objective of stretching the nasal lining and molding alar cartilages presurgically. No primary periosteoplasty was performed, therefore premaxillary contact was not an objective. Long term follow up till adolescence will be shown. 2) Premaxillary vertical excess: age dependent protocol: indications and different options depending on severity and age will be shown as well as long term follow up 3) Orthodontic pre grafting closure of large alveolar bony and soft tissue gaps: orthodontic-surgical cooperation to reduce the burden of surgical care in peculiar patient with soft tissue defects.
Bone-anchored maxillary protraction in cleft patients. Preliminary results
F Lobo¹, M Cimadevilla², B González Meli¹, E Olivares¹
¹Plastic Surgery, ²Orthodontics, Niño Jesus Children's Hospital, Madrid, Spain

Background: In patients with complete cleft lip and palate (CLP), maxillary growth is often compromised by the restrictive forces from the lip and palate repair. Treatment for moderate to severe Class III malocclusion usually relies on LeFort I surgery for maxillary advancement after the end of the growth. Bone-anchored maxillary protraction (BAMP) with miniplates in the maxilla and mandible connected by Class III elastics has been proved effective in patients with Class III malocclusion.

Aims: The purpose of this study is to evaluate if there is improvement in Class III malocclusion with BAMP in cleft patients as well as put on record the complications seen during the treatment.

Methods: 9 patients (6 male and 3 female) were treated with BAMP (1 bilateral complete cleft lip and palate, 2 bilateral cleft lips, 6 unilateral cleft lip and palate). The mean age at the beginning of the treatment was 11.2 years (range 9-13). The choice of this treatment was decided by the orthodontist and the surgeon based on growth potential. All patients were GOSLON 4. Lateral and panoramic radiographs were made before the surgery. Four miniplates were placed in the maxilla at the left and right infrrazygomatic crests and on both sides in the mandible between the roots of the permanent canine and lateral incisor. Three weeks after surgery, Class III intermaxillary elastics were placed with 75 g of force on each side. After one month the force was increased to 150 g on each side. After 2 months the force was adjusted to 250 g per side and remained stable until the end of active therapy. Miniplates were left in place for at least 1 year. New lateral and panoramic radiographs were made when the treatment was finished.

Results: 36 miniplates were used. Facial esthetics and occlusion improved in all unilateral CLP patients except one, where treatment was started late according to growth potential. ANB angle showed an increase between 0.8 and 5.37 degrees. Wits appraisal variation before and after treatment ranged between -1.6 and 7 mm. Overall complication rate was 13.8% (5 complications including 1 wound dehiscence, 2 buried hooks and 2 loosened screws).

Summary/Conclusion: Bone-anchored maxillary protraction is an available orthopedic therapy for patients with CLP with the advantage of achieving much earlier improvement of facial aesthetics and functional occlusion. Patient selection is critical to achieve optimal results. Some patients treated with this therapy will eventually need maxillary advancement. Further studies are needed to confirm that the maxillary advancement will be smaller in patients with non-successful BAMP.
OS 29.3
Pain perception in children affected by craniofacial anomalies: comparison between orthodontic treatments with traditional brackets versus invisalign
S Marrocco1, M Maria Costanza2, A Luca2, C Tortora2
1Maxillo-Facial Surgery, Ospedale San Filippo Neri - Smile House Roma, Roma, 2Maxillo-Facial Surgery, Smile House – S. Paolo and Carlo Hospital, Milan, Italy

Background: Pain is one of the most common concerns regarding orthodontic treatment and it affects a large number of patients undergoing treatment. Many patients seek for more aesthetic treatment, therefore Invisalign (INV) has become very popular in recent years, but there is still little information regarding pain and Invisalign treatment.

Aims: The objective of this study is to compare in a retrospective case-control study the difference in pain perception between a sample of craniofacial patients treated with Invisalign versus traditional orthodontic fixed appliances (FA).

Methods: 98 syndromic caucasian patients affected by various types of craniofacial anomalies were prospectively selected. The study sample, of 49 syndromic patients treated with Invisalign, was compared to a control sample, of 49 patients treated with FA. A modification of the Mc Gill Pain Questionnaire (MMPQ) was adapted to our needs (Mc Gill, 2013).

Results: There is a statistically significant difference between FA and INV in terms of sensory pain parameters (such as throbbing, cutting, pain at the level of cheeks, tongue and lips) and in terms of affective parameters (such as miserable or frustrating feeling). Almost all parameters were in favour of INV. On the other hand, INV was considered significantly more uncomfortable (include the need of remove the appliance for meals) in the female group compared to the male population.

Summary/Conclusion: The data seem to indicate that, in patients affected by craniofacial deformities, who are generally over sensitized to intra oral treatments by previous surgeries and early orthodontics, pain perception is generally higher and seems to be lower with the use of aligners compared to fixed appliances, more so in males than females and in older patients.
Comparison of oxygen saturation during maxillary impression taking in consultation for preoperative presurgical functional maxillary orthopedic treatment in babies with cleft lip and palate

A Agell¹, M Munill¹, N Sierra¹, D Bohorquez¹
¹Pediatric Oral Maxillofacial Surgery, Vall d’Hebrón Universitary Hospital, Barcelona, Spain

Background: The principle objective of presurgical Functional Maxillary Orthopedic (FMO) treatment and nasoalveolar molding is to reduce the severity of the initial cleft deformity. This enables the surgeon taking advantage of the benefits associated with repair of an infant that has a minimal cleft deformity. To perform this procedure, it is necessary to take an impression of the upper maxilla, and according to the Functional Maxillary Orthopedic Technique’s Pannaci Protocol, a monthly intraoral impression is making during consultation, with the patient awake.

Aims: The objective of this study was to evaluate the oxygen saturation levels during intraoral impression of the upper maxilla taking during consultation, as part of the preoperative treatment using Functional Maxillary Orthopedics Pannaci’s Modification and Nasoalveolar molding.

Methods: In our study, 16 unilateral and bilateral clefts babies (mean age 5.28 +/- 3.86 months) were taken 90 impressions during consultation, awake patient. Two types of printing materials were used: irreversible Hydrocolloid with rapid gelation (alginate) and Putty Silicone Addition. Oxygen Saturation Levels were measured in 2 Times: Before and during the Impression.

Results: No significant differences were found between the variations of Oxygen Saturation before and during the impression or between silicone and Alginate.

Summary/Conclusion: Taking of impressions of the maxilla in consultation carried out by trained personal and safety measures such as the correct use of the material and position of the patient is a safe and effective method.
Canine eruption in patients with unilateral cleft lip and palate: cleft side versus non-cleft side
S Rizell¹, Z Alhakim², H Mark³, J Naumova¹
¹Dep of Orthodontics, Institution of Odontology, University of Gothenburg, ²Orthodontic clinic, University Clinics of Odontology, ³Dep of Plastic Surgery, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden

Background: Canine eruption through the bonegrafted cleft area is prone to exhibit an ectopic pathway.
Aims: To compare eruption path for the maxillary permanent canines at cleft side versus non-cleft side in patients with unilateral cleft lip and palate (UCLP).
Methods: One hundred and forty-eight individuals with UCLP, treated in Gothenburg, Sweden were included in this retrospective cohort study. Canine angulation (α-angle), distance to midline and to the occlusal plane were measured on panoramic radiographs obtained at 10 years of age. A comparison of cleft and non-cleft side for the registered parameters was made using paired and un-paired t-test as well as chi-square test.
Results: The cleft side canines exhibited a larger α-angle, were positioned more medially and further away from the occlusal plane compared to the non-cleft canines (p<0.0001).
Summary/Conclusion: This study has shown a considerable difference for canine angulation and position between cleft and non-cleft side.
Dentoalveolar arch dimensions and palate length in boys with unilateral cleft lip and palate (uclp)
T Petrová¹, M Koťová²
¹-, -, -, Kosovo, ²Fakultní nemocnice Královské Vinohrady, Prague, Czech Republic

Background: The authors would like to open the discussion about careful evaluation of each cleft lip and palate patient and the applicability of different treatment timings in each of the cases.

Aims: To compare selected length and width parameters of maxilla in patients with complete unilateral cleft lip and palate and different timing of primary cheiloplasty

Methods: Inter canine and inter molar width was evaluated as well as the length of the palate expressed by a perpendicular from papilla incisiva to tuberal tangent of the maxilla. A group of 16 boys (mean age 8 years) with UCLP and neonatal cheiloplasty and a group of 18 boys (mean age 9 years) with UCLP and cheiloplasty performed in 3 months was evaluated in the first part of the pilot study. Both patient groups were then compared with figures discovered by the same methods in healthy children of czech origin and adequate age (Pěnkava, 1975) and with a group of patients with UCLP with a different timing of operations (Peterka, 1975). The second part of the study will include a group of patients with UCLP before the start of active orthodontic treatment.

Results: Several differences in the observed measurements were discovered while comparing the different groups of UCLP patients together and with the healthy population. The most significant result was observed in papilla incisiva – tuberal tangent measure in neonatally operated patients. It can not be said yet, though, that the neonatal operation has negative effects on the growth of the jaw.

Summary/Conclusion: Insufficient development and growth of maxilla in cleft patients is due to the origin of this congenital anomaly, but also due to earlier termination of growth and shows significant connotations with the results of the primary operations. Therefore it is important to monitor the efficiency of the individual treatment protocols.
Final post-treatment occlusion in patients with unilateral cleft lip and palate

J Bollhalder¹, G Antonarakis², M Eichenberger³, G La Scala⁴, G Herzog⁵, D Wiedemeier⁶, C Staudt⁷

¹Orthodontist, Private Practice, Zurich, ²Division of Orthodontics, University Clinic of Dental Medicine, University of Geneva, Geneva, ³Division of Orthodontics, Center of Dental Medicine, University of Zurich, Zurich, ⁴Pediatric Surgery, HUG, University Center of Pediatric Surgery of Western Switzerland, Geneva, ⁵Orthodontist, Private Practice, Pully, ⁶Statistical Services, ⁷Cleft Lip and Palate Unit, Division of Orthodontics, Center of Dental Medicine, University of Zurich, Zurich, Switzerland

Background: Studies on final post-treatment outcome in patients with clefts are sparse, due to variation in cleft morphology, differences in treatment protocol, long overall treatment time, and lack of standardization of final records.

Aims: To evaluate the final post-treatment occlusion in patients with complete unilateral cleft lip and palate (cUCLP) by comparing the outcome between 1) three centers in Switzerland, 2) males and females, 3) right- and left-sidedness of the cleft, 4) cleft and non-cleft sides.

Methods: Final post-treatment study casts of patients with cUCLP at adult age were obtained from three centers in Switzerland (Geneva, Lausanne, Zurich), the casts were anonymized and the occlusion was scored using the Modified Huddart/Bodenham index (MHB).

Results: 56 patients were studied: 35 males (20 orthognathic surgery (OS), 15 no OS), 21 females (12 OS, 9 no OS), 46 left-sided (30 OS, 16 no OS), 10 right-sided (2 OS, 8 no OS).

The treatment center as well as the sex had no significant influence on the final occlusion. The median MHB score of the total sample was 0 (lower quartile -1; upper quartile 2). When the cleft was on the right side, compared to left-sided clefts, the MHB score was more negative, with a tendency seen for the total arch score (p = 0.055) and with a statistical significant difference for the labial (p = 0.044) and the anterior buccal segments (p = 0.043). On the cleft side compared to the non-cleft side, the MHB score was more negative for the anterior buccal (p = 0.002) and the total buccal segments (p = 0.006).

Summary/Conclusion: This Swiss-wide study has revealed a very satisfactory final post-treatment occlusion in patients with cUCLP using when necessary a combined surgical and orthodontic approach. The more constricted buccal occlusion on the cleft side reflects the difficulty in correcting the more palatally positioned lesser maxillary segment. The influence of cleft sidedness on the occlusal outcome may be explained by initial cleft severity, by the indication for orthognathic surgery or by the handedness of the orthodontist and/or the surgeon.
Dental arch relationships in preadolescent unilateral cleft lip and palate patients

W Urbanova¹, M Kotova¹, E Leamerova²
¹Department of orthodontics and cleft anomalies, Dental Clinic 3rd Medical Faculty Charles University Faculty Hospital Královske Vinohrady, ²Plastic Surgery Clinic, 3rd Medical Faculty Charles University Faculty Hospital Královske Vinohrady, Prague, Czech Republic

**Background:** Assessing the mean GOSLON Yardstick Score for dental arch relationships in unilateral cleft lip and palate patients (UCLP) has been validated in the Eurocleft study and appears to be feasible in this context and sample size. Also many other studies were done using this particular yardstick in preadolescent UCLP children, so the results are easily comparable.

**Aims:** Our aim was to evaluate the dental arch relationships in a preadolescent UCLP patients from Prague cleft center by using the GOSLON score and compare the results with existing GOSLON scores from other studies.

**Methods:** 33 children (born between 2000-2003) from Prague Cleft Center, affiliated with the Faculty Hospital Královske Vinohrady, underwent a two-stage repair: the lip was repaired at 7 months using Tennison-Randall technique, while palatoplasty was done at 36 months using Wardill-Kilner technique combined with vomer flap. The mean age when records were taken was 9.1 years (SD = 0.9; range: 7.3 – 10.2). The gender proportion was 65% males to 35% females. Alveolar bone grafting was not yet performed, all the models used for the investigation were taken right before the procedure. The GOSLON Yardstick was used to rate the dental arch relationship. The models were coded and placed in random order. One rater experienced in the treatment of patients with cleft lip and palate scored the models in the presence of the reference models. Prior to the rating session, a calibration exercise was carried out. In order, to assess intra-rater reliability, 15 randomly selected models were reassessed after a 60-minute break.

**Results:** Intra-rater agreement was very good: the kappa for intra-rater concordance was 0.959. The mean GOSLON score for the preadolescent UCLP patients was 3.21. 33.4% of participants were scored 1 or 2, 21.2% participants were scored 3, and 45.4% participants were scored 4 or 5. The occlusal outcome achieved in Prague is comparable with an average occlusal outcome reported by the UK and American cleft teams, but less favorable than results reported from Warsaw, Vienna and Nijmegen.

**Summary/Conclusion:** The preadolescent UCLP patients treated in Prague cleft center demonstrate relatively unfavorable outcome. Almost half of them is being scored GOSLON score 4 and 5, having negative overjet even before the puberty growth spurt, which is likely to get worse with the subsequent growth. Further research is needed in order to estimate the factors that influence such results (e.g. surgical protocol, ethnical background, orthodontic treatment).
OS 29.9
Pre-surgical functional maxillary orthopedics panacci’s modification: our experience
D Bohórquez Benit¹, A Bohórquez Benítez², T Pannaci², M Munill ²
¹Maxillofacial Surgery, Vall d’Hebron University Hospital , ²Maxillofacial Surgery, Vall d’Hebron Hospital , Barcelona, Spain

Background: There are different orthopedic techniques that can be used to improve the presurgical situation of the cleft patient. The use of a functional maxillary orthopedic plate and nasoalveolar molding progressively reduces the severity of the fissure and consequently could improve the aesthetic result of the surgery.

Aims: The objective of this work is to present our experience with the technique, and the evolution of our protocol since the Functional Maxillary Orthopedics and NasoAlveolar Molding has been implemented in our unit.

Methods: We present in detail a series of 35 patients treated with the Pannaci’s Modification.

Results: We found that the Panacci’s Modification is a reproducible technique. Once the Functional Maxillary Orthopedics treatment is started, the surgery can be done when the orthopedic objectives are achieved. This causes us to change the timing of the surgeries in our protocol, and now, we are performing the primary lip, nose and soft palate repair in one procedure.

Summary/Conclusion: The Functional Maxillary Orthopedics and NasoAlveolar Molding is effective. Reducing the number of surgeries of our protocols is something to have in consideration.
Background: Deviated vomer and twisted premaxilla in bilateral cleft lip and palate patient, compromises nasal airway by asymmetric skeletal findings. The twisted premaxilla can be treated by orthodontic devices, but correction of the deviated vomer and septum often need operative procedures in adulthood. Thus, deviated vomer and twisted premaxilla can results in a permanent upper airway obstruction (UAO) from childhood to adulthood.

Nasal-alveolar-molding (NAM) forces the premaxilla backward and can cause or at least doesn't prevent the deviation of the vomer and septum. Moreover, asymmetric nasal skeletal findings in adulthood make skeletal correction by rhinoplasty more difficult. Even when performing bony and septal dissections, conchotomy and osteotomies as well as cleft related corrections of the nasal dome and columella, the UAO can still exist postoperatively.

Aims: The aim of this study was to present the results of an orthodontic device designed to keep the premaxilla and vomer in the midline position and to prevent vomer and septal folding.

Methods: Since 2015, 6 patients with bilateral cleft lip and palate were included in this study and worn an removable individual made-up orthodontic device to force the premaxilla backward and simultaneously straightening the vomer. This device was incorporated after birth and worn until surgically closure of the palate at 6th month. Endoscopic control of the nasal cavity was done during lip closure at 10th month.

Results: In 5 out of 6 patients, premaxilla and vomer were symmetrical in a midline position without deviation or folding during closure of the palate. In one patient, the vomer was found to be straight but during operation was not in the midline position. Endoscopic evaluation of the nasal cavity before lip closure confirmed the results before cleft palate closure. In the one patient where preoperative treatment failed, the nasal septum was deviated and the nasal floor was found to be narrower on the deviated side. Compared with conventional treated patients with bilateral cleft lip and palate, the time of operation in pretreated patients was approximately one hour shorter.

Summary/Conclusion: Deviated vomer and premaxilla seriously compromises operative closure of the nasal floor and palate in patients with bilateral cleft lip and palate. Preoperative treatment with this orthopedic device correct the deviated position of the premaxilla and straighten the vomer. Moreover, symmetrical nasal floor with non-deviated vomer and septum are key conditions for optimal functional and esthetic nasal rehabilitation.
Quantitative analysis of presurgical orthopedic procedures for patients with orofacial clefts in Europe and North America: a follow up to the Eurocleft Project

J. Hellmann\textsuperscript{1}, A. Jäger\textsuperscript{1}, M. Martini\textsuperscript{2}, C. Breugem\textsuperscript{3}, B. Al-Nawas\textsuperscript{4}, S. Bartel\textsuperscript{5}, B. Braumann\textsuperscript{6}, B. Grayson\textsuperscript{7}, W. Shaw\textsuperscript{8}, F. Stahl\textsuperscript{9}, N. Daratsianos\textsuperscript{1}

\textsuperscript{1}Department of Orthodontics, \textsuperscript{2}Department of Maxillofacial and Plastic Surgery, University of Bonn, Bonn, Germany, \textsuperscript{3}Department of Plastic Surgery, University Medical Center Utrecht, Utrecht, Netherlands, \textsuperscript{4}Department of Oral and Maxillofacial Surgery, University of Mainz, Mainz, \textsuperscript{5}ENT Department, University of Halle, Halle, \textsuperscript{6}Department of Orthodontics, University of Cologne, Cologne, Germany, \textsuperscript{7}New York University Langone Medical Center, New York, United States, \textsuperscript{8}Manchester University Dental Hospital, Manchester, United Kingdom, \textsuperscript{9}Department of Orthodontics, University of Rostock, Rostock, Germany

**Background:** The treatment of orofacial clefts is complex, lasts many years and involves many disciplines. It is usually performed in cleft centres, where specialists can well coordinate the treatment steps. There are no generally accepted treatment guidelines: treatment differs in terms of procedures and their timing. The treatment protocols of European cleft centres were presented and analysed in The Eurocleft Project 1996-2000, about 20 years ago. There is no comparable analysis for North America.

**Aims:** The University of Bonn in cooperation with the German Cleft Palate Craniofacial Association (GCPA) and the European Cleft Palate Craniofacial Association (ECPCA) has set itself the goal of analysing the treatment protocols for patients with orofacial clefts in different European and North American countries. Following The Eurocleft Project 1996-2000, a quantitative analysis of disciplines involved, the procedures, their timing and other aspects was performed.

**Methods:** The contact data of the European and North American cleft centres were collected in a standardised procedure with the support of national and international craniofacial organisations and by manual internet search. An online questionnaire was developed and sent to each cleft team. The questionnaire covered the range of surgical, orthodontic and general questions. The questions were designed to identify the type and timing of the treatment procedures according to the standard treatment protocol of each cleft centre, caseloads, qualification of the specialists and other details. The questionnaire also addressed aspects regarding other disciplines (ENT, speech therapy). No objective quality assessment was made. The poster focuses on the results for the presurgical orthopedic procedures.

**Results:** For presurgical orthopedics, the most frequently used intraoral-only appliances are passive plates. Fixation of the plates is supported mostly by adhesive gel and in some centres there is no fixation at all. The most frequently used combined extraoral-intraoral appliance is the NAM-appliance as described by Grayson, which is used for uni- and bilateral clefts. In most centres, NAM-appliances are used for both mild and severe clefts, with a tendency to severe cases. Extraoral-only devices (in form of lip tapes) are seldomly used. Presurgical orthopedics is mostly used in the first 6 months of life, closely followed by a shorter period of time (only in the first 3 months of life).

**Summary/Conclusion:** The use of presurgical orthopedics has considerably risen compared to The Eurocleft Project 1996-2000.
Orthognathic Surgery

OS 30.1
Clinical refinements in orthognatic surgery
A Rosenberg¹
¹Department of Oral and Maxillofacial Surgery, Center of Excellence in Congenital Orofacial and Dental Anomalies, Utrecht, Netherlands

Abstract Content: Cleft lip palate is a burden in quality of life in young patients. With an incidence of approximately 1:600 new births, it is one of the most common birth deformities. Early in life cleft lip palate patients need correction for a proper functional development. However, corrections early in live may interfere with growth, leading to cleft lip and palate stigmata at older age. Restoration of function is more difficult to obtain at older age, than correction of growth impairment. Growth impairment due to correction at early age is therefore regularly preferred over functional impairment. Growth impairment is corrected at the end of growth, mostly at 18 years of age. A Le Fort I osteotomy is indicated, if a pseudo class III skeletal malocclusion in combination with symptoms of maxillary growth impairment is found. Two principles are followed: Opening of the cleft region in case of transversal collapse, repositioning of the segments, with closure at a later stage and advancement of the segments, and simultaneous closure of the palate. For the patient, parents and for scheduling of the orthodontic treatment it is important to know if a Le Fort I osteotomy will be indicated. If at an age 11.36 years ANB was below 6 there was a 78% chance that a Le Fort 1 osteotomy was required by the age of 18. New 3D computer planning in non-cleft cases makes shared decision-making possible. In cleft patients, planning of the soft tissues is still somewhat unprecise. 3D-printed wafers or even plates may be used for proper positioning of the maxilla instead of wafers.
Abstract Content: For excellence in maxillofacial reconstruction, we prefer prefabricated composite grafts from the scapula and iliac crest. These "like tissues" consist of mucosa-coated bone grafts with already osseointegrated dental implants in a correct three-dimensional anatomical shape, which we first described in 1996. The lecture reports on the use of these grafts in the reconstruction of the "nasomaxillary complex" as tertiary osteoplasty in patients with unilateral and bilateral complete clefts. The "Nasomaxillary Complex" consists of the mucosa coated alveolar process and bony palate, as well as the pyriform section of the maxilla, which is restored in the style of a "like-like" reconstruction with these grafts in size, shape, and tissue-structural identity. In order to meet the respective anatomical conditions, one can combine these grafts with other bone blocks as so-called "stack plasty". Since the increasing extent a maxillary defect results in typical three-dimensional deformations of the upper jaw, such as the midface retraction and the transverse maxillary compression, we combine tertiary cleft osteoplasty with orthognathic surgery. For the three-dimensional correction of the upper jaw, the Distraction osteogenesis is best suited as it can be used to correct the maxillomandibular relations in all spatial directions and at simultaneously open the space for the graft insertion. Far beyond the traditional osteoplastic bridging of the alveolar cleft, tertiary osteoplasty using "like tissues" is thus able to completely reconstruct the anterior maxilla and the "periodontal unit" consisting of bony alvolar process, bone attached gingiva and perio-osseointegrated implants, which is essential for aesthetic implant dentistry in these patients. Restoring the pyriform region as the bony base of the nose provides the best conditions for subsequent septorhinoplasty. With this surgical procedure, a non-discriminatory "smile design" and a good esthetics of the "cental oval of the face" can be achieved, even in patients with extended bilateral clefts. Literature: Vinzenz K, Schaudy C. Complex soft tissue and bony Maxillofacial reconstruction. In: Unfavorable Results in Plastic Surgery: Avoidance and treatment, 4th Edition; Chapter 50, Eds.: S. Thaller, M. Cohen, Thieme USA 2018
Early experience with a new internal le fort i distractor in patients with cleft lip and palate – the trans-nasal device

M Lycka¹, H Hendrick¹
¹Plastic and Craniofacial Surgery, Children's Mercy Hospital, Kansas City, United States

Background: Le Fort I distraction is indicated in a small subset of patients with cleft lip and palate who have severe maxillary deficiency, often in the skeletally immature patient, when the anticipated magnitude of the bony movement is deemed likely to be unstable using conventional orthognathic surgery. Le Fort I advancement by distraction has been performed successfully using both external and internal devices, and is largely dependent on operator preference. The external halo device, while popular among many surgeons for ease of vector control, is objectionable to most patients, for obvious reasons. The alternative, internal devices, while being inconspicuous, suffer from problems such as vector control, trismus from distraction rods impinging on the coronoid process, burying of activation arms in the soft tissues during distraction, and bony anatomic limitations. In an effort to alleviate many of the shortcomings with current internal devices, a new internal trans-nasal Le Fort I distractor was designed.

Aims: It is the aim of this presentation, through clinical and radiographic analysis, to chronicle the experience with a new internal trans-nasal Le Fort I distractor.

Methods: After IRB approval, patients with cleft lip and palate and severe maxillary deficiency (at least 8 mm of negative overjet) who were treated with the trans-nasal Le Fort 1 distractor were retrospectively reviewed. Preoperative virtual simulation of the Le Fort I osteotomy and distraction vector was performed. Distractors were bent on a 3D model preoperatively. A 5 day latency period preceded a distraction rate of 1 mm per day. Cephalometric images were evaluated and compared by the same orthodontist at time point zero (preoperative) and 6 months postoperatively, and when available, after one year. Speech outcomes were measured before and at least 6 months after surgery. Complications were recorded.

Results: Four male patients with bilateral cleft lip and palate (ages 11, 12, 14, and 15) underwent the maximum advancement allowed by the device (25 mm). Follow-up averaged 1.5 years. Consolidation averaged 5 months. Bony union was confirmed in all cases at the time of removal of devices, both radiographically and clinically. All patients achieved positive overjet (-9 mm to 2 mm), that was maintained at one year. Average SNA changed from 75.3 degrees preoperatively to 82.4 degrees postoperatively. Average ANB angle changed from -6.9 to 3.7 degrees, or a tendency to Class 2 overcorrection. There was an overall increase in upper anterior facial height by 6 mm. Families reported ease of turning with minimal discomfort reported by patients. All patients maintained normal mouth opening during and after the distraction phase. Two of the patients developed localized pin site infections after the distraction phase that were treated successfully with oral antibiotics. There were no nose bleeds. Two patients with borderline velopharyngeal function preoperatively developed velopharyngeal insufficiency postoperatively that did not resolve 6 months postoperatively, necessitating further speech surgery.

Summary/Conclusion: The trans-nasal Le Fort I distractor can be an effective device to advance the deficient maxilla, and is well tolerated by patients.
OS 30.4

Comprehensive long term evaluation of a sample of unilateral cleft lip and palate treated in smile house of milan

L Autelitano¹, M Meazzini¹, V Battista¹, A Rezzonico¹, F Biglioli¹, R Brusati¹

¹SMILE HOUSE MILAN, Milano, Italy

Background: A universally accepted protocol for unilateral cleft lip and palate (UCLP) treatment does not exist. Comprehensive assessment of long term results has to be done for a correct evaluation of functional and morphologic outcome of treatments and iatrogenic effects of surgery.

Aims: The aim of this paper was to assess growth, speech and morphological results at the end of the growth of patients with unilateral cleft lip and palate treated with the two stages Milan surgical protocol in Smile House of Milan.

Methods: Craniofacial growth was evaluated with cephalometric analysis and the theoretical need for orthognathic surgery. Nasolabial appearance was qualitatively assessed using the Asher McDade Aesthetic Index. Speech was assessed using the Gos.sp.ass '98 modified for italian language scoring system. Burden of care was recorded in terms of number of secondary surgical procedures and orthodontic treatment burden.

All of the patients were treated and evaluated at Smile House of Milan San Paolo University Hospital. We have recalled for evaluation 52 consecutive patients treated by the same surgeon, 12 patients didn’t come for assessment.

According to the protocol of the Centre, the first surgical step (average age of 6 months) was cheilorhinoplasty (Millard modified Delaire technique) together with soft palate repair (Pigott technique). The second step (average age of 35 months) was hard palate and alveolar repair performed simultaneously with an early secondary gengivo alveolo plasty.

Results: 56% of patients didn’t need any further surgery after the two stage protocol. At the time of evaluation 20% of patients underwent orthognathic surgery, 27,5% secondary rhinoplasty, 7% secondary bone graft, 3 % VPI surgery, 17,5% lip revision

Summary/Conclusion: The two stage surgical protocol of Milan, Smile House, seems to be effective for treatment of UCLP, with good results in term of speech, labial appearance and alveolar cleft management, reducing the total burden of care. Nevertheless, maxillary growth was moderately impaired by the protocol.
Le fort I osteotomy in cleft patients: maxillary advancement and velopharyngeal function
R Lahtinen¹, S Alaluusua¹, J Leikola¹, A Heliövaara¹
¹Department of Plastic Surgery, Helsinki University Hospital, Cleft Palate and Craniofacial Center, Helsinki, Finland

Background: Le Fort I osteotomy is commonly used in the correction of maxillary retrusion and crossbite. Maxillary advancement may affect speech in cleft patients.

Aims: To evaluate whether the amount of maxillary advancement in Le Fort I osteotomy affects velopharyngeal function (VPF) in cleft patients.

Methods: This retrospective cohort study examined 93 non-syndromic patients with unilateral cleft lip and palate (n = 46), bilateral cleft lip and palate (n = 22) and cleft palate (n = 25). All patients had undergone a Le Fort I osteotomy or bimaxillary osteotomy at the Cleft Palate and Craniofacial Center, Helsinki University Hospital. Pre- and postoperative lateral cephalometric radiographs were digitized to measure the amount of horizontal advancement and vertical lengthening of the maxilla during osteotomy. Pre- and postoperative speech was assessed perceptually and instrumentally by experienced speech therapists. To assess reliability, 20 cephalometric radiographs were digitized twice. Student’s t-test and kappa statistics were used in the statistical analyses.

Results: The mean advancement of point A was 4.0 mm horizontally (range: –2.8–11.3) and 3.9 mm vertically (range – 14.2–3.9). The amount of horizontal or vertical movement did not significantly influence the VPF. The interrater reliability was good.

Summary/Conclusion: The amount of maxillary advancement does not affect the velopharyngeal function in cleft patients.
Impact of orthognathic surgery on velopharyngeal function by evaluating speech and cephalometric radiographs

D Impieri¹, K Tønseth¹, Ø Hide², E Brinck¹, H Høgevold¹, C Filip¹
¹Plastic and Reconstructive Surgery, Oslo University Hospital, ²Statped Sørøst, Oslo, Norway

Background: Maxillary retrognathia is a well-known consequence of abnormal facial growth in patients with cleft lip and palate. Orthognathic surgery improves facial appearance and occlusion but may induce velopharyngeal insufficiency.

Aims: The aim of this study was to evaluate the effect of orthognathic surgery on velopharyngeal function by using speech analysis and lateral cephalometric radiographs.

Methods: We conducted a retrospective study of 47 patients who underwent maxillary advancement ± mandibular setback between 2006 and 2016. Preoperative and 1 year postoperative audio recordings were blinded for scoring by three trained speech therapists. Preoperative and 1 year postoperative lateral cephalometric radiographs were used to obtain information about skeletal movement and its relationship with the velopharyngeal area. Correlations between speech outcomes and cephalometric radiographs were determined.

Results: Hyponasality improved significantly after surgery (p < 0.05), whereas hypernasality deteriorated significantly only in patients who underwent maxillary advancement alone (p < 0.05). No difference in speech parameters was found between patients with hypernasality or patients who had a pharyngoplasty preoperatively and the rest of the group. No correlation was found between the amount of maxillary advancement and hypernasality. A significant correlation (r < -0.49, p < 0.05) was found between the preoperative velar length and hypernasality postoperatively.

Summary/Conclusion: Maxillary advancement has a negative impact on velopharyngeal function, whereas bimaxillary surgery seems to protect from deterioration. No difference was found in the amount of maxillary advancement or in velopharyngeal measurements between the Le Fort I group and the bimaxillary group. A short soft palate is a predictor of hypernasality after orthognathic surgery.
Outcome assessment of computer guided mandibular distraction osteogenesis: a clinical study
M Elkassaby\(^1\), Y Elhadidi\(^1\), H Hany\(^1\), H Seleem\(^1\), M Taha\(^1\)
\(^1\)oral and maxillofacial surgery, faculty of dentistry, ainshams university, cairo, Egypt

Background: Distraction Osteogenesis (DO) is an integral part in management of mandibular defects associated with HemiFacial Microsomia (HFM) and post-ankylositic mandibular defects.

Aims: The current study aimed to assess the effect of Three-Dimensionally Constructed Computer Guided Splints on the cosmetic outcome of DO.

Methods: This study extends upon previous conventionally planned cases of HFM and Post-ankylositic patients in the Cleft Care Center and Out-Patient Clinic of our department. The study cases were 6 patients whom had computer guided surgery using prefabricated splints and the control cases are records of 6 patients whom were previously operated in our department.

Results: Assessment of effect of computer planning on mandibular advancement (SNB angle) showed Significant (P = 0.005) improvement in (study) (Mean= 89 %, SD = 7%) compared to (control) (Mean= 61%, SD= 8%). Assessment of effect of computer-guided surgery on chin deviation showed significant (P = 0.005) improvement in (study) (Mean= 63 %, SD = 7 %) compared to (control) (Mean= 18.3 %, SD= 16.9 %). Assessment of effect of computer-guided surgery on ramus height showed a non-significant (P = 0.2) improvement in study (Mean = 89 %, SD=10 %) compared to control (Mean = 72 %, SD=19.7 %). Assessment of effect of computer planning on correction of smile deviation showed non-significant (P = 0.8) improvement in (study) (Mean= 47%, SD = 38%) compared to (control) (Mean= 41%, SD= 14%).

Summary/Conclusion: Conclusion: Computer planning was very successful in improving the chin orientation both antro-posterior and mesio-distal direction (chin deviation). However, it failed in achieving a significant increase in vertical ramal height and in smile deviation correction. Lastly, it was found that patients from both groups would benifit from soft tissue augmentation to enhance the lateral projection of the mandibular body.
Early evaluation of maxillary growth in patients with unilateral cleft lip and palate: comparing Malek modified and Talmant surgical protocols

A Lehn1,2, C Bäumler2, G Captier2, M Bigorre2

1Pediatric surgery, University Hospital of Strasbourg, Strasbourg, 2Plastic pediatric and craniofacial surgery, Hôpital Lapeyronie, Montpellier, France

Background: The aim of palatal repair is to achieve the best possible speech, hearing and midfacial growth as well as social integration. The controversy of the timing and technique for palatal repair to achieve these goals has been discussed for many years.

Aims: The aim of this study was to evaluate maxillary growth prior to orthodontic treatment in patients with unilateral cleft lip and palate (UCLP) operated with two different surgical protocols performed by the same surgical team.

Methods: Patients with a non-syndromic UCLP were included at age 4. Malek modified surgical protocol consisted in a veloplasty at age 3 months, closure of the hard palate and lip repair at 6 months. Talmant surgical protocol associated lip repair, primary septorhinoplasty and veloplasty at age 6 months, and closure of the hard palate between 14 and 18 months. Clinical and dental casts measurements were performed before beginning pre-operative orthodontic treatment to prepare for alveolar bone grafting. Intercanine width, incisor overjet, GOSLON yardstick and dental arch relationships were assessed, after informed consent was obtained.

Results: 67 patients were included: 22 were operated with Malek technique from 1994 to 2006 and 45 with Talmant technique from 2007 to 2014. Talmant’s two-stage palatoplasty reaches mostly GOSLON scores of 1 or 2 (mean 2.1) whereas Malek’s early hard palate closure reaches mostly GOSLON scores of 3 or 4 (mean 3.6). The mean intercanine width was 25.36mm in Malek’s protocol and 26.18mm in Talmant’s protocol. The sagittal growth was improved with 87% of positive incisor overjet in Talmant’s protocol versus 32% in Malek’s protocol.

Summary/Conclusion: Talmant technique shows an improved maxillary growth, both transversal and sagittal. The main differences with Malek technique are the later closure of the hard palate without leaving denuded bone and the early septorhinoplasty restoring the nasal respiration.
**OS 30.9**

**Versatile 3d computer-assisted two-layer and three-layer models of the face**

K Ueda¹, D Mitsuno¹, M Katayama¹

¹Plastic and Reconstructive Surgery, Osaka Medical College, Takatsuki, Japan

**Background:** Recent advances in three-dimensional printing technology are manifest in plastic surgery. The technology is reported to be beneficial for replacing soft tissues. We made realistic three-dimensional, computer-assisted, two-layer elastic model (PRS 2017) and three-layer models (PRS 2018) and used these models for simulation surgery.

And also we reported that AR technology was helpful for evaluation of the body surface in several clinical applications (PRSGO 2017) and devised a method that could align the surgical field and holograms precisely (PRS 2019).

**Aims:** We utilized these two-layer and three-layer facial models for simulation surgery. Our aim is to confirm the usefulness of these models in clinical cases and to explore the possibility of collaboration by these 3D models and AR technology in simulation surgery.

**Methods:** The two-layer elastic model has the surface layer with polyurethane for skin and the inner layer with silicone for subcutaneous tissue and both the layers adhere to each other. And we have made new separable models whose surface layer can be detached from the inner layer (2018 PRSGO). The three-layer model has one more layer representing bone. These models were applied to simulation surgery in many clinical cases with facial bone fracture, congenital anomaly, orbital tumor and etc.

HoloLens from Microsoft, which is a head-mounted mixed reality device can display a precise three-dimensional model stably on the real visual field as hologram.

We tried to display many holograms of skin, facial bones and angiography on the models by using our reported method.

Using separable two-layer elastic models and HoloLens, young doctors and residents analyzed how to display holograms of 2D, 3D or movie of the typical local flap simulation on the models.

**Results:** Using two-layer elastic models, we could teach residents and young doctors how to make several typical local flaps and to perform cheiloplasty.

Using three-layer models of facial multiple fracture cases, we could understand simulation of effective approaching method to the fracture site.

And using three-layer models of congenital anomaly cases, we could guess the size of the bone or cartilage graft and could know the facial profile change of the soft tissue after osteotomy and bone grafting.

Using HoloLens, we could display many holograms of skin, facial bones and angiography on the models.

The residents and young doctors could understand the typical flap design and simulation surgery deeply by using HoloLens and separable two-layer elastic models.

Our model enables residents and young doctors to understand three-dimensional design and flap movement. Especially three-layer models enable us to guess the volume and size of implant, bone and cartilage graft and to know the soft tissue profile change after osteotomy and bone grafting.

And also using HoloLens, we can deeply understand to simulate the operation on face-like structures that have complicated three-dimensional structures.

Collaboration by these 3D models and AR technology has the possibility to give us development in simulation surgery.

**Summary/Conclusion:** We made realistic three-dimensional, computer-assisted, two-layer elastic models and three-layer models and used these models for simulation surgery in many clinical cases. And we explored the possibility of collaboration by these 3D models and AR technology in simulation surgery.
**OS 30.10**  
Periosteal distraction by magnets for cleft palate defects – a finite element study  
P Nalabothu¹, M Dalstra¹, C Verna², A Mueller³  
¹Department of Orthodontics and Pediatric Dentistry, University Dental Centre Basel, ²Department of Orthodontics and Paediatric Dentistry, University Dental centre Basel, Basel, Switzerland, University of Basel, ³Clinic for Oral and Maxillofacial Surgery, University Hospital, Basel, Switzerland

**Background:** Cleft Palate closure by surgery depends on the adequacy of available palatal mucosal tissue. Insufficiency of such tissue is a cause for concern in the closure of severe wide clefts of the hard palate. As tissue mechanics are crucial in the normal physiologic behaviour of cells, other approaches for tissue regeneration may be more successful when mechanical strain is applied.

**Aims:** The aim of this study was to evaluate the mechanical loading during mucoperiosteal tissue expansion in the cleft palate of newborns with unilateral and bilateral clefts

**Methods:** 3D CT scans of newborns with cleft lip and palate were used to create an FE model of the maxilla using dedicated image analysis software. The material properties of the individual elements in the model were linked to the local grey values of the scans edges of an osteodistractive implant across the palatal cleft. The external loading conditions were simulated by attraction forces generated by two magnetic strips, whose magnitude was based on previous in-vitro experiments. The force-distance relationships were experimentally determined for a number of small dental magnets separated by the various thickness of soft tissues. For a number of different magnetic force arrangements, the strains in the bone areas and the periosteum surrounding the palatal cleft were then analysed.

**Results:** The experiments showed that the presence of soft tissue had no or little influence on the force-distance relationships for the magnetes. The periosteal bone strains in the palate were predominantly tensile in the direction of distraction. Furthermore, the associated strain magnitudes were in the order of the adaptive and bone gain windows of Frost’s mechanostat theory. The positions where the magnetic forces were assumed to be applied, did have an influence on the strain distribution in the periosteum and underlying bone: a more concentrated distribution of the magnetic forces would lead to higher strain peak values in the direct vicinity of the implant.

**Summary/Conclusion:** The analyses suggest that the strains in the periosteum and underlying bone in the palate, generated by attractive forces, are of such nature and magnitude that is compatible with adaptive bone formation.
Speech Treatment

OS 31.2
International initiatives to increase opportunities for speech therapy post-cleft palate repair: outcomes and continuing challenges

C Crowley

Abstract Content: Children born with cleft lip and palate have been receiving surgeries through missions and local surgeons for decades. Many of them also need speech therapy post-cleft palate surgery. But, in many places in the world there are simply not enough speech therapists and even in those places that have speech therapists, often they lack the skills to address compensatory cleft palate speech errors. The international cleft palate surgical world now realizes the value of intensive speech therapy to address these compensatory errors post-surgery. Dr. Crowley has worked extensively developing capacity for cleft palate speech therapy. She has worked with several international cleft organizations, with the bulk of her work with Smile Train as its Strategic Partner for speech therapy. Dr. Crowley will present her work in providing in-person trainings and advanced mentorship. She will also share her online initiatives on her website, leadersproject.org, such as her online cleft palate speech therapy course in Spanish, English, or French and her downloadable train-the-trainer course. Her website also provides online speech therapy materials in many languages including Spanish, French, English, Hausa, Yoruba, Amharic, Oromo, Luganda, Mandarin, and Hindi. All these materials are available for free download at leadersproject.org
An evaluation of a parent led, therapist supervised, articulation therapy programme for children with cleft palate speech disorders.

T Sweeney¹, M O'Regan², F Hegarty³, D Sell⁴

¹Speech Therapy, Speech at Home, ²Statistics, Trinity College Dublin, ³Medical Physics, Children's Hospital Group, Dublin, Ireland, ⁴Centre for Outcomes and Experience Research in Children’s Health, Illness and Disability (ORCHID), Great Ormond Street Hospital for Children NHS Foundation Trust, London, United Kingdom

Background: Fifty percent of children with cleft palate have persistent speech problems at 5 years of age requiring speech therapy. However, there is a lack of access to regular targeted therapy in many countries. Although parent training/involvement has positive outcomes in early communication skills in cleft palate and non-cleft speech disorders, little is known about parents undertaking intervention for cleft speech disorders. Connected Health programmes have been used to provide speech therapy services to those who would not otherwise receive therapy.

Aims: To evaluate an alternative method of facilitating improvements in speech by engaging parents.

Methods: Forty-six children, aged 2.9 to 7.5 years, were included in this two-phase, two-centre non-inferiority randomised controlled trial. Informed consent and assent was obtained. In the parent-led arm, parents attended a 2-day training course, were given a speech programme and conducted a 12-week home-based therapy. Parents were supported by a Cleft Speech and Language Therapist using connected health (Facetime) and a one-to-one session. In the control arm, the group received six therapy sessions with a research Speech and Language Therapist comparable to that provided by local services. Speech recordings were undertaken pre- and post-intervention. Percent consonant correct (PCC) scores were the primary outcome measure. Activity and Participation were analysed using the Focus on Outcomes for Children Under Six (FOCUS) and Intelligibility in Context Scale (ICS) scores. Acceptability to parents was evaluated using telephone interviews or focus groups conducted by independent Psychologists/Speech and Language Therapists.

Results: Both groups showed a statistically significant improvement in PCC scores (mean change = 21%; p<0.001; 95% CI 12.27-19.86), the FOCUS scores (mean change = 18.87; p<0.001; 95% CI 10.87-25.73) and the ICS scores (mean change = 2.27 p<0.001; 95% CI 1.41-3.13). There was no statistically significant difference between the groups. Parents reported how they “learned a lot about speech problems and speech therapy strategies”. Overall parents gave positive feedback, reporting a change in their child’s speech and reporting an increase in their child’s confidence, which reinforced their own sense of well-being.

Summary/Conclusion: This trial showed that parent led articulation therapy is effective in changing speech, activity and participation outcomes for children with cleft palate, when supported by a Cleft Speech and Language Therapist using connected health, with implications for an alternative model of service delivery. It provided insights into how parents deliver intervention at home.

The study was funded by Cleft Lip and Palate Association of Ireland, Temple Street Foundation and CLEFT – Bridging the Gap.
European Reference Networks

OS 32.3
The european cleft gateway – a directory of cleft resources in europe
G Davies
Executive Director, European Cleft Organisation, Rijswijk, Netherlands

Background: The European Cleft Gateway was developed as part of the European Science Foundation-funded EUROCleftNet research programme as an online directory of cleft resources in Europe. It was conceived as a successor directory to that of the EU Eurocleft and Eurocran projects that came to an end in 2005. Following the winding up of EUROCleftNet in 2016 the Gateway was redeveloped by the European Cleft Organisation (ECO) and relaunched in Utrecht in 2018.

Aims: The aim was to create a directory of specialist hospitals, health professionals, researchers, patient groups, scientific organisations and NGOs working in the field of cleft lip and palate. A key research element was to provide a listing of all centres wishing to collaborate in studies on prevention and treatment. Important considerations have been free and open access, simple search functionality and the ability to upload own data, subject to approval by administrators.

Methods: There was a two stage data collection process, focusing initially on the data collated by the Eurocran project and then facilitated by voluntary country coordinators. A questionnaire was developed asking details of the cleft centre, team members, case numbers, protocols, funding arrangements, patient groups, research studies and willingness to collaborate in cross centre research. There was an option to return data online, by email or by phone. Results were analysed and posted up to a site managed jointly by EUROCleftNet and ECO.

The first platform for the directory was developed in the UK but the current platform, now managed by ECO and offering increased functionality, was developed in Bulgaria after an open tender process comprising developers in the UK, Bulgaria and Sweden.

Results: The Gateway is live at www.gateway.europeancleft.org and currently lists around 100 cleft teams and over 800 clinicians, 20 scientific organisations and 37 patient groups around Europe. The number of hits is currently around 3500 per month. We are embarking on a new drive to populate and update the directory and to this end it has been promoted during the participant registration process of the 11th European Cleft and Craniofacial Congress.

There has been a high number of Gateway subscribers from Eastern Europe and beyond, which has made the site invaluable in recruiting partners for a number of EU-funded European projects which favour European integration between East and West. Since 2014, nine EU projects have benefited from partners initially identified on the Gateway, including two COST Actions.

Summary/Conclusion: The aims of the project have largely been fulfilled though there need to be continuing efforts to fully populate the directory and ensure all data is kept up to date. This has been an impressive collaboration between clinicians, researchers, patient groups and web developers, facilitated by a European level NGO (ECO). It is hoped it will serve the future interests of the European Cleft Palate and Craniofacial Association, the new European Reference Network for Craniofacial Anomalies, and, above all, help bring about equitable access to good cleft care for patients and their families across Europe.
Quantitative analysis of treatment protocols for patients with orofacial clefts in Europe and North America: a follow up to the Eurocleft Project

N Daratsianos¹, J Hellmann¹, M Martini², C Breugem³, B Al-Nawas⁴, S Bartel⁵, B Braumann⁶, B Grayson⁷, W Shaw⁸, F Stahl⁹, A Jäger¹

¹Department of Orthodontics, ²Department of Maxillofacial and Plastic Surgery, University of Bonn, Bonn, Germany, ³Department of Plastic Surgery, University Medical Center Utrecht, Utrecht, Netherlands, ⁴Department of Oral and Maxillofacial Surgery, University of Mainz, Mainz, ⁵ENT Department, University of Halle, Halle, ⁶Department of Orthodontics, University of Cologne, Cologne, Germany, ⁷New York University Langone Medical Center, New York, United States, ⁸Manchester University Dental Hospital, Manchester, United Kingdom, ⁹Department of Orthodontics, University of Rostock, Rostock, Germany

Background: The treatment of orofacial clefts is complex, lasts many years and involves many disciplines. It is usually performed in cleft centres, where specialists can well coordinate the treatment steps. There are no generally accepted treatment guidelines: treatment differs in terms of procedures and their timing. The treatment protocols of European cleft centres were presented and analysed in The Eurocleft Project 1996-2000, about 20 years ago. There is no comparable analysis for North America.

Aims: The University of Bonn in cooperation with the German Cleft Palate Craniofacial Association (GCPA) and the European Cleft Palate Craniofacial Association (ECPCA) has set itself the goal of analysing the treatment protocols for patients with orofacial clefts in different European and North American countries. Following The Eurocleft Project 1996-2000, a quantitative analysis of disciplines involved, the procedures, their timing and other aspects was performed.

Methods: The contact data of the European and North American cleft centres were collected in a standardised procedure with the support of national and international craniofacial organisations and by manual internet search. An online questionnaire was developed and sent to each cleft team. The questionnaire covered the range of surgical, orthodontic and general questions. The questions were designed to identify the type and timing of the treatment procedures according to the standard treatment protocol of each cleft centre, caseloads, qualification of the specialists and other details. The questionnaire also addressed aspects regarding other disciplines (ENT, speech therapy). No objective quality assessment was made.

Results: Most cleft centres offer a joint consultation with the team up to 4 times a month. In most centres, lip closure for all clefts is performed at 3 months of age with the Millard-technique and in an one-stage approach for bilateral clefts, in most cases simultaneously with primary rhinoplasty or nasal floor reconstruction. The hard and soft palate are mostly closed at 12 months of age using the Von Langenbeck-technique for the hard and Intravelar Veloplasty for the soft palate. About one third of the centres always implements presurgical orthopedics, one third never and the rest in some of the cases. Intraoral-only appliances are mostly used, but combined extraoral/intraoral NAM-appliances are almost equally popular, in particular for bilateral cases. Extraoral-only devices are rarely used. The quadhelix appliance is the most popular device for transversal expansion, typically before later osteoplasty, which is performed by the majority of the centres at 8-9 years of age.

Summary/Conclusion: NAM-appliances seem to be on the rise, but intraoral-only appliances are still used by the majority of colleagues. Compared to The Eurocleft Project, the results concerning the surgical techniques and their timing are similar except for the increased popularity of the Intravelar Veloplasty for the soft palate. The use of presurgical orthopedics has considerably risen compared to The Eurocleft Project 1996-2000.
Nutrition-related epidemiology of children ≤5 years treated for orofacial clefts in low- and middle-income countries: an audit of smile train global database

B Delage¹, P Sheeran², E Stieber³, H de Silva⁴, M Kerac¹
¹London School of Hygiene and Tropical Medicine, London, United Kingdom, ²Smile Train, New York, ³Smile Train, Washington DC, United States, ⁴Guy’s and St Thomas’ NHS Foundation Trust, London, United Kingdom

Background: Children with cleft lip and/or palate experience feeding difficulties caused by structural changes to the oral anatomy essential for suction-based feeding. In resource-poor settings, with reduced access to timely specialist services, orofacial clefts are associated with a variety of health challenges, which carry lasting consequences in those surviving infancy. Moreover, widespread malnutrition, high disease burden, and unmet medical/surgical needs in low- and middle-income countries (LMICs) likely place children born with orofacial clefts at high risk of nutritional deficiency and death. Through partnerships with hospitals across >85 LMICs, the charitable organization Smile Train supports local medical training and free surgery and comprehensive care to alleviate the burden of clefts. However, there is a gap in knowledge regarding the global prevalence of malnutrition in children born with cleft in these settings. To date, only a few small-scale studies have examined the nutritional status of children undergoing cleft repair in LMICs.

Aims: The study was designed to gain a global insight on the epidemiology and nutritional situation of children with orofacial clefts across developing countries in order to fill a gap in knowledge and inform current and future comprehensive cleft care programmes in low-resource settings.

Methods: We conducted a cross-sectional analysis of a dataset of 638,988 children (≤5 years) who underwent surgery between 2008 and 2018 in Smile Train-sponsored local hospitals across the developing world. Patients’ anonymised records included birth date, gender, weight at surgery, ethnicity, country of origin, and date of primary surgery and were analysed with Stata. Underweight prevalence was derived from the generation of weight-for-age z scores using WHO Stata macro and described in relation to cleft epidemiology. For comparisons conducted at country level, the most recent data on underweight prevalence in children aged ≤5 years was extracted from the online Demographic Health Survey program.

Results: The relative proportions of the 3 main cleft types – cleft lip only (CLO), cleft palate only (CPO), and cleft lip and palate (CLP)– varied by gender, ethnic groups, and world regions. Children with CPO had a higher median age at surgery (CPO: 20 months vs. CL/CLP: 8-11 months), and appeared under-represented in the African region (8% vs. 14.7-29.2% of all cases elsewhere). Almost one-third (28.6%) of the children with clefts were underweight at the time of primary surgery. Across all settings except Africa, the prevalence of underweight was lower in children with CPO compared to those with CL/CLP (18.4% vs. 25.5-32.8%). Finally, an analysis across countries showed that the higher the national underweight prevalence estimate in countries, the higher the underweight prevalence among children with cleft ($r_s$ =0.61, $p <$0.001).

Summary/Conclusion: The high prevalence of underweight in our dataset reflects the burden of orofacial clefts in LMICs. Data needs to be further examined to understand why CPO presentation is low in African countries and why the prevalence of underweight is lower among CPO vs. CLO/CLP cases in all settings. The data will inform Smile Train’s ongoing efforts to scale cleft nutrition programs as well as inform the design of interventions to alleviate this burden.
Application to telemedicine by using 3-layer facial models, a mixed reality device “hololens” and skype

M Katayama¹, K Ueda¹, D Mitsuno¹
¹Plastic and Reconstructive Surgery, Osaka Medical College, Osaka, Japan

Background: In recent years, uneven distribution of doctors has become a problem. Although it has been recommended to support for telemedicine with telementoring, there are many problems to be solved such as devices and communication lines. In surgical telementoring, it may be very complicated to transmit sufficient intent only by exchanging voice and video each. Especially in plastic surgery / craniofacial surgery, it is even more difficult because detailed instructions on skin incision design, osteotomy design, bone loss direction and more are required. We had already developed a 3-layer facial model which is covered with skin and subcutaneous tissue similar to the human body on a hard (bone) tissue model. Then, we had confirmed that it will be possible to make a more accurate surgical plan, training doctors, and an explanation to patients by projecting 3D angiograms, facial bones, and target facial contour on the model with a mixed reality device “HoloLens”. So, we considered the application to telemedicine by using the Model, HoloLens and Skype.

Aims: By using the 3-Layer Facial Models, HoloLens and Skype, we aim to find some solutions to these problems accompanying telemedicine using a cheaper and easier way and to demonstrate the usefulness of telemedicine in the field of craniofacial surgery.

Methods: We held a demonstration between Kochi city, Kochi (hospital A) and Takatsuki city, Osaka (hospital B), between that straight-line distance is 250 km. In advance, a mentor surgeon in hospital B got ready a 3-layer facial model of a patient made of polyurethane for the skin surface, made of silicon for the subcutaneous soft tissue, and made of salt for the bone. And a resident in hospital A was given the patient’s information/examination data to study. For a method of real-time video interaction, we adopted Skype (Microsoft Corporation, Redmond, WA) which is popular and freely available for Internet video applications. For a transmission method of nonverbal information, a mixed reality device HoloLens (Microsoft Corporation) was adopted. Given remote guidance, communicating by voice, video of the surgical field, and reference data by the mentor surgeon, the resident wearing HoloLens with a built-in Skype designed incision lines and applied model surgery on the 3D entity model like a simulated patient.

Results: By preparing the 3D entity model preoperatively, connecting each HoloLens of two medical institutions located remotely via Skype and sharing various image information on the model, it became possible to perform more accurate surgical instruction and simulated surgery. There was no delay in voice communication and a delay of <0.5 seconds in the video in telemedicine demonstration experiments. With HoloLens, the resident was able to proceed with surgical operations without transferring the line of sight from the operative field to another. The mentor was able to send appropriate instructions by voice, point out a specific part by telestration function, and draw lines on the 2-dimensional images pasted on the resident operator’s field of vision. And the resident could grasp the situation without the need to outline the specific items by voice.

Summary/Conclusion: By using the 3-layer models, HoloLens and Skype based on patients’ data, it was possible to demonstrate telementoring in the field of craniofacial surgery for future telemedicine support. It is necessary to accumulate improvements and know-how in the future to realize a “realistically feasible and useful” telementoring system.
Craniofacial growth pattern and malocclusion influence upper airway dimensions in treacher collins syndrome.

I Trindade-Suedam, A Ribeiro, F Smith, C Tonello, I Trindade

1Laboratory of Physiology, Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo, 2Department of Biological Sciences, Discipline of Physiology, Bauru School of Dentistry, University of São Paulo, 3Laboratory of Physiology, Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru-SP, Brazil., 4Postdoctoral Research Fellow , University of Colorado Denver Anschutz Medical Campus, School of Dental Medicine, Denver, United States, 5Craniofacial Surgery, Hospital for Rehabilitation of Craniofacial Anomalies, University of São Paulo, Bauru-SP, Brazil

Background: Treacher Collins Syndrome (TCS), a rare craniofacial deformity, is mainly characterized by hypoplasia of the mandible and the zygoma, microtia and, in some cases, a cleft palate. It is speculated that the skeletal pattern, determined by the underdevelopment of the craniofacial structures, may cause upper airway reductions leading to an increased risk for breathing disorders such as chronic upper respiratory tract infections and obstructive sleep apnea.

Aims: This study aimed at addressing the influence of skeletal pattern and malocclusion on the upper airway dimensions of subjects with TCS by means of 3D tomographic assessment.

Methods: Cone beam computed tomography (CBCT) scans of 37 individuals, previously obtained for orthodontic and/or surgical planning, were divided into 2 groups: 1) TCS group: CBCT scans of 19 patients with TCS (10 males and 9 females; mean age: 17 ± 5y), and, 2) CON group: 18 CBCT scans of individuals without any syndromes but with skeletal pattern and malocclusion of the TCS group, i.e., Angle Class II malocclusion and vertical growth pattern (11 males and 7 females; mean age: 32 ± 4y). Pharyngeal volumes (V) and minimal cross-sectional areas (mCSA) were assessed using Dolphin Imaging software. Cephalometric data of growth pattern (FMA), maxillomandibular relation (SNA, SNB, ANB and Pg-NB) and mandibular dimensions (Co-Gn and Co-Go) were assessed on lateral cephalograms obtained from CBCT scans. Statistical analysis included independent sample Student t test for inter-group evaluation. Correlations between the variables were tested with Pearson Correlation Coefficient. Values of p ≤ 0.05 were considered statistically significant.

Results: Mean pharyngeal V of TCS (16.2±7.1 cm³) were significantly reduced when compared to CON (20.0±4.3 cm³). Although reduced, mean values of mCSA for TCS (88.2±52.3 mm²) were not statistically different from CON (99.0±39.2 mm²). On both groups, mCSAs were mostly located at the oropharyngeal level. TCS presented a significantly greater vertical growth pattern (44.3±8.9°) when compared with controls (37.3±7.7°). TCS presented with a more retruded mandible in relation to CON, significantly observed on Pg-NB angle (-5.4±7.6° and 1.2±2.5°, respectively). Finally, mandibular dimensions of TCS were statistically smaller when compared with CON (Co-Gn= 87.3±14.0mm and 108.6±4.4mm / Co-Go= 39.0±10.8mm and 50.4±6.0mm, respectively). A positive correlation was observed between mandibular dimensions and pharyngeal volumes on TCS.

Summary/Conclusion: Pharyngeal volume of individuals with TCS is reduced even when compared with Angle class II and excess face height malocclusion subjects. Growth pattern and the underdeveloped mandible seem to influence the upper airway dimensions on this population.
Costochondral graft in growing patients with hemifacial microsomia: long term results compared with non treated patients

V Battista¹, M Meazzini², F Mazzoleni³, R Brusati², F Biglioli¹, L Autelitano¹
¹Maxillofacial Surgery - SmileHouse, ²Ospedale San Paolo, Università degli Studi di Milano, Milan, ³Maxillofacial Surgery, Ospedale San Gerardo, Monza, Italy

Background: Type III Hemi Facial Microsomia (HFM) is traditionally treated during childhood with costochondral graft (CCG) to establish normal facial proportion in terms of mandibular height and facial contour. These patients usually undergo orthognatic surgery at the end of the growth.

Aims: The aim of this study was to evaluate the long term results of costochondral graft in a sample of growing patients affected by HFM type III and to compare results with a sample of non treated patients affected by the same malformation in order to understand if surgery during growth provides advantages in terms of bony and facial symmetry after an 8 year follow up.

Methods: In the maxillofacial department of the University Hospital of Milan, Smile House, between 1995 and 2006, 33 procedures of ramus condyle reconstruction by CCG were performed. To obtain a homogeneous sample inclusion criteria were: patients affected by HFM type III, costochondral graft for mandibular reconstruction performed at 5/8 years, no associated macrostomia, no associated facial palsy, 10 patients were included (CCG patients group).

A control Group (CG) was retrospectively collected. 10 patients were never offered or did not accept treatment during growth. Selection criteria were: HFM type III with records in the early growing period (6-8 y.o.), never subjected to surgical treatment, records at 15-17 years of age, prior to any type of procedure, no associated macrostomia, no associated facial palsy. Mean follow up was 8 years.

The growth of the CCG (assessed as the ratio between the reconstructed ramus and the healthy ramus) and the occlusal canting was assessed on panoramic X rays. To assess facial symmetry a photometric evaluation on the frontal view was carried out.

Results: CCG patients group: The mandibular ratio decreased from 0.95 to 0.87 after an average follow up of 8 years. A good facial symmetry was achieved after surgery, but was lost in the majority of the cases at the most recent control. CG: occlusal canting slightly increased (from 0.8° to 1.4°) and facial asymmetry was relatively stable during the years.

Summary/Conclusion: In patients with a congenital deformity, restoring the height of the ramus leads to an immediate restitution of facial symmetry, but in the long term there is a return to the asymmetrical ratio in particular for soft tissue defects on the affected side. HFM III patients not treated shows a stable asymmetry during the years with no increase of the facial deformity.
The influence nam has for the surgeon
Philip Kuo-Ting Chen

Various techniques has been using since the 80’s to improve the presurgical cleft condition and help surgeon to achieve better results. A combination of presurgical orthopedics (PSO), lip taping and prone sleep position can effectively reduce the cleft width before surgery. However, several prospective studies failed to show any benefits in feeding, surgical outcome and facial growth. Nasoalveolar molding (NAM) has been introduced since the 80’s. Besides reducing cleft width, the device can improve the nasal shape before surgery. The author has been using NAM since the mid-90’s. With some surgical modification and postoperative maintenance, patient with NAM can achieve a better nasal result comparing to other groups of patients. Although there is no good prospective randomized control study to show the benefit of NAM, that is difficult in clinical practice, the author will still recommend using NAM for better surgical outcome.
Controlled photometric evaluation of the nasal morphology in unilateral and bilateral cleft lip and palate patients after nasoalveolar molding

B Campo¹, M Bendandi², E Fabbri³, A Zarabini⁴, A Baietti⁵, P Morselli³

¹Orthodontics, University of Bologna, ²Orthodontics, IRCCS Institute of Neurological Sciences of Bologna, ³Plastic Surgery, University of Bologna, St. Orsola-Malpighi Polyclinic, ⁴Plastic Surgery, IRCCS Institute of Neurological Sciences of Bologna, ⁵Head of Maxillo Facial and Plastic Surgery Department, IRCCS Neurological Sciences of Bologna, Bologna, Italy

Background: The importance of the use of orthopedic appliances in cleft lip and palate presurgical treatment was first reported by Dr. Pierre Franco in 1561. Later, some other techniques were developed with the aim of retracting the projected premaxilla and aligning the lip segments. The great effectiveness of the Nasoalveolar Molding therapy is to combine the shaping of the nasal cartilage with the manipulation of the alveolar segments and it is widely required by plastic surgeons as a presurgical preparation of bilateral and unilateral cleft lip and palate patients to the primary repair.

Aims: The objective of this study was to evaluate the NAM treatment outcomes on nasal correction and to compare them with an age-matched control sample. Photogrammetry was used to calculate the nasal morphology after the NAM treatment. The comparison between control samples and NAM samples was made on presurgical photographs with the aim of analyzing the efficacy of the NAM without the interference of the surgical technique and the competence of the surgeon.

Methods: In this blinded retrospective study we compared 32 unilateral cleft lip and palate patients that underwent Nasolaveolar Molding with 34 unilateral cleft lip and palate patients that did not receive any presurgical treatment. For bilateral cleft evaluation 15 NAM patients were matched with 18 control patients. The nasal morphology was analyzed from pre-treatment and post-treatment frontal, lateral and basal photographs (i.e. alar base width, alar width, nasal tip protrusion, columella length and width, columella deviation, premaxilla deviation, nostril floor width and height, nasal tip angle and nasolabial angle).

Results: NAM therapy improves the columella length, width and simmetry, the nostril shape and the nasal tip protrusion in both unilateral and bilateral cleft lip and palate. Less efficacy was observed in alar base and alar width reduction for bilateral clefts. The nasolabial angle proved wider in BCLP babies after NAM. In the unilateral control group no statistically significant changes occurred, while in the bilateral control group we observed a worsening of the major nostril height ratio and a reduction of the nasal tip protrusion ratio with an increased nasal tip angle.

Summary/Conclusion: The results of this study determine, at least in our experience, the effectiveness of the Nasoalveolar Molding presurgical preparation, due to the great correction of the nasal dysmophia. The importance of the age-matched control samples was to evaluate the results of the dysmorphic growth without the interference of an orthopedic appliance. Analyzing our data, for the unilateral control sample no significant improvements were observable. While for the bilateral control group the growth alone has led to an evident worsening of the nasal parameters. In this direction, we may conclude that, at least from our evaluation, the NAM treatment improves nasal morphology, facilitates the primary surgical correction and in bilateral cleft lip and palate patients avoids the worsening pattern of dysmorphic growth.
Restructured nasoalveolar molding appliances for cleft lip/palate patients
R Haddad¹, M Saadeh², N Abou Chebel¹
¹Orthodontics, American University of Beirut, ²Orthodontics, Lebanese University, Beirut, Lebanon

Background: To improve esthetic and long-term results, nasal alveolar molding (NAM) reduces the severity of the cleft lip/palate (CLP) presurgically by reshaping the alveolar, lip and nasal segments

Aims: 1- Describe two new modifications of the original NAM appliance for easier manipulation and less irritation, 2- Quantify the nasolabial soft tissue changes after NAM and lip closure surgery, 3- Compare the nasolabial soft tissue changes between CLP patients who received NAM and those who did not before lip closure surgery

Methods: Two groups of 13 CLP patients each were included: Group 1 (mean age: 1.1mos±0.2) consisted of 8 bilateral (B) and 5 unilateral (U) CLP patients treated with modified NAM appliances prior to surgical cleft closure; Group 2 (mean age: 6mos±0.2) included 6 B and 7 U CLP patients treated surgically without prior usage of the NAM appliances. Soft tissue nasolabial segments were measured on pre-NAM (T1), post-NAM (T2) and post-surgery (T3) photographs and measurements were compared

Results: In the NAM group, cleft size was reduced by 66% in 4 to 5 months and a statistically significant (SS) difference in all measurements between between T1 and T2 was found, except for alar length. This accompanied significant improvement in nasal symmetry, including decrease of columellar crest inclination (by 60%), increase in columellar and nostril heights (by 138% and 127% respectively), and decrease of nostril and bialar widths (by 30% and 16% respectively). Both modified appliances achieved elongation of the philtrum of the upper lip by 49.5%. When comparing NAM to non-NAM users at T3, SS differences in the bialar width, columellar height, nostril height and the nasal-alar slope angle were noted

Summary/Conclusion: NAM therapy should be started as early as possible, ideally within the 1st month of life. The modified NAM provided easier activation of the appliance and placement by the parents and significantly reduced or eliminated skin irritation with more promising results
Nursing

OS 35.1
Can the use of implementation intentions improve oral health in children with a cleft?
J Mooney1, P Callery2, C Armitage3, T Walsh4
1Regional Cleft Unit, Royal Manchester Childrens Hospital, 2Division of Nursing, Midwifery and Social Work, 3Division of Psychology and Mental Health, 4Division of Dentistry, University of Manchester, Manchester, United Kingdom

Background: Poor oral health may compromise the long term results that are achievable on behalf of the patient with a cleft. The burden of care both to the child and family is immense in their overall cleft management. Many studies that have examined the oral health status of children with a cleft call for preventive dental health programmes to be introduced and directed towards parents and their children. The use of implementation intention-based interventions has the potential to promote both child and adult behaviour change.

Aims: To evaluate the acceptability and feasibility of the use of implementation intentions designed to change both child and parent oral health behaviour, and generate effect sizes to aid planning for a definitive randomised controlled trial.

Methods: Parents of children aged 5-9 years with a cleft, attending one of three Cleft Units were approached and visited at home by trained Dental Care Professionals (DCP). Informed, written consent was gained from parents and children. The DCP collected baseline data, including Modified Gingival Index (MGI) and Plaque Control Record (PCR). Children were randomised to one of three groups: (A) control group, (B) intervention group in which children and parents were asked to form implementation intentions, or (C) intervention plus booster group where an additional reminder about their implementation intention was sent after 3 months. Following delivery of the intervention, questionnaires and clinical assessments were repeated at 6-month follow-up alongside exit interviews to ascertain acceptability and feasibility. Statistical analysis generated summary statistics and 95% confidence intervals (95%CI).

Results: 58 consented children from 3 Cleft Units completed baseline assessments, were randomised, and received their intervention. 44 (76%) completed exit interviews and of these 42 (72%) provided follow up clinical data. All participants enjoyed involvement in the trial, and those allocated to the two intervention groups additionally believed that forming implementation intentions was effective. In Group C, 86% (95% CI 59, 98) reported the use of implementation intentions in the last month compared to 75% (95% CI 46, 90) in Groups A and B. Descriptive statistics generally showed oral health improvements across all groups although the effects were more marked in the intervention plus booster group. The mean PCR was comparable at the first visit: A: 53.7% (sd 32.4); B: 55.0% (sd 27.8); C: 53.4% (sd 26.1). There was more reduction in plaque measured by the PCR at the 2nd visit, reducing to 49.9% (sd 36.1) in Group A, 39.3% (sd 28.9) in Group B and 29.1% (sd 26.7) in Group C.

Summary/Conclusion: Interventions that seek to change children’s behaviour typically target adults or children, but rarely both. The data collection procedures were acceptable and the intervention feasible to both children and parents. The effect sizes suggest that the intervention plus booster condition has sufficient promise to proceed to a fully-powered randomised controlled trial.
Background: Due to the impact that the first year of life represents in the patient with cleft lip and palate, interdisciplinary management must comply with the criteria and objectives, based on Clinical Practice Guidelines.

Aims: To present to the international community the Clinical Practice Guidelines developed by Mexican experts, in patients with cleft lip and palate during their first year of life.

Methods: Cochrane methodology was used. Relevancy to answer questions was determined by a panel of experts. A comprehensive literature search was conducted in the following databases: MEDLINE (1966-2014), EMBASE (1980-2014), the Cochrane Library (2000-2014), the BVS (1980-2014), the EBSCO Library (1995-2014), the ARTEMIS database (1995-2014), the SCIELO database (1995-2014), in 42 technology assessment agencies, the website of the FDA (Food and Drug Administration, http://www.fda.gov), and the international database for clinical trials (http://www.controlled-trials.com). The abstracts found were read and duplicates were eliminated as well as those who had no relation to each question. All relevant articles were obtained in full text and rigorously reviewed, obtaining the degree of bias, outcomes assessed and a quantitative assessment (meta analysis) was performed. Clinical recommendations were issued to the qualitative and quantitative analysis of scientific evidence through a consensus of 90 Mexican experts of different specialties related to the treatment of cleft lip and palate.

Results: Seven relevant questions in this guide are included: 1) Is the rule of 10 valid for security parameters during the first surgery of patients with cleft lip and palate?, 2) What is the effectiveness of preoperative orthopedics?, 3) At what age should we perform the cheiloplasty?, 4) At what age should we perform the palatoplasty? 5) Should early peri-osteoplasty be performed?, 6) How effective the primary nasal repair is?, 7) Should septoplasty be performed during the primary cheiloplasty?. The clinical recommendations based on Evidence Based Medicine of more than 1,200 articles analyzed will be presented.

Summary/Conclusion: Making decisions based on scientific evidence will allow us to have a more efficient clinical practice. This guide shows Mexico’s efforts to unify criteria and work together in association with all related specialties in the care of cleft lip and palate patients. Participants continue to work in this guide to address the following years of interdisciplinary care.
Drinking and tongue function in children with cleft palate - first ultrasonographic findings
J. Kuttenberger

Background: Newborns with cleft palate are not able to build up a vacuum. Feeding problems are common and breastfeeding is rarely possible. Gain of weight is not sufficient and as a typical sign milk comes out through the nose. Early adaptation to the breast, stimulation of milk production and facilitated feeding are important aims which have to be achieved by a coordinated team approach. The tongue plays an important role in the process of drinking and a thorough knowledge of tongue function is crucial for a deepened understanding of the normal sequence of drinking and swallowing.

Aims: Ultrasonography offers a non-invasive method to study physiological tongue movements during drinking and swallowing. In a pilot study the feasibility of ultrasonography should be tested to examine tongue action in cleft palate patients.

Methods: In four children with cleft palate tongue movements were studied with ultrasonography (Philips iU22, 8,5 MHz sector probe placed in the midsagittal line) during bottle-feeding before and after palatal closure. Tongue action was recorded and analysed qualitatively.

Results: In the ultrasonographic examinations a similar mode of tongue action was found pre- and postoperatively: The teat was compressed by the anterior part of the tongue against the palate or the inserted drinking plate and milk was flowing into the mouth. A characteristic peristaltic movement of the tongue was found, by which milk was transported dorsally. The oral phase of swallowing was completed by elevation of the root of the tongue and the larynx. This pattern of tongue action is demonstrated by typical examples and compared with the literature.

Summary/Conclusion: Ultrasonography provides a feasible non-invasive method to study tongue function during drinking in cleft palate patients. Typical and similar patterns of tongue movements were found pre- and postoperatively. These findings seem to confirm that insertion of a palatal plate immediately after birth and early closure of hard and soft palate, which was performed at 6 months of age, are able to facilitate the development of a physiological tongue function.
OS 35.4
‘A prospective, longitudinal control study of the prevalence of feeding difficulties, birth weight and growth in babies with cleft lip and/or palate’
C Guillaume¹, I de Vries¹, M Verhoeven², S Haverkamp³, A Mink van der Molen¹, M Kon¹, C Breugem¹
¹Department of Plastic Surgery, Wilhelmina Children’s Hospital, University Medical Center Utrecht, Utrecht, ²Department of Pediatrics, Academic Medical Center, Amsterdam, ³Department of Speech Therapy, Wilhelmina Children’s Hospital, University Medical Center Utrecht, Utrecht, Netherlands

Background: To describe a literature-based definition for feeding difficulties (FD) in infants with cleft lip and/or palate (CL/P); to assess birth weight and growth expressed as weight-for-age (WFA) and height-for-age (HFA) in infants with CL/P aged 0-6 months compared to healthy infants; and to investigate the prevalence of FD and the relation to growth in infants with CL/P aged 0-6 months.

Aims: Birth weight, growth and FD (duration of feeding, choking events, parents’ experience, NG-feeding and overall FD).

Methods: We performed a structured review according to the PRISMA guidelines. A definition of FD was developed by means of an expert panel. We performed an one-arm prospective trial including 30 patients with CL/P directed to the cleft team in the Wilhelmina Children’s Hospital, University Medical Center Utrecht, The Netherlands, at the age of 0-4 weeks. Weight, height were measured and a questionnaire was conducted at the ages of 1, 3 and 6 months.

Results: A percentage of 14.3% of infants were small for gestational age. Birth weight was lower in premature infants (p=.012). Respectively, 27.6%, 13.8% and 10.3% of infants had WFA<-2 at 1, 3 and 6 months, while this was 10.3%, 6.9% and 10.3% for infants with HFA<-2. Weight was significantly influenced by the extent of the cleft (p=.048). Overall prevalence of FD was 96.6%, 65.5% and 79.3% at 1, 3 and 6 months. Neither presence of overall FD, nor the individual components of FD significantly influenced weight or height.

Summary/Conclusion: More children with clefts are SGA and/or experience impaired growth compared to their healthy peers. Severity of cleft palate was found to be a risk factor for weight impairment. Feeding difficulties amongst children with clefts are common, but they do not seem to influence growth. No risk factors for FD could be identified.
Background: Cleft lip and palate is one of the most common congenital craniofacial deformity of head and neck region. Feeding problems in infants with deriving from the anatomic defect. Most infants require some mechanical assistance to facilitate milk flow by means of enlarged, cross-cut, or wide based nipples or squeezable bottles.

Aims: In this study, it was aimed to determine the difficulties experienced by pediatric nurses and parents during feeding the babies with cleft palate and to determine solution offers.

Methods: One hundred twenty parents and fourteen nurses who accepted to participate in the study constituted the sample of the study. Data were collected by using nurse interview form and data collection form.

Results: It was determined that the majority of the parents (83.3%) had problems during feeding. The most common problems in feeding were the difficulty in sucking breast (63.2%), milk escaping into the nasal cavity (61.7), excessive air ingesting (50.0%), and feeding longer than normal time (50.0%). In our study, 54.3% of the parents stated that they were not satisfied with the feeding bottles they used for feeding their babies. The most common recommendations of parents to develop feeding bottle was to reduce the price (29.2%), the cover should be added (16.7%) and should be accessible (15.0%). The majority of the nurses (83.3%) stated that infants had problems during feeding. Among the most common solutions suggested by nurses to solve these problems were short-term and frequent feeding (57.1%), intravenous fluid replacement if oral intake is insufficient (35.7%) and family education in feeding frequency, position, and complications (28.4%). The study also revealed that 35.7% (n=5) of nurses were not satisfied with the equipment used in feeding babies.

Summary/Conclusion: In the present study, it was found that most of the nurses and parents experienced difficulties in feeding the babies with cleft lip and palate. In addition, in order to overcome these difficulties, solutions were proposed in the triangle of infants, parents, and nurses.
International collaboration between nurses through an educational module improves quality of cleft care

C Jung¹, A Paganini², B Grollemund³, M Coste⁴

¹Pediatric Surgery, University Hospital Center, Lausanne, Switzerland, ²Pediatric Surgery, Sahlgrensk University Hospital, Gothenburg, Sweden, ³Orthodontics, Pôle de médecine et chirurgies boccodentaires” Hôpitaux Universitaires de Strasbourg, Strasbourg, ⁴Maxillofacial surgery and plastic surgery, Hospital Necker-Enfants Malades, Paris, France

Background: The specialist nurse is an integral part of the multidisciplinary cleft team, and its role have evolved over the years. Seales (2018), in his qualitative study suggests that they are an important and trusted point of contact between parents and team members.

This role requires specific knowledge about facial cleft management, nutrition, pre- and post-operative care, and family-centered care. The cleft nurse are of utmost importance to patients and families from the antenatal period, through birth and infancy and extends into childhood in concurrence with the surgical procedures. Until 2019, there was no specific training in French-speaking Europe. In addition, there is little evidence-based literature available on nursing of patients with facial cleft and their families.

Aims: The aim was to describe how the nursing role has been incorporated into the Faculty of Medicine’s University Degree (DU) "Multidisciplinary Management of Facial Clefts".

Methods: In 2018, the Faculty of Medicine of Strasbourg decided to set up a continuous training course on multidisciplinary management of facial clefts. This training is given for the first time 2019. The training course consists of modules, where each specialty represented in the multidisciplinary team are responsible for the one module each. The nurse-led module covers the aspects of nursing care and support from the antenatal period to the end of growth. Specialist nurses are responsible for sharing knowledge about prenatal counseling, nutrition, pre- and post-operative care and support. They also have to teach how to play a supportive role for families throughout the child’s development and how to collaborate with the various professionals of the team.

The innovative idea of this module is to bring together nurses from three different teams in Europe (France, Sweden and Switzerland) to teach by e-learning. The work was made through email, phone and physical meetings stretching over the fall of 2018.

Results: The collaboration between three specialist nurses from different parts of Europe brings updated and complementary approaches. The exchanges between knowledge allow improvements in the quality of holistic care of the child and his family. Differences in practice were observed and questioned. In order for families to receive from the best quality of care and a harmonized care pathway, the responsible nurses shared knowledge and experience as well as current research.

Summary/Conclusion: This collaboration is the first step towards evidence-based guidelines for facial cleft nursing in Europe, as well as providing a solid teaching material in French. The desire to continue our partnership will lead us to conduct multicenter comparative studies to identify the most efficient and satisfactory care for children and their families. The most challenging practice differences are the post-operative care of the first cleft closure, where great differences were discovered between centers as well as a lack of evidence based research. The urgent need of future studies in the field of nursing of children with cleft and their families was identified and the collaboration will continue to ensure quality of care for all children.
Speech analysis

**OS 36.1**

*Towards Unification on Measuring Speech Outcomes*

Debbie Sell, Centre for Outcomes and Experience Research in Children’s Health, Illness and Disability (ORCHID), Great Ormond Street Hospital NHS Foundation Trust, London, United Kingdom,

Speech outcomes are hugely important in cleft care. The last three decades has witnessed much effort by speech and language therapists to agree best practice in measuring perceptual speech outcomes, especially with regard to outcomes related to surgical interventions. This presentation aims to examine the current state of play focusing on impairment but reflecting briefly on activity, participation and patient reported outcome measures. We will see that although much progress has been made, there remain important differences in approach, still making comparison between studies very challenging. In order to take the field forward, the differences between tools and methodologies will be summarised, and areas requiring more consensus identified. This talk will also report on the more neglected area of measuring speech following therapy intervention, based on novel findings from a recently conducted RCT. The potential serious ramifications of low quality published studies where appropriate speech methodology is not used will be illustrated.

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*A multicenter comparison of speech outcomes: the americleft speech project*

K Chapman¹, A Baylis², K Cordero³, A Dixon⁴, C Dobbelsteyn⁵, J Trost-Cardamone⁶, K Wilson⁷

¹Communication Sciences and Disorder, University of Utah, Salt Lake City, ²Nationwide Children’s Hospital, Columbus, ³Barrow Cleft and Craniofacial Center, Phoenix, ⁴Riley Hospital for Children, Indianapolis, United States, ⁵Dalhousie, Halifax, Canada, ⁶University of California Northridge, Northridge, ⁷Texas Children's Hospital, Houston, United States

**Background:** Multicenter outcome studies have been proposed as a way to judge the overall outcome of treatment protocols. Yet, until recently, multicenter studies were rarely carried out in the United States. However, these studies are needed to provide “insights into the processes and outcomes of treatment of comparable services elsewhere, the establishment of future goals, and the exchange of clearly successful practices” (WHO, 2004).
**Aims:** The primary purpose of the study was to compare speech outcomes of 5- and 6-year-old children with cleft palate (CP) at three different centers in the United States. Additionally, the study was designed to compare the speech of children with cleft palate to noncleft peers of the same age.

**Methods:** One hundred seventy-eight children with repaired cleft palate and 57 noncleft peers were recruited at three centers. Children with diagnosed syndromes, cognitive impairment, developmental delay, sensorineural hearing loss, palate repair after 18 months, or adopted after 12 months of age were excluded from the study. A standard sample of speech including conversation, counting, sentence repetition, and elicited single words was video-recorded. The CAPS-A-AM was used to rate the samples. The ratings were completed by four speech-language pathologists not associated with the three centers.

To compare across the three sites, a multivariable linear regression or logistic regression model was fit separately for each of the study outcomes, depending on level of measurement of the outcome. Age varied across center (but gender and cleft type did not), so it was entered as a covariate. No statistically significant differences were found across the three centers. T-tests and Mann-Whitney U tests were performed to compare cleft and non-cleft children.

**Results:** Although there were no differences for any variables measured across the three centers, the children with cleft palate produced significantly more errors than the noncleft children. Differences were also noted for error types. Acceptable speech was noted for 11% to 17% of children, normal resonance was noted for 45% to 59% of children, and normal speech was noted for 23% to 37% of children at age five.

**Summary/Conclusion:** Similar speech outcomes can be achieved with different approaches to primary palate repair. A majority of children with cleft palate do not have normal speech when they start school at age 5-6 years, and more children are in need of speech intervention than are enrolled in intervention. The findings are similar to those reported in the United Kingdom in terms of speech outcomes at 5 years of age.
Speech outcomes in patients with unilateral cleft lip and palate submitted to two different palatoplasty protocols
M Vasconcelos1, D do Vale1, E Melo1, N Siqueira1, N Alonso3, R Pereira1
1CADEFI, IMIP, Recife-PE, 2CADEFI, IMIP, Recife- PE, 3Craniofacial Surgery Department, FMUSP, São Paulo, Brazil

Background: The goal of cleft palate surgical repair is to accomplish a good speech with preservation of the maxillary growth. Speech and language disorders are an important issue of the treatment failures that cleft palate can leave. During speech, Velopharyngeal Deficiency (VPD) may occur, causing alterations in speech, in production of oral sounds. Some authors relate in patients with Late Hard Palate Closure (LHPC) an increased rate of VPD compared to patients with Early Palate Repair.

Aims: Evaluate the speech outcomes in children with unilateral cleft lip and palate (UCLP) who were operated by a single surgeon in a single-center using two different treatment protocols, one 2 staged repair with Late Hard Palate Closure.

Methods: A randomized, controlled, blinded trial was carried out at the Center for Attention to Defects of the Face of IMIP (CADEFI) of the Instituto de Medicina Integral Prof. Fernando Figueira (IMIP) between January 2010 and October 2016 and was also performed to evaluate the speech characteristics in two groups of subjects with UCLP submitted to different surgical protocols through of auditory-perceptual assessment by three experienced speech-language pathologists (SLP) at a different Institution. The intervention group (IG), had two stages cleft repair with LHPC, between 3 and 4 years of age (32 patients) and the control group (CG) with early single-stage palatoplasty between 9 to 15 months (28 patients). Speech samples were collected and recorded in a standardized manner and consisted of phrases in single words, connected speech and syllable chains with oral sounds and consonants with high pressure with plosives and fricatives phonemes following Brazilian cleft recommendations. For the auditory-perceptual assessment was considered: compensatory articulation disorders (CAD) and hypernasality (present and absent). The unweighted kappa was calculated for the compensatory articulation disorder an hypernasality.

Results: Compensatory articulations were absent in 18(56,25%) patients for the IG and in 19(67,86%) patients in a CG. Intrarater reliability was calculated for the three SLP raters. The findings indicated excellent agreement between the SLP in two groups IG, $k=0.708$ (p<0.001) (DP±0.908-0.508) and GC, $k=0.895$ (p<0.001) (DP±1-0.681). Resonance adequated were verified in both groups: GI=32(46,88%(15), mean=1.5(SD±0.5070), GC=28(60,71%(17), mean=1.6 and SD±0.4973. Kappa statistics revealed moderated agreement, $k=0.420$ (p<0.001, SD±0.634-0.207) for the CG and low agreement $k=0.287$ (p=0.005, SD±0.487-0.087) for the IG among the experienced judges.

Summary/Conclusion: The findings of this study suggested that the compensatory articulation rate was similar in both groups. The agreement level for the three SLP raters showed better results for the Control Group revealed lower frequency of hypernasality. The experience and the calibration of the SLP is very important for the auditory-perceptual assessment in cleft lip and palate population.
The south thames outcomes for cleft - speech and language therapy (stoc-slt)

L. Koutsoulieri¹

¹South Thames Cleft Service, Guy's and St Thomas's NHS Trust, London, United Kingdom

**Background:** It is becoming increasingly important for Speech and Language Therapists to be able to demonstrate the value of our intervention by measuring our outcomes. Stemming from the current financial challenges in the health sector, there is a drive for outcomes-based commissioning of services and therefore it is a vital requirement for clinicians to demonstrate the impact of their practice. There are historical difficulties to measuring outcomes in Speech and Language Therapy (SLT), such as lack of evidence to identify what is best treatment and lack of universally applied means to demonstrate the impact of treatment in every day functioning. In the UK, the Royal College of Speech and Language Therapists has suggested using the Theory of Change (ToC) Model as a framework to illustrate how and why the SLT clinicians’ intervention works and the impact it has on users’ real life functioning.

**Aims:** The aim of this paper is to demonstrate how the ToC Model has been used by the South Thames Cleft Service SLT department to show how the Activities we offer (our interventions), are linked to Interim Outcomes (what the users have been enabled to do), and to Ultimate Outcomes (Policy level outcomes).

**Methods:** The ToC Model was applied by detailing all the tasks undertaken by the SLT department and what these tasks enabled all relevant Stakeholders to achieve. These achievements were then linked to National level indicators of intervention success. We had to provide evidence to prove how our Activities influenced the Interim Outcomes, by using measurable indicators of change. Such evidence has mainly been South Thames Cleft Service SLT data (Service statistics, Questionnaires, Audits), but some relate to the general cleft literature. An important aspect of this work has been to identify gaps in the evidence base linking Activities to Interim Outcomes, while finding ways to produce new evidence.

**Results:** The ToC Model can be used as a customised framework to provide Service specific outcomes and demonstrate the unique value of a SLT Service’s interventions. Applying this Model has also enabled our Service to identify gaps in the evidence base, which has led to the creation of new evidence linking interventions to outcomes. The Model can be flexibly used, depending on which Stakeholder requires evidence of SLT intervention. Importantly, it can also demonstrate how a reduction or an increase in SLT resources can impact on the Service’s outcomes therefore also making it specific to each specialist Service’s needs. The Model has also helped our SLT Service develop team objectives and individual objectives.

**Summary/Conclusion:** By applying the ToC Model as a customised framework, our SLT department has been able to provide evidence for its intervention outcomes for a number of different Stakeholders. The Model also facilitated the production of new outcomes evidence, showing the value of the care provided by this SLT Service. As things change in Service provision and Policy level, this framework can be adapted to reflect these changes. This Model can be used by various Services in the health sector.
Profiling speech/language outcomes of children with cleft palate at 39 months of age
L Beckett¹, K Chapman², M Hardin-Jones³
¹Speech Language Pathology, Riley Hospital for Children, Indianapolis, ²Communication Sciences and Disorder, University of Utah, Salt Lake City, ³Communication Sciences and Disorder, University of Wyoming, Laramie, United States

Background: Results of group studies suggest that children with cleft palate are at risk for speech and language delay. Yet, many children with cleft palate exhibit normal speech and language from a young age. It is not clear however, what percentage of children exhibit problems with speech only, language only, both speech and language delays, or normal speech and language skills at age three. Further, we do not know what early variables are most associated with normal or delayed performance at age three.

Aims: The purpose of this research was threefold. First, to identify the percentage of children with cleft palate at 39 months who fell into each of four speech/language outcome profiles: Profile 1 - normal velopharyngeal (VP) mechanism + normal speech/language, Profile 2 - normal VP mechanism + delayed speech and/or language, Profile 3 - questionable VP mechanism + delayed speech and/or language, and Profile 4 - questionable VP mechanism + normal speech/language (Scherer, Chapman, Hardin-Jones, & D’Antonio, 2005). Second, the study examined the presurgery and postsurgery speech/language skills that predicted membership in each of the four speech/language outcome profiles at 39 months. Third, the study identified risk factors for poorer speech outcomes at 39 months.

Methods: Participants included 43 children with cleft palate. Spontaneous speech/language samples were collected at 9 months (presurgery), postsurgery (approximately 13 months), 21 months, and 39 months of age during an interaction with the caregiver. Speech status at 39 months was classified as normal or disordered based on the Smit et al. (1990) norms for age of acquisition of speech sounds. Language status was determined based on MLU, NDW, and performance on a standardized measure of expressive language. VP status was based on perceptual judgements of resonance. Membership to each of the four speech/language outcome profiles was calculated in percentages. A logistic regression was conducted using selected factors based on the LASSO approach to determine which timepoint(s) and variables were most predictive of the four outcome profiles at 39 months. Chi-square analyses were performed to determine which risk factors were associated with poor speech skills at 39 months.

Results: Thirty-three percent, 41%, 21%, and 5% of the children fell into Profiles 1, 2, 3, and 4, respectively. Results of the logistic analysis indicated that size of true consonant inventory and stop production at 21 months had the highest predictive effects against poor speech outcomes at 39 months. Chi-square analyses indicated significant associations between speech outcomes and three variables; gender, maternal education, and resonance.

Summary/Conclusion: Profile 2 - normal VP mechanism and delayed speech and/or language had the highest membership (41%) followed by Profile 1 - normal VP mechanism and normal speech/language (33%). Variables associated with poor speech outcomes included female gender, maternal education less than a bachelor’s degree, and moderate to severe hypernasality. Early assessment and monitoring of consonant production including growth in the children’s consonant inventory and stop production provide vital information for treatment planning with these children.
Changes in speech after synchronous hard palate closure and alveolar bone grafting in children with unilateral cleft lip, alveolus and palate: a retrospective cohort study

S Hakkesteegt1, H Poldermans1, M Koudstaal1,2, S Versnel1, M Haj1
1Erasmus Medical Center, Rotterdam, Netherlands, 2Karolinska University Hospital, Stockholm, Sweden

Background: Speech disorders and transient speech delays are common in children born with a cleft palate. Palate repair and speech therapy by a trained speech pathologist are necessary steps for optimal speech development. Although there is a general consensus on the timing of soft palate closure and alveolar bone grafting (ABG) in patients with a unilateral cleft lip, alveolus and palate (CLAP), the timing of correction of the hard palate is still being debated. The Erasmus Medical Center – Sophia Children’s Hospital in Rotterdam practices a two-stage approach with soft palate repair between 9 and 12 months followed by a closure of the hard palate combined with alveolar bone grafting when the canine root in the cleft region has reached a two thirds development (between 9-13 years).

Aims: This study aims to evaluate the documented changes in the articulatory patterns and resonance based on pre- and postoperative speech assessments in patients with unilateral CLAP who undergo simultaneous ABG and hard palate closure.

Methods: Patients with non-syndromic unilateral CLAP who underwent synchronous ABG and hard palate closure between 9-13 years of age between 2013-2018 at Erasmus Medical Center - Sophia Children’s Hospital were included. Basic patient characteristics, cleft extent and lateralization, surgical procedures and pre- and postsurgical detailed speech assessments were evaluated. The speech assessments contained evaluation of compensation strategies, presence of articulation or phonetic disorder, voice disorder, nasality and hypernasality degree, facial grimacing and speech intelligibility. Speech evaluations were standardized and carried out before and after synchronous ABG and hard palate closure by a qualified speech-language pathologist who is involved in the cleft care.

Results: A total of 33 patients were included. Twenty were male and thirteen were female. Eight patients (24.2%) underwent velopharyngoplasty before synchronous ABG and hard palate closure due to observed velopharyngeal insufficiency. The synchronous ABG and hard palate closure was carried out by five different oral and maxillofacial surgeons. Speech was assessed at a mean of 4.3 weeks before surgery (SD = 6.4) and at a mean of 15.6 weeks after surgery (SD = 11.4). Phonetic disorders improved in 1/33 patients (p = 0.317) and facial grimacing decreased in 8/32 patients (p = 0.088). The speech intelligibility improved in a total of 20/33 patients (p = 0.000). Hypernasality degree improved in 21/32 patients (p = 0.000).

Summary/Conclusion: After synchronous ABG and hard palate closure a significant improvement in speech intelligibility was observed which can be explained by the significant decrease of hypernasality. There was no significant improvement in phonetic disorders, probably as a result of inadequate articulatory patterns which are incorporated in the speech.
A descriptive study of syllable inventory in 12-months-olds with isolated cleft palate.
Results from 485 infants in an international rct, timing of primary surgery for cleft palate (tops)

C Persson¹, R Cooper², B Conroy², C Gamble², E Willadsen *³
¹Department of Health and Rehabilitation, Speech- and Language Pathology Unit, Institute of Neuroscience and Physiology, the Sahlgrenska Academy at University of Gothenburg, Göteborg, Sweden, ²Clinical Trials Research Centre, University of Liverpool, Liverpool, United Kingdom, ³Department of Nordic studies and Linguistics, University of Copenhagen, Copenhagen, Denmark

Background: TOPS is an international prospective randomised trial investigating the influence of timing of palatal repair on speech in infants born with an isolated cleft palate. 558 infants from 23 cleft centres in five countries (Brazil, Denmark, Norway, Sweden and UK) are included in the trial. All infants are operated by the Sommerlad technique and are randomly assigned to surgery at either 6 or 12 months of age. Syllable inventory in 12-months old infants with cleft palate have earlier mainly been reported from relatively small-scale studies and rarely on infants with isolated cleft palate.

Aims: The aim of this sub-study was to describe the most frequently produced syllables across all infants from each of the five including languages across the group of 485 infants born with an isolated cleft palate.

Methods: Infants were video recorded at 12 months of age during a 45 minutes play session with one of their parents. The recordings were later split into two 22 minutes recordings. Three Speech and Language therapists (SLTs), blinded to the infants’ surgical status, assessed each recording. Half of the infants had received palate surgery while the other half still had an unrepaired cleft. The SLTs listened to the entire recording in real time, i.e. without pausing. No notes were taken. During the assessment, the SLTs annotated syllables as canonical or non-canonical in real time, using a software, TimeStamper, developed for this purpose. At the end of the recording, they listed all syllables they remembered the infant produced with command. For an infant to have been deemed to produce a sound, at least two of the three SLTs had to identify the sound. Sounds identified by two or more SLTs were summed to derive the infant’s consonant inventory. Syllables uttered by at least 10% of infants were presented.

Results: The most common syllables uttered by the infants were /ma, wa, na, ja, ha, da, ga, ba, ηa/. The three most common syllables identified by at least two SLTs /ma, wa, na/ were identified in 67%, 51% and 45% of the infants. The most common syllables overall are the most common syllables within each country, with /ma/ being universally the most common.

Summary/Conclusion: The most common consonants were nasals and approximants, both low pressure consonants. Voiced alveolar/dental plosive followed by voiced velar plosive were the most frequent plosives in the infant’s syllable inventory. The most frequent syllables were almost identical across countries.

*The study is co-authored by the TOPS SLT group.
Development of obstruent correctness from 3 to 5 years in Danish children with unilateral cleft lip and palate

Løg Jensen¹, E Willadsen²

¹Copenhagen Cleft Palate Center, Rigshospitalet, Copenhagen University Hospital, ²Department of Nordic Studies and Linguistics, University of Copenhagen, Copenhagen, Denmark

Background: Speech sound development in pre-schoolers with cleft palate as a group is delayed/deviant. High pressure consonants, also called obstruents, comprise the most vulnerable sound class. Production of these sounds demands increased intra-oral air pressure which is compromised when the palate is completely or partially unrepaired, or in the presence of velopharyngeal dysfunction (VPD). Notwithstanding, no study has to our knowledge examined the development of obstruent correctness longitudinally in a large group of pre-school children with unilateral cleft lip and palate (UCLP).

Aims: The present study aims to investigate the development of obstruent correctness (PCC-obs) from 3 to 5 years of age and the possible influence on outcomes of age, gender, VPD, cleft speech characteristics (CSCs), and developmental speech characteristics (DSCs), in two groups of children with UCLP.

Methods: A total of 126 consecutive Danish children born with UCLP were recruited in the Scandcleft project (Semb et al. 2017). Phonetic transcriptions of audio and video recordings of a naming test from 108 participants (30 girls, 78 boys) were available at both three and five years. All 108 participants had their lip and soft palate closed at four months. Fifty-five participants had early hard palate repair at 12 months (EHPC group), and 53 participants had late hard palate repair at 36 months (LHPC group).

Two speech-language pathologists (SLPs), blinded to the randomisation, transcribed all naming tests. Intra and inter rater agreement was acceptable. At 3 years, disagreement was solved by consensus listening. At 5 years, transcriptions from the SLP with the highest intra-rater agreement were included in the data analyses. Groups were analysed separately because earlier studies showed significant group differences at both 3 and 5 years with the LHPC group demonstrating poorer speech correctness than the EHPC group (Willadsen et al., 2017a; 2017b). Paired t-tests with bias-corrected and accelerated bootstrapping were used to analyse differences between outcomes at 3 and 5 years, and logistic regression analyses were used to investigate predictors of PCC-obs at 5 years.

Results: In the EHPC group, PCC-obs scores increased significantly from age 3 to 5, but effect sizes were small. PCC-obs also increased per sound, but not for early developing sounds. A significant decrease in DSCs was found, but not CSCs. Logistic regression analyses showed that PCC-obs at 3 years, CSCs, DSCs, and gender were significant predictors of PCC-obs at 5 years.

In the LHPC group, PCC-obs scores increased significantly from age 3 to 5 with large effect sizes. PCC-obs increased per sound, except for the velar stop /k/. A significant decrease in both DSCs and CSCs was found. Logistic regression analyses showed that PCC-obs at 3 years and CSCs were significant predictors of PCC-obs at 5 years whereas DSCs and gender were not.

Summary/Conclusion: Both groups of children with UCLP showed significant progress in obstruent correctness from age 3 to 5. However, groups did not only differ in outcomes at 3 and 5 years; they also showed different developmental trajectories. In addition, predictors of PCC-obs scores at 5 years also differed between groups. These results contribute to our understanding of predictors of obstruent correctness at 5 years and further indicates that we need to look beyond CSCs and acknowledge the importance of more overall measures of speech correctness, including DSCs.
Phonoarticulatory analysis of subjects with velopharyngeal insufficiency treated with different surgical techniques: long term speech outcomes

M Brega¹, V Battista¹, L Autelitano¹, M Meazzini¹, F Aceti¹, I Fontana¹, E Todaro¹, A Rezzonico¹
¹Smile House, Department of Maxillo-Facial Surgery, San Paolo Hospital, Milan, Italy

Background: The term velopharyngeal insufficiency (VPI) identifies a condition in which an adequate closure of the velopharyngeal sphincter is physically prevented by an altered anatomical structure, manageable with a surgical approach only. In order to achieve good surgical and functional success a proper choice of an adequate operative method for every single patient is essential. An accurate perceptual speech is the right approach in order to choose the best surgical procedure for each patient. Two experienced speech-pathologists, through the recording and analysis of the phonoarticulatory assessment, can judge the main features of the VPIs speech symptoms, and eventually address the patient to an instrumental evaluation (Kummer, 2003). In order to select the most appropriate surgical technique for each patient, it's necessary for all the variables emerged during the objective evaluation of the velopharyngeal gap and velopharyngeal closure pattern to be taken into account. The lack of a unitary protocol in collecting data and recording surgery outcomes, due to the absence of evidence-based criteria, determines the difficulty in defining the best possible surgical procedure for each patient (Rudnick and Sie, 2008).

Aims: The main purpose of this study is to validate the appropriateness of the selection approach used at the Smile House Center in San Paolo Hospital, Milan based upon the velopharyngeal gap size and VP closure pattern.

Methods: 81 subjects between the age of 4,1 and 24 years with VPI diagnosis were taken into consideration; 51 were subjected to the superiorly-based pharyngeal flap, 18 to the Furlow double-opposing Z-plasty, 9 to the Sommerlad radical intravelar veloplasty and 3 to the Hynes pharyngoplasty. Inclusion criteria were: complete medical records regarding preoperative speech evaluation and a postoperative perceptual evaluation conducted between 4 and 12 months after the surgery. Exclusion criteria were: evidence of postoperative complications and of velopharyngeal dysfunction due to non-structural causes.

The perceptual speech assessment was conducted using the GOS.SP.ASS. modified protocol (Sell et al., 1999), as for the instrumental evaluation the patients underwent a nasopharyngoscopical and a videofluoroscopic examination. Preoperative and postoperative perceptual speech ratings (hypernasality, nasal emission, intelligibility and acceptability) were then analyzed and compared to evaluate the effect of the surgery.

Results: The difference between preoperative and postoperative scores was statistically significant for all the speech parameters taken into consideration (p < 0,01), determining the success of the selection approach used at San Paolo Hospital.

Hypernasality improved in 87,6% of cases, nasal emission in 75,3%, acceptability in 76,5% and intelligibility in 79%.

Summary/Conclusion: The average improvements in each parameter analyzed are in agreement with studies supporting the adequacy of surgical protocols based on velopharyngeal gap size and velopharyngeal closure pattern. These studies reported an average improvement between 62% and 98% among the speech parameters of hypernasality, nasal emission, acceptability and intelligibility, depending on the choice of surgical technique and on the perceptual evaluation protocol used (Sommerlad, 2002; Nabi et al., 2011; Hirschberg, 2012; Pet et al., 2015).
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Quantifying the degree of perceived hypernasality in vowels
N El-Bashiti¹, R Shrivastav²
¹Department of Hearing and Speech Sciences, University of Jordan, Amman, Jordan, ²Vice Dean of Instruction, University of Georgia, Athens, United States

Background: In the case of velopharyngeal dysfunction, hypernasality occurs due to resonances associated with the nasal cavities during the production of oral sounds. The amount of oral or nasal resonance in speech can be estimated by using acoustic analysis. Several acoustic correlates for this property have been reported in previous literature. The major changes in the acoustic signal have been observed in the vicinity of the first formant.

Most instruments available to assess the velopharyngeal function are often hard to administer, costly, and involve a degree of invasiveness. For more effective clinical use, it is important to develop a non-invasive and affordable method to detect and quantify hypernasality.

One way to develop such a tool is through acoustic analysis of speech. Such a tool would need to be highly correlated with perceptual judgments of hypernasality. In this study an acoustic measure was examined in both synthesized samples and natural voice recordings of hypernasal speech. Correlations of this measure with perceptual ratings of hypernasality were obtained.

Aims: The aim of this series of studies is to study the effectiveness of the proposed acoustic measure, A1-Pn, in explaining the variability in the perception of hypernasality in the cleft palate population.

Methods: Two main experiments were conducted. Participants of the first experiment were five native speakers of American English while participants of the second experiment were ten native speakers of Arabic. All participants were undergraduate and graduate students of speech and hearing sciences.

For the first experiment, stimuli consisted of six vowels (/i/, /u/, /ɛ/, /ɑ/, /ʌ/, and /æ/) that were recorded for 4 normal English speakers. MATLAB version 8 was used to edit the recordings and generate a set of synthesized vowels. For the second experiment, forty sentences were recorded from individuals born with cleft palate.

Inter-judge reliability and intra-judge reliability for all perceptual judgements were calculated in both experiments. For the first experiment, to determine the effects of acoustic manipulation of vowels on the perception of hypernasality, a three-way ANOVA with post hoc comparisons using Bonferroni's correction was used. The independent variables of the study were the extra peak location, the extra peak amplitude, and vowel type. For the second experiment, variables including sentence type, speaker group, vowel type were used in a backward stepwise multiple regression analysis to predict perceptual judgments using the proposed acoustic measure.

Results: For the first experiment, significant main effects were found for vowel type and peak location. No significant effect of different amplitudes of the extra peak on the nasality judgment was found.

For the second experiment, the prediction model contained three of the four predictors and was reached in two steps with only one variable removed (sentence type). The model was statistically significant, and accounted for approximately 67% of the variance of perceptual hypernasality ratings.

Summary/Conclusion: The proposed measure, A1-Pn, is shown to be a promising measure of hypernasality. Studies of similar nature must take into account vowel type used in the stimuli, stimuli length, speaker’s gender and age, and the rating scale used to judge hypernasality perception. Due to the complexity of resonance problems, a set of measures would collectively predict perceptual judgments of hypernasality rather than depending on one single measure.
Auriculocondylar syndrome (ACS) is a rare craniofacial disorder, mainly characterized by mandibular hypoplasia and an auricular defect at the junction between the lobe and helix, known as a "Question Mark Ear" (QME). QME can also occur as an isolated anomaly. ACS/QME is a genetically heterogeneous condition. The mode of inheritance is autosomal dominant or recessive, and causal mutations have been identified in the PLCB4, GNAI3 and EDN1 genes. These mutations are thought to result in dysregulation of the endothelin 1 signaling pathway, which is known to play a key role in branchial arch development in animal models.
**Secondary salvage of the unsatisfactory microtia reconstruction**

C Van Hövell Tot Westerflier¹, J Reinisch¹, D Gould³, Y Tahiri¹

¹Pediatric plastic surgery, Cedars-Sinai Medical Center, Los Angeles, United States, ²Pediatric plastic surgery, Wilhelmina Children's Hospital, Utrecht, Netherlands, ³Plastic- and Reconstructive Surgery, Keck School of Medicine of the University of Southern California, Los Angeles, United States

**Background:** Because auricular reconstruction is a complex and relatively uncommon procedure, there are many patients that have had disappointing reconstructions. Total secondary surgery for unsatisfactory results is a challenging procedure because of prior scarring and limited available skin for framework coverage.

**Aims:** To describe our large experience with secondary procedures using an alloplastic porous polyethylene implant in patients with unsatisfactory or failed initial ear reconstruction.

**Methods:** A prospectively maintained database of all consecutive patients who underwent secondary total ear reconstruction from March 1991 to December 2017 was reviewed. Demographic data and outcomes were assessed. Patients with acquired absence of the ear were not included.

**Results:** There were 144 microtia patients that met the inclusion criteria. Patient’s age at the time of the secondary reconstruction ranged from 3 to 59 years of age. Follow-up duration ranged from one year to 21 years. Primary reconstruction was done with rib cartilage in 91 patients, porous polyethylene implant in 47 patients, prosthesis in four patients, and irradiated cadaver rib cartilage in two patients. All secondary reconstructions were done with PPE implants. The alloplastic framework was covered with a temporoparietal fascia flap in 76 patients, an occipital fascia flap in 64 patients and a free fascia flap in four patients (two radial forearms in the same patient, one contralateral TPF and one lateral arm). Fourteen patients (10%) had complications requiring revision surgery. Secondary surgery was successful in all but one patient.

**Summary/Conclusion:** The use of a PPE implant is an ideal method for patients needing total secondary salvage ear reconstruction, because of its minimal morbidity and relatively low complication rate.
Bilateral microtia; analysis of 495 patients
C Van Hövell Tot Westerflier, J Reinisch
1Pediatric plastic surgery, Cedars-Sinai Medical Center, 2Pediatric plastic surgery, Wilhelmina Children's Hospital, Los Angeles, United States

Background: Bilateral microtia is not the presence of two unilateral microtias in the same patient. It differs significantly from the unilateral condition. In addition to being less common than unilateral microtia (9:1 ratio), the possibility of a small jaw with associated sleep apnea and the usual bilateral conductive hearing loss require early evaluation and treatment.

Aims: We aim to present the demographic data of the largest series of patients with bilateral microtia in the literature and to discuss the important distinctions between the bilateral and unilateral conditions.

Methods: We performed a retrospective cohort study of a prospectively acquired database. All consecutive patients with bilateral microtia who had had a consult between 1991 and 2019 were studied. Demographics, clinical characteristics, and surgical outcomes are discussed.

Results: During the studied period, 2737 patients with microtia were included in the database. Of these, 495 (18%) had bilateral microtia (319 males and 176 females). Patients’ age at first consult ranged from 1 week to 56 years (median of 5 years). The largest ethnic group were Asian (37%). Of the studied group, 104 patients (21%) had jaw hypoplasia. Seventy-six bilateral microtia patients (15%) had a known syndrome. Of the 176 bilateral patients who underwent alloplastic ear reconstruction by the authors, 116 patients had bilateral reconstructions, providing a total of 292 ears for analysis. A canalplasty was performed either prior to, or combined with auricular reconstruction in 51% of the cases. In 14% of the cases, a bone conduction hearing device was placed at the time of ear reconstruction. Eleven patients (4%) had complications requiring revision surgery. Revisions were successful in all but one patient.

Summary/Conclusion: In addition to being less common than unilateral microtia, bilateral microtia patients require early intervention for their usual bilateral conductive hearing loss. They have a higher incidence of being syndromic, they demonstrate hypernasal speech, and they have sleep apnea and intubation issues secondary to jaw hypoplasia.
Cost analysis of microtia treatment in the Netherlands
M Kolodzynski¹, C van Hövel¹, M Kon¹, C Breugem¹
¹University Medical Center Utrecht, Utrecht, Netherlands

Background: Ear reconstruction for microtia is a challenging procedure. Although analyzing aesthetic outcome is crucial, there is a paucity of information with regard to financial aspects of microtia reconstruction.

Aims: This study was conducted to analyze the costs associated with ear reconstruction with costal cartilage in patients with microtia.

Methods: Ten consecutive children with autologous ear reconstruction of a unilateral microtia were included in this analysis. All patients had completed their treatment protocol for ear reconstruction. Retrospectively direct costs (admission to hospital, diagnostics, and surgery) and indirect cost (travel expenses and absence from work) were obtained.

Results: The overall mean cumulative costs per patient was €14,753. Respectively direct and indirect costs were €13,907 and €846. Hospital admission and surgery covers respectively 55 percent and 32 percent of the all costs.

Summary/Conclusion: This study analysis the costs for autologous ear reconstruction. Hospital admission and surgery are the most important factors of the total costs. Total costs could be decreased by possibly decreasing admission days and surgical time. This data can be used for choosing and development of future treatment strategies.
Abnormal soft palate movements in patients with microtia
M Kolodzynski1, B van Hoorn1, M Kon1, C Breugem1
1University Medical Center Utrecht, Utrecht, Netherlands

Background: Microtia is a congenital malformation of the auricle, ranging in severity. It can be isolated or associated with (craniofacial) anomalies. Most of these anomalies together are described as being part of the oculo-auriculo-vertebral spectrum (OAVS). Velar abnormalities have been described to occur in patients with OAVS, however the incidence in patients with microtia without OAVS is largely unknown.

Aims: The primary purpose of this study was to examine the prevalence of velar palsy in patients with isolated microtia, and in patients with microtia and signs of OAVS. The secondary purpose was to identify possible risk factors associated with the presence of velar palsy.

Methods: All patients with microtia presented to our department between January 2015 and March 2017 were retrospectively reviewed. Medical images, information of the palate and demographics were subtracted from the patients’ medical files.

Results: Velar palsy was present in 18 out of 42 patients with isolated microtia (43%; no signs of OAVS), and in 33 out of 41 patients with microtia and signs of OAVS (80%). Patients with signs of OAVS were found to be independently associated with a higher prevalence of velar palsy (OR: 4.8; 95% CI: 1.7 to 13).

Summary/Conclusion: This study demonstrates a clear relationship of abnormal velar movement and microtia. We believe that isolated microtia should not be seen as a separate entity but as a part of OAVS. Abnormal velar movement can lead to velopharyngeal insufficiency (VPI), which can affect children’s speech development. Physicians should exam the palate in all microtia patients, not only those with speech difficulties.
Tracking health care services for children with craniofacial microsomia: a comparison of caregiver report and medical chart abstraction
E Lockhorst¹, D Luquetti², A Drake³, B Siebold², C Heike²
¹Medical Faculty, Erasmus University, Rotterdam, Netherlands, ²Craniofacial Center, Seattle Children’s Hospital, Seattle, ³Department of Otolaryngology/Head and Neck Surgery, University of North Carolina, Chapel Hill, United States

Background: Craniofacial microsomia (CFM) has an estimated prevalence of 1:5600 live births and is the second most common congenital facial condition treated by craniofacial teams. The broad CFM phenotypic spectrum is associated with hypoplasia of the ear and/or mandible, along with variable combinations of preauricular tags, lateral oral clefts, eye anomalies and facial palsy. These anomalies can result in hearing loss, airway obstruction, speech impairment and vision loss. Extracranial anomalies of the spine, heart, and kidneys are also common in CFM. For these reasons, most children with CFM undergo many evaluations and surgeries throughout childhood, and treatments may occur at multiple different healthcare facilities. However, there is lack of international consensus about the optimal treatments, and few studies have investigated the healthcare services received by children with CFM. The goal of this study is to identify the relationship between different sources of information about clinical care received by individuals with CFM.

Aims: To evaluate the correlation between data obtained from caregiver report and the medical chart regarding prior healthcare providers, evaluations, and surgeries for children with CFM.

Methods: Ninety-nine individuals with CFM ages 0-21 years were enrolled between 2010-2012 at four US craniofacial centers. All participants completed a caregiver interview, photographs and medical charts were abstracted. Data on phenotype, healthcare providers, surgical interventions, and evaluations were integrated from the interview and medical chart abstraction.

We calculated the type and total number of health care services for each participant based on data that were included either in the interview or the medical chart. We calculated the percentage agreement between the medical chart and caregiver interview for healthcare providers, surgeries and screening evaluations by dividing the number of exact matches between the interview and chart abstraction by the overall number of health care services received.

Results: Participants had a mean age of 8.2 years (range: 0.5-19.6), and 56% of the participants were male. The most common features were microtia (92%), mandibular hypoplasia (85%) and aural atresia (76%). Participants were evaluated by an average of 8.3 (range: 2-18) subspecialists and received care at an average of four healthcare facilities. Eighty percent of participants had a kidney ultrasound, 74% had a computerized tomography scan, 72% had dental radiographs, 63% had spine radiograph and 27% had an echocardiogram. Eighty-five percent of participants had at least one surgery, with an average of 5.2 surgeries per participant.

Agreement between caregiver and medical chart was 54%, 47%, and 60% for surgeries, providers and evaluations, respectively. A higher number of surgeries was associated with a lower level of agreement between the two data sources.

Summary/Conclusion: Children with CFM often require many assessments and interventions and we identified a relatively low level of agreement between the caregiver interview and the medical chart on the participants’ medical history. Future research to identify the optimal sources of information and develop more reliable methods for tracking healthcare received by this population is essential to better evaluate the outcomes of the complex care received by individuals with CFM.
A new way to repair the complete cleft lip: the natural sequence cleft lip repair

Robert Mann¹

¹Pediatric Plastic Surgery, Helen DeVos Childrens Hospital, Grand Rapids, Michigan, United States

Background: The vast majority of cleft lips are repaired in a single operation. Cleft lip deformities range from microform clefts to extremely wide clefts. Essentially all cleft lips are closed with some pattern of geometry; no tissue is added. Presently patients born with incomplete clefts who are missing little tissue can expect consistently good results, but as the embryonic deficiency increases in wider more complex clefts the results become much less consistent. Wider clefts are missing more tissue and the tissue that remains is more dramatically deformed. In the past wider clefts were pulled together with lip adhesions. This operation has been largely abandoned. In the classic lip adhesion compression the bony maxillary segments together at the same time accentuating the cleft nasal deformity. Many surgeons have chosen pre-surgical orthodontics to compress the maxilla to reduce lip tension. This approach achieves closure in one step, but often leaves the lips in an abnormal posture with the lower lip protruding ahead of the upper lip, a classic pout position. This posture somewhat acceptable as an infant is not acceptable as an adult and is difficult to correct, and contributes to classic cleft stigmata of mid facial restriction. Success of a cleft lip repair is not just achieving a closure in one stage. Success is achieved when proper facial balance is achieved as a child and progressing into adulthood. If success is easy in an incomplete cleft, and lip adhesions are not effective and compressive closures leave poor facial balance, then we need to look for a new way to get better results for patients with wider clefts. The Natural Sequence Cleft Lip Repair is a completely new approach. In steps one the complete cleft is transformed into an incomplete cleft, adding tissue intra nasally while positioning nasal base correctly and repositioning the orbicularis muscle with no tension into the lip to allow for expansion of the deficient lateral lip element. Then Step 2 uses the geometric pattern of your choice to address the residual vertical lip deficiency with predictable success resulting in a soft lip and achieving the correct lip nose relation.

Aims: To demonstrate a new way to treat the patients born with a complete cleft lip

Methods: The differences between the anatomy of the incomplete cleft lip and the complete cleft lip will be demonstrated. The Natural Sequence Cleft Lip repair technique will be described and illustrated. A qualitative assessment of cases will be reviewed.

Results: The Natural Sequence Cleft Lip repair appears to deliver an excellent result. The patients demonstrate a soft tension free lip, with good facial balance and inconspicuous scars. With results seen in patients with complete cleft lips are comparable to those achieved when repairing incomplete clefts.

Summary/Conclusion: When using the Natural Sequence Lip repair there is less need for compressive pre surgical orthodontics. The operation can easily place the scars in anatomic positions where they are easy to revise if necessary. The softer lip achieved will not contribute to mid facial compression and the patients consistently achieve better nasal base relation which can make the definitive adult rhinoplasty easier. Although it may take two steps to make up for the embryonic tissue deficiency and the increased nasal deformation seen in patients with a complete cleft lip, you can now achieve the same predictably good results you already see for your patients with incomplete cleft lips.
Cleft lip and palate in ectodermal dysplasia
I. Ganske1, T. Irwin1, O. Langa1, J. Upton III1, W. Tan2, J. Mulliken1

1Plastic and Oral Surgery, 2Division of Genetics and Genomics, Boston Children’s Hospital, Boston, United States

Background: Ectodermal Dysplasia (ED) is comprised of several syndromes affecting ectodermal structures such as skin, hair, nails, and teeth. Some of these disorders with ED also involve orofacial clefting and include autosomal dominant Ankyloblepharon Ectodermal Dysplasia-Cleft Lip (Hay-Wells syndrome; AEC) and its affiliated Cleft Lip/Palate-Ectodermal Dysplasia Syndrome (Rapp-Hodgkin syndrome; RHS), as well as Ectodermal Dysplasia-Ectrodactyly Cleft Lip/Palate (EEC), all of which are associated with TP63 mutations.

Aims: The purpose of this paper is to identify the prevalence and characteristics of cleft lip with or without cleft palate (CL/P) in patients with ED treated at a large, tertiary, academic referral center and to describe outcomes and management considerations in this specific subpopulation of syndromic CL/P.

Methods: This is a retrospective review of all patients diagnosed with any type of ED seen at Boston Children’s Hospital between the years of 1990 and 2016. Patients were identified through a search of the medical records and clinically confirmed as having both ED and CL/P. Demographic information regarding cleft phenotype, surgical procedures, outcomes, and genetic testing were collected.

Results: Of 161 patients with a definitive diagnosis of ED, 15 (9.3%) had CL/P, including 8 (53%) males and 7 (47%) females. Eleven patients (73%) had bilateral CL/P, of which 10 patients had bilateral complete CL/P and 1 had asymmetric bilateral CL/P. One patient (7%) had unilateral complete CL/P. Three patients (20%) had cleft palate alone, and 1 of these had complete absence of the palate. Eleven patients (68.8%) had EEC syndrome based on clinical findings; eight (50%) of whom had ectrodactyly and 3 (18.8%) of whom had other hand findings such as syndactyly. Several aberrations from the typical clinical course were noted: one failure of presurgical dentofacial orthopedics requiring premaxillary osteotomy and set back, one dehiscence of a nasolabial adhesion, one patient with prolonged post-operative admissions for respiratory infections, a patient with total absence of the palate requiring free flap construction, and a high rate of velopharyngeal insufficiency (6/11; 55%). Only six (40%) of the patients identified had genetic testing: three had a mutation in the TP63 gene, one had a variant at 3p24.3 which resides next to TP63, and two patients had non-diagnostic results.

Summary/Conclusion: As with other types of syndromic cleft lip and palate (such as van der Woude and CHARGE), the phenotypic expression of cleft lip and palate in ED is more severe than the general cleft population. Due to a higher risk of respiratory infection, palatal fistula, and wound dehiscence, patients should be carefully monitored in the perioperative period. Further studies are needed to determine if TP63 is the mutation for all patients with ED and CL/P.
OS 38.2
Six nscl/p loci show associations with normal-range craniofacial variation
K Indencleef1, J Roosenboom2, H Hoskens1, J White3, M Shriver3, S Richmond4, H Peeters1, E Feingold2, M Marazita2, J Shaffer2, S Weinberg2, G Hens1, P Claes1
1KULeuven, Leuven, Belgium, 2University of Pittsburgh, Pittsburgh, 3The Pennsylvania State University, State College, United States, 4Cardiff University, Cardiff, United Kingdom

Background: Orofacial clefting is one of the most prevalent craniofacial malformations. Previous research has demonstrated that unaffected relatives of patients with non-syndromic cleft lip with/without cleft palate (NSCL/P) show distinctive facial features, which can be an expression of underlying NSCL/P susceptibility genes. These results support the hypothesis that genes involved in the occurrence of a cleft also play a role in normal craniofacial development.

Aims: In this study, we investigated the influence of genetic variants associated with NSCL/P on normal-range variation in facial shape.

Methods: A literature review of genome wide association studies (GWAS) investigating the genetic etiology of NSCL/P was performed, resulting in a list of 75 single nucleotide polymorphisms (SNPs) located in 38 genetic loci. Genotype data were available for 65 of these selected SNPs in three datasets with a combined sample size of 7,418 participants of European ancestry, whose 3D facial images were also available. The effect of each SNP was tested using a multivariate canonical correlation analysis (CCA) against 63 hierarchically-constructed facial segments in each of the three datasets and meta-analyzed. This allowed for the investigation of associations between SNPs known to be involved in NSCL/P and normal-range facial shape variations in a global-to-local perspective, without preselecting specific facial shape features or characteristics.

Results: Six NSCL/P SNPs showed significant associations with variation in normal-range facial morphology. rs6740960 showed significant effects in the chin area ($p = 3.71 \times 10^{-28}$). This SNP lies in a non-coding area. Another SNP, rs227731 near the NOG gene, showed a significant effect in the philtrum area ($p = 1.96 \times 10^{-16}$). Three SNPs showed significant effects on the shape of the nose. rs742071 ($p = 8.71 \times 10^{-14}$), rs34246903 ($p = 6.87 \times 10^{-12}$), and rs10512248 ($p = 8.4 \times 10^{-9}$). Respectively, these SNPs are annotated to PAX7, MSX1, and PTCH1. Finally, rs7590268, an intron variant of THADA, showed an effect in the shape of the supraorbital ridge ($p = 3.84 \times 10^{-7}$).

Summary/Conclusion: This study provides additional evidence NSCL/P-associated genetic variants influence normal-range craniofacial morphology, with significant effects observed for the chin, the nose, the supraorbital ridges and the philtrum area.
The diagnostic odyssey for medically complex children with cleft lip and palate

A Kaye, S Hughes, M Tracy

Children’s Mercy Kansas City, Kansas City, Missouri, United States, Genetics, Plastic Surgery Research, Children’s Mercy Kansas City, Kansas City, Missouri, United States

Background: Cleft care for syndromic patients with multiple medical problems requires highly individualized treatment plans. Having a unifying diagnosis for a constellation of physical findings can help clinicians provide more accurate prognoses for developmental outcomes. For families, having a diagnosis provides an identity and access to a community of similarly affected children for support. The journey to arrive at an official diagnosis is not easy; genetic testing is a lengthy process due to technological limitations and patients’ evolving symptomology.

Aims: This study aims to assess the time it takes to establish a genetic diagnosis for our population of medically complex children with cleft lip and/or palate.

Methods: This study is a retrospective review of genetic evaluations and diagnoses of cleft team patients with a history of multiple medical issues in addition to a diagnosis of cleft lip and/or palate.

Results: A retrospective review of genetic evaluations and diagnoses of cleft team patients with a history of multiple medical issues in addition to a diagnosis of cleft lip and/or palate was performed. 133 patients were identified with orofacial clefts co-presenting with 36 different named syndromes, 22 unique chromosomal abnormalities, and 30 unidentified/unconfirmed constellations of anomalies. There are 72 patients (54.1%) for whom diagnoses have been made or confirmed by laboratory testing including high resolution chromosomes (n=20), microarray (n=19), gene studies (n=24), and FISH probe testing (n=9). Five additional patients had their diagnoses confirmed by brain MRI. Of those with confirmed diagnoses, 7 (5.3%) were diagnosed prenatally via amniocentesis or fetal MRI. Eighteen patients (13.5%) were diagnosed at birth. Another 22.6% were diagnosed within the first year and the remainder (16.5%) after 12 months of age. The mean time to diagnosis is 10.9 months (SD 18.6, range 0-148.7 months). For those not diagnosed prenatally or at birth, mean time to diagnosis is 24.0 months (SD 33.8). Forty patients (30%) maintain a clinically-derived diagnosis because specific testing does not exist, or the diagnosis is suspected clinically but confirmatory genetic testing has remained negative. Lastly, there are 16 patients (12%) for whom no unifying diagnosis has been proposed or decided based on clinical and laboratory findings. For the 56 patients with unconfirmed or undefined diagnoses, 32 had normal chromosome testing, 30 had normal microarray testing, 20 had a variety of normal gene-specific testing, and 21 had normal FISH testing.

Summary/Conclusion: Patients with CL/P and complex medical conditions often present with confusing or difficult-to-define diagnoses. More than half our patients have received confirmatory testing of their syndromic diagnoses, but less than 20% were diagnosed at birth. Complex genetic testing is often negative and may take several months or more to confirm a diagnosis. For more than 40% of patients a diagnosis has not been confirmed or identified at all. Protracted delays in confirming a diagnosis can be very frustrating for both clinicians and families who desire closure as well as prognostic information to provide more comprehensive treatment for affected patients.
The outcome of targeted next generation sequencing in a cohort of 144 patients with craniosynostosis
A Topa1, A Rohlin2, L Lovmar2, G Stenman1, L Kölby3
1Department of Pathology and Genetics, University of Gothenburg, The Sahlgrenska Academy, 2Clinical Genetics, Sahlgrenska University Hospital, 3Department of Plastic Surgery, University of Gothenburg, The Sahlgrenska Academy, Gothenburg, Sweden

Background: Approximately 1 in 2000 children are affected by craniosynostosis. During the last few years, mutations in several genes have been identified as cause of early closing of the cranial sutures. The list increases constantly as new genes with relation to craniosynostosis are detected. Craniosynostosis occurs isolated or associated with other symptoms including malformations as part of several syndromic disorders.

Aims: The aim of our project was to study the prevalence and spectrum of the genetic disorders which are associated with craniosynostosis in a unique cohort of individuals addressed for surgery intervention at the department of Plastic Surgery in Gothenburg.

Methods: The mutational screening of our patient cohort has been performed by using a custom-designed NGS-enrichment panel including 63 craniosynostosis-related genes selected from OMIM.

Results: In approximately 63% of cases mainly represented by syndromic forms of uni- or bicoronal synostosis, either a known previously reported pathogenic variant or a likely pathogenic variant has been detected. As expected, the majority of variants have occurred in the craniosynostosis “core genes”: FGFR2, TWIST1, FGFR3, TCF12, EFNB1 and POR. However, novel likely pathogenic variants have been observed also in IL11RA, KMT2D and SKI-genes. Pansynostosis with Crouzon-like phenotype has been noted in all three patients with variants in IL11RA.

Summary/Conclusion: Our study shows that a broad genetic screening using a targeted NGS assay have a high diagnostic yield in a large cohort of patients with mainly syndromic forms of craniosynostosis.
OS 38.5

How genetic data can help us to understand the causes and consequences of being born with an orofacial cleft

S University Of Bristol¹, G Sharp², E Stergiakouli³, L Howe⁴, C Dardini¹, A Davies², K Humphries², Y Wren², J Sandy²
¹Population Health Sciences, ²Bristol Dental School, University of Bristol, Bristol, ³University College London, London, United Kingdom

Background: The majority of clefts are multifactorial in origin, with both genetic and prenatal environmental risk factors many of which are still unknown. In developed countries the cleft is usually repaired in the first year of life, however often further surgeries are required as the child grows, coupled with other interventions to improve speech, hearing and psychological outcomes amongst those born with a cleft. In addition, there is evidence that children born with a cleft have lower educational attainment than those without a cleft. It is not clear if this is due to shared genetic factors or factors related to having a cleft (such as anaesthesia or absences from school) or confounding factors (eg parental lifestyle factors). A better understanding of the causes and consequences of orofacial clefts will be essential to inform prevention strategies and reduce the risk of co-morbidities.

Aims: To use genetic data to determine; a) modifiable causal risk factors for non-syndromic cleft lip and/or cleft palate (nsCL/P) b) biological pathways to nsCL/P and c) the pathways by which adverse outcomes associated with having a cleft arise.

Methods: To use genetic data to determine; a) modifiable causal risk factors for non-syndromic cleft lip and/or cleft palate (nsCL/P) b) biological pathways to nsCL/P and c) the pathways by which adverse outcomes associated with having a cleft arise. Using a large population based birth cohort we; 1) used polygenic risk scores to determine whether there was a genetic overlap between nsCL/P and related face shape variables 2) carried out Mendelian randomization analyses using a single genetic variant associated with alcohol intake to determine whether moderate maternal alcohol intake during pregnancy influences face shape and 3) used Mendelian randomization and LDscore regression to determine whether common genetic variants that increase the chance of being born with nsCL/P also influence educational attainment.

Results: We found strong evidence of a genetic overlap between nsCL/P and philtrum width in the general population; a 1 S.D. increase in nsCL/P PRS was associated with a 0.10 mm decrease in philtrum width (95% C.I. 0.054, 0.146; P = 2x10⁻⁵). But we found little evidence that exposure to low levels of alcohol in utero influenced 6 facial features or that genetic liability to nsCL/P was correlated with lower educational attainment.

Summary/Conclusion: Genetic data can provide a powerful alternative to questionnaire data (where that data is missing or to avoid measurement error and confounding) and can be used to identify modifiable factors which influence face shape and nsCL/P. Such data can also help to determine the pathways by which genetic variants for nsCL/P cause clefts (ie by altering philtrum width) and to determine whether outcomes associated with having a cleft are due to shared genetic or environmental risk factors.
Associated malformations in children with isolated cleft palate
S Larsdotter¹, H Mark¹, P Boivie¹
¹Sahlgrenska Academy, Gothenburg, Gothenburg, Sweden

Background: Cleft lip and/or palate is one of the most common birth defects among children, affecting 2/1000 in Sweden annually. This study focuses on isolated cleft palate (ICP). In comparison with other cleft types, ICP is described to be more frequently associated with malformations. However, there is a wide variability in international studies regarding risk factors, gender, heredity and the frequency of associated malformations.

Aims: The aim of this study is to chart demographic incidence of ICP and associated malformations and to explore how associated malformations vary between children with ICP in the soft palate only and both soft and hard palate.

Methods: In this retrospective study, data on children born between 1985-2016 with ICP, and treated at the Department of Plastic surgery at Sahlgrenska University Hospital, was investigated. A total of 875 children with ICP, unilateral or bilateral cleft were included. Demographical data and surgical outcomes were obtained from standardized medical records and statistical analyses were accomplished.

Results: In the ICP-group, the incidence of associated malformations were 37%, most frequently located in musculoskeletal organs, the circulatory and craniofacial systems. The study further confirms the tendency towards female prevalence in ICP patients and shows significantly higher rates of female presence in children with bigger clefts. In this group, higher rates of associated malformations were seen in female patients. Also, findings showing a tendency of more numerous associated malformations in ICP patients with bigger clefts. We are still processing the incidence of associated malformations in children with unilateral and bilateral clefts and data will be presented.

Summary/Conclusion: This study provides essential insights into demographic data and factors that may impact on the presence of associated malformations in cleft patients. In ICP-patients, the length of the cleft tends to influence the presence of associated malformations.
OS 38.8

Is there a role for akt in palatogenesis

D Tophkhane¹
¹- -, -, Kosovo

Background: Cleft palate (CP) has a polygenic and multifactorial aetiology and it can occur alone or in association with cleft lip (CL). Development of this anomaly is a result of defect in mechanisms responsible for development of palate during embryogenesis. Complete fusion of the two lateral palatal shelves following midline epithelial seam (MES) disintegration is one the crucial step in formation of secondary palate. Cell migration, epithelial to mesenchymal transformation (EMT) and apoptosis are the three controversial mechanisms that have been proposed for determining the fate of the MES. Multiple signaling pathways are activated during these events but the TGFβ3 signaling pathway has been associated with MES disintegration and palatogenesis for more than three decades. TGFβ3 is a cytokine that signals via two pathways Smad-dependent and Smad-independent to bring about cellular changes. The PI3K/Akt pathway is a Smad-independent pathway which is activated by TGFβ3 when the Smad-dependent pathway is inactivated or blocked. PI3K/Akt pathway activates Akt which has been associated with cell survival, cell proliferation, cancer metastasis, cell migration and EMT but very less information is available about its role in palatogenesis.

Aims: TGFβ3 induces cell migration, EMT, and apoptosis by phosphorylation and activation of Akt through Smad-independent PI3K/Akt pathway.

Methods: An in vitro model for cell migration, EMT, and apoptosis was developed using medial edge epithelial (MEE) cell line derived from single unfused palatal shelf isolated from palatal organ culture of CF-1 mouse embryo. The effect of exogenous human recombinant TGFβ3 and Akt inhibitor MK-2206 on the cell migration, EMT, and apoptosis was investigated. Scratch assay, scatter assay, µ-slide Chemotaxis 2D assay, 3D Biomatrix three-dimensional cell culture assay, immunofluorescence staining for E-cadherin, vimentin and PARP-1 antibody, SDS PAGE and Western blot were performed following standard aseptic laboratory protocol.

Results: Increase in cell migration, EMT, and apoptosis were observed in cells treated with exogenous recombinant TGFβ3. Cell migration, EMT along with early and later apoptotic changes were observed under phase contrast microscopy and 24 hours time-lapse microscopy. In the immunofluorescence staining, the expression of E-cadherin was found to be down-regulated while the expression of vimentin was up-regulated in the cells treated with TGFβ3. The expression of apoptotic marker (PARP-1) was high on the day 7 after treatment with TGFβ3. The cell migration, EMT, and apoptotic changes decreased in the presence of Akt inhibitor MK-2206. The levels of S473 phosphorylation were considerably reduced after incubation with MK-2206.

Summary/Conclusion: The results established that TGFβ3 plays a role in cell migration, EMT and apoptosis by activation and phosphorylation of Akt. MK-2206 blocks phosphorylation of Akt at S473 residue.
Unique phenotype in a brazilian girl with syndromic robin sequence and a 5.6 mb deletion in Xp11.4p11.3.

N Kokitsu-Nakata1, F Jehee2, R Sandri1, S Piffoli1, C Alvez1, R Monteiro3, J Mazzeu4, R Zechi-Ceide1, A Richieri-Costa1
1Rehabilitation Hospital for Craniofacial Anomalies, Bauru, Brazil, 2Clinical Genetics, Erasmus Medical Center, Rotterdam, Netherlands, 3Institute of Education and Research of Santa Casa de Belo Horizonte, Belo Horizonte, 4Federal University of Brasília, Brasília, Brazil

Background: The Pierre Robin sequence is a rare condition characterized by micrognathia, glossoptosis, and upper airway obstruction. Almost half of the cases has associated multiples anomalies, being classified as syndromic Robin sequence or as Robin Plus sequence. The etiology of syndromic cases is heterogeneous and frequently unclear. Clinical follow up and molecular genetic tests are essential for a proper diagnosis.

Aims: To investigate the genetic etiology of the atypical phenotype of an patient with syndromic Robin sequence.

Methods: Clinical genetic evaluation and SNP array

Results: The patient is a 5–year-old Brazilian girl born to normal and nonconsanguineous parents. Clinical evaluation at age 7 months showed microtrigonocephaly, abnormal structural nervous system (Dandy-Walker variation), left microphthalmia, ocular hypertelorism, broad nasal bridge, abnormal skin crease in nasal tip, Robin sequence, abnormal ears, partial syndactyly of 2nd and 3rd fingers, dysphagia, heart defect (atrial septal defect and bicuspid aortic valve) and developmental delay. The clinical follow-up at 5 years old showed worsening of the craniofacial phenotype with arched eyebrows, long palpebral fissures, arched upper lip, and low-set prominent and abnormal ears. She also had a short neck with pterygium coli. She showed a moderate developmental delay: she was able to walk with support, maintain eye contact, socially smile and understand simple orders. SNP-Array showed a heterozygous deletion in the X chromosome: arr[hg19] Xp11.4p11.3(40,648,211–46,310,832)x1. Parental samples were not tested.

Summary/Conclusion: The patient has a pattern of clinical anomalies that do not fit in a known syndrome. The deletion found by SNP-array include two X-linked dominant genes (KDM6A and CASK) that could, at least in part, contribute to the atypical phenotype. For instance, haploinsufficiency of KDM6A results in X-linked Kabuki syndrome. The patient shows clinical findings compatible with Kabuki syndrome, such as long palpebral fissure, abnormal ears, dysphagia and developmental delay. Additionally, MICPCH syndrome (Mental retardation and microcephaly with pontine and cerebellar hypoplasia) is caused by heterozygous mutation or deletion in the CASK gene. Although some features of MICPCH are compatible with the phenotype of our patient, such as microcephaly and hypertelorism, the brain and eye anomalies, sensorineural hearing loss and epilepsy do not match the symptoms of our patient. This variable phenotype could be due to the X-inactivation pattern. Furthermore, the overlap of our patient’s phenotype with other syndromes with midline defect such as Baraitser-Winter syndrome could point to a possible influence of other gene(s) variation(s) in the full phenotype. Atypical clinical presentations may be the expression of blended clinical phenotypes arising from independent pathogenic events at two loci. Whole exome sequencing could help elucidate this intricate syndromic Robin sequence phenotype.
Orthodontics

OS 39.2
Dental and orthodontic treatment outcome of clp patients
P Saele¹, M Mustafa Sharafeldin¹
¹Center for CLP Bergen Norway, Oral Health Center of Expertise/Western Norway, Bergen, Norway

Background: To study the dental and orthodontic treatment outcome of 15-16 years old Norwegian patients born with cleft lip and or palate between 1995 and 2005 enrolled in the CLP archive in Bergen Norway.

Aims: To evaluate the received surgical-orthodontic treatment outcome among cleft lip and or palate patients (CLP) and the need for dental care-treatment after the age of 15 years compared to the Norwegian population without CLP.

Methods: Five hundred consecutive patients born with all subgroups of CLP registered in the CLP archive of Bergen Norway were included. All patients were treated and followed according to the same protocol throughout their growth. Exclusion criteria were missing cooperation and records. All patients were clinically and radiographically examined at the age of 15 years. The dental status was evaluated from OPG, lateral cephalometry and dental casts. The study had focus on three parameters: Growth and intermaxillary relations based on ANB. The dental arch, levelling and need for dental restorations, and the occlusion and need for orthognathic surgery.

Results: All patients were treated by fixed appliance except 2% of the patients who denied treatment. The intermaxillary relations, the ANB was less than 0 degrees among 22% of the patients. Unsatisfactory occlusion was reported in 15% of the patients and they were appointed for orthognathic surgical evaluation at the age of 19.

Fifty percent of the patient still had a need for some orthodontic, orthognathic and prosthodontic treatment. Additional levelling of the arch after the age of 15 is also indicated for certain patients. Agenesis of one or more teeth was observed in 35% of the study participants and 20% of the patients did need dental restorations like implants or porcelain bridges due to missing teeth.

Summary/Conclusion: The need for orthodontic treatment among the Norwegian population without CLP is about 34%, and the average agenesis rate is 6%. Our study showed that all patient with CLP had a dental diagnosis that indicated need for orthodontic treatment, but 50% of the patients still need some dental adjustments after the age of 15 due to missing teeth, intermaxillary relations or need for further orthodontic levelling.

Conclusion: Patients with a CLP have a significant higher need for orthodontic treatment and dental service than the Norwegian population without CLP.
Analysis of craniofacial growth in colombian individuals with and without cleft lip and palate
C Téllez Conti1, M Gonzalez Carrera11, I Mora11
1Universidad El Bosque, Bogota, Colombia

Background: Craniofacial growth is a dynamic and unpredictable process influenced by genetic and environmental factors, presenting phenotypic and gender differences. Poor control of cell growth, migration, differentiation, and apoptosis may lead to the development of clefts. The craniofacial growth pattern in patients with cleft lip and palate (CLP) causes changes in the facial structure and function, and determines the treatment required for craniofacial reconstruction.

Aims: The aim of this study was to analyze and compare the differences in craniofacial growth and development in patients with complete unilateral and bilateral CLP, and without CLP, classified by gender and age.

Methods: Eight cephalometric measurements on profile radiographs, five on linear radiographs, and three on angular radiographs were performed. The linear measurements were sella to nasion (S-N), anterior nasal spine to posterior nasal spine (ANS-PNS), gonion to pogonion (Go-Pog), nasion perpendicular to the Frankfort plane to point A (N perpendicular A), nasion perpendicular to the Frankfort plane to pogonion (N perpendicular Pog). The angular measurements included A-nasion-B (ANB), sella-nasion and sella-basion (SN-SB), and sella-nasion and gonion-pogonion (SN-GoPog). A single calibrated researcher plotted the data and compared cephalometric data of 540 profile radiographs of 127 patients with unilateral CLP, 126 with bilateral CLP, and 287 without CLP (controls). The patients were classified by age (3 to 18 years or until the end of craniofacial growth) and gender. The enrolled patients of affected groups had a history of CLP correction surgery without nasoalveolar molding with orthopedic and orthodontic treatments. Descriptive statistics and comparison tests were performed.

Results: There were significant differences (p < 0.05) between the groups of patients with and without CLP, between types of clefts, and between genders. The bone structures of patients with CLP were smaller than those of control patients but improved with craniofacial growth. The clefts occurred in the middle third of the face, and the vertical measurements were larger in patients with CLP than in controls. There were significant differences in craniofacial growth and surgical outcomes between patients with different types of CLP. For example, unilateral CLP patients presented flat profiles and predominant class III malocclusions, whereas bilateral CLP patients had higher variability in the size and position of the maxilla during craniofacial growth with predominant class II malocclusions and orthognathic profile by the end of craniofacial growth. The mandible of CLP patients presented postero-inferior rotation as compared to controls.

Summary/Conclusion: The dynamism of craniofacial growth due to variations in individual and family traits produced differences between subjects with and without CLP. The development of a cleft in the middle third of the face causes intrinsic limitations in craniofacial growth, and even post-treatment, certain structural and functional features remain that are markedly different from non-CLP individuals.
Pharyngeal airway changes following maxillary protraction after using alternate maxillary expansion and contraction protocol in cleft patients

E Dogan

Orthodontics, Faculty of Dentistry, Izmir, Turkey

Background: This study is about evaluating the airway dimensions on patients with unilateral cleft lip and palate, after the maxillary protraction treatment.

Aims: The aim of this study is to evaluate the pharyngeal airway changes following the maxillary protraction after using alternate maxillary expansion and contraction protocol in patients with unilateral cleft lip and palate compared to non-cleft patients having Class I malocclusion.

Methods: This prospective study is done with 17 patients (mean age: 12.09 ± 0.83 years) who have unilateral cleft lip and palate with skeletal and dental Class III malocclusion. Alternate maxillary expansion and contraction protocol was applied before facemask treatment. Control group was consisted of 15 patients (mean age: 12.50 ± 0.89 years) having Class I malocclusion. The lateral cephalometric X-rays were evaluated by using Dolphin Imaging Software Version 11.7 and Arnett Gunson FAB analysis was used to evaluate the pharyngeal airway changes. Student t test was used to evaluate the collected data.

Results: In cleft group, after treatment (A/G)-A and (A/G)-Mx1 were increased when compared to control group (p<0.001). Pharyngeal airway dimensions at (A/G)-B and (A/G)-Pog were similar in both groups. Soft palate lengths were different between the groups statistically (p<0.05).

Summary/Conclusion: Maxillary protraction after alternate maxillary expansion and contraction protocol on cleft patients causes changes on upper airway dimensions. The treated cleft patients' airway dimensions become nearly similar with the patients having Class I malocclusion. The development of the upper airway dimensions, may prevent cleft patients to have obstructive sleep apnea.
“only” cleft of the lip. An evaluation of 32 consecutive patients born with an isolated cleft of the lip and no visible defect of the dental jaw at birth.

P Saele¹, H Vindenes¹, B Morken¹, S Granrud¹
¹Center for CLP Bergen Norway, Oral Health Center of Expertise/Western Norway, Bergen, Norway

Background: Study the effect on the hard tissue like bone and dentition in cases where the patients are born with isolated cleft of the lip.

Aims: To assess the dental pattern, the need for dental follow up and for the surgical interventions on patients born with isolated cleft of the lip.

Methods: 32 consecutive patients born with an isolated cleft of the lip and registered in the CLP archive of Bergen Norway were included. All patients were at birth given the severity of the cleft diagnosis. If needed the diagnosis were adjusted at the primary lip surgery. Patients without implement of the dental jaw were included in the study. All patients were treated and followed according to the same protocol throughout their growth. All patients were clinically and radiographically examined by OPG, CBCT and intraoral at the age of 6 and 16 years.

Results: All patients had need for orthodontic treatment. All were treated by fixed appliance except of 3 patients that did not want any treatment. 28% of the patients had agenesis of one or more teeth. All patients had need for orthodontic treatment. All were treated by fixed appliance except of 3 patients that did not want any treatment. 28% of the patients had agenesis of one or more teeth.

Summary/Conclusion: Traditionally we believe that patient born with a soft tissue defect of the lip do not have an increased need for treatment of the hard tissue. In our study, we had one patient with the need for bone grafting and several of the patients showed anatomically bone deviations at the cleft side seen from CBCT. Our study describe at the same time statistically much higher dental abnormal findings than among the normal population. This is a crucial information for the CLP team when planning treatment and degree of follow up of this group of patients.
Presurgical nasal molding using oxygen canula in infants with cleft lip and palate
S Nissan¹, E Botzer¹
¹Pediatric Dentistry, Tel Aviv Medical Center, Tel Aviv, Israel

Background: Cleft lip and palate can arise with variation in severity and form. Wider clefts are associated with more significant nasolabial deformity. These clefts, deficient in hard and soft tissue elements, present a significant surgical challenge to achieve a functional and cosmetic outcome. Even a mild incomplete unilateral cleft lip in the absence of a cleft palate can be associated with a nasal deformity. The principal objective of pre-surgical nasoalveolar molding (NAM) is to reduce the severity of the initial cleft deformity. This can help achieving a finer surgical scar, good nasal tip projection, and more symmetrical and precisely defined nasolabial complex. Nasoalveolar Molding consist of using a plate with nasal stent that extend from the anterior flange of an intraoral molding plate.

Aims: To display a technique for nasal molding using oxygen cannula. case series.

Methods: We used oxygen cannula a base for nasal stent without the need for intraoral plate. By adding soft acrylic to the oxygen cannula we created a nasal stent. The addition of the soft acrylic is similar to the NAM method by Grayson.

Results: The technique is suitable for unilateral incomplete cleft lip with nasal deformity when there is no need for alveolar molding. And in cases that needs alveolar aliment at the beginning of the treatment it can be combine with alveolar molding plate before adding the nasal stent. In unilateral incomplete cleft lip with nasal deformity, we add soft material every week until we achieved nasal symmetry. Since the baby is growing, we follow-up the patient every 2-3 week and add more soft acrylic if needed. the advantages of the technique: It can be done at the clinic immediately with no need to take an impression and wait for laboratory work. It is cheap (oxygen cannula, soft acrylic and tapes), the baby can breast feed. The limitations of the technique the oxygen cannula needs support from the lower part of the nostril, partial cleft or good adhesion of the lips. In cases that needs alveolar aliment, nasoalveolar molding require first alveolar aliment with molding plate and only after it achieved it is possible to add the nasal stent to correct the nasal deformity.

In cases that needs alveolar aliment, we start the treatment with oxygen cannula and molding plate. After aligning the alveolar segments, we add the nasal stent like in any other nasoalveolar molding treatment. The advantages of the technique are starting treating the nose at an early age while the nose cartilages are more soft and can be mold easily. Moreover, in bilateral cleft lip and palate it allows starting elongating the collateral at earlier age. Therefore, it can make the treatment time shorter (3 months instead of 4-6 months in bilateral cases)

Summary/Conclusion: Using oxygen cannula for nasal molding can be used in incomplete unilateral cleft lip without the need of intraoral plate and also in combination with intraoral plate to achieve shorter treatment period.
The art of custom orthodontic surgical corrections; how to achieve the best results for your patients
N Warshawsky

Surgery, University of Illinois, Glenview, United States

Background: Today orthodontics is not considered a luxury, but more likely a proactive attempt to create better oral health through improving facial harmony and esthetics. Occlusion and functional issues such as mastication, breathing, speech, and self esteem are now issues that patients understand and want to improve. However, there is a huge disparity in the variety of treatment mechanisms that a patient may utilize, often limited not by what the patient wants, but rather to mechanics that the doctor knows or uses daily. This issue gets even more complicated when the patient has congenital anomalies such as a cleft or a syndrome and their anatomy is not considered normal.

Aims: In an effort to improve patient experience as well as outcomes, the UIC Craniofacial center adopted a digital workflow to improve our surgical case submissions. In addition, we feel that planning our cases digitally will help our cases to treat more efficiently and accurately.

Methods: This presentation will highlight our evolution of going digital and it will demonstrate a variety of methods we currently utilize to achieve orthognathic correction. Technology such as 3D printing, virtual surgical planning, and virtual reality are of few of the items that we use to design and implement surgery. Both labial and lingual braces will be documented showing that regardless of a patient's desire, it is capable to correct them surgically while providing them exactly what they desire. By adopting a patient centric model where the mechanics and the experience match the patients desires, our goal is create happier patients which achieve their goals quicker.

Results: A multitude of case types will be demonstrated to indicate the value of the digital workflow when planning to correct the transverse, vertical or antero-posterior dimensional issues. Both adolescent and adult cases will be featured to demonstrate how all ages will benefit from patient centric treatment planning.

Summary/Conclusion: Our presentation will demonstrate how we are creating better surgical results through adopting a digital workflow to design, process, and implement patient care.
A preliminary outcome assessment for bilateral cleft lip and palate: presurgical nasoalveolar molding (pnam) versus surgery alone
M Rachwalski¹, T Nayak², S Bitra², P Shetty², K Bonanthaya²
¹Department of Maxillofacial and Plastic Surgery, Hopital Universitaire Necker-Enfants Malades, Paris, France, ²Smile Train
Cleft Lip and Palate Center, Bhagwan Mahaveer Jain Hospital, Bangalore, India

Background: Currently there is no internationally accepted measurement tool available to reliably evaluate the treatment outcomes for complete bilateral cleft lip and palate (CBCLP). Furthermore, there is still debate in the field regarding the effectiveness of nasoalveolar molding.

Aims: Our aim was therefore to compare the postoperative morphology of nose, lip and scar in patients with CBCLP who underwent presurgical nasoalveolar molding (PNAM) versus no presurgical nasoalveolar molding with our newly developed numerical evaluation scale.

Methods: Our cohort comprised 121 Indian subjects with CBCLP who underwent surgical repair a modified Millard technique and were divided into two equally matched groups: Group A: 59 patients with PNAM therapy versus Group B: 62 patients, no PNAM therapy. A panel of three professionals evaluated each subject’s outcome of the bilateral cleft lip repair six months post-operatively on two-dimensional (2D) photographs in frontal and worm’s eye view. A simple two-point rating system was applied to separately analyze a total of twelve components of lip, nose and scar.

Results: The results and mean scores for the analyzed anatomical areas were 2.2±1.01 (max=3) for nose, 5.4±1.54 (max=8) for lip, and 1.9±1.3 (max=3) for scar, with a total score of 7.7±2.21 (max=12) indicating a good surgical outcome. The inter-examiner ICC for nose, lip, scar, and total score was calculated at 0.836, 0.889, 0.723, and 0.927, respectively and indicated a strong level of repeatability and reliability that was highly significant (p<0.001). Statistical analysis (unpaired t test) showed significant difference between the two groups (PNAM vs. no PNAM therapy) with respect to lip, lip+scar and nose (p<0.001) whereas the difference was not statistically significant for scar alone (p=0.080).

Summary/Conclusion: In conclusion, we were able to develop and test a scoring system for measuring outcomes in CBCLP that warrants simplicity of use, reliability and reproducibility. Patients with bilateral cleft lip and palate exhibited superior nasal and lip esthetics when treated with PNAM prior to cheioplasty as evaluated 6 months postoperatively. The results of this study can only be considered to be preliminary, as reevaluation after growth cessation still has to confirm long-term effects on facial morphology.
Growth of the maxillary dental arch in children with cleft lip and palate.

M Soots, R Lutter

Maxillofacial surgery, University of Tartu, Tartu, Estonia

Background: Every year about 20-22 newborns with congenital cleft lip and/or palate are born in Estonia. Disorders of maxilla growth is one of the principal problems in treatment of cleft lip and palate patients. During the last decades a significant improvement of treatment techniques and quality has been achieved, there is a lot of discussion left on the treatment time, tactics and methods. Despite the general opinion that the deficiency of tissue, probably seen as the width of the alveolar cleft, and the position of maxillary segments are essential variables affecting the growth of the maxilla following lip, palate and nose repair.

Aims: To analyse the development of the maxilla arch growth after surgical treatment of cleft lip and palate patients.

Methods: Thirty-four patients with cleft lip and palate were enrolled into this retrospective study. Analyses of the development of the maxilla arch was performed from plaster casts at age 5 months, 1 year and 3 years. As male and female data did not differ for any age (Student’s test), pooled values were considered. The width, length and symmetry of the maxilla arch were assessed in the canine and molar regions.

Results: Mean anterior maxilla arch width in unilateral CLP patients was 30.96mm at the age 5 months, 29.35mm at 1 year, and 27.75mm at age 3 years. Mean posterior maxilla width was 35.81mm at age 5 months and it increased from 39.74mm at year 3. Differences in maxilla width changed significantly over time both in the anterior and posterior parts of the maxilla. Maxilla length on the unaffected side was 28.99mm, 29.44mm and 30.97mm, respectively.

Summary/Conclusion: Maxilla length growth was more intensive than width growth. Differences in maxilla width changed significantly over time both in the anterior and posterior parts of the maxilla. During the first 3 years the width of the anterior part of the maxilla decreased due to surgical interventions; growth of posterior part increased steadily.
The application of stimulators in the treatment of syndromic cleft lip and palate
J Radojicic¹, B Trifunovic², A Radojicic³, A Radojicic⁴
¹Orthodontics, Faculty of Medicine, University of Nis, Nis, ²University Children’s Clinic, Belgrade, ³Faculty of Medicine, University of Nis, ⁴Orthodontics, Ortodent, Nis, Serbia

Background: The clinical picture of a newborn with a syndromic cleft lip and palate is severe. The orthodontic and surgical treatment of cleft is complex and long-term. It is further complicated by various birth defects which can be life-threatening for a newborn or can make the therapy itself more difficult. The induction of a newborn into total anesthesia with a view to performing the surgery of a cleft is often made difficult or time-limited.

Aims: Aim of the paper is to showcase the benefits of active treatment with stimulators for pre-surgical preparation of patients with three types of cleft.

Methods: This paper presents pre-surgical orthodontic therapy in newborns with three severe types of cleft, unilateral cleft lip and palate, bilateral cleft lip and palate and median cleft lip which occurred within three rare syndromes Goldenhar syndrome, lobar holoprosencephaly with a median cleft lip and trisomy 13 (47XX+13). Pre-surgical orthodontic therapy was conducted by means of RBJ stimulators without extra oral fixation, whose construction was conditioned by the type of cleft.

Results: Results: With active treatment of RBJ stimulators, the cleft area in all three types of cleft was significantly reduced, as well as the protrusion of the premaxilla in bilateral cleft lip and palate. By directing the growth of cleft segments of newborn’s upper jaw, the most approximate shape to a healthy newborn’s jaw shape is achieved.

Summary/Conclusion: All three types of described stimulators used in the therapy of syndromic cleft lip and palate enabled primarily the feeding of newborns, and thus their survival. With their orthopedic treatment they created optimal conditions for successful performing of surgical care of syndromic cleft lip and palate.
Hearing in cleft and craniofacial anomalies

OS 40.1
Bone anchored hearing aids in aural atresia: improving outcomes in periabutment skin inflammation
R Stokroos
UMC Utrecht, Utrecht, The Netherlands

**Introduction:** Hearing loss in unilateral and bilateral aural atresia can be rehabilitated successfully using bone conduction devices. These devices are usually coupled to the implant via a percutaneous abutment. This percutaneous abutment frequently causes an inflammatory reaction in the surrounding skin, resulting in discomfort, less usage and in some cases, to implant loss.

**Materials and Methods:** A periabutment coating of the titanium implant using hydroxyapatite was developed aimed at reducing periabutment pocket formation and cicatrisation formation, thereby enabling the natural defence mechanisms to defend against periabutment formation since a skin-abutment integration was aimed for. This was evaluated using molecular and microbiological assays and subsequently evaluated by a multicenter trial in several european centers. Secondly, the surgical procedure to implant bone anchored hearing devices was changed to a more minimally invasive procedure, aimed at preserving integrity of the surrounding tissue, thereby reducing periabutment inflammation also. This procedure was evaluated separately in another prospective clinical trial.

**Discussion and conclusions:** Hydroxylapatite abutment coating lead to a close coupling of the skin-abutment interface, but in daily clinical practice, no effects on periabutment inflammation could be demonstrated. A minimally invasive surgical technique lead to better cosmesis, less loss of sensibility in the skin surrounding the abutment and a reduction in surgical time, but did not lead to a lower prevalence in periabutment inflammation.
Vestibular dysfunction is a manifestation of 22q11.2 deletion syndrome
A Willaert¹, C Van Eynde¹, N Verhaert¹, C Desloovere¹, V Vander Poorten¹, A Swillen¹, K Devriendt¹, G Hens¹
¹KU Leuven, Leuven, Belgium

Background: The 22q11.2 deletion syndrome (22q11.2DS) is the second most common cause of developmental delay after Down syndrome. Impaired cognitive development is highly prevalent, but also motor abnormalities such as hypotonia and delays in achieving motor milestones are described. Instability is frequently detected in children, adolescents, and adults and is mostly attributed to their limited motor performance. Until now, vestibular function has not been investigated in these patients, despite the growing evidence that they often have inner ear malformations.

Aims: The aim of this prospective study was to identify the presence and character of vestibular dysfunction in 22q11.2DS.

Methods: We investigated 23 subjects with proven 22q11.2DS, older than the age of 12. We performed caloric testing and pendular rotation chair tests with videonystagmography, cervical vestibular-evoked myogenic potential (c-VEMP)-testing, and posturography. Additional otoscopy and audiometry were performed on all subjects. This study was approved by the local Ethical Committee of the KU Leuven—University of Leuven. All participants or their parents gave written informed consent on beforehand to participate in the study. The study conforms to the recognized standards (Declaration of Helsinki).

Results: We found a unilateral caloric hypofunction in 55% of patients, a certain absent c-VEMP response in 15% of ears, an inconclusive c-VEMP response in 33% of ears, and abnormal posturography in 68% of patients, of whom 42% displayed a typical vestibular pattern. Remarkably, 90% revealed uni- or bilateral weak caloric responses, independent of caloric symmetry.

Summary/Conclusion: Vestibular dysfunction is frequent in subjects with 22q11.2DS. This knowledge should be taken into account when assessing motor performance in these patients. Additional larger studies are needed to determine whether this dysfunction implicates a therapeutic potential.
**OS 40.3**

**A new solution to treat conductive hearing loss in infants - the adhesive bone conduction hearing system adhear**

K. Neumann¹, S. Dazert²

¹Dept. of Phoniatrics and Pediatric Audiology, Clinic of Otorhinolaryngology, Head and Neck Surgery, St. Elisabeth-Hospital, ²Clinic of Otorhinolaryngology, Head and Neck Surgery, Ruhr University Bochum, Bochum, Germany

**Background:** Bone conduction hearing devices as integrated in softbands (BCD-S) are frequently not well accepted by children with conductive hearing loss due to pressure on the head, sweating, or cosmetic stigma. The non-surgical hearing system ADHEAR uses a new bone conduction concept. It consists of an audio processor connected to an adhesive adapter fixed behind the ear.

**Aims:** This study evaluated the short- and long-term clinical efficacy of the ADHEAR in children compared with conventional BCD-S.

**Methods:** For 10 children with conductive hearing loss (age: 8 months to 9.7 years) the ADHEAR was compared with a BCD-S. Aided and unaided pure tone/behavioral observational audiometry and—if applicable—speech audiometry in quiet and noise were performed initially and after 8 weeks of ADHEAR use. The subjective hearing gain, usage of the ADHEAR, and patients’ as well as parents’ satisfaction were assessed by questionnaires. First long-term results and data of four additional children are available.

**Results:** The functional gain with the ADHEAR (t-test, \( p = .012, n = 11 \)), averaged over 0.5, 1, 2, and 4 kHz, exceeded that one achieved with BCD-S (34.7 dB HL ± 14.1 SD vs. 27.7 dB HL ± 14.7 SD). Speech perception in quiet and noise improved in the aided situation similarly for both hearing devices. All parents except two evaluated the ADHEAR as useful for their child. After an average use of 13 months \((n=4)\) the functional gain with the ADHEAR remained stable or even improved, as did the adhesion time of the adhesive adapter and the wear acceptance. Eight weeks after first fitting six children used the ADHEAR permanently, one year later eight children and the additional four children.

**Summary/Conclusion:** Meanwhile, 11 of 13 children use the ADHEAR permanently, among them three with multiple disabilities. Initial problems regarding wear comfort, fixation of the adhesive, skin irritations, and feedback noise have been largely overcome by technical and handling optimization. One child received an active middle ear implant and one continued in using a softband-integrated BCD.

The ADHEAR system is a favorable technical solution for children with conductive hearing loss or chronical draining ears.
Reliability of speech variables and speech related quality indicators in the swedish cleft lip and palate registry
K Klintö1, K Brunnegård2, E Hagberg3, C Havstam4, Å Jonasson5
1Skåne University Hospital, Malmö, 2Umeå University, Umeå, 3Karolinska University Hospital, Stockholm, 4Sahlgrenska University Hospital, Gothenburg, 5Uppsala University Hospital, Uppsala, Sweden

Background: The agreement within and between assessors is crucial for the reliability and usefulness of the results in cleft lip and palate (CLP) speech research. Also in quality registries evaluating medical care the data need to be reliable. In 2019 and beyond, the results of quality indicators in the Swedish CLP registry will be published continuously on internet. Thus, there was a need to assess the reliability of the speech variables and the speech related quality indicators in the Swedish quality registry for CLP.

Aims: The purpose was to test the reliability of the speech variables and speech related quality indicators in the Swedish quality registry for CLP.

Methods: A total of 93 children participated, 41 with bilateral CLP and 52 with unilateral CLP. Data in the registry on percent oral consonants correct (POCC), percent non-oral errors, and perceived velopharyngeal competence (VPC) were compared with reassessments of audio recordings by four independent judges. Inter judge agreement of percent consonants correct (PCC) and the reliability of three speech related quality indicators was also assessed. Agreement was calculated by the intraclass correlation coefficient (ICC), quadratic weighted kappa, and percentage agreement point by point.

Results: Agreement between data in the CLP registry and the reassessments was excellent for POCC, (single measures ICC 0.94; CI 0.91-0.95), fair for non-oral errors (single measures ICC 0.79; CI 0.74-0.85); and good for VPC (kappa values between 0.66 and 0.75). Inter-judge agreement was good for PCC (single measures ICC 0.85; CI 0.79-0.89). For the quality indicator proportion of children without non-oral speech errors, exact agreement between registry data and the SLPs varied between 90 and 95%, and for proportion of children with competent or marginally incompetent velopharyngeal function, between 89 and 93%. For proportion of children with PCC >86%, agreement between judges was 72% when compared at the same time.

Summary/Conclusion: Registry data on POCC and VPC seem reliable, as well as the quality indicators proportion of children without non-oral speech errors and proportion of children with competent or marginally incompetent velopharyngeal function. The quality indicator proportion of children with PCC >86% needs further development, in order to strengthen the reliability, before open comparison of results between centres can be performed.
Hearing loss in children with orofacial clefts before age 3 years
E Gallagher1, D Wu2, E Christianson3, P Formsma3, B Siebold4, K Sie5
1Pediatrics, University of Washington, Seattle Children's Hospital, 2School of Medicine, University of Washington, 3Audiology, Seattle Children's Hospital, 4Seattle Children's Research Institute, 5Otolaryngology, University of Washington, Seattle Children's Hospital, Seattle, United States

Background: Numerous studies have shown that children with orofacial clefts have lower academic outcomes compared to unaffected peers. Many potential factors likely contribute to these observed differences, one of which is hearing loss. Children with clefts that involve the palate often develop serous otitis media and conductive hearing loss, yet the range of severity of hearing loss in this population before palate repair has not been reported previously.

Aims: The primary objective of this study was to describe the degree of hearing loss in a large sample of children with cleft palate before and after palate repair. A secondary objective was to examine the number of audiology assessments in this patient population.

Methods: We conducted a retrospective review of patients with cleft palate (with or without cleft lip) who received palatoplasty at an academic medical center. At this institution, tympanostomy tubes are typically placed for the first time with the palate repair, around 12 months of age. Degree of hearing loss was assessed before and after palate repair, until age 3 years. We included results from brainstem auditory evoked response (BAER) or behavioral pure tone audiometry tests. The number of audiology tests per patient and test-re-test reliability were also examined.

Results: The study included 503 patients born between 2008 and 2015 with clefts that involved the palate. Of these patients, 61 received a BAER at an average age of 2.7 months, with 49.2% demonstrating mild hearing loss and 11.5% showing moderate or greater hearing loss at this age (remainder of tests were normal). Behavioral audiograms were completed in 259 patients before palate repair, at an average age of 11.0 months, with 32.0% showing mild hearing loss and 30.5% showing moderate or greater hearing loss. Pre- and post-palate repair audiograms were completed in 151 patients, and 92.7% of patients had improvement in hearing after palate repair. Among patients who completed behavioral audiograms after palate repair, many required more than one test to obtain a valid result (average 2.2 tests), and test-re-test reliability improved after 10 months of age.

Summary/Conclusion: In this large sample of patients with clefts that involved the palate, the number of audiology tests per patient was high, and nearly one-third developed moderate or greater hearing loss prior to palate repair. The impact of this degree of hearing loss on academic outcomes has not been clearly established. In many craniofacial centers, hearing loss is not treated before palate repair and tympanostomy tube placement. Given the range of severity of hearing loss during the first year of life, further studies are recommended to determine whether interventions to improve hearing during this time may result in improved academic outcomes in this patient population.
Is there a relationship between conductive hearing loss and cleft speech characteristics in children with cleft palate
S Baker¹, Y Wren², F Cooper³, Z Fei³, H Extence¹
¹The Welsh Centre for Cleft Lip & Palate, Abertawe Bro Morgannwg University Health Board, Swansea, ²Cleft Collective
UK, University of Bristol, Bristol, ³Cardiff School of Health & Sports Sciences, Cardiff Metropolitan University, Cardiff, United
Kingdom

Background: Children with cleft palate are at high risk of developing both conductive hearing loss (Flynn et al., 2009) and
cleft speech characteristics (CSCs) (CSAG, 2001). Whilst there is evidence that a conductive hearing loss can affect speech
perception skills (Zumach et al., 2010) there is limited research to understand whether hearing loss specifically affects
speech development alongside a cleft palate resulting in CSCs. At present there is inconclusive evidence regarding the
management of hearing loss in children with cleft palate (Ponduri et al., 2009). It is important to ascertain whether hearing
affects the development of CSCs to be able to measure whether management for hearing loss modifies the speech outcome
for these children. This study used data from the Cleft Collective. This is a national study in the UK collecting information
from families of children born with a cleft lip +/- cleft palate. The study was designed to answer parents’ key questions, as
identified by research from the James Lind Alliance (2012). There is a speech and language study nested within this. Cleft
centres across the UK collect data related to speech and language development from those recruited to the study (Wren et
al., 2018).

Aims: This study used data from the Cleft Collective Speech and Language (CC-SL) study and examined whether there
was a relationship between those with a history of diagnosed hearing loss and presence of CSCs in children with cleft palate
(+/- cleft lip) aged 18-24 months.

Methods: Speech and Language Therapists (SLTs) across the UK completed standardised assessment forms for those
recruited to the CC-SL study. They reported history of diagnosed hearing loss, intervention provided for hearing loss, and
presence of CSCs. A consonant inventory for each participant was also completed. In total, 123 participants were included
in this study. Statistical analysis of relationships between hearing loss and CSCs was completed. Analysis of the consonant
inventories was also undertaken to give further information related to speech acquisition.

Results: There was a statistically significant relationship between history of diagnosed hearing loss and presence of CSCs
(p <0.05). Further analysis of confounders, including whether grommets or hearing aids have a modification effect, is
ongoing and will be reported.

Analysis of the consonant inventories highlighted that children with diagnosed hearing loss used a reduced number of oral
consonants compared to those with normal hearing. Fewer children used alveolar consonants in the hearing loss group
than those with normal hearing which was statistically significant (p<0.05).

Summary/Conclusion: This study provides some evidence that a conductive hearing loss can affect the development of
speech consonants in children with cleft palate resulting in CSCs. Therefore, these children should be prioritised by SLT
services for close observation and early intervention. These children should also be closely monitored by audiology to
ensure that hearing is optimised for speech and language development. Continued data collection will provide additional
evidence regarding the impact of how this hearing loss is managed. Further research using CC-SL data at age 3:0 would
provide additional evidence regarding the longer-term impact of hearing loss on CSCs.
Marking of phoneme boundary in cleft palate speech
I Thammaiah
Department of speech science, JSS institute of Speech and Hearing, Mysore, India

Background: In cleft lip and palate speech, phonemic representation has been tested in the well known phonetic dimension of voice onset time or VOT. VOT is one of the strongest phonetic cues for consonant discrimination and refers to the time interval between the release of the articulatory occlusion and the onset of vocal-fold vibration.

Aims: The aim of the present study was to measure the VOT continuum and mark the occurrence of phoneme boundary in speech of children with cleft lip and palate and typically developing children.

Methods: The speech sample obtained from individuals with cleft lip and palate were collected and synthesised further using VOT continuum technique for voiced and voiceless stop consonants namely bilabial sounds (p, b), palatal sounds (t, d) and velar sounds (k, g). VOT truncation at +/- 10msec, +/- 30 msec was done. The synthesized stimulus was presented to 15 trained and untrained listeners. The participants had to indicate the percentage of phoneme crossing

Results: Results suggested a significant identifiable pattern of upper phonemic boundary and Lower phonemic boundary in CLP speech.

Summary/Conclusion: The present study can further applied for phonetic structure marking for cleft lip and palate individuals.
OS 40.8
Canonical babbling and early consonant production in Swedish infants born with unilateral cleft lip and palate treated with early soft palate closure in a two-stage palate repair procedure
E Hagberg1, J Nyberg2, A Lohmander3
1Functional Area Speech and Language Pathology and Stockholms Craniofacial Team, Karolinska University Hospital, Stockholm, Sweden, 2Division of Speech and Language Pathology, CLINTEC, Karolinska Institute, Functional Area Speech and Language Pathology and Stockholms Craniofacial Team, Karolinska University Hospital, 3Division of Speech and Language Pathology, CLINTEC, Karolinska Institute, Stockholm, Sweden

Background: Occurrence of canonical babbling at 10 months, and frequency and complexity in early consonant production is important markers in the typical speech and language development. The consonants used in babbling has shown to be phonetically similar to the sounds in the child’s early words. There are indications that infants with unoperated cleft palate have lower occurrence of canonical babbling and high pressure consonants than infants who had early palate surgery, even if only the soft palate was repaired. More data on the impact of surgical method and timing of palate repair on babbling development is needed since prevention of atypical babbling may have an impact on later speech development. This is beneficial since between 30-50% of the children born with cleft lip and palate are still reported to have speech difficulties at 5 years of age.

Aims: To investigate the presence of canonical babbling and aspects of the consonant production in Swedish infants born with unilateral cleft lip and palate (UCLP) treated with early soft palate closure in a two-stage palate repair procedure.

Methods: A consecutive series of 27 children born with UCLP between 2014-2016, treated at Stockholm Craniofacial Team, Karolinska University Hospital, were included. Due to additional malformations and missing data, 22 children at 10 months and 23 children at 18 months were included. Lip and soft palate closure were performed at 5-9 months (mean age 6 months). None had undergone hard palate closure. Data were collected using standardized babbling observation at 10 and 18 months of age by the team speech and language pathologist during a 30-45 minutes interaction between the child and a parent at the clinic. Presence of the valid variables canonical babbling, high pressure consonants/oral stops, and dental/alveolar stops were noted at 10 months and oral stops, dental/alveolar stops and the consonant inventory at 18 months of age.

Results: According to the observation at 10 months, 19 children had canonical babbling (86%). Nine (41%) had high pressure consonants/oral stops and three of these (14%) had dental/alveolar stops. At 18 months, 19 children (83%) had oral stops and 11 (50%) had dental/alveolar stops. The number of different consonants at 18 months varied between 1 to 9 (mean 5). The three children without canonical babbling at 10 months did not have dental/alveolar stops at 18 months and exhibited lower number of different consonants than the group mean.

Summary/Conclusion: Almost all the children had canonical babbling at 10 months of age. Most children had oral stops at 18 months, and half of them with dental/alveolar placement. The variation in number of different consonants present at 18 months of age raises questions about impact on future speech and language development which will be investigated longitudinally and also compared with a Swedish subgroup included in the ScandCleft project.
Speech disorders in cleft patients. A new classification
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

Background: Speech disorders in the fractured patient despite their reconstruction are usually present to a greater or lesser degree. In general, Velopharyngeal insufficiency has been referred to as the most common disorder in patients with this condition, in other occasions reference is made to a bad position or poor muscular function of the elevator, that many of us call velopharyngeal incompetence. These two disorders are defined as an altered anatomical and functional feature, until then the diagnosis suggests what the disorder is and at the same time what its treatment would be. Other speech disorders are no longer defined by their functional anatomical alteration but by their sound characteristics such as Hyper nasality, hypo nasality, glottic strokes, rubs, compensatory articulation, rotacism, etc. already without following a logical anatomofunctional pattern. All this leads to confusion to better understand the speech disorders of the fissured patient.

Aims: The objective of this work is to look for a classification based on anatomopathological disorders that affect the alteration of speech at different levels of modulation of the same, which at the same time indicate which is the pathology and simultaneously which would be the appropriate treatment. All this would achieve to unify concepts, clarify the pathology and prescribe better treatments depending on each disorder.

Methods: It has been determined on the experience of Fonoaudiólogas and based on the literature on sounds in speech and its modulation at different levels of the oropharyngeal tract to segment the pathology of speech in the fissure and this allows grouping seven speech pathologies in fissured patients: A VELOFARINGEO LEVEL. - Its poor production can be due to two pathologies very well known by all of us: VELOFARINGEA INSUFFICIENCY, VELOFARINGEA INCOMPETENCE AT DENTOPALATINO LEVEL. - The alteration of the sounds due to their cause should be called as DENTOPALATIN INSUFFICIENCY, DENTOPALATIN INCOMPETENCE A NASOLABIAL LEVEL. - Its inadequate structure will produce disorders such as: NASOLABIAL INSUFFICIENCY and NASOLABIAL INCOMPETENCE. Finally the consequence of speech disorders not corrected in time ends in a vicious alteration that seeks to compensate for the defect as the compensatory articulation, which in this classification scheme we would call it HYPERCOMPETENCE either Velofaringea, dentopalatina or naso labial, depending on the disorder that gave its origin.

Results: The scheme proposed in this work, has been put into practice among Plastic Surgeons, Residents, Language Therapists and other volunteers of Operation Smile Ecuador, Peru, Paraguay, Morocco, which has allowed a better understanding, classification of disorders of the specks with a classification based on anatomofunctional disorders.

Summary/Conclusion: The need for a better understanding of the speech disorders that affect the fissured patient, before and after the corrective surgeries, has motivated us to look for standard parameters and a grouping of the sound disorders as a characteristic of a deficiency at different levels of the oral tract. achieving better understanding of the pathology, and searching for the best adapted therapy for each case. On the other hand, this distinction will allow more extensive, specific statistical studies and achieve a greater involvement of professionals in the understanding and treatment of speech disorders.
OS 40.10

Speech in 5-year-olds born with unilateral cleft lip and palate – a prospective swedish intercentre study

K Klintö1, K Brunnegård2, C Havstam3, M Appelqvist4, E Hagberg5, A Taleman6, A Lohmander7

1Skåne University Hospital, Malmö, 2Umeå University, Umeå, 3Sahlgrenska University Hospital, Gothenburg, 4Uppsala University Hospital, Uppsala, 5Karolinska University Hospital, Stockholm, 6Norrköping kommun, Norrköping, 7Karolinska Institutet, Stockholm, Sweden

Background: Studies on the impact of cleft palate surgery on speech with stringent methodology are called for, since we still do not know the best timing or the best method for surgery.

Aims: The purpose was to report on speech outcome for all Swedish-speaking 5-year-olds born with a non-syndromic unilateral cleft lip and palate (UCLP), in 2008 to 2010, treated at the six different cleft palate centres, and to compare speech outcomes between centres.

Methods: Informed consent was obtained. The speech of the 57 children was assessed with percent consonants correct adjusted for age (PCC-A), based on phonetic transcriptions from audio recordings by five independent judges. Also, hypernasality and perceived velopharyngeal function were assessed.

Results: The median PCC-A for all children was 93.9, and medians in the different groups varied from 89.9-96.8. In the total group, 36 children (63%) had no hypernasality, 12 children (21%) mild, 7 children (12%) moderate, and two children (4%) severe hypernasality. Twenty-two children (38.5%) were perceived as having competent/sufficient velopharyngeal function, 25 (44%) as having marginally incompetent/insufficient velopharyngeal function, and 10 children (17.5%) as having incompetent/insufficient velopharyngeal function. No significant differences among the six groups, with eight to ten children in each group, were found.

Summary/Conclusion: The results were similar to those in other studies on speech of children with UCLP, but poorer than results in normative data of Swedish-speaking 5-year-olds without UCLP. Indications of differences in frequency of surgical treatment and speech treatment were observed.
Robinson Sequence

OS 41.1
The breadth of feeding problems and feeding experiences in infants with robin sequence
K Evans

Infants with Robin sequence (RS) can experience a variety of feeding problems. We will review the multiple factors that can contribute to feeding problems in infants with RS, including glossoptosis (posteriorly positioned tongue), oropharyngeal dysfunction and weak sucking mechanics. Upper airway obstruction can lead to disordered suck swallow breath, and the concomitant presence of a cleft palate introduces challenges with nasal regurgitation, suction and flow. Primary swallowing dysfunction leading to aspiration can further compromise respiratory sufficiency in the infant with RS. As the etiopathogenesis of RS is heterogeneous, the presentation and functional consequences of feeding and swallowing issues vary and can be complex. Multiple tests and investigations designed to evaluate the quality of feeding and the feeding experience for infants and caregivers have been described, and we will discuss applications to this population. A multidisciplinary approach that includes experts in feeding, swallowing, breathing, nutrition and neurodevelopment is essential. In this presentation, we will review the science behind feeding problems in infants Robin sequence. We will discuss the available tools to assess feeding and swallowing in this population. As we discuss possible treatment algorithms to address feeding in RS, we will highlight the feeding and nutritional metrics that can be collected to understand and assess feeding outcomes for children with Robin sequence.
Pierre robin sequence (prs) in a centralised network in the uk: report on the incidence and outcome in these patients.

S Van Eeden¹, V Beale², M Bowden², H McClements¹, S McMahon¹, D Phare², H Robson²

¹Cleft, Alder Hey Children's Hospital, Liverpool, ²Cleft, Royal Manchester Children's Hospital, Manchester, United Kingdom

Background: Babies born with Pierre Robin Sequence (PRS) are a low incidence high need group of patients. Centrality of care in the UK has given rise to higher numbers of patients presenting for treatment in each network meaning that meaningful audit on these patients can be carried out.

Aims: To understand the incidence of PRS in a centralised regional service and the outcomes for these patients.

Methods: Retrospective case note review of all babies born with a cleft palate between 2007 and 2016 in a centralised regional service in the UK to identify those with PRS and to review their outcomes.

Results: 701 babies born with a cleft palate were identified of which 159 (23%) were designated as having PRS as defined as those with airway obstruction, micrognathia and glossoptosis. Gestational age, birth weight, hospital stay, airway management, feeding, timing of surgery, speech outcome and development will be reported.

Summary/Conclusion: The incidence of PRS in this cohort is higher than that reported in the literature. Airway management appears to differ significantly from that reported previously in the literature. Diagnosis of PRS however is not an accurate science and over-reporting may account for the high incidence in this study and for the airway management outcomes.
Background: Pierre Robin Sequence (PRS) is a congenital deformity characterized by microgenia, mandibular retrognathia and glossoptosis and possibly cleft palate. Consequently, the children suffer from upper airway obstruction, which results in intermittent hypoxia, disturbed sleep and failure to thrive. In the literature, the neurologic development is therefore often described as impaired.

Aims: We assumed that children treated with the preepiglottic baton plate do not suffer from a relevant oxygen deficiency and therefore not from an impaired neurologic development. In the present prospective monocentric survey study we compared the neurologic development of children with PRS and cleft palate with age-matched children with isolated cleft palate (CP).

Methods: We included children aged 5 to 6 years with isolated cleft palate and with PRS and cleft palate. Cleft palate closure was performed until the age of 18 months. All PRS children were treated with the preepiglottic baton plate. We excluded children with further diseases, associated syndromes and children that were treated longer than 1 year with the preepiglottic baton plate. The neuropsychological function was assessed with the WPPSI-III (Wechsler Preschool and Primary Scale of Intelligence) and K-ABC-SED (Kaufman Assessment Battery for Children - Skala einheitlichen Denkens).

Results: From 2015 until 2018 forty-four consecutive children (22 PRS, 22 CP) were included. Mean age was 72.9 months (± 8.0). All children showed an age adequate physical and neurologic development within the test specific reference values. Furthermore, we did not observe any neurologic or cognitive developmental delays in the PRS group.

Summary/Conclusion: The present results support the hypothesis that PRS children can be treated successfully with the preepiglottic baton plate. PRS children treated with this technique show a normal physical and neurologic development. More invasive interventions like tracheostomy, mandibular distraction or glossoptosis can be avoided.
OS 41.4
Surgical and long-term speech outcomes of cleft palate repair in patients with robin sequence versus isolated cleft palate.
B Logjes¹, S Upton², B Mendelsohn³, C Breugem⁴, W Hoffman⁵, J Pomerantz²
¹Plastic and Reconstructive Surgery, University California San Francisco Medical Center, United States & University Medical Center, Utrecht, Netherlands, ²Speech and Language Pathology Craniofacial Center, University California San Francisco Medical Center, San Francisco, United States, ³Pediatrics & Medical Genetics and Genomics, University California San Francisco Medical Center, San Francisco, United States, ⁴Plastic and Reconstructive Surgery, University Medical Center, Utrecht, Netherlands, ⁵Plastic and Reconstructive Surgery, University California San Francisco Medical Center, San Francisco, United States

Background: The Robin sequence (RS) associated cleft palate (CP) differs from isolated-CP (ICP) in terms of cleft anatomy, timing of repair, and other patient characteristics.
Aims: To compare surgical and long-term speech outcomes and identify outcome predictors.
Methods: All consecutive RS-patients (1990-2016) were retrospectively reviewed and compared to ICP-patients. Surgical protocol included straight line repair with intravelar veloplasty (SLIV) or Furlow repair depending on airway and CP-characteristics. Outcomes included oronasal fistula (ONF), velopharyngeal insufficiency (VPI), speech operations, cleft-speech characteristics, and sleep apnea.
Results: Seventy-five RS- and 83 ICP-patients underwent CP-repair, mean age 13.7 (SD 5.3) and 11.3 (SD 5.1) months, respectively (p = 0.004). RS-patients had wider and more severe CP’s (p = 0.001). Syndromic RS was identified in 55%. Fistula occurred in 5% RS vs. 0% ICP (p = 0.049). Speech evaluation was available in 44 RS- and 47 ICP-patients, median follow-up of 8.2 years. Velopharyngeal insufficiency (VPI) occurred in 41% RS vs. 17% ICP (p = 0.012) and in 60% syndromic RS vs. 16% isolated RS (p = 0.005). Sixteen RS- vs. 4 ICP-patients had secondary Z-plasty, and 3 RS- vs. 1 ICP-patients underwent a third speech operation to resolve VPI. Severe respiratory distress in RS was not associated with VPI (p = 0.307). Sleep apnea occurred in 17% RS vs. 2% ICP-patients (p = 0.031).
Summary/Conclusion: This CP-protocol adjusts timing and repair type based on individual patient characteristics, which included wider and more severe CP-anatomy, and airway compromise in RS. Our data indicate that this is safe from an airway standpoint. A more severe CP-anatomy was not associated with VPI. Isolated-RS demonstrated similar VPI rates compared to ICP. Syndromic RS-patients should be expected to attain similar long-term speech outcomes but with higher rates of secondary Z-plasty to resolve VPI. Obstructive speech operations such as sphincter pharyngoplasty or pharyngeal flap can be avoided while achieving normal VP function in RS patients.
National review of early management and speech outcomes in children with robin sequence (uk)

J Maraka¹, M Fullarton², F Kapasi³, N Brierley⁴, S Martin⁵, P Hodgkinson¹, D Sainsbury¹, J Miller⁶, S Van Eeden⁶, C Collaborative⁷

¹Plastic Surgery, The Royal Victoria Infirmary, Newcastle, ²Maxillofacial Surgery, Alder Hey Children's Hospital, Liverpool, ³Maxillofacial Surgery, Oxford University Hospitals, Oxford, ⁴Plastic Surgery, Leeds Teaching Hospitals NHS Trust, Leeds, ⁵Plastic Surgery, Royal Belfast Hospital for Sick Children, Belfast, ⁶Cleft Speech and Language Therapy, The Royal Victoria Infirmary, ⁷Cleft Mdt Collaborative, Newcastle, United Kingdom

Background: Children born with Robin Sequence (RS) and cleft palate have been reported to be at higher risk of speech difficulties. However, there is very little longitudinal data in the literature with regard to their speech and language development and its relationship to early management.

Aims: The aim of this project is to gather data from a number of different centres in the United Kingdom (UK) to provide a review of longitudinal speech and language outcomes in RS with cleft palate.

Methods: As part of a Cleft Multidisciplinary Collaborative (CMC) project data is being collected nationwide in the UK on a group of patients with RS and cleft palate born between 1st January 2005 - 31st December 2009. Data will include the following:

- Basic demographics
- Airway management
- Feeding management
- Speech outcomes at 18m, 3y, 5y and 10y
- Language outcomes at 18m and 3y

Results: Data collection is ongoing but results will provide a descriptive overview of speech and language outcomes in patients with RS. These will be compared against national data standards in the UK where they exist. Speech and language data will be compared to early airway and feeding data.

Summary/Conclusion: This is the first longitudinal overview of speech and language outcomes in children born with RS across the UK. Children with RS are at high risk of persistent speech and language problems. The impact of this warrants more research as does investigation into possible aetiologies and comorbidities. Speech and language is a key outcome measure for any child with RS and cleft palate. They require close monitoring by specialist speech and language therapists (SLTs) throughout their childhood.
Long-term outcomes of cleft palate repair in Pierre Robin sequence: a case control study

M Schwaiger¹, H Cook², S Edmondson², K Echlin², D Atherton², P Haers², N Timoney², L Ferguson²
¹South Thames Cleft Service, Department of OMFS, Medical University of Graz, Austria, Guy’s and St. Thomas’ Hospital, Department of OMFS, Medical University of Graz, Austria, ²South Thames Cleft Service, Guy’s and St. Thomas’ Hospital, London, United Kingdom

Background: Pierre Robin Sequence (PRS) is a congenital condition, consisting of micrognathia, glossoptosis and upper airway obstruction. In up to 90.4 percent of subjects, diagnosed with PRS, palatal clefting can be found. The overall incidence of PRS in Northern Europe is reported to be between 7.1 and 12.4 per 100,000 live births. The majority of published PRS studies in the current literature have focused on the emergent management of this patient cohort during the neonatal period. However, in recent years, there has been increasing interest in the evaluation of long-term outcomes in patients with PRS, post palatoplasty, in terms of well-defined, standardised clinical and functional parameters. Despite this, such data remains limited.

Aims: The aim of our study was to assess patients with Pierre Robin Sequence in comparison with a matched cleft palate-only patient cohort, treated in a single institution during the years 2005-2012, in terms of the National Speech outcome measures in the United Kingdom, the oronasal fistula rate and the rate of secondary speech surgery.

Methods: 64 Patients with a cleft palate, diagnosed with Pierre Robin Sequence (1. airway obstruction, 2. micrognathia and 3. glossoptosis), who were treated within the South Thames Cleft Service for primary palatoplasty from 2005 to 2012, were included in this study. Outcome measures were: 1. 5-year audit speech assessment; 2. postoperative oronasal fistula rate and 3. rate of secondary speech surgery. Speech was assessed at the routine 5-year audit point using the GOS.SP.ASS (Great Ormond Street Speech Assessment) and then analyzed using the Cleft Audit Protocol for Speech-Augmented (CAPS-A). The outcomes in the PRS cohort were compared to a Cleft Palate only (CPO) cohort.

Results: 53 PRS patients were included in the final analysis. Intravelar veloplasty was the standard palatoplasty technique used. According to the CAPS-A grading system (dark green, light green, amber and red), significant differences were found between the PRS cohort and the CPO (Double green: 47% PRS vs 80% CPO). Ongoing hypernasality was found in 37% in the PRS group and in 13% in the CPO cohort. Secondary speech surgery was significantly more frequent in the PRS group (31% PRS vs. 10% CPO). There were no differences in rates of postoperative fistulae occurrence.

Summary/Conclusion: Patients with PRS were found to have significantly worse speech outcomes at age 5 review, when compared to a CPO cohort, treated in the same institution, during the same time period. Additionally, a higher rate of secondary speech surgery was found in the PRS group.
OS 41.7

Temporomandibular joint space and condyle morphology in robin sequence before and after mandibular distraction osteogenesis

A Singh1, A T Wilson1, K Bruckman1, C Resnick2, D Steinbacher1

1Department of Surgery, Section of Plastic Surgery, Yale University School of Medicine, New Haven, CT, 2Department of Oral and Maxillofacial Surgery, Boston Children’s Hospital, Boston, MA, United States

Background: Changes in temporomandibular joint (TMJ) space and condyle morphology associated with mandibular distraction osteogenesis (MDO) in infants with Robin sequence (RS) are unknown. Complications attributed to MDO such as mouth opening limitation and TMJ ankylosis may be impacted by changes in TMJ anatomy.

Aims: The purpose of this study was to characterize early morphologic changes in TMJ space and condyle morphology from computed tomography (CT) scans before and after MDO in infants with RS.

Methods: This is a retrospective, cross-sectional analysis of infants with RS who underwent MDO. Pre- and postoperative 3-dimensional CT scans were analyzed in 3-matic and Mimics software (Materialise). The boundaries of the “TMJ space” were defined by planes connecting the articulating surfaces of the condyle and fossa. The primary plane was placed such that it divided the anterior articulating surface from the posterior articulating surface, thus occupying the superior joint space (SS). Rotation of the SS plane 20° anteriorly and 20° posteriorly resulted in anterior joint space (AS) and posterior joint space (PS) planes, respectively. Euclidean distances were measured at five intervals along each plane, from the lateral to medial pole of the condyle. Separately, the surface area and volume of each condylar head was measured and expressed as a ratio compared to the entire mandibular surface area/volume. Differences in TMJ space, condyle surface area, and condyle volume were assessed using Wilcoxon signed-rank test. Statistical significance was set at p<0.05.

Results: The study sample included 14 patients (28 sides). Pre- and postoperative CTs were obtained at mean ages 7.9±10.8 and 23.7±12.4 weeks of life, respectively. Distraction was performed across an oblique osteotomy in the bilateral mandibular bodies using a horizontal vector. From pre- to postoperative images, the TMJ space increased at the medial boundary of each joint space plane: SS (0.8336 mm, 95% CI 0.1556 to 1.5754 mm; p<0.002), AS (0.87825 mm, 95% CI 0.3292 to 1.5756 mm; p<0.001), and PS (0.60015 mm, 95% CI 0.3046 to 1.36 mm; p<0.007). There was a postoperative decrease in condylar surface area ratios (-0.3914286 mm², 95% CI -0.5379537 to -0.2449034; p<0.001), but no change in condylar volume ratios. Intragroup analyses showed no differences between left and right sides.

Summary/Conclusion: There is a significant increase in temporomandibular joint space at the medial aspect of the joint and a decrease in condylar surface area, but no significant change in condylar volume after MDO in infants with RS. Further analysis of age-matched controls is needed to determine the role of MDO in these morphologic changes.
**OS 41.8**

**Highlighting feeding disorders as a predictor factor of a functional approach in robin sequence: a thirty-year single center experience**

F Blanc¹, O Plan², I Harrewijn², M Bigorre¹, G Captier¹

¹Service of Pediatric Orthopedic Plastic Surgery, Lapeyronie Hospital & University of Montpellier, ²Department of Neonatal Medicine and Pediatric Intensive Care, Arnaud de Villeneuve Hospital & University of Montpellier, Montpellier, France

**Background:** The Pierre Robin sequence (PRS) associates retrognathia, glossoptosis, and a cleft palate. It can lead to respiratory and feeding disorders due to upper airway obstruction and sucking–swallowing incoordination. Although declining, the mortality rate for children with PRS is up to 16.6% according to latest reports.

**Aims:** The aim of this study was to describe the evolution and outcomes of the initial treatment of children with PRS in a tertiary single center. Indication and results of the use nasopharyngeal tube are emphasized. We also analyzed the predictive factors of the severity of respiratory obstruction.

**Methods:** Retrospective single-center study of 138 children with PRS consecutively followed in the tertiary Hospital of Lapeyronie & University of Montpellier between 1986 and 2017. We used the nasopharyngeal tube since 2001. Group 1 comprises children without specific respiratory management, group 2 children requiring prone positioning, and finally, group 3 children requiring nasopharyngeal tube or non-conservative surgical treatment.

**Results:** 138 children were included. 46% (n=63) belong to Group 1, 36% (n=50) to Group 2 and 18% (n=25) to Group 3. In this last group, the floor of mouth release was required for 7 children, cephalic traction for 3, and use of a nasopharyngeal tube for 18. No child required tracheostomy in our center for PRS since 1986. Pre-term birth, birth weight, or associated congenital malformations were not significantly different between the three groups. However the age of oral exclusive alimentation was significantly different : 1 day (quartiles : 0-3) for Group 1 ; 11 days (quartiles : 1 - 28) for Group 2 ; 39 days (quartiles : 19 - 111) for Group 3 (p<0.0001). This data seems to confirm the associated risk of severe respiratory treatments for children facing severe neo-natal swallowing disorder.

**Summary/Conclusion:** This study describes the evolution of the initial conservative and non-conservative treatments of children with PRS since 1987 in a single center. In our experience and through the use of the nasopharyngeal tube, surgical intervention is necessary for a limited number of children. Invasive therapy is associated with initial swallowing disorder requiring nasogastric tube and/or gastrostomy. Thus, nasopharyngeal tube seems promising in PRS. Prospective studies are required to confirm the indications and the outcomes of the nasopharyngeal tube in PRS.
OS 41.9

Improvements in oral feeding after mandibular distraction for pierre robin sequence

A Kaye¹, M Foster², M Tracy²
¹Plastic Surgery, ²Plastic Surgery Research, Children’s Mercy Kansas City, Kansas City, Missouri, United States

Background: Pierre Robin Sequence (PRS) consists of the triad of micrognathia, glossoptosis, and airway obstruction. Associated feeding difficulties are common and infants are at risk for failure to thrive. Mandibular distraction is now a well-accepted technique for treatment of infants with PRS and moderate to severe airway obstruction. With proper patient selection relief of airway obstruction is highly successful. Mandibular distraction also frequently helps improve feeding, but it is unclear when normal feeding resumes. Some patients will still require gastrostomy tube feedings post-operatively.

Aims: This study aims to identify the relationship of pre- and post-operative feeding trends in infants with PRS treated with mandibular distraction.

Methods: This study was performed as a retrospective review of patients with PRS treated with mandibular distraction from 2010–2016 to evaluate pre- and post-operative feeding and need for G-tube supplementation.

Results: There were 42 infants identified who underwent mandibular distraction, 36 of whom (85.7%) had surgery before 5 months of age. For these 36, mean age at distraction was 26.2 days (SD 29.8) with a mean in-hospital post-operative stay of 23.6 days (SD 16.7). Prior to surgery, 23 patients (58.3%) did not tolerate any oral feeds, 11 tolerated some oral feeds, and only 2 (5.6%) were taking full oral feeds. After distraction, 55.6% were taking full oral feeds prior to hospital discharge. Ten patients (27.8%) ultimately required G-tubes at discharge for feeding. Of those taking partial oral feedings prior to surgery, only 2 went on to require G-tube feedings after distraction. The remainder were feeding 100% orally prior to discharge. Of those infants with no pre-operative oral intake 9/23 improved to 100%, 5/23 improved <100%, and 9 did not change. For those not feeding prior to distraction the mean time to full feeds was 18.9 days (SD 11.9) compared to 10.55 days (SD 7.2) for those with only partial oral feeds. Mean age at surgery for those successfully feeding prior to discharge was 30 days (SD 37.9) versus 21.6 days (SD 11.2) for those requiring supplementation. There was no significant difference between pre-operative weight, z-score, or presence of a syndrome for feeders and non-feeders. Accounting for the 6 infants older than 5 months age at time of distraction, none were tolerating oral feeding prior to surgery and all 6 required G-tubes for feeding at discharge.

Summary/Conclusion: Neonates having mandibular distraction for PRS-related airway obstruction can be anticipated to have improvements in feeding after surgery. More than half were able to advance to full oral feedings prior to discharge. Almost 28%, however, required G-tube placement for long-term feeding supplementation. Patients with at least some ability to take oral feedings prior to distraction almost always advanced to full feedings prior to discharge. Those without ability to tolerate oral feedings prior to surgery are more likely to require G-tube supplementation at the time of discharge.
Speech outcomes of tongue lip adhesion in patients with robin sequence: a matched case-control study
J Mermans1, R Logjes12, M Coerts3, C Breugem2, J Don Griot1
1Plastic-, Reconstructive-, and Handsurgery, Amsterdam UMC, Amsterdam, 2Plastic-, Reconstructive-, and Handsurgery, UMCU, Utrecht, 3Department of Speech and Language Pathology, Amsterdam UMC, Amsterdam, Netherlands

Background: Tongue-lip adhesion (TLA) is commonly used as a surgical treatment for upper airway obstruction (UAO) in patients with Robin sequence (RS) if conservative therapy fails. The effect on speech outcomes after TLA and subsequently cleft palate (CP) repair in patients with RS is insufficiently investigated.

Aims: The aim of this study is to assess the speech outcomes in RS patients who underwent TLA and subsequently CP repair and to compare these results with a matched case control group of isolated cleft palate (ICP) patients. This abstract presents the preliminary results of our study.

Methods: All consecutive RS-patients (1995-2014) who underwent CP-repair at the Amsterdam UMC (Vrije Universiteit Amsterdam) were retrospectively reviewed. The control group of ICP patients was matched on CP-type with the RS patients. Complete CP-closure using the Von Langenbeck technique with intravelar veloplasty was performed around 9 months of age. If involvement of the anterior hard palate and complete closure was not achievable in one stage, a separate closure was performed at a later age. TLA in RS patients with severe UAO is comprehensively described by Bijnen et al. Variables analyzed were sex, CP-type, age at TLA, associated syndromes with RS, and age at CP repair. The multidisciplinary cleft team of our center assessed the binary speech outcomes at around 4 years of age when the normal acquisition of speech and languages is completely developed. These outcomes included: Secondary speech operation, diagnosis of velopharyngeal insufficiency (VPI), hypernasality, and articulation errors (including: 1. nasal emissions, 2. glottal-pharyngeal realizations, 3. labial, alveolar and palatal disorders). In addition, open mouth posture and weakness of the lower lip musculature were assessed.

Results: A total of 43 RS patients (15 males and 28 females) underwent CP-repair (mean age: 9.7 months, SD± 2.6) and had sufficient speech follow-up, of which 24 (56%) RS patients had a TLA at a mean age of 27 days (SD±30). Cleft palate characteristics were: 28% soft palate only, 25% soft palate + partial hard palate, and 47% soft + hard palate. Nine RS patients needed closure of the anterior hard palate at median age of 7.6 years (range: 3.6-10.0 years). A total of 24 (57%) patients had syndromic RS versus 18 (43%) patients who had RS as an isolated entity. Median age at time of speech evaluation was 4.5 years (range: 3.3-6.2 years). VPI was diagnosed in 20 RS patients (55% TLA vs. 45% no TLA, p = 0.920) of which 19 (44%) RS patients underwent a pharyngeal flap for the treatment of VPI (mean age 4.5 years, SD±1.3).

Summary/Conclusion: Our data demonstrates that the need of a TLA in the neonatal period is not associated with VPI. Further conclusions/results regarding articulations errors and the comparison with the matched ICP group will be presented in June.
Assessment of health-related quality of life in robin sequence: a comparison of mandibular distraction osteogenesis and tongue-lip adhesion

B Logies¹, Y Mermans², E Paes¹, M Muradin³, P Don Griot², C Breugem¹
¹Plastic and Reconstructive Surgery, UMC Utrecht, Utrecht, ²Plastic and Reconstructive Surgery, Amsterdam UMC (Locatie VU), Amsterdam, ³Cranio-Maxillofacial Surgery, UMC Utrecht, Utrecht, Netherlands

Background: Numerous studies have proven the efficacy of mandibular distraction osteogenesis (MDO) or tongue-lip adhesion (TLA) in Robin sequence (RS) infants with upper airway obstruction. However, none have compared health-related quality of life (HRQoL) outcomes post-MDO to post-TLA.

Aims: To compare HRQoL outcomes post-MDO to post-TLA in the surgical treatment of Robin Sequence.

Methods: In the present retrospective study, RS-infants younger than 1-year-old, who underwent MDO or TLA, were included (2006–2016). The infants’ caregivers were asked to complete a questionnaire based on the Glasgow Children’s Benefit Inventory (GCBI).

Results: The response rate was 71% (22 of the 31 questionnaires, MDO 12/15 and TLA 10/16), median follow-up was 5.9 years (range:1.3–10.5 years), and median age at surgery was 24 days (range: 5–131 days). The median total GCBI-scores post-MDO and post-TLA were 21.9 (9.4 IQR) and 26.0 (37.5 IQR) respectively (p = 0.716), indicating an overall benefit in HRQoL from both surgical procedures. Positive changes were observed in all subgroups (emotion, physical health, learning, and vitality) for both MDO and TLA. In syndromic RS, both surgical procedures demonstrated a lower positive change in HRQoL compared to isolated RS (19.8 MDO and 16.7 TLA versus 24.0 MDO and 35.4 TLA, p = 0.303).

Summary/Conclusion: Both MDO and TLA, demonstrated an overall benefit in HRQoL-outcomes, with no significant differences between these two surgical techniques. Our findings contribute to the debate regarding the use of MDO versus TLA in the surgical treatment of RS; however, long-term outcome studies evaluating HRQoL in larger RS-cohorts are necessary to identify which procedure is likely to be best in each individual RS-infant.
Cleft Long-term Outcome (Growth)

OS 42.2
Long-term, longitudinal follow up of individuals with uclp after gothenburg two stage palate closure. A comparison between hard palate closure at 3 respectively 8 years of age – dental arch relation

M Najjar Chalien¹, H Mark², J Lilja², C Havstam³, S Rizell¹
¹University Clinics of Odontology, Dep of Orthodontics, University of Gothenburg, ²Department of Plastic Surgery, Sahlgrenska University Hospital, ³Department of Otorhinolaryngology, Division of Speech and Language Pathology, / Sahlgrenska University Hospital, Gothenburg, Sweden

Background: Between 1975 and 1993 delayed hard palate closure was practiced in unilateral cleft lip and palate (UCLP) in Gothenburg, closing the soft palate at 6 months and hard palate at 8 years. A long term followed-up series has previously been evaluated regarding growth, occlusion, speech and appearance which demonstrated good results. However, a high degree of retracted oral articulation was present due to the residual cleft. Therefore a modified two stage palate closure was introduced in 1993 including soft palate closure at 6 months and hard palate closure at 3 years of age.

Aims: The aim was to longitudinally compare dental arch relationship in children born with UCLP who had hard palate closure at 3 (HPC3) respectively 8 (HPC8) years of age

Methods: Twenty-eight individuals born with UCLP who were consecutively operated according to Gothenburg protocol of two stage palatal closure with soft palate at 6 months and hard palate at 3 years were compared with 59 individuals born with UCLP who had two stage palatal closure with soft palate closure at 6 months and hard palate at 8 years. The material consisted of 321 cast models obtained at 5, 10, 16 and 19 years of age and were evaluated with GOSLON yardstick by four calibrated raters.

Results: Intra- and interrater reliability were analyzed using weighted kappa statistics and found to be very good (0.79 to 0.84) respectively good (0.6 to 0.78). There was a statistical significant difference between the two protocols in all age groups except for 16 years of age. At 5 years of age 75% were scored 1/2 in HPC3 group and 89 % in HPC8 group (p=0.006). At the final age, 19 years, 60 % were scored 1/2 in HPC3 group and 79 % in HPC8 group (p=0.003). The mean GOSLON score for the entire sample in HPC3 was 2.17 and 1.98 in HPC8 group.

Summary/Conclusion: Both groups, HPC 3 and 8, have demonstrated very good results for the dental arch relationship. However statistically significant difference was found between the two groups with better GOSLON scores in the HPC8 group. The results will be discussed with regard to growth data and to surgical timing.
Long-term, longitudinal follow up of individuals with UCLP after Gothenburg two stage palate closure: surgical and speech outcome of closure of soft palate at six months and hard palate at three years

H. Mark¹, J. Lilja¹, S. Rizell², M. Najar Chalien², C. Havstam³, C. Persson⁴

¹Department of Plastic Surgery, Sahlgrenska University Hospital, ²University Clinics of Odontology, ³Department of Otorhinolaryngology, Division of Speech and Language Pathology / Sahlgrenska University Hospital, ⁴Institute of Neuroscience and Physiology, Speech and Language Pathology Unit, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden

Background: In Gothenburg, Delayed Hard Palate Closure (DHPC) has been practiced in UCLP patients between 1975 and 1993. The soft palate was closed at six months and a delayed palate closure at 8 years. Good long-term results regarding growth, occlusion, speech and appearance have been presented. However, a high degree of retracted oral articulation was present due to the residual cleft. In order to close the hard palate earlier, a modified two stage palate closure was introduced 1993 including soft palate closure at six months and hard palate closure at three years.

Aims: The purpose of the present study is to present the surgical results and the effect on the retracted oral articulation of this method.

Methods: Between 1993 and 1996 a consecutive series of 28 patients with UCLP at Sahlgrenska University Hospital, Gothenburg, Sweden was surgically treated with a two stage palate closure including soft palate closure at six months and hard palate closure at three years. The patients have been followed on a regular basis regarding surgical and speech outcome until 19 years of age. Speech recordings of sentences and spontaneous speech from the ages 5, 10, 16 and 19 years of age of 23 participants (14 males and 9 females) was analyzed blindly and independently by two local and one external speech-language pathologists. Intra- and inter rater reliability was calculated.

Results: Mean age of soft palate closure was 6.25 months and median age 6 months Mean age of hard palate closure was 3 years and 4 months and median age 3 years and 3 months.

Mean age of bone transplantation was 7 years and 10 months and median age 7 years and 8 months. Re-bone transplantation occurred in 2 patients, orthognathic surgery in 1. Fistula closure occurred in 2 patient (7%) and speech improving surgery in 6 (28%). Retracted oral articulation was prevalent to some degree for about 22% of the 5 year olds and had been resolved at 10 years of age.

Summary/Conclusion: Long-term, longitudinal follow up of individuals with UCLP after modified Gothenburg two stage palate closure including soft palate closure at six months and hard palate at three years show a safe surgical method with a low rate of surgical complications as the previous method, Delayed Hard Palate Closure (DHPC). The result also show on less retracted oral articulation but increased frequency of speech improving surgery compared to DHPC. The surgical and speech result will further be analyzed and discussed in detail.
OS 42.4
Maxillofacial growth in patients with cleft lip and palate submitted to a late surgery: case-control study
E Melo¹, L Caceres², J Leal², R Pereira³
¹CADEFI, IMIP, Recife- PE, ²Cirurgia Plastica, ³CADEFI, IMIP, Recife-PE, Brazil

Background: Primary surgeries in patients with unilateral cleft lip and palate (UCLP), normally performed during the first year of life and many variables can interfere with maxillofacial growth and speech outcomes. In a large number of Cleft centers, palatoplasties are performed in the first and second year of life bringing benefits to the patient's speech. However, many studies show that late surgical intervention after three years occurs at a stage where there is a decrease in maxillary-mandibular growth development, which minimizes the changes on bone structures and favors maxillofacial growth.

Aims: To analyze the relationship of primary palate surgeries on the facial growth of patients with unilateral cleft lip and palate submitted to one-stage primary palatoplasty early and late operated in the Center for Attention to Defects of the Face of IMIP (CADEFI) of the Instituto de Medicina Integral Prof. Fernando Figueira (IMIP) between 1999 and 2009.

Methods: This case-control study evaluated 99 patients, twenty-nine of them were submitted to a late procedure, after three years of age, and 70 operated between 9 and 24 months. The surgical techniques used were modified Von Langenbeck or Veau with Braithweite procedures. Multivariate analysis was performed for the association between late palatoplasty and maxillofacial growth. For the trend of maxillofacial growth in both groups, models of the dental arches were made using stone gypsum and evaluated by the Goslon index through the degree of agreement Kappa.

Results: Kappa general agreement among the evaluators was 0.712 (SD +,- 0.608-0.816), with p<0.001. The distribution of Goslon scores in the case group was 51% for a good growth tendency (scores 1 and 2) and 31.04% unfavorable to growth (scores 4 and 5), with a mean of 2.6 (SD +,- 1.22). In the control group, the distribution of scores was 38.7% for a good growth with a Goslon mean of 2.9 (SD +,- 1.14). In the multivariate analysis for late palatoplasty and growth, the result was OR=0.9913 (95% IC, 0.3986-2.4652), RAP = - 0.008776.

Summary/Conclusion: In this sample, the late palatoplasty presented a positive odds ratio for the trend of maxillofacial growth.
Unilateral cleft lip and palate severity: there's any association related to outcomes?

R Pereira¹, D Vale², D do Vale², E Melo², E Melo², N Siqueira³, N Siqueira², N Alonso⁴, J Leal⁶, N Alonso⁶

¹CADEFI, IMIP, Recife-PE, ²CADEFI, IMIP, Recife-PE, ³CADEFI, IMIP, Recife, ⁴Craniofacial Surgery Department, FMUSP, São Paulo, ⁵Cirurgia Plastica, IMIP, Recife-PE, ⁶Cirurgia Cranifacial, FMUSP, Sao Paulo-SP, Brazil

Background: The initial dimensions of the unilateral cleft lip and palate (UCLP) are varied and may present wide differences between each patient and was reported by some authors that it can be responsible for less favorable outcomes related to cleft severity.

Aims: The aim of this study was to establish possible associations between the width and dimensions of the cleft in children with complete unilateral cleft lip and palate, between 0 and 6 months of age and outcomes related to facial growth, after surgical treatment, evaluated between 4-5 years of age.

Methods: This was a prospective study nested in Clinical Trial Randomized with a group (GI) of patients submitted to a surgical protocol with Late Closure of the Hard Palate (UCLP) between 3-4 years of age and another control group (CG) submitted to palatoplasty 9 and 15 months of age.

LOCAL: The study was conducted at the Center for Attention to Defects of the Face (CADEFI) of the Institute of Integral Medicine Prof. Fernando Figueira in Recife, PE, Brazil, between January 2010 and October 2016.

METHOD: Dental artery study models were obtained at CADEFI enrollment and after 5 years of age in the sixty-four patients with consecutive non-syndromic unilateral cleft lip and palate. An assessment of the width of the newborn's cleft was performed. Anterior and posterior widths of the cleft were measured and the transversal and anteroposterior dimensions of the maxilla were also evaluated (anterior, posterior and alveolar arch lengths). In the models in the deciduous dentition the transversal and anteroposterior dimensions of the maxilla and the maxillo-mandibular relationship (FYOI).

Results: The width / amplitude of the cleft evaluated initially presented a similar distribution in both groups (GIxGC). The correlation between the width / amplitude of the fissure thus evaluated and the maxillo-mandibular ratio evaluated by the FYOI was not evidenced by the Spearman method.

Summary/Conclusion: The outcomes demonstrate a great variation in the severity of the cleft lip and palate unilateral at birth. It was not found any evidence of an association between the less favorable results of FYOI evaluation and larger dimensions of the fissure. There was also no association between the use of lateral liberating incisions in palatoplasty and less favorable results of facial growth.
The orthodontic outcomes of 42 consecutive patients with a repaired bilateral cleft lip and palate treated by one operator

H Bellardie1, A Nur Ashikin Abd Rahman3, D Stonehouse-Smith4, J Mooney1

1Regional Cleft Unit, Royal Manchester Children's Hospital, Manchester, United Kingdom, 2Orthodontics, University of the Western Cape, Cape Town, South Africa, 3University Teknologi MARA, Selangor, Malaysia, 4Maxillo-facial surgery, Aintree University Hospital, Liverpool, United Kingdom

Background: The Peer Assessment Rating (PAR) Index was developed to provide a single summary score for all the occlusal anomalies which may be found in a malocclusion. The score provides an estimate of how far a case deviates from normal alignment and occlusion. The difference in scores between the pre- and post-treatment cases reflects the degree of improvement and, therefore, the success of treatment. The components of the PAR Index have been weighted to reflect current British orthodontic opinion and is flexible in that the weightings could be changed to reflect future standards and those currently being achieved in other countries. The PAR Index offers uniformity and standardization in assessing the outcome of orthodontic treatment. (S Richmond et al).

The current UK Audit Standard for those born with Unilateral Cleft Lip and Palate is 69% reduction in PAR score with 7.5% of cases in worse or no different category. There are no published Audit Standards for those born with Bilateral Cleft Lip and Palate.

Aims: A retrospective study to assess the orthodontic outcome for patients with a bilateral cleft lip and palate (BCLP) attending the Cleft Lip and Palate Unit, Royal Manchester Children's Hospital (RMCH), Manchester, UK, treated by one Consultant Orthodontist.

Methods: 42 consecutively treated fixed appliance cases, including those where orthognathic surgery was undertaken, were assessed using the peer assessment rating (PAR) index on orthodontic study models which were taken at the start of treatment and at deband. Exclusions included syndromic patients, those who were awaiting or declined orthognathic surgery, poor compliance or had incomplete records. A total of 11 patients were excluded from the original leaving 31. Data, including missing, ectopic, supernumerary, hypoplastic and extracted teeth were recorded retrospectively from patient records and radiographs. Treatment time and number of visits were also recorded.

Results: The mean start PAR score was 43.42, the mean end of treatment PAR was 8.42. The mean reduction in PAR score was 80.6%. The data demonstrated 29(93.5%) cases being greatly improved and 2(6.5%) improved. The percentage of cases where the score was worse or no different was 0%. The treatment time, from bond up to deband, was 27.6 months. 26(83%) patients had missing teeth, 15(48%) required extractions for orthodontic purposes, 7(22.5%) reported ectopic teeth, 2(6.5%) supernumeraries, 11(35.5%) peg lateral incisors, 13(42%) hypoplastic upper right central incisors and 13(42%) left central incisors.

Summary/Conclusion: The recommended final PAR score for a Cleft Unit for UCLP is 69% with 7.5% of cases in worse or no different category. The RMCH mean percentage PAR reduction, for BCLP cases, of 80.6% with 93.5% greatly improved, 6.5% improved and 0% worse or no better compares well with other reports on outcome. This is especially so considering the greater number of dental anomalies in this (BCLP) group of patients. All audit standards for Non-cleft and UCLP were met.
Evidence for midfacial-growth for patients with complete unilateral non-syndromic cleft lip and palate treated by one-stage and two-stage palatal repair

I Zepa\textsuperscript{12}, I Akota\textsuperscript{13}, I Bagante\textsuperscript{13}, I Maulina\textsuperscript{12}

\textsuperscript{1}Riga Cleft Lip and Palate centre, Institute of Stomatology, \textsuperscript{2}Department of Orthodontics, \textsuperscript{3}Department of Oral and Maxillofacial Surgery, Riga Stradins University, Riga, Latvia

Background: There were two surgical methods for hard palate closure used at the Riga Cleft Lip and Palate centre - one-stage (ref?) and two-two-stage palatal repair (ref?). Surgical protocol used for cleft repair plays significant role for the outcome in facial growth and thereof the resultant facial proportions. Factors who influence facial growth adversely (resulting in very poor maxillary growth) had been identified previously - e.g., aggressive surgical technique leaving denuded bone (Friede 2007).

Aims: The aim of the present study was to evaluate the midfacial-growth results for 16 year old patients with complete non-syndromic unilateral cleft lip and palate (UCLP) treated by one-stage and two-stage palatal repair surgery at Riga Cleft lip and Palate Centre, Latvia.

Methods: This was a retrospective cohort study. We evaluated 98 consecutive patients born between 1980 and 2004 with non-syndromic complete UCLP (60 male and 38 female) operated either by one-stage (n=50) or two-stage palatal closure protocol (n=48). We analyzed lateral cephalograms of patients with UCLP at 16 years of age. Cephalometric data were used to determine midfacial-growth results. Incidence of re-operation for residual defects was counted. Mann Whitney U test was used to compare the significance of differences in the cephalometric results of the two samples. Level of significance was set at 95%.

Results: Most parameters studied were similar except more vertically deficient growth of maxilla for one-stage palatal repair group. Upper face height was shorter for one-stage palatal repair group. Prevalence of residual palatal defects was 10.6% for one-stage palatal repair group and 10.3% for two-stage palatal repair group.

Summary/Conclusion: At the mean age of 16 years midfacial-growth results were clinically similar for patients with complete non-syndromic UCLP operated either by one-stage or two-stage palatal repair surgery.
The orthodontic outcomes of 84 consecutive patients with a repaired unilateral cleft lip and palate treated by one operator.
H Bellardie1,2, A Nur Ashikin Abd Rahman3, D Stonehouse-Smith4, J Mooney1
1Regional Cleft Unit, Royal Manchester Children’s Hospital, Manchester, United Kingdom, 2Orthodontics, University of the Western Cape, Cape Town, South Africa, 3Orthodontics, Universiti Technologi MARA, Selangor, Malaysia, 4Maxillo facial surgery, Aintree University Hospital, Liverpool, United Kingdom

Background: The Peer Assessment Rating (PAR) Index was developed to provide a single summary score for all the occlusal anomalies which may be found in a malocclusion. The score provides an estimate of how far a case deviates from normal alignment and occlusion. The difference in scores between the pre- and post-treatment cases reflects the degree of improvement and, therefore, the success of treatment. The PAR Index offers uniformity and standardization in assessing the outcome of orthodontic treatment. The components of the PAR Index have been weighted to reflect current British orthodontic opinion and is flexible in that the weightings could be changed to reflect future standards and standards currently being achieved in other countries. (S Richmond et al).

The current UK Audit Standard for those born with Unilateral Cleft Lip and Palate is 69% reduction in PAR score with 7.5% of cases the in worse or no different category and 75% in the greatly improved category.

Aims: This retrospective study assessed the orthodontic outcome for consecutive patients with a repaired unilateral cleft lip and palate (UCLP) attending the Cleft Lip and Palate Unit, Royal Manchester Children’s Hospital (RMCH)Manchester, UK, treated by one Consultant Orthodontist.

Methods: 84 consecutively treated fixed appliance cases, including those where orthognathic surgery was undertaken, were assessed using the PAR index on orthodontic study models taken at the start of treatment and at deband. Patients with a syndrome, or poor cooperation, those awaiting orthognathic surgery or had declined orthognathic surgery, and patients with incomplete records were excluded, a total of 14 from the original 84 leaving 70. Data, including missing, ectopic, supernumerary, hypoplastic teeth and teeth extracted were recorded retrospectively from patient records and radiographs. Treatment time and number of visits were also recorded.

Results: The mean start PAR score was 41.82. The mean end of treatment PAR was 6.66. The mean reduction in PAR score was 84.07%. The data demonstrates 64(91.5%)cases were greatly improved and 6(8.5%) improved. The percentage of cases where the score was worse or no different was 0%. The treatment time, from bond up to deband, was 23.2 months. 49 (70%) patients had congenitally missing teeth, and 25 (36%) required extractions for orthodontic purposes. 4 (4%) reported ectopic teeth, 10 (14%) supernumeraries, 16 (23%) right peg lateral incisors, 7 (10%) left peg lateral incisor, 5 (7%) hypoplastic upper right central incisors and 6 (8.5%) left central incisors.

Summary/Conclusion: The recommended final PAR score for a Cleft Unit for UCLP is 69% with 7.5% of cases in the worse or no different category and 75% in the greatly improved category. The RMCH mean percentage PAR reduction of 84.07% with 93.5% greatly improved, 6.5% improved and 0% worse or no better compares well with other reports on outcome. All audit standards for UCLP were met.
Sella turcica in patients with unilateral cleft lip and palate: a cephalometric morphometric study
G Antonarakis¹, L Huanca Ghislanzoni¹, G La Scala², D Fisher³
¹University of Geneva, ²University Hospitals of Geneva, Geneva, Switzerland, ³The Hospital for Sick Children, Toronto, Canada

Background: The relationships of the sella turcica with the pituitary gland may be important when evaluating growth and pathology. No attempt has been made to provide normative data with regard to the size and shape of the sella turcica, in subjects with clefts, using geometric morphometrics. This method has the advantage that differences in orientation of both the lateral cephalometric radiograph and the sella turcica itself do not influence neither the measurements of size nor the definition of shape.

Aims: The aim of the present study was to use geometric morphometric methods to investigate the size and shape of the sella turcica in children with unilateral cleft lip and palate (UCLP) using lateral cephalograms.

Methods: A consecutive sample of lateral cephalograms from 56 otherwise healthy children (13 females; 43 males), between the ages of 5 and 12, with UCLP was obtained retrospectively from an institutional cleft database. All radiographs were imported into specific cephalometric software and adjusted for magnification. Conventional measurements were initially carried out, namely sella height (anterior, posterior, and median), width, and area. To assess sella shape, the contour of the sella turcica was traced. Nine additional equally-spaced points were defined by the software along the contour. Within this contour, the most posterior, most anterior, and deepest points of the sella were defined using the Frankfort horizontal plane as the reference plane. Sella shape analysis was performed using the 11 points defining the sella turcica contour, using morphometric analysis. Procrustes superimposition was used to register all sella contour tracings in order to calculate an average shape. Principal component analysis (PCA) was used to assess shape variation.

Results: Sella height varied from 3.4mm to 9.6mm with an average of 6.1mm. Sella width ranged from 4.8mm to 11.8mm with an average of 8.6mm. Sella area ranged from 20.6 to 70.1 square mm, with an average of 42.7 square mm. 95% of the cumulative variance of sella turcica shape, as described by PCA, was attributed to three principal components (PC), with PC1 describing one third of the variance.

Summary/Conclusion: The results of the present study constitute quantitative reference data that could be used for objective evaluation of sella size and shape in individuals with UCLP.
Cranial Facial - Cleft Surgery

OS 43.1
Soft tissue management of craniofacial clefts – our experiences
G Srinivas Reddy

Abstract Content: Craniofacial clefts take many forms and involve bony and soft tissue defects. They are considered a result of failure of fusion of the facial processes. Craniofacial cleft is rare and has an incidence of 1.4 – 4.9 per 100,000 live births. Further to this, the severity of each craniofacial cleft varies and no two are the same. The surgical management of craniofacial clefts involve bony reconstruction, and reinsertion of facial muscles and reconstruction of cutaneous defects. The range of techniques for managing soft tissue in craniofacial clefts include primary closure, local flaps, Z-plasty and V-Y advancement flaps. Local flaps may result in tissue that has different colour, thickness and texture of skin, however improved aesthetic results have been described when combined with tissue expanders. Z-plasty have been reported to increase the ‘patchy’ effect of the face and may leave multi-directional scars, although it enables lengthening of the scar. The Pfeifer technique is utilised in the soft tissue management of craniofacial clefts at the GSR Institute of Craniofacial and Facial Plastic Surgery, Hyderabad. Long-term clinic outcomes of a representative sample of these patients are presented.
The normal evolution of the cranium in 3d
J Meulstee¹, G de Jong², W Borstlap¹, T Maal¹, H Delye²
¹Oral and maxillofacial surgery, ²Neurosurgery, RadboudUMC, Nijmegen, Netherlands

Background: During the first years of a newborn’s life, the cranium grows very rapidly. Insight in this development is essential for monitoring cranial development of new-borns, identify cranial abnormalities and for the follow up of craniosynostosis surgery.

Aims: This study presents a three-dimensional (3D) evaluation of the normal cranial shape by the use of 3D stereophotographs and CT scans.

Methods: A total of 130 3D stereophotographs and 94 CT scans of new-borns without abnormalities were used to evaluate cranial evolution. All 3D photos were acquired between 3 and 24 months of age with a fixed interval of 3 months. This dataset was used to calculate the cranial length, cranial width, cranial index, cranial circumference and volume. Distance maps of the complete 3D morphology were produced to create a 3D perception of the cranial evolution.

Results: Evaluation of the 3D distance maps revealed a more prominent growth of the anterior part of the skull between 3 and 12 months. The posterior part of the skull developed more rapidly between 12 and 15 months compared to the rest of the skull and an almost uniform growth was seen between 15 and 24 months.

Summary/Conclusion: This 3D analysis of the normal cranial shape provides a valuable insight of the evolution of the cranium during the first 24 months of life. This study presents data which can be used for monitoring cranial development and for the follow-up of craniosynostosis surgery.
Bws protocol base on cephalometric and photometric findings
C Tortora¹, M Besana², M Meazzini¹, M Ferrari¹, L Autelitano¹
¹Smile-House, Santi Paolo e Carlo Hospital, Milan, ²San Gerardo Hospital, Monza, Italy

Background: Macroglossia is the predominant finding in BWS, with a percent approximating 90% of patients. It is also highly specific for the syndrome. It can be of variable degree and, in most cases, it is of modest entity, requiring no treatment. It is suggested that after 2-3 years of age, the growth of the tongue in a number of children with BWS gradually slows down and the tongue is increasingly contained within the oral cavity.

Aims: The first part of the study compared photometrically the craniofacial characteristics of BWS patients with the craniofacial characteristics of non affected individuals (Farkas normative data) at different ages. The second part of the study, focused the attention on the differences in craniofacial growth of patients affected by BWS when glossectomy was performed and when it wasn't.

Methods: The sample was composed of 62 patients retrospectively collected. 26 of these patients have undergone glossectomy during the first 2 years of live, while the others patients had no surgery. Pre and postoperative data were collected in all patients in order to assess either the long-term effect of glossectomy, the spontaneous or the orthodontically supported improvement of jaw relationship.

Results: The photometric findings showed that there was no significant difference in preoperative severity. Operated patients seem to have a more harmonic craniofacial growth from 1 to 4 years in terms of early profile improvement, than patients who did not undergo glossectomy. This difference, though, was measured only till 5 years of age. Interestingly, maxillo-mandibular relationship in nonoperated patients improved photometrically and cephalometrically at 6-7 years of age, more so if an orthodontic treatment had been carried out, and there were no differences between groups at 12 or 16 yeras of age.

Summary/Conclusion: Glossectomy, though often unavoidable, should not be the first-line treatment when it does not inhibit feeding or breathing. Further studies are needed to understand whether different genetic mutations have different craniofacial growth prognosis.
Background: The surgical techniques used for the primary surgical repair of cleft palate are evolving. Medium to long term outcomes of the different surgical techniques are lacking.

Aims: To review the medium term surgical outcome of patients who had received primary palatoplasty in a single center in Hong Kong in the past ten years.

Methods: Medical records of patients who had primary palatoplasty in our center since 2009 to 2018 were retrospectively reviewed.

Results: 269 patients' data was reviewed. 26 (10%) of them were submucous cleft palate and 19 (7%) had Pierre Robin Sequence. For the surgical technique employed in the primary palatoplasty: 90 patients (33%) received Furlow double opposing Z-plasty while 179 (67%) patients received von Langenbeck with intra-velar veloplasty. At the time of review, 22 (8%) patients were documented to have hypernasality likely secondary to velo-pharyngeal insufficiency. Of the 22 patients with hypernasality: 4 (18%) of them are from the Furlow group and 18 (82%) are from the von Langenbeck group. There were 29 re-operations on the palate: 6 (21%) had received primary palatoplasty in other institutions, for the remaining 23 (79%) palatal revisions: 6 (21%) were for indicated for correction of VPI, 17 were indicated for palatal dehiscence or palatal fistula.

Summary/Conclusion: Furlow palatoplasty produced overall less velo-pharyngeal insufficiency when compared to the von Langenbeck palatoplasty, although the difference did not reach a statistical significance (p = 0.16). A longer term follow up would further validate these findings.
**OS 43.7**

**Structural fat grafting in craniosynostosis patients**

D Leshem¹, S Zismann¹

¹Plastic & Reconstructive Surgery, Tel Aviv Sourasky Medical Center, Tel Aviv, Israel

**Background:** Craniosynostosis, the premature fusion of cranial sutures, a relatively common condition that can appear both independently and in the context of syndromic conditions, results in a range of aesthetic and developmental sequelae. Structural fat grafting is a well-known technique, which has been used and promoted widely in the last few years. The new era of fat grafting pioneered by Dr. Coleman offers a valuable tool to address patient for cosmetic procedures and reconstructive surgery. This pediatric craniofacial unique population requires techniques for reconstruction and final refinement of surgery. Structural fat grafting is an ancillary method for these patients.

**Aims:** This study examines the use of autologous fat grafting in pediatric patients as a solution for the repair of aesthetic defects following correction of craniosynostosis.

**Methods:** We retrospectively reviewed structural fat grafting in 10 patients, younger than 16 years of age at Tel Aviv Sourasky Medical Center in the years of 2005-2018. Using the Coleman fat grafting method, standard harvesting sites: abdomen and inner thighs. We assessed complications, cosmesis, and functional results for a follow up period of 1 to 5 years. All patients were under pediatric Plastic Surgery follow up.

**Results:** This series included 10 patients, 4 males and 6 females, after cranial vault surgery. No complications occurred in the immediate post operation period or later. Most of the patients required 1-2 sessions of fat grafting. Excellent cosmetic results where achieved in all patients.

**Summary/Conclusion:** The pediatric craniofacial patients are a population with special requirements, minimal and long lasting procedures in order to avoid psychosocial problems and good acceptance by the surrounding. Structural fat grafting is a safe procedure in this population. Exceptional results raise the question if fat grafting in this young population has superb results in comparison to older patients.
**OS 43.8**

**Palatal cleft: results after 18 years, one surgeon, one primary technique**

A De Buys Roessingh¹, O El Ezzi¹, G Herzog¹, M Broome¹, L Medinger¹, C Zbinden-Trichet¹, C Béguin¹, C Richard¹, S Fries¹, J Despars¹, Y Robert¹, G Dimitropoulos¹

¹CHUV, Lausanne-CHUV, Switzerland

**Background:** This study reviews the results 18 years later of our surgery and follow-up for children born in our hospital with cleft palate (CP).

**Aims:** Evaluation of 18 year-old youngsters born with CP, operated at the time by the same surgeon following the same primary surgical procedure (von Langenbeck).

**Methods:** We analyzed files of children born from 1998 to 2001 with a cleft and operated in our hospital. Operations for CP were performed by the same surgeon, following the von Langenbeck procedure adapted according to the children's age/weight. The evaluation was based on the results of the primary surgeries, ENT intervention, maxillo-facial surgery and final phonatory results. All syndromic children were excluded.

**Results:** Seventy-nine files from children born with a cleft were reviewed: 34 were taken in consideration for CP, among them 5 Pierre Robin Sequence (PRS). Children were operated at a median age of 5.6 months (3-8), and 8 months for the PRS. 19% had a fistula whose closure required one operation per child. 47% had grommets, of which 60% required several operations. None had a Lefort osteotomy. For the phonatory score, 15% were operated for a pharyngeal flap following the Sanvenero-Rosselli technique, 3 at the age of 16, and 2 at the age of 7. Thirty-six (36)% have a normal phonation with no nasal air emission (phonation I), 32% have a good phonation with intermittent nasal air emission but good intelligibility (phonation I/II) and 32% a phonation with continuous nasal emission but good intelligibility and no social discomfort (phonation IIb). A median of 5.5 multidisciplinary consultations were realized with a median number of general anaesthesia of 2.4 (1-6).

**Summary/Conclusion:** This retrospective study is based on one primary surgical technique by the same surgeon. It shows that the von Langenbeck procedure done at 6 months is related to a risk of fistula but good long-term phonatory results. The number of grommets used is low and must be confronted with otitis media. No surgical intervention was realized for mandibular or maxillary growth.
How to improve our results in the primary labial repair of unilateral cleft
J Saboye
surgery, cleft center medipole garonne toulouse france, toulouse, France

Background: Primary repair of cleft lip and palate is a nasal and lip repair. The Millard's technique (1) has been used for many years but some points are not so efficient. We must correct all anomalies and extend our dissections, find new ways for improving our results.

Aims: It is necessary to analyze its own results on the lip and nose, to judge them and to look for technical improvements. Our main goals are to lengthen the length of the cleft lip under the nasal alar, obtain a symmetry with both sides, create a philtral column, to have a horizontal cupid's bow, to obtain a continuity of Roll white roll, of red lip-white lip, of wet/dry red lip, to obtain a median Volume in red lip, to create symmetrical nasal holes with a projected and symmetrical nose, and few scars.

Methods: after an analysis of our results on more than 100 cases operated, we changed the first incisions, using the technique describe by david Fischer (2)we changed our way to operate the nose and the septum. Six different points are analyzed and explained.

Results: Changing some points in our initial technique learned by Ralph Millard, allows us to improve our results on unilateral cleft primary surgery. To improve our results some major changing are necessary. Remove a future scar, change the paradigm of septal dissection.

Summary/Conclusion: First of all, we have to analyze our results, we have to be critical, seeing what is wrong, we have to change, evolve, listen, read,...we have to think that it is never enough and always try to live up to the hopes we carry on us.

Being humble and realistic, the perfect result does not exist...And, twenty times on the trade hand over your work (Nicolas Boileau)

Aesthetic Outcome

Comparing ralph millard and ron pigott’s aesthetic outcomes with non cleft children using symnose.
F Maggiulli¹, C Hinton², L Simpson³, S Gujural³, T Su⁴, I Timmins⁵, R Pigott⁶, B Richard⁴
¹Plastic Surgery, Great Ormond Street Hospital, London, ²Plastic Surgery, Birmingham Children Hospital, Birmingham , ³Plastic Surgery, Birmingham Children Hospital, ⁴Plastic Surgery, Birmingham Children's Hospital, Birmingham, ⁵Children's Hospital of Birmingham, University of Leicester, Leicester, ⁶Cleft Service of Birmingham Children's Hospital, Birmingham Children's Hospital, Birmingham, United Kingdom

Background: The SymNose Computer program, developed by Pigott and Pigott in 2010, has been proven to be a rapid and objective method for the quantitative assessment of the lip and nose symmetry in normal and CLP patients.

Aims: The aim of this study is to compare non-cleft children lip symmetry measurements, with cohorts of UCLP children at age 5, using Symnose. The normal results will be compared to patient cohorts from two famous cleft surgeons Prof D Ralph Millard and Mr Ron Pigott.

Methods: We acquired two historical cohorts of complete UCLP post op photographs from two of the world’s most renowned cleft surgeons. A series of 48 patients operated by Millard, 92 patients operated by Pigott and 91 non cleft children. The images have been assessed by 5 independent investigators, each tracing their batch of images twice. This was analyzed to provide intra and inter rater reliability scores by ICC (95% CI). The median symmetry was then compared between each batch.

Results: The average lip percentage mismatch for Millard’s series is 15.12% (IQR= 6.36). The average lip percentage mismatch for Pigott’s series is 9.10 (IQR=5.56). Both series were compared using Wilcoxon signed rank to normal 4.9% (IQR=2.39) to give P=<0.001. The average lip percentage mismatch is significantly different between Millard’s and Pigott’s series.

Summary/Conclusion: The greatest asymmetry difference was shown between the Cleft cohorts and the Normal, with some crossover between each cohort. The normal cohort shows the most lip symmetry. The two cleft cohorts show a similar, but clearly distinguishable degree of asymmetry. In nearly all Pigott’s cases, at least one revision was done before the age of 5. Millard and Pigott's cohorts represent work spanning their careers, with changes in practice and surgical technique as expected.
A global evaluation of surgical techniques and results of unilateral cleft lip repairs
A Volk¹, R Dempsey¹, E Buchanan¹
¹Plastic Surgery, Texas Children's Hospital, Houston, United States

Background: Smile Train, an international children’s charity, provides safe, high-quality cleft care throughout the world by partnering with local healthcare systems. The organization’s sustainable global health model ameliorates the backlog of cleft disease and increases surgical capacity by empowering cleft care teams through training and educational opportunities. There is no universally accepted technique for cleft lip repair or system to evaluate aesthetic outcomes. As part of their quality assurance monitoring process, Smile Train has developed lip grading criteria to ensure successful results are achieved after cleft lip repair operations performed by partnering surgeons across the world.

Aims: The aim of this study is to present Smile Train’s lip grading criteria and to evaluate outcomes of unilateral cleft lip repair operations based on the surgical technique used.

Methods: Smile Train’s lip grading criteria assigns a pre- and post-operative grade from an uncropped, frontal photograph. Grading is performed by experienced independent plastic and reconstructive surgeon reviewers. Reviewers assign a score based on the severity of the pre-operative defect (grade 1-5) and the final post-operative result (grade 1-5), where 1 is the lowest severity and poorest outcome, respectively, and 5 is the highest severity and optimal outcome, respectively. Values are multiplied, producing a final composite grade. Acceptable results include a composite grade of ≥"3" for a pre-operative grade of “1” and composite grade of ≥"6" for pre-operative grades “2-5”.

A retrospective review was conducted of unilateral cleft lip repair operations performed by Smile Train surgeons spanning the geographic regions of Africa, Americas, Asia Pacific, Europe and Middle East between 2014-2018. Cleft laterality, completeness, and involvement of the alveolus and/or palate were recorded. Surgical techniques included the rotational advancement, straight line repair and its variations, z-plasty variations, and “other”. Using the lip grading criteria, acceptable outcomes were identified and compared with the surgical technique used.

Results: 8,041 unilateral cleft lip repairs were reviewed; 65% left- and 35% right-sided. 60.5% were complete cleft lips, 63.3% involved alveolar clefting and 42.5% involved clefting of the hard and/or soft palate. Overall, the majority of repairs were performed with the rotational advancement technique (n=5,541, 68.9%) with a statistically significant highest percentage of acceptable outcomes (92.1%), followed by an acceptance rate of 91.9% with variations of the z-plasty technique, 90.5% with the straight-line repair and its variations, and 88.4% using “other” techniques (p<0.05). No difference was seen between outcome and technique used for incomplete cleft lips, however, the rotational advancement had the highest acceptable outcomes for complete cleft lips (94.7%, p<0.0001), associated alveolar clefting (94.2%, p<0.05), and concomitant clefting of the hard and/or soft palate (94.3%, p<0.05).

Summary/Conclusion: Regardless of surgical technique used, Smile Train surgeons maintain high rates of successful surgical outcomes. The rotational advancement technique was the most commonly utilized with the highest overall acceptable outcomes, especially for more severe clefts. Results of our study will help complement and support the existing medical infrastructure and training systems Smile Train employs for surgeons working with comprehensive cleft care teams around the world.
Background: Cleft lip and palate (CLP) is associated with difficulties in tooth-status, eating, speaking, hearing and appearance. Thus, children with CLP are early included in a multidisciplinary program of surgery and therapy lasting through childhood, adolescence and into young adulthood. After finishing the standard care program, little is known of the impact of the CLP and its treatment on adult life.

Aims: To explore and describe the experience of CLP in adults.

Methods: Face to face interviews were conducted as part of a larger multidisciplinary follow up of adults (n=21), who were 40 years of age, and treated for unilateral CLP. Giorgi’s phenomenological method was used for analysis. The study was approved by the Regional Committee for Medical and Health Research Ethics REC West (p. number 2016/269). All participants gave their written consent to participate.

Results: The trajectory of growing up with a CLP meant to become aware of a bodily otherness and possible reactions from peers early in life, later complicating the striving for inclusion outside the close family. Then, the reflected image, through mirrors, windows and photos led to bodily adjustments recognizable into adulthood. The trajectory of treatment was not questioned during childhood, accepting the parents’ and experts’ decisions on care. Needs for corrections and treatment later in life were results of a balancing between risks and benefits, and a more hesitant view on the possible benefits of additional surgery.

Summary/Conclusion: Overall, growing up having CLP did not contradict opportunities for achieving goals and satisfaction in life.
Three dimensional virtual planning of premaxilla osteotomy in bilateral cleft
A Matthews-Brzozowski¹, J Kraeima², R Schepers², J Jansma²
¹Department of Oral and Maxillofacial Surgery, Medical Centre Leeuwarden, Leeuwarden, ²Department of Oral and Maxillofacial Surgery, University of Groningen, University Medical Centre Groningen, Groningen, Netherlands

Background: Premaxilla osteotomy is used to correct the severely malpositioned premaxilla in bilateral cleft. This procedure occurs mostly in combination with the alveolar bone grafting before eruption of the canine. Usually, the premaxilla is fixated in the new position using a manually fabricated dental splint. This is a time consuming and variable process.

Aims: We aimed to show 3D virtual planning of the premaxilla osteotomy and dental splint.

Methods: A novel 3D surgical planning method of the premaxilla osteotomy based on the CBCT data was performed in two patients. A developed CAD-CAM workflow including the fabrication of a custom dental splint was applied in order to align the premaxilla conform the virtual planning.

Results: A fully 3D planned premaxilla osteotomy with a dental splint showed high accuracy of the alignment of the premaxilla and excellent fitting of the dental splint.

Summary/Conclusion: This novel 3D workflow for premaxilla osteotomy planning and perioperative positioning provides a predictable result.
OS 44.5

Exceptional appearance outcomes of the nose and lip after unilateral cleft lip and palate repair: judgment by professionals, patients and laypeople

R Tan¹, D Mosmuller¹, H De Vet², R Schwirtz¹, J Don Griot¹

¹Plastic, Reconstructive and Hand Surgery, Amsterdam UMC, location VUMc, Amsterdam, Netherlands, ²Department of Epidemiology and Biostatistics and the Amsterdam Public Health research institute, Amsterdam UMC, location VUMc, Amsterdam, Netherlands

Background: There is still a need for a simple and easy-to-use instrument to evaluate nasolabial outcome after unilateral cleft lip and palate (UCLP) repair. Hence, the Cleft Aesthetic Rating Scale (CARS) was recently developed. This instrument uses a photographic reference scale to grade ‘typical’ nasal and lip appearance outcomes. These typical outcomes evolve out of scar contraction and result in an a-symmetric higher vermillion border and a wider nostril (flaring ala) both on the repaired cleft-side. On the contrary, a lower vermillion border and a more narrow nostril can be denoted as more exceptional appearance outcomes. In addition, a whistling deformity can equally be considered as an exceptional outcome as well. Since the CARS does not include photographs of these exceptional outcomes, controversy among raters emerged while using this instrument.

Aims: To investigate how the three exceptional appearance outcomes are ranked compared to the typical outcomes. Evaluation was performed by professionals, UCLP patients and laypeople. Based on the findings, suggestions to further improve the CARS were proposed.

Methods: An online survey was developed containing three series of typical and exceptional nasal and lip photographs and was sent to 30 professionals, 30 UCLP patients and 50 laypeople. The participants were asked to rank order the photographs from excellent to severe based on overall appearance. Agreement between professionals, patients and laypeople was assessed with an ANOVA analysis. Difference in rank score between typical and exceptional results were assessed using a T-test.

Results: Differences in rank order of the photographs was not statistically significant (P=0.87) between professionals, patients and laypeople. A nose with a wider cleft-side nostril was ranked significantly (P=0.00) better than a nose with a more narrow nostril. A lip with a high cleft-side vermillion border was ranked significantly (P=0.02) better than a lip with a low vermillion border. A lip with a whistling deformity was ranked significantly worse than a lip with a high (P=0.00) and a low vermillion border(P=0.03) on the cleft-side, respectively.

Summary/Conclusion: The findings in this study gave insight in the interpretation of exceptional post-operative nasal and lip appearance outcomes compared to typical outcomes. Professionals, UCLP patients, and laypeople are in agreement when grading these. This gives input to modify the CARS to include these exceptional appearance outcomes.
**Background:** Although efforts to improve access to care for patients with cleft lip deformities in the developing world have grown tremendously, there is a lack of data regarding the aesthetic outcomes.

**Aims:** The purpose of this study was to investigate early aesthetic outcomes and severity-outcome relationships following unilateral cleft lip repair in resource limited environments.

**Methods:** 1,823 patients treated at surgical missions and surgical centers in low-and middle-income countries were assessed using the unilateral cleft lip severity index and surgical outcomes evaluation scale for unilateral cleft lip. Three separate evaluators scored each case for a total of 5,469 total independent evaluations. Evaluations were based on pre- and postoperative photographs. 56% of the patients had incomplete cleft lip deformities (severity 1-2) and 44% were complete clefts (severity 3-4).

**Results:** For severity grade 1, the mean aesthetic outcome was 6.9 on a scale from 0-8. For severity grade 2 the mean aesthetic outcome was 6.44 and for severity 3 and 4 the mean outcome was 5.89 and 5.83 respectively. The differences in aesthetic outcome was statistically significant (p<0.005) between all groups except between severity group 3 and 4.

**Summary/Conclusion:** The unilateral cleft lip severity index and surgical outcomes evaluation scale are valuable tools for standardized evaluations of large cleft lip cohorts. The results in this study provide normative outcome scores for each severity grade and shows that more severe cleft deformities are associated with lower outcome scores.
Validating a asher mc-dade score to assess facial aesthetic outcomes in twenty two consecutive complete bilateral cleft lip repairs

R Thomson1, D Drake2
1Plastic Surgery, 2Maxillofacial Surgery, Welsh Centre for Cleft Lip & Palate, Swansea, United Kingdom

Background: Assessment and judgement of facial aesthetics is subjective and varies according to individuals, culture and society. Facial symmetry is often an important determinate of this especially when asymmetries are close to the midline. The complex nature of bilateral cleft lip defects means that patients frequently score lower in assessments of facial aesthetics and tend to have higher levels of dissatisfaction with appearance. Scoring systems used in assessing facial aesthetics in cleft patients lack consistency, with no internationally agreed system. The most widely used and validated tool is the Asher-McDade assessment, used to assess unilateral cleft lip repair. There are currently no reports of its use in scoring outcomes after bilateral cleft lip repair.

Aims: We describe the outcomes from twenty two consecutive bilateral cleft lip repairs assessed using the Asher-McDade five point scale. We aim to validate the Asher-McDade assessment tool for bilateral cleft lip repairs.

Methods: Retrospective review was undertaken of 22 consecutive patients with bilateral cleft lip repairs performed at our centre. Each patient underwent bilateral lip repair (advancement rotation repair) at 3-4 months of age with a vomer flap on one side. This was followed by repair of remaining hard and soft palate cleft (intravelar veloplasty) 3 to 4 months later. Standardised photographs were taken 5 years following repair as part of standard 5 year old cleft audit. Anterior/posterior (AP) view and AP lips Worms eye views were taken and cropped to isolate the nasolabial component, then randomised in a photographic slide show. Each image was shown for 90 seconds. 11 members of the cleft team were asked to rate each image on a 5-point scale. This was repeated a second time 6 months later. Statistical analysis was performed using a Two-way ANOVA test. Blind analysis was run by two independent analysts who agreed on the final result and interpretation.

Results: 22 consecutive patients with complete bilateral cleft lips were photographed. There was an overall mean score rating of 3.2, ranging from 4.3 to 1.8. The Two-way ANOVA test shows that the 11 raters account for 11.23% of the total variance between the scores (p<0.0001). Difference in patient appearance (aesthetic outcome) accounts for 44.51% of the total variance between the scores (p<0.0001).

Summary/Conclusion: We demonstrate that variance in the patients scores is significantly related to a true difference in aesthetic outcome with only a small percentage of variance due to inter-observer variation. This has been found in other studies when assessing unilateral cleft lip repairs (Asher-McDade et al 1991, Mercado et al 2011, Johnson et al 2003) but is not described for the assessment of bilateral cleft lip repairs. We show that the Asher-McDade scoring system is a valid tool to use when assessing bilateral cleft lip repairs. It’s a quick and can easily be used by clinicians and lay assessors. It is hoped that the use of Asher-McDade scoring may promote accurate auditing of aesthetic outcomes in bilateral cleft lip repair and used to compare outcomes within and between cleft centres.
**OS 44.8**

**Comparing ccuk aesthetic outcomes with non cleft children using symnose**

C Hinton¹, L Simpson¹, I Timmins², T Su³, B Richard¹

¹Cleft Lip and Palate Service, Birmingham Children’s Hospital, Birmingham, ²University of Leicester, Leicester, ³University of Manchester, Manchester, United Kingdom

**Background:** Symnose was developed by Pigott and Pigott to provide an objective measurement for facial aesthetic outcome post unilateral cleft lip and palate repair. It has been subject of recent research to establish its use, in this paper the largest UK cleft database (CCUK) has been traced and measured in entirety.

**Aims:** Using the recently published cohort of Non cleft children Symnose lip symmetry measurements, it is possible to compare them with cohorts of Cleft children at age 5. The null hypothesis states that there will be no significant difference between the normal and UCLP post op photographs.

**Methods:** We acquired the CCUK cohort of UCLP post op photographs and compared their aesthetic outcome against the recently published Symnose measurements of non-cleft children. Two observers traced the lip of 240 images twice. This was then compared against the previously established normal reference.

**Results:** Intrarater agreement is ICC (90% CI) 0.86 (0.82-0.89) for rater one and 0.89 (0.85-0.91) for rater 2. Interrater agreement 0.87 (0.83-0.9). Lip mismatch % median (IQR) for CCUK was 9.34 (6.4) and for normal 4.9 (2.39). Wilcoxon signed rank test comparing CCUK to normal is p <0.001.

**Summary/Conclusion:** The intrarater and interrater agreement is comparable to that shown previously in the literature to be considered reliable. The null hypothesis is rejected, results show that UCLP post op photographs are significantly different to normals. Symnose is superior to human raters in its ability to reliably quantify this difference.
Cleft lip segmentation based on fusion of colour and contour detection
H Liu¹, Y Liu², J Shen¹, L Yuan³, B Richard⁴
¹Computer Science and Technology, Harbin Engineering University, Harbin, China, ²Computer Science, Edge Hill University, Ormskirk, United Kingdom, ³Key Laboratory of Nondestructive Testing, Nanchang Hangkong University, Nanchang, China, ⁴Birmingham Children’s Hospital, Birmingham, United Kingdom

Background: SymNose was developed by Pigott and Pigott to provide an objective measurement for facial aesthetic outcome post unilateral cleft lip and palate (CLP) repair. It represents a new way of evaluating archived CLP photographs. But it requires a human to trace the contour of lip manually. There are several methods where a computer can automatically detect and segment the lips.

Aims: Develop an algorithm that can detect and segment the lips from CLP facial images automatically. The segmented lips could then be used in CLP aesthetic outcome assessment software.

Methods: A cleft lip segmentation method based on fusing colour and contour detection was developed. Firstly, 3 basic colour-based lip detection approaches (HSV-based, LAB-based, and NMF-based) and one contour-based lip detection approach were implemented. Then, a vote-based fusion algorithm is proposed for fusing the above lip detection results. In this algorithm, each basic detection result will vote with a weight to determine whether a pixel in the CLP facial image is lip-pixel or not. The pixels with more than half of the votes are identified as lips and segmented.

Results: 25 CLP facial images are used for testing the proposed algorithm and comparing the performance with the basic methods. Three performance indexes, precision, recall, and F-score, are computed based on the manually segmentation results as ground truth. The precision, recall, and F-score of proposed method are 0.82, 0.89, and 0.84 respectively. The best results of the basic methods are 0.71, 0.80, and 0.78 respectively.

Summary/Conclusion: The test results show that the proposed algorithm works well and it can obtain the better performance than the basic methods. The segmented lips can be used by software for CLP aesthetic outcome assessment.
Three-dimensional assessment of philtral ridges in repaired unilateral cleft lip

I Ganske¹, O Langa¹, J Mulliken¹
¹Plastic and Oral Surgery, Boston Children's Hospital, Boston, United States

Background: In rotation advancement repair of unilateral cleft lip, the projection of a philtral ridge is established by the reorientation and repair of the orbicularis oris muscle layer. Laser scanning and three-dimensional photogrammetry has been used to assess the depth of the philtral dimple in unaffected patients, but has not been used to measure the symmetry and projection of the philtral ridge in patients with repaired cleft lip.

Aims: This study aims to evaluate the symmetry of philtral ridges in patients with repaired unilateral cleft lip using three-dimensional photogrammetry.

Methods: This is a retrospective review of 34 nonsyndromic patients with unilateral cleft lip ± palate who underwent repair by one surgeon at Boston Children's Hospital's from 2000-2013. Patients were divided into subgroups by phenotype: complete cleft lip/palate repaired by two-stage rotation-advancement (n=9); incomplete cleft lip repaired in a single-stage (n=10); incomplete cleft lip repaired in two stages (n=9); and microform cleft repaired by double unilimb Z-plasty (n=6). Three-dimensional images (VECTRA 3D VAM, Canfield Scientific, Parsippany NJ) were taken at a mean age of 9.25±4.54 years, prior to alveolar bone grafting. The projection of the cleft side and non-cleft side philtral ridges was measured directly above the Cupid's bow and at the mid-height of the philtrum.

Results: In patients with repaired complete cleft lip, the cleft philtral ridge was on average 0.423mm (p=0.109) lower relative to the non-cleft ridge at Cupid's bow and 0.220mm (p = 0.92) higher at the mid-height of the philtrum. Patients with single-stage repair of incomplete cleft lip had a lower philtral ridge at the Cupid's bow by 0.016mm (p= 0.914) and higher by 0.265mm (p=0.283) at the mid-height. Patients with two-stage repair of incomplete cleft lip had a cleft-side ridge that was 0.082mm (p=0.355) more projecting relative to the non-cleft ridge at Cupid's bow and 0.500mm (p=0.089) more projecting at the philtral mid-height. Patients with microform cleft lip had a 0.350mm higher philtral ridge on the cleft side at the Cupid's bow (p= 0.090) and 0.488mm (p=0.049) higher at the mid-philtrum. The only statistically significant difference was greater projection of the cleft-side philtral ridge on the cleft-side at the mid-height of the philtrum in patients with microform cleft lip.

Summary/Conclusion: Poor philtral projection is a common stigmata following repair of unilateral cleft lip. This study identified that it is possible to construct a philtral ridge that is not significantly different in projection from the non-cleft side. There was a trend toward greater projection of the cleft-side philtral column at the mid-height of the lip compared to the Cupid’s bow, possibly related to more eversion of the orbicular repair at that level. Greater muscle thickness has been documented by ultrasonography following nasolabial adhesion. In this study, there were no significant differences in cleft-side philtral column height in patients with incomplete cleft lip following a single or a two-stage repair.
The effects of nasoalveolar molding on nasal proportions at the time of nasal maturity
S Maliha1, R Kantar1, M Gonchar1, B Eissemann1, D Staffenberg1, P Shetye1, B Grayson1, R Flores1
1Hansjörg Wyss Department of Plastic Surgery, NYU Langone Health, New York, United States

Background: Nasoalveolar molding (NAM), a form of passive presurgical orthopedics, has been associated with greater nasal symmetry in patients with unilateral cleft lip and palate (UCLP) through several studies by multiple institutions. However, these reports have been limited by short to medium-term analysis and no study to date has been conducted in patients at the age of nasal maturity.

Aims: This study assesses the effects of NAM versus no-NAM on nasal morphology in patients with UCLP at the time of nasal maturity.

Methods: A retrospective, single-institution review was conducted on all non-syndromic patients with UCLP. Nasal maturity was defined as 14 years of age. Inclusion criteria included age greater than or equal to 14 years, unilateral cleft repair at time of infancy, and adequate photography taken at or past the age of nasal maturity and prior to definitive rhinoplasty. Exclusion criteria included age less than 14 years, syndromic diagnosis, incomplete clinical records, and rhinoplasty prior to nasal maturity. Photos utilized were taken prior to any form of orthognathic surgery. Ten parameters were measured on frontal, worm’s eye view, and sagittal view photographs: alar base width, columellar height, columellar angle, nasal tip deviation, cupid’s bow deviation, alar cant, nasolabial angle, nasofacial angle, vertical alar height, and alar height angle. These measurements were performed twice using the Dolphin Imaging Software for establishment of intrarater reliability. Subjective analysis was achieved through completion of the Asher McDade grading scale (nasal form, nasal symmetry/deviation, nasal profile, and vermillion border) by three expert cleft practitioners using frontal, worm’s eye view, left profile, and right profile photographs.

Results: 7 of 10 nasal parameters were significantly more symmetric in patients who had undergone NAM compared with no-NAM: Columellar height (0.88 ± 0.09 vs. 0.73 ± 0.17; p=0.002), columellar angle (3.55 ± 2.64 vs. 1.61 ± 7.17; p=0.01), alar cant (2.57 ± 2.54 vs. 4.50 ± 2.99; p=0.032), vertical alar height (0.92 ± 0.10 vs. 0.83 ± 0.08; p=0.005), alar height angle (3.70 ± 2.39 vs. 5.54 ± 2.85; p=0.032), nasofacial angle (21.72 ± 5.29 vs. 26.43 ± 5.65; p=0.009), and nasolabial angle (87.82 ± 10.91 vs. 68.17 ± 17.43; p=0.001). Asher McDade analysis of NAM vs. no-NAM groups revealed that patients who underwent NAM had significant improvement in all scores including: nasal form (2.02 vs 3.03, p = 0.001), nasal symmetry/deviation (2.1 vs 3.03, p = 0.001), nasal profile (2.12 vs 2.92, p = 0.001), vermillion border (1.63 vs 2.70, p < 0.001), as well as overall score (1.97 vs 2.92, p < 0.001). Intrarater reliability was rated as good to excellent for all measured parameters.

Summary/Conclusion: The use of pre-surgical NAM during infancy improves nasal symmetry and nasal proportions at the time of nasal maturity.
**OS 44.12**

Comparative evaluation of aesthetic outcomes in unilateral cleft lip repair between mohler’s and fisher’s repair techniques – a prospective, randomised observer blind study.

G Deshpande1,2, S Natraj1, M Deshmukh1

1Oral and Maxillofacial Surgery, MGM Dental College and Hospital, Navi Mumbai, India, 2Plastic and reconstructive Surgery, PennState hershey School of Medicine, Hershey, United States

**Background:** Cleft Lip and palate is the most common congenital anomaly of face (CFA), affecting 1 in every 600 new born babies worldwide. Globally, the birth prevalence for cleft lip (CL) with or without cleft palate (CP) ranges from 3.4 to 22.9 per 10,000 live births, and for CP alone ranges from 1.3 to 25.3 per 10,000 live births2. The size of a cleft reflects differing contributions of tissue deficiency and/or tissue displacement. Cleft-lip repair has evolved largely in the past century to its modern form and has seen both revolutionary changes and subtle technical refinements. “All cleft lip surgeons have their favourite surgical technique for repairing the unilateral cleft lip. It is usually a hybrid of training experience and imagination.” Although this is true to a certain extent, basic principles of cleft lip repair have been laid in the past which should be followed. Over the past five decades, several different repair techniques have gained greater popularity over others. This study makes a correlation between pre-operative cleft severity and post-operative aesthetic outcome of Mohler and Fisher cleft lip repair techniques.

**Aims:** To make a comparative evaluation of aesthetic outcomes in unilateral cleft lip repair between Mohler and Fisher cleft lip repair techniques.

**Methods:** A total of 50 patients with unilateral cleft lip with or without cleft palate were taken as the sample size for the study. Patients were randomised consecutively irrespective of age, gender, caste or creed. All necessary pre-operative photographic records were maintained for these patients. Cleft severity was evaluated based on the Unilateral Cleft Lip Severity Index. Post-surgical aesthetic outcome was evaluated based on 4 components by The Surgical Outcomes Evaluation Scale by 3 laymen to eliminate bias.

**Results:** There was a statistically significant negative correlation between cleft severity and aesthetic outcome. A one-way analysis of covariance was performed to determine the relation between the technique of repair and the aesthetic outcome using technique as independent variable and aesthetic outcome as dependent variable with the means adjusted using cleft severity as the covariate. But there was no significant difference in the mean aesthetic outcome and technique of repair even when means were adjusted for initial cleft severity.

**Summary/Conclusion:** Although Fisher’s technique gave an overall better aesthetic outcome than Mohler’s technique of cleft lip repair, it could not be ascertained if any of the technique would be better for a particular grade of cleft severity.
Parent experiences of the ‘parent led, therapist supervised, articulation therapy (plat) programme’ for cleft articulation therapy intervention

D Sell¹, C O’Rourke², K Powell³, L Deasy⁴, T Sweeney⁵

¹Centre for Outcomes and Experience Research in Children’s Health, Illness and Disability, Great Ormond Street Hospital NHS Foundation Trust, London, United Kingdom, ²Research Department, Amarach, Dublin, Ireland, ³Children’s Speech and Language Therapy, Haringey NHS Foundation Trust, London, United Kingdom, ⁴RIP, previously Speech at Home, ⁵Speech at Home, Dublin, Ireland

Background: Although parent training/involvement has positive outcomes in early communication skills in cleft palate and non-cleft speech sound disorders, little is known about parents undertaking intervention for cleft speech disorders. Our RCT set out to compare parent led articulation therapy (PLAT) with standard intervention for children with a cleft-related speech disorder.

Aims: This study aims to report parents’ experiences of PLAT which included training followed by intervention with their child supported by a specialist therapist and connected health (FaceTime).

Methods: Forty-six children, aged 2.9-7.5, were included in a two-phase, two-centre randomized controlled trial. In the parent training arm, parents attended an in depth 2-day training course, received a detailed speech programme and conducted home based SLT for fifteen minutes a day, five days/week, for 12 weeks, making decisions about what to do and how to do it. They were supported by a specialist therapist through two FaceTime sessions at weeks 3 and 9 and one face-to-face session at week 6. Post intervention, the parent training group completed a questionnaire which investigated their experiences of the 12 week programme. Nineteen parents (83%) participated in telephone interviews or focus groups conducted by independent Psychologists/Speech and Language Therapists. Analysis was carried out by an independent psychologist using thematic analysis.

Results: Parents reported how they “learned a lot about speech problems and speech therapy strategies”. Parents praised the content, flexibility and pace of the intervention programme devised by the therapist. They commended FaceTime, particularly when tasks were not working. Challenges included how they felt “somewhat daunted when taking on the course” but retrospectively “were very happy to have participated”. They fed back that they were at times overwhelmed by the materials and that time was needed to plan and establish the therapy routine. They expressed concern as being seen by their child as a teacher. Making PLAT a routine was difficult especially for working parents. Parents provided excellent critical feedback on how to improve training and fine-tune the therapy programmes. Most parents reported an improvement in their child’s speech and an increase in their child confidence, which reinforced their own sense of well-being.

Summary/Conclusion: Parent experiences of the Parent-Led Articulation Therapy Programme were largely positive. They gained new knowledge and skills about speech and intervention. Parents reported an increased sense of empowerment and control in relation to their child’s therapy. They mostly reported improvements in their child’s speech and confidence over the 4 month period. The focus groups and interviews provided the researchers with constructive criticism on how the programme could be improved for the future.

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**Surgical and speech outcomes in children with cleft lip and/or palate with van der woude syndrome: a comparative study**

M Farid¹, C Hillyar ², L Cafferky ³, B Fitzpatrick³, R Slator¹

¹Cleft and Craniofacial Surgery, Birmingham Women’s and Children’s Hospital, Birmingham, ²Barts and The London School of Medicine and Dentistry, London, ³Speech and Language Therapy, Birmingham Women’s and Children’s Hospital, Birmingham, United Kingdom

**Background:** Van der Woude syndrome (VdW) constitutes 2% of cleft lip and/or palate (CL/P) cases and has an incidence of 1 in 50,000 worldwide. The mode of inheritance is autosomal dominant related to a mutation in IRF6 and there is variable phenotypic expression.

**Aims:** Previous studies have shown a greater incidence of bilateral cleft lip and palate (BCLP) in children with CL/P and VdW syndrome than in children with non-syndromic CL/P. The aim of this study was to examine speech outcomes for children with VdW and CL/P in terms of nasality, cleft speech characteristics (CSCs) and secondary speech surgery, and to try to identify whether the high incidence of BCLP contributed to worse speech outcomes.

**Methods:** All children with CL/P in our centre have a standardised speech assessment at the age of 5 years which includes information about secondary surgery for speech. A retrospective cohort study of a consecutive series of children with VdW and CL/P over 10 years was conducted using this data. Informed consent was not required. A control group of non-syndromic CL/P patients were compared to the VdW group for speech outcomes (nasality and CSCs), and for rates of secondary speech surgery at age 5 years using two-tailed Mann-Whitney and Chi-square tests. The differences between the two groups were also analysed at the level of cleft type (unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP) and cleft palate (CP)).

**Results:** There were 18 children in the VdW group and 168 in the non-syndromic CL/P group. The cleft type distribution for children with VdW was UCLP 28%, BCLP 33%, CP 39%, and in the non-syndromic group UCLP 36%, BCLP 16%, CP 48%. Overall, the nasality outcome was worse in the VdW group compared to the non-syndromic group (p = 0.0018). This difference arose from the UCLP subgroup. VdW patients also had a higher rate of secondary surgery compared to the non-syndromic group (p<0.0001). This difference arose from the CP subgroup (p=0.0016). There was no overall difference in articulation between the 2 groups, and no differences in articulation between the different cleft types.

**Summary/Conclusion:** Children with CL/P and VdW syndrome in this study, as in previous studies, had a higher incidence of BCLP than the group of non-syndromic children. They presented with worse speech outcomes for nasality and secondary surgery compared to children with non-syndromic clefts. Analysis of the outcomes by cleft type suggests that differences in velopharyngeal competence at age 5 years between those with VdW syndrome and those with no syndrome arise from the cleft types, UCLP and CP. No differences between the two groups were identified in children with BCLP. Interestingly, despite poorer velopharyngeal function, outcomes for cleft speech characteristics were similar between the two groups. We consider the difficulties in studying outcomes in children with CL/P and rare syndromes, and whether underlying anatomical features for VdW clefts may be the reason for poorer outcomes.
Assessment and comparison of the speech in patients with complete unilateral cleft lip and palate operated by two palatoplasty techniques

Y Markova¹, Y Anastassov², K Gigov², R Velikova², M Kazakova¹

¹ALA – Association for Facial anomalies of Bulgaria, ²Plastic and craniofacial Unit, Medical University of Plovdiv, Plovdiv, Bulgaria

Background: Exploration of speech production, the standard common protocol by the specialists working with this contingent, and the development of a commonly agreed speech assessment protocol is a necessity that has arisen in our recent practice. The data collected in the Electronic Medical Record for Facial Anomalies in recent years is significant and their analysis will optimize the work of the Team.

Aims: To summarize and compare the collected speech production data in two patient groups and to assist the surgeon in the evaluation of the surgical technique.

Methods: The study included 100 patients with unilateral complete cleft lip and palate between 4 and 15 years. They were operated in the Plastic and Craniofacial Unit and divided into two groups according to the operating technique used: I group (80) - operated by Veau-Wardill-Kilner technique with transversal section of the nasal layer and the II group (20) – with releasing lateral incisions technique (modified palatoplasty with transversal section of the nasal layer). The study involves auditing and evaluating patient speech video records by summarizing the data and making a statistical analysis. Records contain nonstandard material - counting, days of the week, naming random pictures. The study is conducted in two stages. In the first stage 11 speech therapists (Group of Trusted SLP by ALA) aimed to develop a consensus protocol for a sufficient review and an objective evaluation of the video records. In the second stage 4 speech therapists (Group of Independent Experts) blinded made speech assessment independently of each other and with confidentiality review the records of all patients without having other data except hearing the records. The protocol data completed by the 4 experts for the entire patient group was summarized and compared, and statistical analysis of the speech output data was performed by comparing the speech output in the two operative techniques.

Results: There is no significant difference in speech outcomes between the two groups according to the operating technique with regard to measures of consonant inventory, place of articulation, manner of production, nasal resonance, nasal emission, cleft type characteristics.

Summary/Conclusion: This work defines the essential criteria for the evaluation of speech, compares the speech outcomes in the two operative techniques, confirms the already adopted model for standard case description and use of the Electronic Medical Record. The speech test used so far, which includes counting to ten and repeating the days of the week, is not sufficiently reliable in accurate speech assessment. It is necessary to elaborate and standardize a more detailed instrument, consistent with the phonetic features of the Bulgarian language.
Speech outcome in young adults born with bilateral cleft lip and palate - a descriptive and comparative study
P Boivie¹, O Lundwall², R Stenlund², C Persson²
¹Department of Plastic Surgery, Sahlgrenska University Hospital, Institute of Clinical Sciences, ²Institute of Neuroscience and Physiology, Division of Speech-Language Pathology, Sahlgrenska Academy, Gothenburg, Sweden

Background: Bilateral Cleft Lip and Palate (BCLP) is a rare malformation and BCLP is a heterogenous group. The surgical treatment of BCLP is often based on experience from treatment of Unilateral Cleft Lip and Palate (UCLP). Few previous studies have evaluated the speech outcome of young adults born with and treated for BCLP.

Aims: The aim of the present study was to evaluate speech in young adults born with BCLP and to compare speech outcome with age-matched patients born with UCLP, and with a with age-matched control group (born without Cleft Lip and Palate).

Methods: Non syndromic patients with BCLP, born between 1989 and 1993 were included in the study. Patients with incomplete bilateral cleft lip was included, however all patient had bilateral alveolar clefts. All patients had completed the standard program of care (19 year) and all surgery was performed at Sahlgrenska University Hospital, with two-stage closure of the cleft palate. The BCPL cohort was compared to age-matched patients, born with UCLP and to the control group born without CLP. Speech recordings of the Swedish Articulation- and Nasality Test (SVANTE) and Swedish Intelligibility test (SWINT) collected at 19 years was used for evaluation. Two listeners transcribed all target sounds from SVANTE by phonetic transcription and the words from SWINT by orthographic transcription. Velopharyngeal function was rated by three speech and language pathologists and nasalance score was obtained with Nasometer™.

Results: Participants born with BCLP had significantly lower results in articulation, velopharyngeal function and intelligibility in words, compared to participants without cleft. Participants born with BCLP had significantly more nasal air leakage and anterior articulation errors compared to participants born with UCLP. The control group did not differ substantially.

Summary/Conclusion: The results indicate more deviant articulation and velopharyngeal function in young adults born with BCLP, compared to young adults without cleft. However further research regarding the speech outcome of patients born with BCLP is required.
Low-level laser therapy in recovering the orofacial sensitivity after orthognathic surgery on the individuals with repaired cleft lip and palate: a pilot study.

C De Almeida¹, R Martinelli de Carvalho ², J Carlos da Cunha Bastos Junior², R Paciello Yamashita¹

¹Laboratory of Physiology, ²Department of Oral and Maxillofacial Surgery, Hospital for Rehabilitation of Craniofacial Anomalies , University of São Paulo, Bauru - SP, Brazil

**Background:** Individuals with cleft lip and palate frequently present dentofacial alterations and need orthognathic surgery for the functional and aesthetic correction. However, postoperative disturbances of the orofacial sensitivity are common after the surgery. The bioestimulation of the tissue through the low-level laser therapy has been used in the regeneration of the neural sensitivity after the orthognathic surgery.

**Aims:** To investigate the effectiveness of the low-level laser therapy in the early recovering of the orofacial sensitivity after the orthognathic surgery in individuals with repaired cleft lip and palate.

**Methods:** This study was approved by the institutional review board. Pilot study randomized controlled trial carried out with 12 patients with repaired cleft lip and palate, aged 23 years on average, undergone to orthognathic surgery, divided into 2 groups: experimental group (6 patients) and control group (6 patients). All the patients were undergone to the orofacial myofunctional evaluation for the sensitivity test of the tongue, lips, incisive papilla and mental region, 2 days before (preoperative), and 3 months later (post-operative) the surgery. The sensitivity test was performed through theesthesiometer consisting of a set of six monofilaments of colored nylon and different diameters which touch the tested area and produce different pressures. A score is assigned for each monofilament from 0 (thinner filament) indicating best sensitivity to 5 (thicker filament) indicating worse sensitivity. Only experimental group was undergone to low-level laser therapy after inferior alveolar nerve exposition during orthognathic surgery, and in four post-operative sessions with interval of 12 hours between each application. The results were analyzed through descriptive analysis considering the percentage of changes of the orofacial sensitivity in both groups after surgery.

**Results:** Before surgery, all patients (6) of the experimental group presented adequate sensitivity in tongue, lips, mental region and cheeks and, 2 (33%) patients presented mild altered sensitivity on the papilla. After surgery, there was an improvement in the sensitivity of the papilla area. One (16%) patient changed to adequate sensitivity and one maintained mild altered sensitivity. In the control group, before surgery, all patients (6) presented adequate sensitivity in tongue, lips, mental region and cheek, and 2 (33%) patients presented mild/moderate altered sensitivity in papilla. After surgery a higher number of changes in the orofacial sensitivity was observed in the control group as compared to the experimental group. Three (50%) patients changed the sensitivity in the papilla area: 1 (6%) changed from mild to mild/moderated altered sensitivity and, 2 patients (33%) changed from adequate to mild/moderate altered sensitivity. In the mental region, 66% (4) patients presented mild altered sensitivity. One of these patients also presented mild altered sensitivity in papilla and cheek.

**Summary/Conclusion:** These preliminary results showed that low-level laser therapy was effective in recovering orofacial sensitivity in a short term (3 months) after orthognathic surgery in patients with repaired cleft lip and palate.
Practical use of the electronic medical record for facial anomalies in Bulgaria (EMRFA) for patients and specialists: a 5 years experience

Y. Anastassov¹, K. Gigov¹, Y. Markova², R. Khater¹, P. Petrov³, R. Velikova², H. Zhelyaskov¹, A. Guylev⁶, T. Petkova⁶, P. Tsarvulanova², K. Bojikova⁷, N. Slaninkova⁷, N. Pareva⁷, M. Kazakova⁷

¹Plastic and craniofacial Unit, MEDICAL UNIVERSITY OF PLOVDIV BULGARIA, Plovdiv, ²ALA-Asociation facial anomalies of Bulgaria, Sofia, ³Maxillofacial Surgery, ⁴Neurosurgery, Medical University of Plovdiv, Bulgaria, ⁵Ear nose and Throat, ALA-Asociation facial anomalies of Bulgaria, ⁶Imaging Dentistry, Medical University of Plovdiv, Bulgaria

**Background:** The EMRFA has started in 2013 enabling the Centre to control the work of the trusted specialists on a national level and to communicate with the patients improving their follow up.

**Aims:** This study analyse the level of use of this tool by the specialists and the patients –parents on a national level from 2013 to 2018.

**Methods:** In the end of 2018 are registered a total of 1032 patients, 836 have personal electronic address. The trusted specialists in the country are 65, 7 of them are in the Centre. The EMRFA gives us a variety of dynamic information and also statistical analysis of a concrete question (new cases born in the country each year, the number of posts from patients-parents and specialists, risk factors, loss of view cases, etc.)

**Results:** The number of posts from patients and parents is 890 and 7877 from the specialists. The total number of posts from patients in the period 2013-2018 are 958 (394 contain uploaded pictures or videos). For the same period the number of posts from the specialists outside of the Centre is 216 (137 of these posts contain uploaded pictures or videos). Analysing the psychosocial risk factors, we have 20 patients with high and 60 with medium risk. 1 patient older than 1 year has not been operated for his cleft lip, 9 had non operated palate and were aged more than 1.6- to 3 years and 14 patients did not come for consultation between 1 and 4 years of age.

**Summary/Conclusion:** In this study we have seen how increase the use of the EMRFA by the users - parents-patients and the specialists. We discuss the use of some new functionalities as individual reminders (tasks) and the reminders by age for a better documentation of each case.
The effects of perioperative pain management strategies on recovery after primary palatoplasty: a multi-surgeon retrospective study

I Ganske¹, O Langa¹, N DiTullio¹, J Meara¹, C Rogers-Vizena¹, J Mulliken¹
¹Plastic and Oral Surgery, Boston Children's Hospital, Boston, United States

Background: With a growing opioid crisis in the United States, much focus has been paid to the ways in which pain can be managed with less narcotic. Preference in perioperative pain management following palatoplasty varies among cleft surgeons. Assessment of pain in infants is difficult despite established severity scales. Thus, strategies for postoperative pain management are typically guided by a surgeon’s preference and experience.

Aims: The purpose of this study is to define the perioperative pain management variables that affect demand for postoperative narcotic and decrease time to resumption of oral intake.

Methods: This is a retrospective chart review of non-syndromic patients undergoing primary palatoplasty between 8-14 months of age from 2015-2018 at Boston Children’s Hospital. Pain management variables included: type of local anesthetic, preoperative ropivacaine pterygopalatine ganglion nerve block, placement of a palatal pack, and postoperative use of ketorolac, dexamethasone, and nursing controlled analgesia (NCA) opioid dosing. Outcome measures for postoperative pain included time to resumption of oral intake and morphine equivalence (mg/kg/hour) for each patient. Regression analyses were performed for each variable.

Results: Of the 102 patients who met study criteria, palatal phenotypes were: Veau I (30%), Veau II (21%), Veau III (31%), Veau IV (16%), and submucous cleft (2%). There was no statistical significance between cleft severity and postoperative narcotic consumption or time to resumption of oral feeding, except between Veau I and II, where the morphine equivalence (mg/kg/hr) was significantly higher in Veau II patients than Veau I patients, likely reflective of differences in pain following two-flap versus Furlow palatoplasty techniques. Neither age nor weight had a significant effect on morphine equivalents used or time to resumption of oral intake. Nor was the type of local anesthetic injection at time of palatoplasty. The difference in postoperative opioid requirement was significantly lower in patients who had an intraoperative ropivacaine peripheral nerve block. Postoperative pain management variables such as use of ketorolac or dexamethasone or the placement of a palatal pack, were not statistically significant in reducing opioid use or time to resumption of oral intake. Nursing controlled analgesia (NCA) was associated with nearly 60% higher quantity of administered post-operative narcotic with no significant difference in time to resumption of oral intake or duration of inpatient stay.

Summary/Conclusion: This study showed that for primary palatoplasty, several perioperative pain management techniques produced comparable times to oral intake and quantities of post-operative narcotic. NCA is not recommended as a standard regimen for post-operative palatoplasty patients. Other strategies achieve the same level of patient comfort with equivalent time to resumption of oral feeding and readiness for discharge. Further studies are needed to assess the risks, benefits, and costs of performing a peripheral nerve block at the time of palatoplasty.
An analysis of phonatory and articulatory results in two different surgical approaches for unilateral cleft lip and palate

P Todaro¹, F Aceti¹, I Fontana¹, L Autelitano¹, M Meazzini¹, A Rezzonico¹
¹Smile House S.Paolo Hospital, Milano, Italy

Background: Until 2006 at the San Paolo Hospital in Milan, UCLP patients were treated with the “two stages” protocol (lip and soft palate at 6 months – hard palate at 24 months). Since 2006 UCLP patients with a cleft width narrower than 10-12 mm at the hard palate and gap have been treated according to the “all in one” protocol with simultaneous closure of hard palate together with soft palate (-9 months).

Aims: The aim of this study is to compare phonation and articulation at 5 years among subjects treated with two different surgical approaches: the “all in one” protocol and the “two-stage” protocol.

Methods: For this retrospective study, 112 UCLP patients were selected. Patients with syndromes and multiple malformations were excluded. All patients were operated between 1997 and 2012 by the same senior operator at the San Paolo Hospital. Speech outcomes were evaluated by three speech and language pathologists experienced in cleft lip and palate, using the GOS.SP.ASS ‘98 protocol modified for the Italian language. The parameters assessed were resonance, nasal emission, turbulence, grimaces, articulation, intelligibility and acceptability. As far as the phonation was concerned, the patients with velopharyngeal insufficiency were given special attention. The patients were divided into two groups: the first group consisting of 61 subjects treated with the “all in one” protocol, and the second group consisting of 51 subjects treated with the “two-stage” protocol. For this study the perceptual speech evaluation was carried out at the mean age of 5.3 years. If VPI was present, nasal endoscopy and videofluoroscopy were performed as additional examinations.

Results: In the two observed groups no statistically significant difference was observed in phonatory and articulatory parameters. From a qualitative point of view, the amount of VPI is lower in the patients group treated with “all in one” protocol (6.6%) than in the two stage one (13.7%). Both the percentages comply with data in the literature.

Summary/Conclusion: This study shows similar results in phonation and articulation in both groups; considering the reduction of burden of care due to a single surgery, from a logopedic point of view, we recommend this approach. Nevertheless this result must take into consideration the maxillary growth that may be affected by an early closure of the hard palate, for which a later assessment is needed.
Family-centered care, how to apply the model to cleft nursing?
C Jung\textsuperscript{1}, A de Buys Roessingh\textsuperscript{1}, O El Ezzi\textsuperscript{1}
\textsuperscript{1}Pediatric Surgery, University Hospital, Lausanne, Switzerland

\textbf{Background:} The role of the clinical nurse specialists (CNS) in multidisciplinary teams dealing with facial clefts is evolving in Europe. It covers several aspects including family support. These nurses offer a point of contact in the hospital for the families. According to Campbell (2003) and Weish, Fisher and Baird (2002), the illness of a family member can create stress in the family and cause disorganization. The announcement of the arrival of a baby with a cleft lip and/or palate, for instance, can have repercussions on the whole family. This is when the CNS should provide family-centered care and not just individual care. In order for the clinical nurse specialists to define and widen their role, structure their intervention, and collaborate with other team professionals, it is necessary to refer to a model of care, for instance the Calgary models.

\textbf{Aims:} Describe how CNS can use the Calgary models to improve family support and interprofessional collaboration.

\textbf{Methods:} Calgary models are useful for evaluation and intervention in family systems. It guides the CNS in the collection of data about family structure, development and functioning. It helps to develop hypotheses and plan interventions. The role of the CNS is also to strengthen family skills by working on the process of adaptation and promoting autonomy. From the first meeting with the parents the CNS has to investigate strengths and problems within the family. It is therefore necessary to pay attention not only to the parental couple but also to the whole family system. With the help of genogram and ecocard, the nurse can evaluate the composition, and medical history of the family, its social interactions and communication, and its beliefs and resources in relation to the event that they must face. In prenatal and pre-operating consultations this approach makes it possible to find the right way of addressing each family individually and to detect difficulties of adaptation. The need to refer the situation to the psychologist of the team is also evaluated.

\textbf{Results:} The use of this model permits a structured analysis of the family that the CNS meets, as well as a systematic screening for difficulties of adaptation. The model can be adapted to the context, and the nurse will judge which category has to be thoroughly evaluated. Parents feel listened to and understand that their reactions are normal. They can express their fears and expectations. The referral to the psychologist or another professional of the team will be faster and more personalized.

\textbf{Summary/Conclusion:} The whole family must be considered when a baby is born with a cleft. The CNS need a model to structure their approach. Personalized care enhances the relationship of trust between the family and the multidisciplinary team. The Calgary models appear to be a tool well adapted to the needs of specialist cleft nurses. It would be interesting to evaluate the satisfaction of families in regard to this support in a future study.
Pierre robin sequence patients' osa - the role of positioning treatment
H Kukkola¹, T Kirjavainen¹, P Vuola², A Saarikko³, P Salminen³, M Seppä-Moilanen⁴
¹Pediatrics, ²Plastic surgery, ³Pediatric surgery, ⁴Pediatric neurology, HUS, Helsinki, Finland

**Background:** Pierre Robin sequence (PRS) is referred to the association of micrognathia, glossoptosis, and airway obstruction with or without cleft palate. PRS is one of the most common birth defects in infants. Depending on the severity of PRS it can cause a variety of breathing and eating disorders. Airway obstruction may lead to obstructive sleep apnea (OSA). When the child is in the supine position, micrognathia and glossoptosis cause the tongue to fall back into the hypopharynx or to be trapped in the cleft palate, which leads to airway and feeding problems. First line treatment to resolve these problems has been prone or lateral positioning. Prone positioning is based on the hypothesis in which the narrow pharyngeal space in PRS can be corrected by the effect of gravity moving the mandible forward in infants sleeping prone. Prone positioning allows the tongue to move forwards thus freeing the airway. Recommendations for sleep position for infants has changed significantly in the 1990s. Nowadays infants are recommended to be put to sleep in supine position because it reduces the risk for sudden infant death syndrome (SIDS). The impact of positioning treatment to polysomnography results has not been widely described. The PSG studies priorily done have not shown objectively that position treatment is a safe option and can be recommended for infants with PRS.

**Aims:** We studied the effectiveness of sleep positioning in the treatment of OSA in infants with Pierre Robin Sequence. Management of OSA in these infants vary between treating centers and the severity of OSA, and include prone or lateral positioning, high flow pressure support, nasal CPAP, nasopharyngeal airway tube (NPA) and in severe cases tracheostoma. Positioning has known to be sufficient treatment to resolve breathing difficulties for most of these infants.

**Methods:** We evaluated during 2009 – 2018 done polysomnography (PSG) recordings’ results of PRS patients. PSG recordings are routinely done in our clinic in three different positions supine, prone and lateral if possible. We evaluated positioning’s significance to obstructive apnea hypopnea index (OAHI), to obstructive desaturation index (ODI3) and to work of breathing (WOB) in total sleep time and in REM sleep. Patients were collected from Children’s hospital’s sleep laboratory’s archive based on polysomnography referrals. The local ethics committee approved the study protocol. The statistical analysis was performed with IBM® SPSS® Statistics software version 22. We performed all the analysis as pairwise comparisons, for which we chose to use the repeated measures of ANOVA. The patients acted as their own controls. The level of significance was set at $P < 0.05$.

**Results:** The New Children’s hospital treats approximately 90% of PRS patients in Finland. We had 67 patients that had their first PSG done under the age of 3 months. Sixtyfive patients had cleft palate. We found that OSA is very common in this patient group. We found that there is statistically significant difference in the PSG parameters (OAHI, ODI3) depending on the position of the infant. The PSG measurements showed that prone position treats OSA in these patients effectively.

**Summary/Conclusion:** OSA is a key problem in infants with PRS. Most infants can be managed by positioning treatment alone. Our study shows the efficacy and safety of positioning treatment objectively.
The cleft nurse specialist: a key building block in the cleft team
S Martin¹, E Slevin², C Hill³
¹Cleft Department, Royal Belfast Hospital for Sick Children, ²RBHSC, Belfast, United Kingdom, ³Cleft Department, RBHSC, Belfast, United Kingdom

Background: The cleft nurse specialist (CNS) plays a key role in counselling parents from the diagnosis, through birth and along the pathway of cleft treatment for years to come.

Aims: To determine the benefit of the cleft specialist nurse in the regional cleft unit.

Methods: The cleft database was used to locate babies born in 2010/2011 and 2013/2014. Parents were contacted by phone and completed a questionnaire on the care they received following the diagnosis of CLP (Antenatal/postnatal) and the support they received from the cleft team in the early days and around the time of primary surgery.

Results: Parents of 38 babies completed the survey. In the 2010/2011 cohort only 21% of parents had an antenatal diagnosis compared to 47% in 2013/2014. 2011/2012: 3UCLP, 3 BCLP, 4 Lip only, 9 palate only. 2013/2014: 5 UCLP, 7 BCLP, 7 palate only. 2013/2014: 84% were counselled by the CNS following diagnosis. 53% were seen within 48 hours and 47% 3-7 days after birth. 2011/2012: 68% were counselled by a surgeon. The majority (42%) were seen >1week after birth, with some up to 6-weeks later. Parents in 2013/2014 felt more supported by the cleft team throughout pregnancy and the early days, with home visits being particularly advantageous.

Summary/Conclusion: The introduction of the CNS to the cleft multi-disciplinary team has significantly improved the pathway for parents and is a key link with the wider cleft team. Counselling occurs at an earlier stage and enables preparation for the birth. Home visits give added support to new parents who may struggle in the early days.
P-001
The use of acellular dermal matrix in primary palatoplasty with intravelar veloplasty technique
H Abdali¹, H davari¹
¹craniofacial and cleft research center, Isfahan university of medical sciences, Isfahan, Iran, Islamic Republic Of

Background: There are different methods in repairing cleft palate with variable rate of fistula and recurrent. Intravelar veloplasty may be consider one of the best techniques with low fistula occurrence. In newer technique, the use of Alloderm has considerably improved the outcome and lowered the rate of fistula occurrence.

Aims: To investigate the use of Alloderm or primary palatoplasty

Methods: This prospective study compared forty two patients as a case group who underwent the new technique with Alloderm with forty two patients as a control group undergoing the same technique without the use of Alloderm. A 6 month post-operation follow up was undertaken for each group of patients for comparison.

Results: Result will be presented which discuss the occurrence of fistula and other related issue

Summary/Conclusion: The use of Alloderm in palatoplasty is without any problem and safe and it is with reduce of risk of fistula formation post operation.
P-002
Final surgical touches in the adult cleft lip patient.
B Abdulrauf
1PLASTIC SURGERY, KING FAISAL SPECIALIST HOSPITAL & RESEARCH CENTER, JEDDAH, Saudi Arabia

Background: In this presentation, we would like to share a case series of Adult Cleft lip and Nasal Correction. There are 2 types of such Adult cleft lip patients frequently encountered for consultation: First type are those asking for specific area correction, usually the nose or lip or both. The second type are those who had very poor care early on (Neglected category), asking for major external correction or make over.

Aims: From this case series of Adult Cleft patients in our personal experience, 1. It is important to be able to analyze the problem(s), prioritize them keeping in mind the patient's wishes. 2. Identifying the patients who need Nasal correction and pursuing it prior to any lip final correction. 3. Finding out what techniques consistently provide the best possible cosmetic outcome.

Methods: Approximately 14 Adult Cleft patients would be shown in part during this presentation. 7 underwent Unilateral cleft Rhinoplasty; 4 underwent only Lip reconstruction with Abbe flap, and 3 neglected cases of Bilateral cleft underwent Nasal Reconstruction followed by Abbe flap reconstruction.

Results: 1. The Unilateral Cleft Rhinoplasty is a technically demanding procedure due to the pre-existing congenital asymmetric memory of the tissues, therefore there is a limit how much improvement can be achieved. 2. Abbe Flap provided an excellent aesthetic change in all patients who had it. 3. In the Bilateral neglected cases, the protocol of using probalial tissue to build the columella (with the Rib graft) as first stage followed by Abbe flap was found to be optimum to follow.

Summary/Conclusion: 1. In adults, Nose and Lip secondary correction must always be staged, beginning with the Nose. 2. Adult Unilateral Cleft Rhinoplasty is more challenging compared to the Bilateral one due to difficulty in achieving symmetry. 3. Rib Graft as a columellar strut in essential in majority of Adult Cleft Rhinoplasty. 4. Abbe Flap with its various versions remains to be an excellent operation for optimum aesthetic outcome in the Bilateral cleft lip reconstruction.
Dental treatment of cleft patients under general anaesthesia
M Alhassani1, J Smallridge1
1Addenbrooke's Hospital, Cambridge, United Kingdom

Background: The case records of children with clefts undergoing dental treatment under general anaesthesia (GA) across UK cleft centres were reviewed. The characteristics of the admission and dental care undertaken were recorded. Findings were assessed against standards of care from the Royal college of Anaesthetics, Royal college of Surgeons, Department of Health and Service Specification for cleft care relating to dental treatment for children under GA.

Aims: To review and compare the characteristics of dental treatment under GA among children with clefts from three regional cleft centres in the UK.

Methods: This was a retrospective review of case records over a 24 month period relating to hospital admissions for GA within regional cleft teams. Results were analysed overall for compliance with the gold standards and by region to see if care was equitable across the UK.

Results: Times on waiting list varied from 1-32 weeks, most patients were treated as day cases, the mean age at time of treatment was 6.3 years, children were having to travel up to 91 miles for this type of care, treatment was planned by specialist/consultants in Paediatric Dentistry. A full range of dental treatments was undertaken. 35% of teeth were treated by extraction.

Summary/Conclusion: Children were having to travel long distances to access appropriate care but the care received compared favourably with the standards of good care as per the guidelines. The average age at admission reflects the fact that a lot of these children were seen at 5 years for the first time by the paediatric dental services and the high percentage of teeth needing extraction demonstrates a need for closer monitoring of these vulnerable children.
Cleft lip repair with geometrical technique. A new technique.
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

**Background:** The Geometrical technique, using a new form of measurement and adding elongation processes, manages to lengthen the lip and avoid making incisions that cross the filtrum, the crest of the filtrum and other areas. Without making unnatural incisions Geometrical Technique achieves the elongation of the cleft lip with adequate symmetry and balance.

**Aims:** Within the technique a new conceptual element is incorporated to deepen the filtrum and exalt the delicacy of the reconstruction of the unilateral lip with straigh line scar.

In case of bilateral cleft of the lip, the geometric technique offers the possibility of achieving a narrow filtrum, with simultaneous elongation of the columella and the typically short prolabio.

**Methods:** Once the consents of the parents have been obtained who have been duly informed of the technique that is being applied in their children and after explaining to the Surgeons of the medical team of D’Artis Plastic Surgery Clinic and Operation Smile Ecuador, under general anesthesia and infra-orbital block with Bupivacaine 0.25%, in all cases of complete fissures required the repair of the nasal floor with the Sommerland technique and suture with 6/0 vicryl. In this phase the deattachment nasal wing of the piriform opening is performed without dissection of the alar cartilages, after remove the unnecessary skin and mucosa segments, then the minimum dissection of the orbicular muscle and disinsertion of its anomalous fixation. The orbicular muscle is divided into two topographic segments, the cutaneous segment and the vermilion segment until subcutaneously crossing the filtral crest of the healthy side. Once the tissues of their anomalous insertions and adequate hemostasis are released, the labial mucosa is repaired with absorbable suture of vicryl 6/0. At this moment at the mid-point of the filtrum it is fixed with a 5/0 absorbable suture towards the mucosa, which allows to deepen the filtrum and elongate the prolabium by the Rose-Thompson effect. The orbicular muscle is performed with vicryl 5/0. Finally, the skin is repaired with a continuous subcuticular suture of 5/0 nonabsorbable material, which is removed after 8 days.

32 cases of geometric repairs were studied, of which 19 were male and 13 female. The periodic controls were obtained until 6 months and the results were evaluated.

**Results:** The evaluation of the 32 cases was made by the senior author a case of partial dehisencia was determined that did not need reintervencion, two cases of hypertrophy in the scar and a case of minimal retraction of the lip. The other cases were rated as good to very good results with a comparable straight scar and sometimes higher than the results with the Fischer technique obtained by the same surgeons previously.

**Summary/Conclusion:** The Fischer’s technique is the most representative of straigh lines, but when he uses the small triangle on the white roll he loses the philosophy of not making cuts on the surface of the filtrum. The geometric technique offers the alternative of creating a deep lip without unnatural scar and elongate the lip using geometric resources on the skin and muscle on the Rose-Thompson effect. We will need more representative and multicentric studies at a longer distance to determine the effectiveness of the Geometrica technique as a new option.
Background: Very wide and attractive regional techniques have been described for the repair of a cleft palate, starting from the technique of minimal incisions until reaching large surgeries with extended release of two palatal flaps. Both achieve the primary objective that is the repair of the two mucous membranes of the cleft, but in few cases very aggressive techniques have been applied for small defects or on the contrary, conservative incisions in extensive defects leading to excessive scarring in the first cases and a possible dehiscence in the second option.

Aims: The intention of this work is not to describe one more technique for the repair of the cleft palate, but to recommend a reasoned philosophy in each case to get to repair the cleft palate using the ideal technique in each situation and avoiding unnecessary incisions and dissections that will lead to an already altered facial development alteration in the fissured patients. "Cut as you go", has been applied in this work and this has allowed us to grasp that in the plane of reconstructive surgery the most frequent and recommended is to use asymmetric techniques that get the repair saving unnecessary scarring to the patient.

Methods: In the last two years, the author carried out 87 palatoplasties distributed 52 for male patients and 35 for female patients between complete and incomplete cases, unilaterally or bilaterally, applying the concepts of surgical palate philosophy and the results were: Minimally incisions palatoplasty 52 cases Minor incisions plus contralateral vonlagenbeck 25 cases Minimal incisions plus hemi-wardill 7 cases Veaw Wardill standard 3 cases

Results: Our index of palatal fistulas with this method was maintained in the order of 7%, but the most important thing was to note that the asymmetric technique 32 cases, that is, 36.8% of cases, would have performed two flaps technique and it was possible with this principle of "cut as you go" to save in those patients cicatrization that in the future would determine retardation of the maxillary growth.

Summary/Conclusion: Most cleft surgeons are accustomed to perform symmetrical techniques when repairing a cleft palate, that is to say Minimal incisions or Von Lagebeck, or Bardach or Veaw Wardill among others, each with advantages and disadvantages over the others. With this concept of "cut as you go" in 36.8% that were our index of asymmetric techniques, we avoided making unnecessary cuts, protecting the patient's facial growth and development, which in other circumstances would have been necessary to proceed to larger incisions and cuts.
Cleft palate index. A new concept
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

Background: There is no pattern or protocol to follow for cleft palate surgery that is applied routinely in the surgery services for fissure, usually the surgeon performs the standard technique and his training with which he is familiar. Sometimes with good results and others with complication, trying to understand why the cases are complicated led us to the conclusion that the technique would not work with a certain degree of severity but with another, so we have applied an algorithm that does not allow choose safely the best procedure for each case.

Aims: Within the reviewed bibliography there is no adequate and standardized parameter to define the severity of a cleft palate and perhaps only at the end of the surgical procedure according to what was seen by the surgeon can be said whether the case was severe or not. We try with this Fissure indice to provide the surgeon with a guide and algorithm to diagnose and recommend a proven surgical procedure that provides the security of success in the surgery for each case.

Methods: We took 87 cases of palatoplasty performed by the senior author of our service, where millimeter measurements were made at the level of the palatal gap at the juxtaproximate points of the junction of the hard and soft palate, usually the wider site of the cleft, this value was divided for the summation in millimeters of the emergence of the palatine artery to the point of junction of the hard palate with the soft one at the level closest to the fissure. This mathematical relationship allowed us to divide according to the index of cleft palate in four grades according to its severity in:
- MILD 0 to 0.25
- MODERATE 0.26 to 0.5
- SERIOUS 0.51 to 0.75
- SEVERE 0.76 or more

Results: This algorithm will allow us to have a logical basis to classify the cracks not according to the experience of the surgeon, but with a previously established mathematical parameter. On the other hand, the algorithm derived from the severity index of the cleft palate will allow us to choose the surgery that has proven the most safety according to the author's experience, being more effective in each degree of severity.

Summary/Conclusion: At the time of this work, it has not been possible to find in the literature review a method for classifying the severity of palatal fissures, which at the same time recommends a safe surgical technique, depending on the severity of the fissure. Not all grades of cleft palate can be treated with the same technique. The cleft palate index algorithm mainly allows the young surgeon to have an instrument to adopt a surgical technique proportional to the severity of the fissure, and on the other hand this index may be the basis for other types of statistical studies related to the severity of the fissure. Palatal fissures that nowadays do not have a support parameter to classify them.
Lingual deep reverse tongue flap. A new technique
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

Background: Many times the need for repair of a palatal fistula and other defects require tissues that are sometimes nonexistent locally, and it is necessary to look for regional tissue alternatives. The tongue is an extraordinary source of tissue for this purpose, but its bulky and fibrous appearance makes this option a not very desirable alternative. The present work achieves a refinement of the original technique that provides tissue easily and without previous negative observations.

Aims: We seek to provide a technique that gives the surgeon an option to have non-scar tissue of smooth appearance and without associated deformities such as the lingual flap in its original description by Guerrerosantos did. The multiple attempts of unsuccessful closures of fistulas leave only scar tissue surrounding in the fistula that complicates the intension of its repair. The deep reverse lingual flap based on the artery of the same name is a new technique that solves the problem in two surgical times.

Methods: Six patients between 7 and 32 years were selected with multiple attempts to repair fistula that failed and on average their sizes ranged from 1 x 1.5 cm to 5 x 3 cm. After they or their representatives voluntarily and informed accepted to undergo the procedure, they underwent surgery under general anesthesia where they first proceeded to get an adequate repair of the nasal floor with the extended nasal flap reported by us, finally the flap was stalked mucosa of the deep face of the tongue and the defect was repaired with vicryl 5/0, after 3 weeks the pedicle was freed and with its remnant the gingival defect was repaired when necessary.

Results: Of the six cases taken for the report of this work, the resolution of the integral fistula was achieved in 5 cases and partial resolution in one of them due to dehiscence evidenced 5 days after their primary surgery.

Summary/Conclusion: Undoubtedly the biggest challenge in a cleft lip and palate surgery is the resolution of large recurrent fistulas, and having surgical alternatives will provide natural resources with biological tissues that can solve the problem without falling into the most simple and practical as It is a sealing plate with its limitations as well and does not even reach a much more demanding microsurgical technique. The deep reverse lingual flap provides reconstruction of up to 70% of the entire palate with regional tissue and minimal sequelae in the donor area.
Third generation for veloplasty. A new technique
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

Background: One of the goal sought in the repair of the palate after individualizing the oral and nasal cavities, is the search for an adequate repair of the anatomical structure as the basis for phonation. In our study series, the velopharyngeal insufficiency index is in the order of 53%. The third generation veloplasty seeks to solve primarily the problems of pharyngeal vein insufficiency and incompetence without causing structural disorders in other areas as generated by techniques such as Wardill-Kilner, Furlow, San venero Rosselli, Antia.

Aims: The objective of the third generation Veloplasty is to provide a surgical resource that allows to extend the contact surface of the levator muscle of the palate and lengthen the posterior half of the soft palate, which contributes with an adequate sufficiency of the soft palate.

Methods: Once the consents of the parents have been obtained who have been duly informed of the technique that is being applied in their children and after explaining to the Surgeons of the medical team of D'Artis Plastic Surgery Clinic and Operation Smile Ecuador, we proceeded to the infiltration of the palate with a 0.125% Bupivacaine solution in a maximum amount of 1 cc per kilo of weight. First, we proceed to a de-epithelization of the hemi-valves in their medial part and a transversal incision of the uvula base so that we geometrically generate a rhombus whose final suture makes it possible to gain an average of 1 cm of effective length of the soft palate important in phonation. Once the release and relaxation of the structural elements of the soft palate has been completed, raffia is first performed on the mucosa of the nasal side with 6/0 vicryl until the corresponding segment of the hard palate is completed. Afterwards, we repair the staminate Uranus muscle back forwards, followed by the lifting muscle of the veil and aponeurosis of the veil tensor with vicryl 5/0. Finally the oral mucosa repair with vicryl 6/0.

Results: This technique has been applied on the soft palate in all our palatoplasties independent of the palatoplasty technique of the hard palate, usually minimal incisions. In a total of cases distributed in incomplete, complete and revisional by VPI. The following findings were found: 4% of fistulas at the level of the junction of the hard palate with the soft palate, the rest of the cases healed adequately. The results in speech were appreciated until 6 years of age and the results in speech were compared with similar ones in which another technique of soft palate reconstruction was applied. 77% of cases with our technique obtained an adequate speech quality compared to 53% of the other cases.

Summary/Conclusion: The quality of the pharyngeal veil sounds depends on an adequate muscular function (competence) and an adequate length of the soft palate (Sufficiency). Some techniques seek to achieve the two mechanisms, but most of them at the expense of generating problems at another structural level of the palate. The third-generation veloplasty solves the two problems of proficiency and competition without creating problems in other areas, and phonation quality was measured with our Speech Score.
**Background:** Scar outcomes in cleft patients is difficult to resolve completely, because scar tissue is an individual, genetically controlled process. The physiological contractions caused by scar tissue may determine bad aesthetic results, asymmetries, depressions of the affected site, can cause a bad function of the lip, and, although it is still controversial, may interfere with maxillary development.

**Aims:** This study focuses on the results obtained using the lipofilling technique both to improve the quality of the scar and to increase the volume of a hypotrophic or retracted region on the cleft side.

**Methods:** We included in the study cleft patient undergoing secondary scar revision in the San Filippo Neri Hospital in the last 5 years. All patients were photographed before, 6 months and 12 months after the operation. In collaboration with the Smile House Milano we performed a qualitative and quantitative assessment of the subjects included in the study using the standardized photographs taken before and after the surgery using Burstone esthetic soft tissue analysis and with an Asher McDade. Measurements concerning the increase in projection of the upper lip on the sagittal plane were calculated and we evaluated the quality of the scar six months after lipofilling. It was thus possible to quantify the extent of resorption.

**Results:** Photometric analysis showed that the lipo-aspirate graft corresponds to a visible increase in scar quality, symmetry and projection which however shows a 6-months resorption of about 30%. The donor site has never presented any complications of any kind. A second lipofilling operation has to be performed in the same region to compensate for the physiological resorption of the graft.

**Summary/Conclusion:** Lipofilling is a simple, versatile and easily reproducible method that allows to obtain visible qualitative and quantitative improvement results of scars in the secondary treatment of cleft patients. Further scientific studies will allow us to obtain a more reliable predictability of the result to reduce the number of re-interventions or to broaden the indications for the use of this method.
Background: Secondary bone grafting is a well established technique, iliac crest is worldwide considered the first choice as donor site, because it provides a large amount of cancellous and cortical bone for reconstructing the volume, is a very easy and safe procedure. The major drawbacks are the scar and postoperative pain. Many different donor sites have been proposed. We present a case series of alveolar reconstruction using vomerine bone.

Aims: The idea at the base of the procedure is to join the bone graft with nasal septum deviation correction, solving at the same time respiratory problems, that typically afflicts cleft patients, and reconstructing the alveolar defect using the bone of the vomer.

To our knowledge, this bone source has never been used before for alveolar reconstruction.

Methods: The sample consisted of 20 patients operated at the Smile House in San Paolo Hospital of Milan, that underwent alveolar graft with vomerine bone and septoplasty during the same procedure. Alveolar ossification was evaluated after 1 year from surgery with panoramic X-ray and CBCT. A modified Bergland scale on panoramic X-ray, and axial measurements of alveolar thickness on CBCT were the methods of evaluation.

Results: Alveolar thickness and height were ideal or good in the majority of patients. Few patients had insufficient bone volume after vomerine graft.

Summary/Conclusion: Alveolar graft with vomerine bone can be considered in all the unilateral cases that have a septal deviation and when the alveolar defect is small enough, in our experience narrower than 1 cm at the nasal floor and 2-3 mm at the lower alveolar level.
Background: Common practice uses multiple surgeries for unilateral cleft lip and palate (UCLP) repair. However, a stepwise approach has two inevitable drawbacks. First, it brings repeated burden. Second, it leaves an area of secondary wound healing at the junction between the repaired and unrepaired cleft area in the first operation. In the following operation, the same area needs re-exposure to allow gapless suturing. The one-stage protocol of UCLP repair facilitates operating within tissue that is entirely unaffected by previous surgical intervention and thus, scar formation in the overlapped regions can be minimized. Further, the alveolar-palatal junction zone can be freely accessed for a continuous two-layer closure if the lip closure can be performed at the end of the primary procedure.

Aims: The aim of the study was to assess the final outcome of craniofacial development of patients born with complete UCLP operated on according to the one-stage method.

Methods: The study evaluated mature, consecutively treated non-syndromic UCLP patients that received a one-stage repair at one center between 1993 and 2000. The patients who underwent alveolar bone grafting before the 6th year of life were excluded in an aim to avoid the potential inhibitory influence on maxillary development. The presurgical cephalograms of patients who underwent Le Fort I maxillary osteotomy were not excluded from the evaluation. The skeletal maturity of each patient was confirmed by using the cervical vertebral maturation method. The surgical protocol in the study group consisted of the one-stage primary cleft repair performed between 6 and 16 months of age. The surgical method comprised: a two-layer hard palatal closure with bilateral bipedicle flaps, a soft palate repair, and a lip repair with an inferior triangle lengthening of the medial cleft side during single operation. No presurgical infant orthopedics and no gingivoperiostplasty were performed. All patients received secondary interventions according to their clinical need if required.

Results: Finally, the study group consisted of 78 patients (50 males and 28 females) with a mean age of 18.4 years at cephalometric evaluation. The sagittal maxillary development observed in the study group (SNA 78.1, ANB 0.34, Co-A 77.5) lies below healthy mean data (e.g. Michigan growth study, SNA 82, ANB 3) but compares favorably to adult UCLP patient treated by multi-stage protocols (e.g. Eurocleft, best values at 17y years: SNA 75.7 and ANB -0.1). 22.4% of patients fulfilled the objective cephalometric criteria of maxillary deficiency. The percentage of patients treated by orthognathic surgery, pharyngoplasty and oronasal fistula repairs were 6.6%, 6.6% and 18.4%, respectively. The mean number of surgical interventions under general anesthesia was 3.0 at the time point of completion of treatment.

Summary/Conclusion: A mild inhibition of maxillary growth (comparable to what is perceived in multistage procedures) with a simultaneous benefit of a low total number of surgeries was achieved with the one-stage protocol. The one-stage protocol offers a chance to reduce the burden of care for patient, families and health-care provider with no evidence for compromised long-term craniofacial growth outcome.
Is there a relationship between the age of a cleft patient at secondary alveolar bone grafting and the donor site morbidity, surgical duration or hospitalization?

A Brudnicki¹, M Rachwalski², L Wiepszowski¹, E Sawicka³

¹Maxillo-facial Surgery Unit at the Department of Surgery of Children and Adolescents, INSTITUTE OF MOTHER AND CHILD, Warsaw, Poland, ²Department of Maxillofacial and Plastic Surgery, National Reference Center for Cleft Lip and Palate, Hôpital Universitaire Necker-Enfants Malades, Paris, France, ³Department of Surgery of Children and Adolescents, INSTITUTE OF MOTHER AND CHILD, Warsaw, Poland

Background: Although the autogenous bone tissue harvested from the iliac crest remains the most frequently used material for secondary alveolar bone grafting in the surgical treatment of clefts defects, still there is no consensus regarding the optimal timing of this procedure. The recent tendency to perform secondary alveolar bone grafting at an earlier age raises the questions about potentially higher complexity of bone harvesting procedure or risk of donor site-related morbidity at earlier age (especially concerning cleft patients much younger than 6 years old), hence potentially longer surgical time and hospitalization.

Aims: We aimed to investigate the potential correlation between the age of patients during surgery and donor site-related symptoms/complaints, surgical time and hospitalization following bone harvesting procedure.

Methods: The outcome of 195 consecutive alveolar bone graftings carried out over one calendar year (2012) among different age groups (≤3, 4-6, 7-9, 10-12, ≥13) was retrospectively assessed based on a chart review and purpose-prepared report forms. The material consisted of 126 male and 69 female patients who were born with various cleft malformations and had secondary alveolar bone grafting between 1.8 and 40.5 years of age (mean age of 7.1 years). All patients were operated on at one center according to analogous surgical technique regardless of the patient’s age. Only the cleft cases with the iliac crest as donor site were included. The association between age, gender and hospitalization following bone harvesting was tested by Spearman rank correlation, while relationships (i.e. between age and pain) were evaluated by logistic regression.

Results: The most frequent donor site complaints included: pain equal to or exceeding that of the recipient site (93%) and gait disturbances/reluctance to walk (92.5%) occurring immediately after the procedure. Chronic complaints included: iliac contour alteration (40.1%), unsightly scar (23%) and recurring discomfort (2.1%). Statistical analysis showed no correlation between donor site symptoms, their duration or hospitalization time following surgery at different ages, except a higher incidence of significant pain immediately after bone harvesting in older females (r = 0.268; p = 0.030).

Summary/Conclusion: The results of the present study imply that performing alveolar bone grafting/bone harvesting at an earlier age for sure does not increase donor site-related morbidity, surgical duration or hospitalization following surgery.
Background: Initial repair of a cleft palate, a procedure often performed in cleft centers all over the world, belongs to the basic skills of a cleft surgeon. The speech outcome is always of the main concern in cases with palatal involvement.

Aims: The aim of this study was to present the surgical technique of our one-stage method of palatoplasty in patients suffering from isolated cleft palate and to evaluate its short and long term morbidity from the long-term perspective (around 10 years of age).

Methods: The study included all the patients born with isolated cleft palate who were operated consecutively within a time span of 24 months (between January 2005 and December 2006) according to the same surgical method. The assessment was based on medical documentation recorded up to 10 years of age.

Results: The study evaluated medical documentation of 207 patients (38.6% males and 61.4% females) – both syndromic (34 cases) and non-syndromic (173 cases) who were born with isolated cleft palates. The non-syndromic patients operated on during the second half of the first year of life were 41 (34.2%) males and 79 (65.8%) females. The assessment was performed on average 9.3 years postoperatively. The following mean values were calculated in regard to the moment of the procedure: age was 8.6 months, duration of surgery was 66 minutes (range: 30-135), and length of hospitalization following operation was 2.6 days (range: 1-8). The incidence of severe velopharyngeal insufficiency, oronasal fistulas and persistent otitis media with effusion requiring surgical intervention was 2.5%, 3.3% and 18.3%, respectively. Two patients had surgical wound dehiscence, one of them revealed upper airway infection instantly following surgery.

Summary/Conclusion: The morbidity of the presented palatoplasty is relatively very low. Its surgical technique is simple, not time consuming and does not require any special equipment, whatsoever. The low burden of secondary surgical repairs registered in the evaluated group of patients with isolated cleft palate indicates high efficiency of presented method since these patients have been under control of speech therapist. The present study indicated the second part of the first year of patient’s life as the optimal timing for applying the primary palatoplasty. The need for subsequent pharyngoplasty among the non-syndromic patients operated on at this age was only 2.5%.
The role of prophylactic antibiotics in primary cleft surgery: reducing the duration does not increase early complications

A Burke-Smith, R Nicholas, K Echlin, D Atherton, N Timoney

1Evelina London Children's Hospital, 2Guy's and St Thomas' NHS Foundation Trust, London, 3Birmingham Children's Hospital, Birmingham, United Kingdom

**Background:** The effectiveness of peri-operative prophylactic antibiotics requires evaluation in the face of increasing bacterial resistance. While florid infection after a cleft lip or palate repair is unusual, it may lead to worse aesthetic outcome and is a factor in the formation of post-operative oro-nasal fistulae. A recent survey found that all UK cleft units currently give peri-operative antibiotics but there is little evidence on which to base prescribing.

**Aims:** On 1st April 2018, the Evelina Children's Hospital changed antibiotic protocol from an extended course of 6 days (3 intravenous doses and 5 additional days of oral co-amoxiclav) to a short course of 24 hours (3 intravenous doses only) for patients undergoing repair of cleft palate or cleft lip with vomerine mucosal flap reconstruction. We present a completed audit cycle reviewing rates of early complications namely surgical-site infection or fistula formation following this change. We will also present rates of antibiotic associated adverse events.

**Methods:** Patients undergoing surgery between 31st August 2017 and 31st December 2018 were included. This spanned the protocol change on 1st April 2018; those in a transition period from 1st April 2018 to 14th April 2018 were excluded. A retrospective review of patient records was performed for surgical site infections and palatal fistulae. Fistulae were recorded according to Pittsburgh classification system. One of three cleft surgeons in the department did not change protocol and patients under his care were used as a control group to ensure any changes were not the result of seasonal or external factors.

**Results:** A total of 147 patients were included. Of these, 91 had an extended course of antibiotics and 56 had a short course. For patients who underwent repair of cleft lip with vomerine mucosal flap, 25% of those who had an extended course of antibiotics postoperatively developed surgical site infection, whereas infection rate was 13% with a short course of antibiotics. This was not significant (Chi² 0.47, p 0.49). There was no significant difference observed in the rate of fistula formation between those who had an extended-course of antibiotics (26%) and those who had a short-course (11%) (Chi² 2.2, p0.14). Within the control group, the fistula rate was 13% prior to 1st April 2018 and 17% after the 14th April (p 0.78).

**Summary/Conclusion:** Reducing the duration of prophylactic antibiotics for primary cleft surgery did not significantly increase early complications in our case series. This corresponds to the current literature which we review. This provides additional evidence to reduce unnecessary prescribing of prophylactic antibiotics.
P-015
A report of a preliminary survey within the UK to identify the numbers of children with intentionally unrepaired cleft palate between 2007 and 2017.
S Butterworth¹, D Sainsbury¹, S Van Eeden², P Hodgkinson¹, C Cleft Multidisciplinary Collaborative²
¹Plastic Surgery, ²NHS, Newcastle upon Tyne, United Kingdom

Background: A small number of children born with cleft palate do not undergo palate repair. Such children often have complex healthcare needs, including conditions increasing the risk of general anaesthesia. To not repair a child’s cleft palate is often a challenging decision for the cleft MDT; some parents/carers may struggle to understand the reasoning.

Aims: To estimate the numbers of children born in the UK, between 2007-2017, who had an intentionally unrepaired cleft palate.

Methods: The Cleft Multidisciplinary Collaborative identified individuals interested in completing a survey within their cleft teams.

The following information was requested:
- · estimated numbers of patients with an unrepaired cleft palate
- · documented reason(s)
- · presence of a syndrome
- · developmental level, including speech and language
- · feeding status

Results: Seven completed surveys were received. Two centres had 3-5 patients, three centres had 6-8 patients and two centres had over 9 children with intentionally unrepaired cleft palates over the time period. The following reasons were noted:
- · Risk from general anaesthesia
- · Global developmental delay
- · Neurological problems
- · Cardio-respiratory problems

Nearly all of the children had a diagnosed syndrome, were non-verbal/pre-verbal and had PEG or NG tube feeds.

Summary/Conclusion: Many reasons were identified for not repairing a cleft palate; the primary factor being the potential risk of general anaesthesia. Many cleft teams reported experiencing pressure from parents to repair their child’s cleft palate. We aim to supplement these findings with data from other cleft centres to enable professionals caring for these children to give parents and carers more information about leaving their child’s cleft palate unrepaired.
Alveolar bone grafting with chin bone in cleft and lip palate
R Carvalho¹, T Okada Ozawa²
¹Cirurgia Bucomaxilofacial, ²Ortodontia, Hospital de Reabilitação de Anomalias Craniofaciais - Universidade de São Paulo, Bauru - SP, Brazil

Background: Although chin bone has been consolidated as a good option as donor site for alveolar bone grafting in cleft lip and palate patients, only during the past two years we tried to used it in our practice, based on Borstlap papers.

Aims: This study intended to compare the success index of alveolar bone grafting in cleft lip and palate patients with chin bone, iliac crest bone and recombinant human bone morphogenetic protein (rhBMP-2), as donor sites.

Methods: Two hundred and forty unilateral cleft lip and palate patients were submitted to alveolar bone grafting surgery during their rehabilitation course. Each eighty of them were selected from a period of time according to the donor site: cancellous bone from iliac crest (gold standard), rhBMP-2, and, most recently, chin bone.

The results were evaluated through periapical radiographs from cleft area.

Results: The images were analysed by three judges using Chelsea scale and showed very similar and good results, when consider interdental septum height.

Summary/Conclusion: Although harvesting chin bone is not an easy task sometimes, new bone formation in alveolar grafting has showed very good results to bone bridge.
Long term results of our experience with recombinant human bone morphogenetic protein (rhBMP-2)
R Carvalho¹, T Okada Ozawa²
¹Cirurgia Bucomaxilofacial, ²Ortodontia, Hospital de Reabilitação de Anomalias Craniofaciais - Universidade de São Paulo, Bauru - SP, Brazil

Background: Recombinant human bone morphogenetic protein (rhBMP-2) were used as alveolar bone grafting in cleft lip and palate from March 2011 to August 2015 in our service, as a substitute for autogenous iliac crest bone repair of congenital clefts.

Aims: Because of administrative matters, the only option we had for alveolar bone grafting during a period of time was rhBMP-2 in a collagen sponge. Once we have had good results in a previous study, we accepted to use this material in our day by day.

We intend to present our experience with a “new” material in a long term perspective to alveolar bone grafting.

Methods: Four hundred patients with cleft lip and palate were submitted to alveolar bone grafting with rhBMP-2 during these four years by the same surgeon. Some of them were complete clefts and others, don’t; some were unilateral and others, bilateral; some surgeries were performed before canine eruption and others, much later than this.

The patients have been evaluated clinical and radiographically through all these eight years.

Results: Successful osseous union was achieved in most of them. The constructed alveolus performed clinically as normal bone and responded to natural tooth eruption and orthodontic movement.

Summary/Conclusion: Although this is a controversy material for alveolar cleft lip repair, our experience tells us that this should be considered as a very good treatment option.
P-018
Redefining the median tubercle during cleft lip repair: our learning experience.
D Chattopadhyay¹, M Vathulya¹
¹Burns and Plastic Surgery, AIIMS Rishikesh, Rishikesh, India

Background: The reconstruction of median tubercle of lip has not been highlighted in the different techniques of cleft lip repair. But the presence of a normal looking median tubercle is extremely important for the aesthetics of the upper lip. Thus we adopted this modified technique of lip repair, preserving the tissues of the median tubercle.

Aims: To refine the technique of cleft lip repair to produce a visible median tubercle on table

Methods: Since April 2018, we have used this technique in 7 cases (3 bilateral and 4 unilateral) of cleft lip repair. I am hereby presenting the 6 months follow up results of the cases. In unilateral cleft lips, the excess tissue from lateral vermillion was de epithelised and placed under the median tubercle from the medial side. In bilateral cleft lips, the prolabial tissue was utilised to form the median tubercle

Results: In profile photographs after 6 months, all the patients showed normal looking median tubercle with nice fullness of upper lips. Lateral photographs showed good protrusion of upper lip.

Summary/Conclusion: Various techniques of cleft lip repair aim to produce an aesthetically normal looking lip. That the median tubercle is a very important aesthetic unit of the upper lip has been recognised for long but no technique emphasizes on its reconstruction during cleft lip repair. In a recent case series Resnik et al have used dermis fat graft to augment the tubercle. In contrast, this technique uses the native tissue, is simple but elegant and provides beautiful results. In the first bilateral lip where we used the technique, we were hesitant to deviate from the textbook norms but the on table result was simply beautiful. The idea behind presenting this series is to highlight the reliability and reproducibility of this new technique which redefines the median tubercle of the lip during cleft lip repair.
Obstructive sleep apnoea following velopharyngeal surgery in children with cleft palate: a literature review

M Chin¹, M Haj², H de Gier³, S Versnel⁴, H Poldermans⁵, M Koudstaal⁶

¹Oral and Maxillofacial Surgery department, ²Oral and Maxillofacial Surgery Department PhD, StR, ³Ear, Nose, and Throat Surgery (ENT) department, ⁴Plastic and Reconstructive Surgery department, ⁵Speech Therapy department, Erasmus MC, Rotterdam, Netherlands, ⁶Craniofacial diseases, Oral and Maxillofacial Surgery department, Karolinska University Hospital, Stockholm, Sweden

**Background:** Velopharyngeal insufficiency (VPI) is a common problem after a soft palate closure in children affected by a cleft palate with an estimated frequency of 5-20%. VPI commonly results in functional limitation in breathing, eating and speech. Various surgical techniques are in use to treat VPI after a primary repair of the palate, but the optimal treatment remains debatable. Sleep apnea is a common complication following velopharyngeal surgery and may lead to developmental deficits or occurrence of sudden infant death syndrome.

**Aims:** This paper attempts to review the prevalence of airway obstructive outcomes (short-and long term) related to different surgical modalities and prompts to gain more insight in the optimal treatments.

**Methods:** A search was performed in Embase, Medline Ovid Cochrane library, Google scholar and Web of Science for articles on: “sleep disorder breathing”; OSA (obstructive sleep apnea); non-syndromic cleft; pharyngoplasty. Studies that included information on the occurrence of OSA following velopharyngeal surgery to improve speech in children with a repaired non-syndromic cleft palate were included.

**Results:** Twenty-seven articles were included. Of these only nine studies collected long term data by use of polysomnography. The Pharyngeal Flap (PF), Sphincter Pharyngoplasty (SP) and Palatoplasty (PP) were the most common procedures amongst the surgical modalities. Although the techniques varied in between studies, PP and SP resulted in the lowest prevalence of post-operative obstructive airway (OA) symptoms in respect to PF (respectively 12.0%; 14.4%; 36.3%). However, PF is reported to result in better speech outcomes. Several other surgical methods were identified that seemed promising, but lacked the long term follow up data of OA outcomes.

**Summary/Conclusion:** VPI can be treated using various surgical methods. However, each patient should be assessed on speech, closure pattern of the pharynx and OA before choosing a treatment strategy. More studies are needed that provide pre-operative and long term post-operative data on obstructive airway problems. Also, the collection of data needs to be standardized, this also accounts for the classification of OSA.
**P-020**

Holistic outcome at adulthood period and comprehensive care of patients with frontoethmoidal encephalomeningocele

P Chowchuen¹, B Chowchuen², N Patjanasoonthorn³, C Thanapaisal⁴

¹Department of Radiology, Faculty of Medicine, ²Plastic Surgery Division, Department of Surgery, Faculty of Medicine, ³Department of Psychiatry, Faculty of Medicine, Khon Kaen University, Khon Kaen, ⁴Burapha University, Cholburi, Thailand

**Background:** Frontoethmoidal encephalomeningocele (FEEM) is common occurrence in Southeast Asia, particularly in Thailand, and other regions of the world with limited information of long-term management and holistic outcomes.

**Aims:** To report long-term holistic outcomes and comprehensive management at the adulthood period of the patients with FEEM.

**Methods:** Holistic outcomes were analyzed from 28 patients with FEEM, (>18 years of age), treated between 1993 and 2011 at the Tawanchai Center, Srinagarind Hospital, Thailand. The unsatisfied outcomes and bully experiences were analyzed by the patients’ gender, FEEM type, age of primary surgery, surgical approach, number of operations, educational level, and IQ.

**Results:** The majority of patients were female (60.71%). 71.43% of all patients had nasoethmoidal (NE) and combined types. 60.71% were uneducated or completed grade 6 educational level (primary school). 96.43% of the patients satisfied with overall treatment. The mean aesthetic overall score was 1.48 and aesthetic score of nasal shape was 1.59. The majority of patients had moderate IQ at low level with the mean score of 62.71. Unsatisfied outcomes were found significantly higher in NE and combined types (p-value 0.044), and combined surgical approach (p-value 0.009). 85.71% of the patients had abnormal MRI.

**Summary/Conclusion:** Low educational level, low IQ, unsatisfied outcomes of NE and combined types and combined approach, and abnormal MRI are main challenges. The children with FEEM have significant higher rates of learning problems and less academic successful. Comprehensive care is needed for provision of better health-related quality of life or functioning patients and better long-term outcomes.
Early speech and language outcome and predictive factors of palatoplasty with intravelar veloplasty in children with cleft lip-palate

B Chowchuen¹, P Sakarinpanichakul², B Prathanee³, P Surakunprapha⁴, K Jenwitheesuk⁵

¹Plastic Surgery Division, Department of Surgery, Faculty of Medicine, Khon Kaen University, Khon Kaen, ²Buriram Hospital, Buriram, ³Department of Otolaryngology, ⁴Department of Surgery, ⁵Plastic Surgery Division, Department of Surgery, Faculty of Medicine, Faculty of Medicine, Khon Kaen University, Khon Kaen, Thailand

**Background:** The goal of cleft palate surgery is to gain normal functions of speech and language, ear function, and maxillary growth provided by coordinated interdisciplinary team management. The appropriate early assessment and intervention are important.

**Aims:** This study purposed to evaluate speech and language outcomes of children with cleft palate after palatoplasty with intravelar veloplasty (IVV) at 2 years of age, and analyze the factors associated with unsatisfied outcome.

**Methods:** A time-series, prospective study was performed during April 2011 to June 2013. All patients with non-syndromic cleft lip-palate or cleft palate (CLP or CPO) who underwent primary palatoplasty using the 2-flap palatoplasty with IVV, and when indicated, myringotomy was performed according to the Tawanchai Center Protocol were included. Speech and language competencies were assessed at 2 years of age, using receptive language quotient (RLQ), expressive language quotient (ELQ) and combine language quotient (CLQ) of the Thai Speech and Language Test for Thai children (TSLT2). The Oronasal fistulas (ONFs) were recorded. Predictive factors, comprised of gender, cleft types, age at surgery, myringotomy, and surgeons were analyzed using linear regression to determine the associated unsatisfied outcome.

**Results:** There were 40 patients with the average follow up post operatively of 10.85 (4-15) months, 25 males (57.5%) and 17 females (42.5%). Hearing loss and abnormal tympanogram were presented in 8 patients (28.57%) and 26 patients (65%), respectively. Myringotomy performed in 26 patients (65%). The average time at speech evaluation post operatively was 4-15 months. There was no complication e.g. severe bleeding, airways difficulties, oropharyngeal or ear infections, re-operation and re-admission. ONFs occurred in 3 of 40 patients (7.50%). The overall mean scores of RLQ in patients with CLP or CPO was significantly lower than normal children (p-value= 0.013) but there was no significant difference in ELQ and CLQ. The unsatisfied outcome was 25% in RLQ category; 95% CI [13, 41], 15% in ELQ category; 95%CI [6, 30] and 25% in CLQ category; 95% CI [13, 41]. Males, CPO, delayed group and ONF had more unsatisfied outcome. There was no significant difference between the female and male groups, the cleft type (cleft palate only- Vau I and other types), hearing loss, age at surgery (≤15 months and ≥16 months), the operation by plastic surgery staffs and residents, myringotomy, and oroanasal fistula with unsatisfied outcome.

**Summary/Conclusion:** The overall mean scores of RLQ in children with CLP or CPO were significantly lower than normal children (p-value= 0.013). The delayed speech and language development was diagnosed by RLQ (10 children-25%), ELQ (6 children-15%) and CLQ (10-children-25%). Children with non-syndromic CLP or CPO may be at risk for early delay in both language and cognitive development. Unsuccessful cleft palate repair, craniofacial anomalies, or neurological problems may lead to unsatisfied speech and language outcome and need appropriate treatment by interdisciplinary team.
Management of cleft maxilla
A Chuq¹, G Reddy¹
¹Department of Dentistry and Craniomaxillofacial Surgery, All India Institute of Medical Sciences, Rishikesh, Uttarakhand, Rishikesh, India

Background: Maxillary hypoplasia is a common developmental problem in cleft lip and palate deformities. These deformities have traditionally been corrected by means of orthognathic surgery. Management of skeletal deformities has been an important challenge for maxillofacial surgeons. Distraction osteogenesis is a surgical technique that uses body's own repairing mechanisms for optimal reconstruction of the tissues and has been used for management of cleft maxilla patients. This talk describes an update on the current concepts and principles in the treatment of patients with cleft lip and palate. Sequencing and timing of orthodontic and surgical treatment in infancy, early mixed dentition, early permanent dentition, and after the completion of facial growth will be discussed.

Aims: The management of patients with cleft lip and cleft palate requires prolonged orthodontic and surgical treatment and an interdisciplinary approach in providing them with optimal esthetics, function, and stability.

Methods: The cleft patients showed anteroposterior maxillary hypoplasia with class III malocclusion and negative overjet resulting in a concave profile according to preoperative cephalometric measurements, dental relationship, and soft tissue analysis. The patients treated by orthognathic Le Fort I osteotomy and by maxillary distraction osteogenesis with rigid extraoral devices (RED) connected after a Le Fort I osteotomy will be presented. The rate of distraction was 1 mm per day to achieve Class I occlusion with slight overcorrection and to create facial profile convexity.

Results: The occlusion changed from class III to class I. The profile of the face changed from concave to convex. A complete overview of cases treated by both distraction and osteotomy will be presented.

Summary/Conclusion: This talk describes an update on the current concepts and principles in the treatment of patients with cleft lip and palate. Sequencing and timing of orthodontic and surgical treatment in infancy, early mixed dentition, early permanent dentition, and after the completion of facial growth will be discussed.
The use of magnetic resonance imaging in the evaluation of velopharyngeal sphincter function

T Yıldız¹, F Çınar¹, S Baghaki¹, A Demiröz¹, Z Polat², N KOÇER³, Y Aydın¹
¹PLASTİC,RECONSTRUCTİVE AND AESTHETİC SURGERY, İSTANBUL ÜNİVERSİTY-CERRAHPAŞA, İSTANBUL, ²Audiology, Cerrahpaşa, Cerrahpaşa, ³Radiology, İSTANBUL ÜNİVERSİTY-CERRAHPAŞA, İSTANBUL, Turkey

Background: One in every 1000 newborn encounter with cleft palate problems, and they need palatal repairing surgery. That kind of surgery has its own complications, and velopharyngeal insufficiency is a complication which can be corrected by secondary interventions. In velopharyngeal insufficiency, Leakage of the air from oral cavity to the nasal cavity creates phonation problems during speech, and it decreases the quality of pronunciation of oral explosive sounds.

Aims: While we were beginning our new research, we know that there are many previously defined methods for evaluation of velopharyngeal insufficiency and quality of speech. Perceptual analysis, Pittsburg Weighted Speech Scale (PWSS), nasometry, and nasoendoscopy, are previously defined evaluation methods for examination of velopharyngeal insufficiency. Some of them are still being used while the use of other methods such as use of video fluoroscopy has been limited due to its harmful radiative effects. In our study, our aim was to evaluate velopharyngeal insufficiency objectively while minimizing the harmful effects and downsides of other methods.

Methods: Since some studies have shown that structural characteristics in static position are insufficient for understanding the velopharyngeal function; for a better understanding we had an idea that dynamic MRI can help us by displaying movement of velopharyngeal sphincters. The study group of this research includes a radiologist, speech therapist and plastic surgeons to evaluate different aspects of velopharyngeal insufficiency. Seventeen patients with repaired cleft palate by single surgeon were enrolled in this study. Quantitative velopharyngeal measures from the oblique coronal plane and midsagittal plane in static and dynamic positions were collected. Patients’ speech intelligibility was evaluated by using Pittsburgh Weighted Speech Scale and nasalance score was also measured. Correlation analysis methods were used for evaluating relation between MRI gathered measurements and speech intelligibility scores for determining consequential data.

Results: Our study shows that the velar knee-posterior pharyngeal wall distance measurement while explosive sound production is the most related data with speech intelligibility.

Summary/Conclusion: Although our study group is not big enough, we think MRI is a very helpful method in providing reliable information. MRI is a very successful method in visualizing dynamic imaging of contractile function of velum especially in the cases of diagnosis of submucous palate. MRI is a very successful method in visualizing dynamic imaging of contractile function of velum especially in the cases of diagnosis of submucous palate. On the other hand, according to the results of our study, we think that MRI is a very helpful method in providing information about the anatomical and functional status of velum. It can also be concluded that MRI is an effective tool in diagnosing patients with submucouspalate cleft without clinical findings.
Challenger to the surgeon and their surgery methods: 4 case reports

F Çinar¹, B Dağhan¹, T Yıldız¹, A Demiröz¹, İ Yıldırım¹, Y Aydın¹

¹PLASTIC,RECONSTRUCTIVE AND AESTHETIC SURGERY, İSTANBUL UNIVERSITY-CERRAHPAŞA, İSTANBUL, Turkey

**Background:** Craniofascial abnormality arises because of growing deformity of head and face bones. This abnormalities exist from the birth and they are seen in a wide spectrum from little differences to really serious deformities. It seems like that the anomalies appear as common results of genetic anda environmental factors and the deficiency of some molecules (folic acid, vit b12 etc.) needed for development rather than a single factor.

**Aims:** We purpose to present 3 cleft faces and 1 probozis lateralis case which is heavy that even an experienced plastic surgeon slog and their surgery methods.

**Methods:** Between 1995 and 2000, four cases "challenger to the surgeon" is attached to the study which is done its' first operations by Prof. Dr. İbrahim YILDIRIM and still followed by same surgeon. All the facts have congenital facial abnormalities. All the patients' first surgeries are came true in their first three months.

**CASE REPORTS**

**CASE 1.**
Teissier type 30 (mandibula) cleft face + hamartoma in mental area.
- In first year hamartomatosis lesion is determined. Cartilaginous structure in lesion is used for filling the bone gap.
- At 7 age bone grafting is putted to the bone defect in mandibula.
- At 15 age , opening of neck's contracture, reorganizing of soft tissue transfer and scar revision is done.

**CASE 2.**
Right unilateral Tessier type four cleft face
- In two years, multi-transposition flaps and lower palpebrae reconstruction is done. At the same session, bone grafting is putted for orbita floor reconstruction.
- Repairment of dexture is done
- Fixing of macrostomia operation is applied.

**CASE 3.**
Patient who has right tessier type 4+10 and left tessier type 3+11 cleft face
- In first year, lower and upper palpebrae creating and repairing, nose shaping and lip reparining is done.
- After first year, repairement of cleft palate + same session pharyngeal flap is done.
- Enucleaction is done due to exposure keratitis which grows on right eye.
- Within Abbe flap and atrichosis area reconstruction on upper lips contour correction process is applied.

**CASE 4.**
Patient who has one nostril, right probozis lateralis and rudimentary right half of the nose.
- In sixth month, reviving of lower eyelift, right side of the nose operation is done.
- In ninth month fistula excision in probozis stem is done.
- In twelfth month flap is diminished.

**Results:** Craniofascial abnormalities management is congenital deformity which require experince. Face is functionally very important area because of entry of digestive and airway and carrying organ of sight on it. Besides it is one of the most anatomical area which identifies and carries big importance in sedial relations. People recognize and remember each other with their faces in a first stage. Non-standard beauth or deformity in the face is caused remembering. When viewed from this aspect, looks out of the normal will be stigmatize (marker, remembering factor) for a person. Facial abnormalities' management gains huge social and functional importance in this concept.

**Summary/Conclusion:** We think that presenting rarely seen facial anomaly cases' treatment and late period consequences will be a guide for treatment in tough cases when we will face in the future.
Background: Baker and Millard first described the intraoperative suction test as a predictor of velopharyngeal competence in 1993. They described a 92% positive predictive value (PPV) for not requiring further surgery and an 83% PPV for the development of adequate speech. In AlderHey Children's Hospital, this test represents a variation in practice. It is a routine procedure in all primary palate repairs operated on by Surgeon 1 but not used by Surgeon 2.

Aims: To compare these results with a five year cohort of Surgeon 1’s primary palate repairs. Does the suction test provide reassurance in this group of adequate speech development and no further need for surgery?

Methods: All primary palate repair patients (n=80) born between January 2008 and December 2012 were included in the study. Data collected included: demographics, cleft type, primary palate repair technique, positivity of suction test, presence of fistula, need for further velopharyngeal surgery and results of cleft audit protocol for speech (CAPS-A) at age 5 between the years January 2013 and December 2018.

Exclusion criteria: incomplete or missing CAPS-A records

Results: To be presented to include PPV of suction test in relation to speech and further velopharyngeal surgery.

Summary/Conclusion: Can Surgeon 1 reliably use the suction test to inform their outcomes?
Early alveolar cleft repair with hydroxyapatite filling: 16 years follow-up.

J Cortes-Araya¹, A Diaz-Munoz²
¹Cirugia y traumatologia bucal y maxilofacial, ²Universidad de Chile, Santiago, Chile

Background: There is currently possible to achieve aesthetic and functional results, as ventilatory function, phonation and an acceptable size and position of the maxillary skeleton as result of the primary reconstruction of a cleft lip and palate. However, the morphofunctional weakness persists in the results of the primary reconstruction of the premaxillary. This problem is evident at the time of performing an advancement Lefort 1 surgery in which we check a skeletal discontinuity of the alveolar process, the maxillary base and the affected nasal floor.

According to Delaire, the soft tissues of the area involved in patients with cleft lip and palate are atrophic, deformed and displaced. The correct treatment to allow the functional interactions that define a normal skeleton implies that they must be identified, dissected and repositioned. The premaxillary cleft involves the anterior bony palate and its fibromucosa, the alveolar process, its periosteum and covering mucosa, the nasal floor with its skin, mucosa and bone and the piriform rim. The last has several muscle attachments and is subject to tensions derived from ventilation, phonation, mime and other functions. Most published periostioplasties articles emphasize alveolar closure and reconstruction, while ignoring the piriform rim, cleft and reconstruction of the nasal floor corresponding to the palatal vault. This way of proceeding also achieves that the palatal vault and the alveolar arch evolve structurally fragile.

Aims: Against that background, the decision was to perform a surgical treatment -Posnick procedure- earlier, in order to reconstruct the alveolar process, including the nasal floor and the contour of the piriform rim, thanks to the restoration of the tissue envelopes covering the three faces of the premaxillary skeleton -following Delaires’s rationale- and the placement of hydroxyapatite filling the surgical site.

Methods: In 10 children with a diagnosis of unilateral lip and palate cleft and treated according Delaire techniques and chronology for lip, and palate, between age of 1.5 to 2.5 years they were practiced a premaxillary reconstruction following these steps: 1.- General anesthesia, preparation of the operative field and injection of local anesthesia with vasoconstrictor. 2.- design the surgical accesses necessary for the premaxillary exposure, including the alveolar process and the nasal floor. 3.- Reconstruction of the nasal, palatal and alveolar mucosa. 4.- Filling of the bony space created with granulated hydroxyapatite and closure of the vestibular access.

Results: As 17 years later results, a maxillary’s advance surgery was performed. Once the maxilla is exposed, the complete reconstruction of the intervened premaxillary area is checked.

Summary/Conclusion: The technique proposed by Posnick associated with the functional reconstruction of Delaire allows a complete premaxillary skeletal reconstruction.
**P-027**

**Hemifacial hyperplasia – a case series and review of the literature**

A Dattani¹, A Heggie²

¹Oral and Maxillofacial Surgery, University of Birmingham, Birmingham, United Kingdom, ²Maxillofacial & Plastic Surgery, Royal Children's Hospital, Melbourne, Australia

**Background:** The manifestations of condylar hyperplasia are known to result in facial asymmetries and are a well-recognised group of unilateral mandibular enlargements. Condylar hyperplasia has also been sub-classified into hemi-mandibular hyperplasia and hemi-mandibular elongation. A much rarer disorder, hemifacial (hypertrophy) hyperplasia (HH) is a rare congenital malformation with a reported incidence of 1:86,000 live births. It is characterised by unilateral overdevelopment of the hard and soft tissues of the face. The affected side grows at a faster rate than the non-affected side creating a marked asymmetry. HH is usually identified at birth and progresses towards puberty but is not thought to alter throughout the lifetime of affected individuals.

**Aims:** To present a retrospective review of 5 cases of HH with respect to the clinical features and to discuss their individual surgical management. The more recent identification of possible genetic mutations that may be responsible for HH and related overgrowth disorders is discussed.

**Methods:** Patients who suffered HH were identified and data was collected from the Royal Children’s Hospital (RCH) and a Melbourne Oral and Maxillofacial Surgery practice. Individual retrospective case analysis of each patient record was carried out to ascertain the type and extent of HH.

**Results:** A total of 5 cases were identified with true HH that ranged between 6-68 years of age. Three cases were male. Left-sided asymmetry was seen in 3. Five of cases had features of tongue involvement and 4 had a midline shift. One case had hemihypertrophy of the body. One patient declined surgery whereas three cases underwent orthognathic surgery with other individualised surgery and the remaining patient underwent mandibular recontouring and genioplasty.

**Summary/Conclusion:** HH is a rare developmental deformity that may be misdiagnosed. Presentation of HH varies from mild to severe in presentation. Surgical management is via an individualised approach in correlation with clinical features. The aetiology of hemifacial hyperplasia is poorly understood, but more recently, it has been reported that facial overgrowth may result from down-regulation of phosphate tensin homolog (PTEN). It is also speculated that depending on genetic factors, the disorder may be progressive in specific cases as seen in our case series.
Unilateral cleft lip and palate: results after 18 years, one surgeon, one primary technique

A De Buys Roessingh¹, O El Ezzi¹, G Herzog¹, M Broome¹, L Medinger¹, C Zbinden-trichet¹, C beguin¹, C richard¹, S fries¹, J despars¹, Y Robert¹, G Dimitropoulos¹

¹CHUV, Lausanne-CHUV, Switzerland

Background: This study reviews the results 18 years later of our surgery and follow-up of children born in our hospital with unilateral cleft lip and palate (UCLP).

Aims: Evaluation of 18 year-old youngsters born with UCLP, operated at the time by the same surgeon with the same primary surgical procedure (Malek).

Methods: We analyzed files of children born with a cleft between 1998 and 2001 and operated in our hospital. Operations for UCLP were performed by the same surgeon following the Malek procedure, adapted according to children's age. The evaluation was based on the results of the primary surgery, ENT intervention, maxillo-facial surgery and final phonatory results. All syndromic children were excluded.

Results: Seventy-nine files of children born with a cleft were reviewed: 27 were taken into consideration for UCLP: 12 right and 15 left. Among the 27 UCLP, 84% were operated in two stages following the inverse Malek procedure, at a median of 3.2 months for the veloplasty and 5.9 months for the labio and palatoplasty; 16% were operated in one stage at 4 months. Sixty per cent had a fistula whose closure required a median of 1.5 operations. Eighty-eight (88)% had grommets, of which 4 required several operations. Ninety-seven (97)% had an alveolar graft at a median age of 9 (5-10) and 22% underwent a Lefort osteotomy at a median age of 15 years (13-16). Based on the phonatory score, 7% children were operated at the age of 6 following the Sanvenero-Rosselli technique for a pharyngeal flap. For the 27 children, fifty-five (55)% have a normal phonation with no nasal air emission (phonation I), 30% a good phonation with intermittent nasal air emission and good intelligibility (phonation I/II), and 15% a phonation with continuous nasal emission but good intelligibility and no social discomfort (phonation IIb). A median of 5.7 multidisciplinary consultations were realized with a median number of general anaesthesia of 7 (4-13).

Summary/Conclusion: This retrospective study considers the long-term results of one primary surgical technique by one surgeon. It shows that the Malek procedure (in two inverted stages) is related to a high risk of fistula but good long-term phonatory results. The number of operations needed for these children is high and this information should be transmitted to the parents from the beginning of care.
Comparison of palatal length, speech outcome and surgical complications with use of von langenbeck and bardach two flap technique for cleft palate repair: a prospective randomized study.

G Deshpande1, S Natarajan1, Y Oswal3

1Oral and Maxillofacial Surgery, MGM Dental College and Hospital, Navi Mumbai, India, 2Plastic and Reconstructive Surgery, PennState Hershey School of Medicine, Hershey, United States, 3MGM Dental College and Hospital, Navi Mumbai, India

Background: Is there a difference in palatal length, speech outcome and surgical complications between Von Langenbeck and Bardach two flap technique? Is there a difference in palatal length, speech outcome and surgical complications between Von Langenbeck and Bardach two flap technique?

Aims: To compare the outcomes of two palatoplasty techniques, Bardach and Von Langenbeck in palatal length, Complications and speech.

Methods: This study included 32 non-syndromic cleft palate patients between 4 to 30 years with repaired cleft lip or without cleft lip. They were randomly divided into Group ‘A’ (n = 16) who underwent Von Langenbeck and Group ‘B’ (n = 16) who underwent Bardach palatoplasty. Sommerlad’s Intravelar Veloplasty was performed in all patients. Each patient was evaluated for palatal and cleft measurements, speech and surgical complications. Effect of confounding factors such as age, cleft width, palatal flap width, Veau’s & Randall’s class on speech parameters and complications was also studied. This study included 32 non-syndromic cleft palate patients between 4 to 30 years with repaired cleft lip or without cleft lip. They were randomly divided into Group ‘A’ (n = 16) who underwent Von Langenbeck and Group ‘B’ (n = 16) who underwent Bardach palatoplasty. Sommerlad’s Intravelar Veloplasty was performed in all patients. Each patient was evaluated for palatal and cleft measurements, speech and surgical complications. Effect of confounding factors such as age, cleft width, palatal flap width, Veau’s & Randall’s class on speech parameters and complications was also studied.

Results: A significant amount of palatal lengthening was achieved by Bardach as compared to Langenbeck technique. Except articulation error which was corrected to a greater extent in Bardach group no other speech parameter was affected by palatoplasty technique. Increased age and severe veau’s class patients showed poor articulation. No correlation was obtained between Randall’s classification, cleft width, flap width and speech parameters. 2A complication rate of 12.5% was encountered. All the complications occurred in patients having a cleft width > 18.6 mm. A significant amount of palatal lengthening was achieved by Bardach as compared to Langenbeck technique. Except articulation error which was corrected to a greater extent in Bardach group no other speech parameter was affected by palatoplasty technique. Increased age and severe veau’s class patients showed poor articulation. No correlation was obtained between Randall’s classification, cleft width, flap width and speech parameters. 2A complication rate of 12.5% was encountered. All the complications occurred in patients having a cleft width > 18.6 mm.

Summary/Conclusion: Although Bardach technique achieved a significant palatal length no difference was observed when considering the overall speech outcomes. Intra Velar Veloplasty re-established the velopharyngeal sphincter and improved the quality of speech. Confounding factors such as age & Veau’s class can alter the speech outcomes and increased cleft width can increase the complications of palatoplasty.
Background: Cleft lip and/or palate (CL/P) anomalies are common congenital defects in the United States (1:900 live births). Pediatric and adult patients with these diagnoses may have many associated medical and physical challenges including speech impairments, difficulty hearing, issues with social integration, and challenges with self-worth and appearance. While some data exist on access barriers to primary craniofacial care, the specific medical and social needs of the adult CL/P population need to be more clearly elucidated. This study highlights the presence of facial dysmorphia in CL/P patients and is the first to quantify this phenomenon in the United States.

Aims: To identify specific psychological burdens related to living as adult with CL/P and to characterized the degree of FDD symptoms in an adult craniofacial population.

Methods: Patients were consented for our study under the University of North Carolina-Chapel Hill Institutional Review Board protocol 16-1632. All participants were administered a 12 question Body Dysmorphic Disorder-Yale Brown Obsessive Compulsive Scale (BDD-YBOCS) questionnaire to assess for both presence and severity of BDD symptoms (n=20 non-CL/P patients, n=30 CL/P patients). Qualitative, semi-structured interviews were also conducted with a total of all adult CL/P participants. Interviews were focused on social issues related to self-identity, life experience living with an orofacial cleft, socioeconomic access, barriers to healthcare, and needs and desires of each patient with respect to medical and dental management.

Results: There were no significant differences in control or CL/P patients mean ages (40.3 versus 42.2 years), sex (75% versus 66% female), or ethnicity (20.0% versus 25.9% non-white). Patients with CL/P had a significantly higher facial dysmorphic disorder score (mean 13.3) compared to control participants (mean 4.2; p<0.001) as well as higher obsession (2.9-fold increase, p<0.001) and compulsion subscores (3.3-fold increase, p<0.01). Qualitative interviews revealed three major themes present in CL/P patients experiences, including: access to cleft-care professionals, perception of physical appearance and the effect of CL/P on social relationships (3.3-fold increase, p<0.01).

Summary/Conclusion: Adult patients with cleft anomalies routinely experience both medical and social barriers to receiving care. Among our patient population, we have identified that patients with CL/P have significant facial dysmorphic disorder symptoms compared to non-CL/P participants. Further, we have identified several high-need specialties to be integrated into a multi-disciplinary adult cleft clinic, including: facial plastic surgery, specialized dental care, and psychosocial support. Addressing residual surgical and dental needs appears to be a realistic goal or outcome for adults who present with clefts. Integrating these services into a single, recurring clinic appears to have impact on the health and wellness of the adult CL/P population.
Treatment of proboscis lateralis: a novel surgical approach using tape elongation and a one-stage tunneling procedure

O Engström¹, M Hakelius¹²
¹Plastic Surgery, Uppsala University Hospital, ²Department of Surgical Sciences, Plastic Surgery, Uppsala University, Uppsala, Sweden

Background: Proboscis lateralis is a rare craniofacial anomaly in which the nasal soft tissues have formed a tubular, trunk-like structure, and is caused by an incomplete fusion of the midface. It is a rare condition, occurring in less than 1 in 100,000 births, and may be associated with other facial malformations.

Surgical treatment of proboscis lateralis is mainly based on two different techniques, either using the de-epithelized skin island from the proboscis lateralis and suturing it to the lateral nose wall or de-epitelizing the proboscis before tunneling it and suturing it to the alar rim.

Aims: Here, we present a case of proboscis lateralis in a young girl being treated using a novel surgical technique combining tape elongation and one-stage tunneling.

Methods: An 18-month-old adopted girl, originating from China, was referred to our institution for treatment of her proboscis lateralis, 5 months after her arrival to Sweden. She presented with a solid conical structure protruding from the right supramedial canthal region, consistent with group III according to Boo-Chai. The appendage measured 1 cm in total length and 1 cm in width at the distal end and was seated in the supramedial canthal area. She also had a coloboma of the right medial upper eyelid, a caudally displaced medial canthus on the right side and an absence of a nasal cavity and maxillary sinus on the right side. Her development was normal, but she suffered from severe snoring.

In order to treat the condition, decision was made to use a one-stage tunneling method. Due to the high position and relatively short length of the proboscis, she was treated with preoperative elongation using surgical tape. The tape was applied around the pedicle of the proboscis and after 4 months of elongation treatment the proboscis reached a length of 3.5 cm. The patient was able to undergo surgery at 2.5 years of age.

The tubular structure was de-epithelized, and the pedicle of the proboscis was mobilized with great caution to avoid damaging the pedicle. A tunnel was dissected subcutaneously on the right side of the nose and a triangular incision was made for the new opening. The distal end of the tubular structure was tunneled to the location of the right nostril and sutured.

Results: The patient was discharged on the second postoperative day. Further procedures have included primary and secondary medial canthopexy, dacrocystorhinostomy, tonsillotomy and a reconstruction of the back of the nose using a dermis graft from the groin. The patient is now 10 years old and no further surgery is scheduled.

Summary/Conclusion: Proboscis lateralis, also known as half-nose, is a rare congenital anomaly. Several aspects need to be addressed when planning for the reconstruction, including timing of the procedure, availability of tissue and the nature of the defect. Our patient presented with a relatively short appendage, providing insufficient length for a reconstruction using previously described techniques. By first using tape elongation and then tunneling the proboscis in a one-stage procedure, we were able to create a sufficient amount of tissue to reconstruct the right side of the nose. To our knowledge, this is the first case combining these techniques. Given that a contralateral airway is present, we conclude that our novel tape-elongation technique in combination with one-stage tunneling provides a good treatment alternative.
P-032
Objective and subjective evaluation of velopharyngeal dysfunction (VPD) following surgical re-repair of the cleft palate using the Furlow palatoplasty
N Fallico¹, M Rizzo¹, O Rajabtork Zadeh¹, S Vagnoni³, A Palmieri⁴, M Zama¹
¹Plastic and Maxillofacial Surgery Dept, Cleft and Craniofacial Malformation Center, Bambino Gesù Children Hospital, Rome, Italy, ²South Thames Cleft Service, Guy's and St Thomas’ Hospital, London, United Kingdom, ³Speech therapy, Bambino Gesù Children Hospital, ⁴Statistics, Istituto Superiore della Sanità, Rome, Italy

Background: Velopharyngeal dysfunction (VPD) diagnosis and speech surgery outcomes are currently based solely on subjective evaluation criteria consisting of perceptual speech assessment and high-compliance functional imaging.
Aims: This study describes an objective and comparable method in VPD assessment and investigates the concurrence between the objective and subjective evaluations.
Methods: The present study included 20 paediatric patients presenting with VPD after primary repair (intravelar veloplasty) of cleft palate. Our protocol was based on computerised analysis of voice parameters and nasal emission by means of two objective tools: spectrography with MDVP software and rhinomanometry. The protocol also included perceptual evaluation by speech therapist and phoniatrician (consensus listening), and parents. This is a single centre experience and all patients underwent a secondary Furlow’s palatoplasty. Assessments were performed pre and postoperatively and upon completion of speech therapy. Results were compared using the two-tailed t student test for paired data. Statistical significance was set for p-values <0.01.
Results: Spectrographic analysis and rhinomanometry confirmed an improvement in velopharyngeal closure after surgery and speech therapy consistently with the results of perceptual evaluations.
Summary/Conclusion: The study confirmed the availability and reliability of an objective method for VPD evaluation based on the analysis of voice parameters and nasal emission with examinations that are of simple execution, already present in hospitals, require low compliance and provide quantitative data to compare pre and postoperatively along with perceptual assessments.
A multidisciplinary approach on premaxillary downward protrusion in complete bilateral cleft lip and palate post cheiloplasty and palatoplasty: a one year follow-up case report

P. Gianni¹, P. Kreshanti¹, J. Pancawati²

¹Department of Surgery, Medical Faculty, Universitas Indonesia- Cipto Mangunkusumo Hospital, Plastic Reconstructive and Aesthetic Surgery Division, ²Department of Oral and Dentistry, Dental Faculty, Universitas Indonesia, Orthodontics Division, Central Jakarta, Indonesia

**Background:** Severe premaxillary protrusion is commonly found in patients with bilateral cleft lip and/or palate (BCLP), this condition may become an obstacle to plastic surgeon. This patient was presented with premaxillary downward protrusion with a collapsed dental arch. A collapsed dental arch can be a problem to perform presurgical preparation before conducting a corrective surgery.

**Aims:** To present the importance of multidisciplinary approach towards a complex cleft and craniofacial case.

**Methods:** This case report was conducted between March 2018-March 2019. A 4-year old male patient was presented to the cleft and craniofacial center with severe premaxillary protrusion in complete bilateral cleft lip and palate who underwent lip repair and palate surgery in charity setting by the age of 1. Functional disability in this patient including deep bite and nasal speech caused by palatal fistula. This patient had not associated with any other anomalies or syndromes nor familial history of cleft. Our center used the dual palatal expander which allows the enlargement of the gap. After the gap between the palate reached the optimal width, then we performed the surgical repair using the premaxilla set back osteotomy, fixation with absorbable plate and screw, then mucoperiosteal flap (gingivoperioplasty).

**Results:** After the surgery, the patient shows better occlusion, adequate lip closure and aesthetically improved in appearance, enhancing patient's level of confidence.

**Summary/Conclusion:** This dentofacial anomaly requires multidisciplinary approach including presurgical assessment by the orthodontist to ease the surgical correction resulting the optimal result and also by speech therapist to assist the nasal speech problem. The timing of the surgery might vary as compare to previous study to achieve an optimal midfacial growth whilst maintaining social function approaching school age.
A review of the incidence and outcomes for children with cleft lip/palate and charge syndrome

M Farid1, L Cafferky2, C Hillyar3, R O’Sullivan2, M Kershaw4, J Kirk4, R Slator1
1Cleft and Craniofacial Surgery, 2Speech and Language Therapy, Birmingham Children’s Hospital, Birmingham, 3Barts and The London School of Medicine and Dentistry, London, 4Department of Paediatric Endocrinology, Birmingham Children’s Hospital, Birmingham, United Kingdom

Background: CHARGE syndrome is rare with a prevalence of 2 per 10,000 live births and an autosomal dominant pattern of inheritance. Cleft lip +/- palate (CL+/-P) is a recognised feature of CHARGE syndrome but there is little published literature to guide management of the cleft in this group of patients.

Aims: The aim of this study was to identify all children in our hospital with a diagnosis of CL+/-P and CHARGE syndrome, and review their cleft characteristics and care in comparison to nonsyndromic children with CL+/-P.

Methods: Medical case notes and electronic data were used to identify all children diagnosed with CHARGE syndrome and CL+/-P between 1989 and 2019 in a Children’s Hospital with a Regional Cleft Centre serving a population of 6 million. The diagnosis of CHARGE syndrome was based on genetic testing for CHD7 mutation and clinical features. Hospital records were used to identify the cleft type, and the timing of cleft lip and palate repair. Reasons for ‘delayed’ surgical repair were noted. Difficulties with and methods of feeding and communication were also recorded.

Results: 23 children with CL+/-P and CHARGE syndrome were identified. 8 had complete bilateral cleft lip and palate (35%) and 2 incomplete bilateral cleft lip and palate (9%), 7 had complete unilateral cleft lip and palate (30%), 1 had a very mild incomplete cleft lip only (4%), and 1 had a unilateral cleft lip and alveolus (4%). 2 children had a complete cleft palate only (9%), and 2 had a submucous cleft palate (9%).

All but one patient had primary repair of both lip and/or palate. One patient with a complete unilateral cleft lip and palate had only cleft lip repair and no palate repair. The mean age for cleft lip repair was 14 months (range 5-66 months) while for palate repair it was 27 months (range 9-56 months).

Recurrent chest infections appeared to be the predominant reason for delayed or cancelled cleft surgery. Multi-modal forms of communication (including signing and speech), and of feeding (Oral, Enteral, and Parenteral) were noted throughout the treatment journey.

Summary/Conclusion: This study suggests that CHARGE syndrome occurs in <1% of the children with CL+/-P that we see. However, children with CHARGE syndrome have more severe forms of CL+/-P with 41% having bilateral CL+P in this group. Overall bilateral CL+/-P in the United Kingdom accounts for only ~10% of the cleft population.

Most children with CHARGE syndrome had their primary cleft lip and palate repair later than children with non-syndromic clefts, but apart from one child, they did all have their cleft lip and palate repaired. Delays in surgery were usually due to ill health in the first year of life.

Children with CHARGE syndrome had a greater range of problems with feeding and communication than non-syndromic children with CL+/-P.

The outcomes of this study are in line with the paper published by Isaac et al in 2017. The information gained will help us in consultations with families, and in guiding expectations for outcomes.
Current UK trends in primary surgical interventions for cleft lip and palate
M Fell\textsuperscript{1}, A Davies\textsuperscript{2}, Y Wren\textsuperscript{2}
\textsuperscript{1}North Bristol NHS Trust, \textsuperscript{2}Cleft Collective, Bristol, United Kingdom

\textbf{Background:} The James Lind Alliance prioritised research to investigate the best protocol for primary repair of both the lip and palate including technique, timing and sequence. The CSAG and CCUK reports both described variation in surgical cleft protocols in the UK. It is important that surgical practice is recorded so that trends and outcomes can be analysed.

\textbf{Aims:} This study describes primary repairs performed on a large cohort of UK patients with cleft lip and/or palate.

\textbf{Methods:} Data was obtained from the Cleft Collective Birth Cohort, a national longitudinal cohort study. Surgical forms, completed by the operating surgeon at the time of the primary surgery, provided data on the cleft pathology, the primary surgical operation and adjuncts used during the operative period. Demographic data on participants was collected via parental questionnaires.

\textbf{Results:} To date, 1268 surgical forms have been completed, of which, 1186 detail a primary lip (N=598) or primary palate repair (N=588). The data shows that the mean age at primary lip repair is 4.29 months and 9.43 months for primary palate repair. Further descriptive statistics on cleft pathology and techniques used across the UK will be presented.

\textbf{Summary/Conclusion:} The centralised nature of UK cleft care means that it is possible to identify current practice, analyse outcomes and facilitate international comparison. This descriptive analysis contributes to our understanding of current UK practice and will lead to analysis of the effectiveness of protocols used.
**P-036**


M CORTEZ LEDE\(^1\), C FENOLLAR QUEREDA\(^2\), M VIÑAS PINEDO\(^3\), V VILLANUEVA SAN VICENTE\(^1\), M RODRIGUEZ GONZÁLEZ\(^1\)

\(^1\)ORAL AND MAXILLOFACIAL SURGERY, HOSPITAL CLÍNICO UNIVERSITARIO VIRGEN DE LA ARRIXACA, \(^2\)ORTHODONTIST, PRIVATE PRACTICE, MURCIA, \(^3\)ORTHODONTIST, ASSISTANT PROFESSOR ORTHODONTICS UCM, MADRID, Spain

**Background:** Orthopedics in clefts patients is usually performed preoperatively before closing the lip, modeling the affected nostril to achieve aesthetic improvement in both the lip and nose. Most of the articles focus on analyzing the nasal symmetry and the projection, others focus on the relationship of presurgical orthopedics with the growth of the maxilla. There are many differences, in the type of plates and in the closing protocol of both the lip and the palate, which can affect maxillary growth. The first publication on presurgical orthopedics was made by McNeil in 1950 and there are still detractors of the technique.

**Aims:** The objectives are focused on evaluating with three-dimensional analysis how the maxillary development is affected performing a functional maxillary orthopedics and how surgeries can affect to it.

**Methods:** We proceed to analyze using a dental scanner a series of cleft palate cast. The patient had a left unilateral complete cleft lip and palate that has been treated from the third day of birth to the present through orthopedic plates based on the McNeil studies modified according to Dr. Pannaci. We have done monthly impressions from birth to 12 months and then every two months. Analyzing the differences between the first model, the fifth (before performing a frenectomy), before and after the cheiloplasty (7th and 8th), before and after the soft palate surgery(11-12th) and the last one. The orthopedic plate has to generate pressure points on the palate and must be loose or not very tight in the mouth, to get a mild and intermittent stimulus, in this way we can model the palate.

**Results:** Digital models where used to assess treatment effect. The anterior cleft width diminished 13.13 mm. The distance between tuberosity points remained stable. After lip repair the premaxilla rotate very fast, and the maxillary dental arch had a better form. We observe that before we closed the soft palate the patient didn’t use the plate and we have a recurrence of the lesser segment rotating up and collapsing it in the anterior part, that could be due to the pressure of the tongue. The soft palate closed do not alterate the posterior width of the maxillae. By maintaining functional orthopedics we manage to avoid transversal maxillary compression and improve the shape of the arch.

**Summary/Conclusion:** We believe that the restoration and maintenance of the maxillary arch until the primary dentition is completed and the stimulation of the growth generated by orthopedics together with the delay in closing the hard palate can improve the growth of the maxilla. More exhaustive studies are needed to corroborate the results.
Treatment algorithm for sequelae in adolescents and adults with cleft lip and palate. Three-dimensional surgical approach.

M CORTEZ LEDE¹, V CABALLERO ILLANES¹, A CAMACHO LUNA¹, D MARTIN BOTELLA¹, V VILLANUEVA SAN VICENTE¹, A SORIANO¹, E TERUEL HERNANDEZ¹, M RODRIGUEZ GONZÁLEZ¹
¹ORAL AND MAXILLOFACIAL SURGERY, HOSPITAL CLÍNICO UNIVERSITARIO VIRGEN DE LA ARRIXACA, MURCIA, Spain

Background: The sequelae in multioperated clefts patients are a challenge for the surgeon. Bone deficiency, hypoplasias compensated dentally, the consequences of bone loss due to maxillary advances, retractable scars, etc, complicate the approach of adult patients. Aims: Explain how is our algorithm to decide the most appropriate approach according to the sequel presented by the patient and develop what treatments the patients have had and the aesthetic improvement obtained. Methods: Four clinical cases are presented. The first patient is a teenager with sequelae of unilateral cleft lip due to trauma after surgery. It is approached through a Millard technique with Mulliken modifications and a primary rhinoplasty with septoplasty. The second patient is an adult with bilateral cleft lip and alveolus who had been made an Abbe flap to rebuild the philtrum and proceeded to perform a secondary rhinoplasty with rib graft and retouching of the lip scars with dermofat graft in the upper lip. The third patient presents a cleft of the right full unilateral lip with left facial paresis, proceeding to perform a cheiloplasty using the Millard technique with Mulliken modifications and a closed rhinoplasty with rib graft and facial lipofilling. The fourth patient presented a left unilateral palatal cleft lip to which tertiary rhinoplasty was performed with costal graft, facial, palate and pharyngeal walls lipofilling. Results: The photographic study and the CT Scan of the patients allows us to have a three-dimensional idea of the defects and use both osteochondral and costochondral grafts to reconstruct the defects in a three-dimensional way. We also rely on dermofat or lipofilling grafts to improve the projection and labial volume. The lipofilling can be used to restructure the facial projection and improve the aesthetics, both for maxillary and malar hypoplasia and for alterations and asymmetries derived from facial paralysis. Summary/Conclusion: We would like to share the way we approach the cases and the results that we are obtaining by carrying out complex treatments based on a three-dimensional restructuring of the defects of our patients.
Ten years of palatoplasty: an analysis of 667 primary cases
Alessandro Giacomina, Gian Luca Gatti, Renata Salvadorini, Beate Kuppers, Brita De Lorenzo, Luca Rosato

1Plastic Surgery, Hospital of Pisa - Dept. of Plastic Surgery, 2Plastic Surgery, Hospital of Pisa, 3Speech Therapy, Stella Maris Foundation IRCCS, 4Anesthesia, Hospital of Pisa, Pisa, 5Plastic Surgery, Hospital of Piacenza, Piacenza, Italy

Presentation Background: Clefts Palate are different regarding anatomy, wide and complexity; if related to cleft lip they may need more aggressive procedures than soft palate clefts. Aims: In our experience the two most important goals of surgery are: the optimal lengthening of the velum and the anatomical closure of all layers, with muscular repositioning and reconstruction. Methods: in the “Percorso Labiopalatoschisi” of Pisa the major techniques are two: Bardach’s palatoplasty, which is performed in complete CLP and in clefts that need a strong lengthening, and “minimal lateral incisions” or “no lateral incisions” palatoplasty. In both of them the reconstruction is achieved with complete closure of all layers of palatal anatomy (nasal, muscular, oral). The timing is between 6 and 7 months; in some selected cases surgery is performed at 5 months with cheiloplasty procedure. Patients can eat before and after the surgery. In rare cases (mostly PR patients) we use a nylon 3-0 at the basis of tongue to safely pull-out it when needed. Main points of technique are Ernst space dissection, Pigott’s eversion sutures and other minor tricks Results: from 2009 to 2018 we have treated 667 patients with primary cleft palate; 252 patients had a Veau I-II clefts, 415 patients had a Veau III-IV clefts. The percentage of P-R syndrome or sequence is between 20% and 30% (of Veau III). No further treatments were needed (mandibular distraction/tongue-lip adhesion). Fistula percentage is about 10%, nearly all of type V-VI of Pittsburgh class. (in Veau III-IV). Complications were 4 (1 bleeding, 2 dehiscence with reoperation, 1 necrosis of palatal flap), 0.91%. Hyperrinophonia (VFI Test – Glazer mirror, air paddle and auscultatory tube) is present in less than 25% of patients (aged from 4 to 18 years), compared with 50% of patients coming from other Hospitals (for different procedures). Summary/Conclusion: surgery of cleft palate is a difficult challenge for all surgeons. The speech assessment and the facial growth are the most important outcomes to evaluate during years. So many techniques historically were proposed as the best one but we believe that complete knowledge of palatal anatomy and dynamic and the skilled hand of surgeons are the keystone of a successful treatment.
The effect and timing of surgical closure on postoperative complications in children with cleft palate: a randomized, prospective study

C Guillaume1, I de Vries1, T Hamelink1, S Haverkamp2, A Mink van der Molen1, M Kon1, C Breugem1
1Department of Plastic Surgery, 2Department of Speech Therapy, Wilhelmina Children's Hospital, University Medical Center Utrecht, Utrecht, Netherlands

Background: Cleft lip and palate is a world-wide common birth defect with an incidence of approximately 1 in 750 live births. Although the prevalence of oral clefts in the Netherlands has been decreasing over the past two decades, it is still a major part of all congenital deformities. There has not been reached a consensus about the perfect timing of the palatal closure. In our unit, the soft and hard palate are being closed simultaneously before the age of 12 months. We would like to investigate whether the palatal closure can be done at an even earlier age (6-8 months) without putting the child at a higher risk of surgical complications. Possible surgical complications are postoperative bleeding or infection, or the occurrence of a fistula. Younger children could be at a higher risk for immediate postoperative airway problems after palate surgery.

Aims: The aim of the study was to determine if earlier surgical closure of the palate in children with cleft palate puts the child at a greater risk for postoperative complications.

Methods: A randomized controlled intervention trial was performed of 70 patients visiting the Pediatric plastic surgery division at Wilhelmina Children's Hospital, University Medical Center Utrecht, The Netherlands. Between 2012 and 2019. The study population was randomly divided in an early group (N=36) with surgical closure at the age of 6-8 months, and a late group (N=34) with surgical closure at the age of 10-12 months. Data were collected for postoperative complications such as infection, bleeding or fistula, and also length of hospital stay and duration of surgery.

Results: 70 children with a palate cleft were included in the study. Group A contained one participant with a fistula and four participants with other postoperative complications. Four participants in group B developed a fistula and three participants developed another postoperative complication. Postoperative complications and fistulization were not significantly different in group A compared with group B.

Summary/Conclusion: Surgical closure of a cleft palate at an age of 6-8 months, in comparison with closure at an age of 10-12 months, does not result in a significantly higher occurrence of postoperative complications, such as infection or fistula.
Van der Woude syndrome; fistula & velopharyngeal surgery: a retrospective study
C Guillaume1, B Binkhorst1, M van den Boogaard2, A Mink van der Molen1, M Kon1, C Breugem1
1Department of Plastic Surgery, Wilhelmina Children's Hospital, University Medical Center Utrecht, Utrecht, 2Department of Human Genetics, Wilhelmina Children's Hospital, University Medical Center Utrecht, Utrecht, Netherlands

Background: Van der Woude syndrome (VWS) is one of the most common syndromes associated with oral clefts. Little is known about the clinical outcome after surgical treatment.

Aims: In this study an analysis was performed on fistula rate after cleft palate closure, and the incidence of secondary velopharyngeal surgery.

Methods: A retrospective cohort study was performed at a tertiary care center. All subsequent cases of Van der Woude syndrome known at the cleft team and department of human genetics at the Wilhelmina Children's Hospital between 1970 and 2016 were included. This group consisted of 22 patients (11 men, 16.8y mean age). The main outcome measures were post-operative fistula, quantity of velopharyngeal surgery and speech (nasality and articulation).

Results: Twenty patients were included in the study. 2 patients: with no cleft, 2 patients had a solitary cleft lip/alveolar cleft. 18 patients were identified with cleft lip and/or palate. Veau type 1: 6 (33.3%), Veau type 2: 2 (11.1%), Veau type 3: 0, Veau type 4: 10 (55.6%). 17 patients needed a primary palatal closure. Three (Veau type 4) out of 17 patients (17.6%) developed a palatal fistula after palatal closure, while in 10 (Veau type 1: 2 patients, Veau type 4: 8 patients) out of 17 patients (58.8%) secondary velopharyngeal surgery was needed to improve speech abnormalities.

Summary/Conclusion: The present study describes fistula rates and velopharyngeal surgery after surgery of VWS patients. We observed that a rather large amount of the VWS patients have a Veau type 4 oral cleft. This could explain the high rate of fistulas and the high rate of velopharyngeal surgery needed in this group.
P-041

Pierre robin sequence: incidence of speech-correcting surgeries and fistula formation
C Gustafsson¹, P Vuola¹, J Leikola¹, A Heliövaara¹
¹Department of Plastic Surgery, Cleft Palate and Craniofacial Center, Helsinki University Hospital, Helsinki, Finland

Background: Velopharyngeal insufficiency (VPI) is common in children with corrected cleft palate. Pierre Robin sequence (PRS) is a rare condition in infants, comprising micrognathia, glossoptosis, and airway obstruction. It is often associated with a wide cleft palate.

Aims: The study’s purpose was to determine the long-term incidence of speech-correcting surgeries (SCS) and fistula rates in PRS after primary palatoplasty and the differences between affecting factors.

Methods: A retrospective single-center, observational chart review study. The cohort comprised 78 non-syndromic children with PRS (48 female) born between 1990 and 2009 and treated at a single center. Affecting factors compared included gender, surgeon, age at primary palatoplasty, surgical technique, airway obstruction in infancy, and cleft severity. The outcome was analyzed at age 8 years and at data retrieval.

Results: SCS were performed to 34 children (43.6%) by age 8, 6 (7.7%) of the 19 (24.4%) post-operative fistulas underwent closure. By data retrieval, 37 children (47.4%) had undergone SCS and 8 (10.3%) had a fistula closure. Median age at SCS was 6 years and at follow-up 14 years. The results showed no significant association for gender, surgeon, age at primary palatoplasty, surgical technique, cleft severity, or airway obstruction in infancy regarding incidence of SCS, fistulas, or repaired fistulas.

Summary/Conclusion: PRS in children is associated with a high incidence of SCS and fistula formation, which necessities accurate clinical follow-up and observation of speech development. The development of VPI in PRS is complex and challenging to estimate at the time of primary surgery.
Hints for repair of cleft palates with furlow's technique

E Güvercin1, G Taylan Filinte2, S Oz1, B Kanık1, S Çıldır1

1Plastic, reconstructive and aesthetic surgery, İstanbul Dr. Lutfi Kirdar Kartal Training and Research Hospital, 2İstanbul, İstanbul, Turkey

Background: Cleft palate is a frequently seen congenital malformation worldwide and it creates financial, medical, psychological and social burden. Genetic and environmental factors are equally responsible for the etiology of cleft palate. In addition to aesthetic problems, cleft palate cause disfagia, inadequate feeding, maxilla-facial development failure, ear infections and speech impairment.

Aims: There are currently various techniques in cleft plate repair. New techniques are developed due to high incidence of fistula formation, maxillary retrusion, and velo-pharyngeal deficiency with the old techniques. Furlow introduced the Double opposite Z-plasty technique and this technique became the most widely used today. Hence in this study we aimed to share our experience with the Furlow technique.

Methods: In this study nine patients who underwent Furlow Z Plasty between the years 2017-2019 were analyzed. In all patients mucomuscular anterior and posterior flaps were established, and the defect is closed by transposition. All patients are examined by videofluroroscopy and transferred to speech therapist postoperatively.

Results: The mean age of the patients were 20.5 months (range: 8-52). The mean follow up period was 68.7 month (range: 9-117 month). No major complications as palatal fistula, speech impairment and velopharyngeal insufficiency were seen postoperatively.

Summary/Conclusion: The main advantage of Furlow palatoplasty is that it provides elongation without the use of hard palate. This technique allows total dissection palatal aponeurosis, precise dissection of muscles and enables transvers orientation. Other than doing a straight incision, zig zag incision provides better functional outcomes for the utilization of soft palate. Fistula rate is relatively less than the other techniques. In addition to result in less velopharyngeal insufficiency, it is also used for the repair of velopharyngeal insufficiency. Moreover palatal competence is better with this technique.

Although it seems simple, the success rate is based on experience and it has a steep learning curve. It should be practiced in the senior years of the residency and our clinical practice is in this manner. Flap angels should be planned close to 60 degrees posteriorly and 90 degrees anteriorly otherwise the closure of the donor site would be difficult. The post-operative care should be attentive. Horizontal mattress sutures should be employed for the closure and a lot of knots should be utilized. Preoperative planning is a necessity otherwise other techniques would not help the closure of the defect. The operative time is longer than the other techniques. In wider defects, application of back cut technique and the release of mucoperiostal flaps might necessitate. The disadvantage of zig zag incision is that without the dissection of the muscles the reopening of the soft palate is sometimes a drawback.

This technique is widely used currently due better elongation to palate, low incidence of postoperative fistula rate and better speech development. However it has a steep learning curve and preoperative planning and postoperative attentive care is a necessity for optimal results.
Dental infection cause of flap loss in cleft palate repair: a rare occurrence
G Deshpande¹, R Jagtap²
¹Oral and Maxillofacial Surgery, ²Periodontics and Implantology, MGM Dental College and Hospital, Navi Mumbai, India

Background: Flap necrosis is a very common complication encountered after cleft palate repair, especially in uni-pedicled flaps. Many causes have been attributed to this complication but very limited data is available in the literature on dental infection as the cause of flap necrosis. This report of a case describes loss of flap caused due to a periapical granuloma of dental origin impinging on the pedicle causing suspected thrombosis of the pedicle resulting in flap necrosis. Since this is an important cause that can be easily prevented, it is reported.

Aims: To discuss the preventive measures for flap loss due to dental infection

Methods: Rare case reported discussing the causes of flap loss due to dental granuloma

Results: Successful rehabilitation can be achieved even after the loss of flap, but prevention of such complication is more important

Summary/Conclusion: Although, very rare and never reported, dental infection can cause flap necrosis and can be prevented by simple intervention before the surgery
Palatal growth changes one year after early neonatal cheiloplasty in different cleft types
L Jaklová¹, J Borský², M Jurovčík², E Hoffmannová¹, J Dupej¹3, J Velemínská¹
¹Department of Anthropology and Human Genetics, Charles University, ²Department of Otorhinolaryngology, 2nd Faculty of Medicine, Charles university and Motol University Hospital, ³Department of Software and Computer Science Education, Faculty of Mathematics and Physics, Charles University, Prague, Czech Republic

Background: Early neonatal cheiloplasty performed in the first week of life has been applied in the Czech Republic for 13 years. This modified surgery protocol has many advantages and our previous studies furthermore confirmed that it has no negative effects on the growth of the palate in bilateral (BCLP) or unilateral (UCLP) cleft lip and palate patients.

Aims: Goals are to compare palatal growth trends in complete and incomplete BCLP and UCLP newborns one year after early neonatal cheiloplasty and put these results into context with published data of cleft neonates operated by later operation protocol (realized between 3–6 months) and noncleft groups.

Methods: The sample of 26 BCLP and 66 UCLP individuals was analysed using classical (2D) and geometric (3D) morphometry. Each patient provided two dental models: T0 before early neonatal cheiloplasty (BCLP 5 ± 5 days, UCLP 5 ± 4 days) and T1 before palatoplasty (BCLP 12 ± 6 months, UCLP 10 ± 2).

Results: Within classical morphometry significant growth differences between one-year-old patients with complete BCLP and UCLP were found in selected dimensions, such as palatal length, intercanine width and anterior basal angle. Incomplete BCLP and UCLP newborns varied significantly in all analyzed dimensions except palatal length immediately after birth. These differences were reduced during the observed period and after that these individuals differed significantly in anterior basal angle only. Three-dimensional palatal growth visualization created by pair analyses showed the most intensive growth changes in the anterior part of the upper dental arches in both BCLP or UCLP neonates. In analyzed cleft types alveolar clefts were narrowed during the 12 months as a result of anterior growth of the palate combined with natural formative effect of the lip suture. Early neonatal cheiloplasty did not restrict the palatal growth in posterior direction in any cleft types, thus there was no decrease in the dentoalveolar arch. The more favorable growth tendencies in anterior and posterior direction were noted in less serious UCLP defects as well in incomplete variants interconnected by tissue bridges of both cleft types. Comparison of selected dimensions in observed groups demonstrated that individuals with incomplete BCLP and UCLP grew equally to noncleft group in intertuberosity width and palatal length. Complete BCLP and UCLP patients exhibited similar growth tendencies as later operated neonates in those same dimensions. In intercanine width UCLP patients showed same growth trends as noncleft group only, while BCLP neonates grew in the same way as later operated group. Palatal variability was enormous in complete BCLP individuals and decreased during the monitored period. In incomplete UCLP and BCLP patients the variability of more severe BCLP approximated UCLP variability.

Summary/Conclusion: Combination of 2D and 3D methods is suitable for detailed evaluation of palatal development. Although more positive growth tendencies were observed in incomplete variants of cleft defects, it can be concluded that early neonatal cheiloplasty does not affects the palatal growth in BCLP nor in UCLP individuals in any direction and can be considered as an acceptable method to the later operation protocol.
**P-045**

**Pierre robin sequence; mean and long term follow up: is it justified to operate at 3 months?**

I James, T Kraft, A Goyet, P Pozard, J Godeneche, C Chamard

1 pediatric surgery, clinique du val d'ouest, Ecully, 2 pediatric surgery, hopital femme mere enfant, Bron, 3 maxillofacial surgery, clinique du val d'ouest, Ecully, 4 speech pathology, hopital mere enfant, bron, 5 speech pathology, 6 orthodontics, clinique du val d'ouest, ecully, France

**Background:** Pierre Robin sequence represents 2 major early challenges: feedings and respiratory troubles, and late issues: speech results and facial growth. Actually, persistent questions, which answers could be different between teams, are related to breathing assistance, feeding process and optimal time to repair the cleft.

**Aims:** The Authors report 33 cases with a mean of 8 years follow up, to provide an answer to these questions

**Methods:** They found: 22 girls 11 boys, 7/33 syndromic forms. The severity was divided in 8 grade I, 12 grades II and 13 grades 3; All had a palatal plate before surgery. We describe means of feeding, oral stimulations and possible needs for nasogastric tubes. All had pre and post-operative polysomnographic study, Polysomnographic data: apneas/hypopneas index, percentage time <88% oxygen saturation, mean carbon dioxide rate are analyzed

Means of breathing assistance used are: Ccap 16, nasopharyngeal tube 1 or both together 4; we never used tracheostomy, labio glossopexy or distraction. All of them has been operated at 3 months of age by the same surgeon: 27 times the cleft was totally closed in one time, 6 in 2 times because of the width of the cleft, to avoid flap translation of the oral mucosa, what we know is deleterious for maxillary growth

**Results:** Results are analyzed with suitable follow up: > 1 year for respiratory and feeding results; >3 years for speech and early orthodontics results >7 years for mean orthodontics and school learning results

We note that for syndromic cases the prognosis is more related to the syndrome that to the Pierre Robin sequence

The most important information in that study is: in non-syndromic cases, pre-operative severe OSA (obstructive sleep apnea) stopped after surgery to allow the remove of breathing assistance; 17/21 cases are weaned immediately after surgery and the remaining 4 cases kept cpap or nasopharyngeal tube for a maximum of 2 months.

In primary teeth at the first pluridiciplinary consultation we find 13 Class I, 18 Class II before orthodontic treatment

In Mixt and adolescent teeth: 14 class II, 10 class I, 1 class 1->III et 2 concave profile due to pression by Cpap mask over 6 months

5 need pharyngoplasty at a mean age of 6

4 cases, who had an diagnosis of isolated Pierre robin, had learning difficulties, attention troubles, dyslexia or cognitive troubles

**Summary/Conclusion:** In conclusion, this early surgery is possible without increasing respiratory problems; it had instead allowed to improve respiratory status of the children; Our theory, is that early palate repair leads to a better short latency reflex of suction and swallowing that benefits for feeding and breathing conditions. This is only a retrospective study and in the futur we would like to share these data with other teams to build a prospective work on large series of patients.
P-046

lip reduction surgery with autologous lip dermal fat graft augmentation in secondary notching lip deformities

P Jittilaongwong

1Plastic surgery, Punisa Lip and Plastic surgery unit, Bangkok, Thailand, Bangkok, Thailand

Background: Lip reduction surgery can improve symmetry and undesirable shape of congenital lip deformities. Patients who have undergone a lip reduction surgery by Dr. Pusit technique, “Seagull wing incision”, which is a specific technique to bring the best result of lip reduction surgery for both aesthetic purposes and abnormality correction. Autologous lip dermal fat grafts of removal lip tissue have been used to augmentation the free border of the lip, correction of volume loss and secondary notching lip deformity.

Aims: The advantages of lip reduction surgery with autologous lip dermal fat graft are the predictability of the augmentation can bringing more natural look and most importantly making it functional as usual.

Methods: A total of 15 patients were studied and followed up at Punisa Clinic lip and plastic surgery unit between January 2015 to April 2018 in Bangkok, Thailand. The case files of these patients were analysed for sex, age and type of cleft lip deformities. All patients who underwent lip reduction surgery with using autologous free lip dermal fat graft augmentation were evaluated.

Results: All patients showed cosmetic benefit from lip reduction surgery with free lip dermal fat grafting. No complication postoperatively, including graft resorption. The satisfaction rates as determined by the patient and the surgeon at 1, 3, 6 and 12 months after the surgery was high statistically when using this technique.

Summary/Conclusion: Autologous lip dermal fat graft has number of advantages include the availability of adequate amount of tissue, lack of morbidity and the excellent adaptability of the tissue. Lip reduction surgery can improve patients lip shape, size and many type of congenital lip abnormalities. Both of this procedure is an effective and reliable method for lip augmentation to correction of undesirable lip volume loss especially secondary notching lip deformity with a low complication rate. An appropriate patients selection, careful surgical technique and a good follow-up are essential for satisfactory long-term results.
Middle ear fluid in neonates with cleft palate

M Jurovcik¹, J Borsky¹, P Dytrych¹, L Jaklova², M Cerny³, M Maciak⁴, J Skrivan¹, J Velemnska²

¹ENT, University Hospital Motol, Charles University, ²Department of Anthropology and Human Genetics, Faculty of Science, Charles University, ³Department of Neonatology, 2nd School of Medicine, Charles University, 2nd School of Medicine, Charles University, ⁴Department of Probability and Mathematical Statistics, Charles University in Prague, Prague, Czech Republic

Background: Almost all patients with cleft palate suffer from dysfunction of the Eustachian tube. The result is usually the development of secretory otitis and conductive hearing loss.

Aims: The goal was to describe the first signs of the middle ear’s pathology in newborns with a cleft defect that were operated on in the early post partum period and their follow-up.

Methods: The audiological examination (standard and high frequency tympanometry and otoacoustic emissions) of middle ear is performed one or two days prior to the actual procedure. In indicated cases an aspiration of the middle ear fluid is performed. Then the actual procedure follows – reconstructive plastic surgery of the lip. Long term monitoring is required.

Results: We operated on 300 patients with unilateral and bilateral cleft lip or cleft lip and palate at the age of 1 to 10 days after birth from June 2010 to December 2018. 168 of the cases were cleft lip and palate, 132 were cleft lip only. 73 % of patients with cleft lip and palate were diagnosed with pathology middle ear effusion. All patients with isolated cleft lip had no effusion. Patients with both cleft lip and palate are much more likely to develop the effusion in their ears and this difference is statistically significant considering a critical value of 5 %. The only significant predictor of the middle ear fluid presence is the Hz1000 tympanometry (p-value <0.0001). We performed palatoplasty in 76 patients at a distance of 6-8 months. 65% had secretion in both ears during the first surgery. After palatoplasty, further surgery was indicated in 47 patients. In 31%, the finding of the secretion was negative.

Summary/Conclusion: Occurrence of middle ear effusion is observed in the first days in cleft palate patients. All cases with isolated cleft lip we found negative. HF tympanometry is a reliable method for examination of middle ear in newborns. During the follow-up, we observed 31% of the negative finding in patients whose secretions were aspirate in the neonatal age.
Outcomes associated with malar onlays: a review of 78 implants placed by one UK consultant

A Kana¹, K French ¹, M Gormley¹, P Revington¹
¹Oral and Maxillofacial Department, University of Bristol Hospital Foundation Trust, Bristol, United Kingdom

Background: Alloplastic malar onlays have been used by surgeons to correct or enhance the midfacial skeleton for over 40 years. A literature search demonstrated respectable results using different alloplastic materials in various regions of the face, however most studies had small sample sizes and limited follow-up.

Aims: This paper aims to highlight the success rate of malar onlays in a large cohort with long-term follow-up.

Methods: Malar onlay patients were retrospectively identified using clinical notes, theatre logbooks and records held by the Stryker® representative. They were all placed by a single Oral and Maxillofacial surgeon. Success was defined as the implant not having been removed during this follow-up period.

Results: In total we found 122 malar onlays in 61 patients, placed over a 14 year period, 39 of which were cleft cases. The success rate in our cohort was 96.72%.

Summary/Conclusion: We demonstrated a high success rate for malar onlays in this cohort, suggesting they are a predictable option to correct or enhance the midface in appropriate cases, using the method described.
A challenging period after repair: investigation of postoperative follow-up rates and etiology in intensive care unit of patients with cleft lip and palate after surgical intervention

M Kara¹, M Çalış¹, Ö Canbay², F Özgür¹
¹Plastic, Reconstructive and Aesthetic Surgery, ²Anesthesiology and Reanimation, Hacettepe University Faculty of Medicine, Ankara, Turkey

Background: The cleft lip and palate (CLP) can vary according to geographical regions, but it is considered to be one in 700 to 1000 births. The frequent occurrence of CLP and the longer follow-up time make this clinical situation important. For this reason, each step should be illuminated in the follow-up and treatment process.

Aims: There are many publications in the cleft literature about repair technique, the timing of repair, speech outcomes, and maxillary development. However, there are limited publications related to intensive care follow-up after cleft surgery. At this point, the aim of our study is to reveal the etiology and follow-up rates of patients in the intensive care unit after repair of cleft lip and palate.

Methods: We retrospectively reviewed the data of the patients who underwent primary cleft repair between 1 August 2016 and 1 August 2018 at Hacettepe University Cleft Lip and Palate Team. Patient data include age, gender, cleft type, concomitant anomalies, syndromic diagnosis, the age of surgical repair, repair technique, postoperative follow-up in intensive care unit (ICU), follow-up reasons in ICU, follow-up period in ICU and conditions that prolong this period were recorded. The causes of follow-up in the ICU were grouped as concomitant anomalies, syndromic diagnoses, difficult intubation, difficult extubation and postoperative conditions.

Results: In the last two years, a total of 7587 surgical interventions were performed in our clinic, including those taken from the emergency department. 5142 of these surgeries were performed under general anesthesia, and 26% (1348 patients) of these surgeries were primary CLP surgery. 4.7% (241 patients) of all patients operated under general anesthesia were followed up in the ICU after surgery. 24% (58 patients) of patients followed up in ICU were primary CLP patients. 4.3% (58 patients) of 1348 CLP patients were followed-up in the ICU for various time periods after surgery. 64% of the patients who were followed in the ICU were isolated with cleft palate, 33% were cleft lip and palate, and 3% were isolated cleft lip. In addition, 23% of these patients (13 patients) were diagnosed as syndromic, 7% (4 patients) were preliminarily diagnosed as syndromic but the diagnosis process continued and 48% (28 patients) had concomitant anomalies. The mean follow-up period in the intensive care unit was 29.9 hours (range 1 to 336 hours), and 20.7% (12 patients) of the patients were found to have prolonged intensive care follow-up (> 24 hours). Fever, tachycardia, respiratory problems and respiratory tract infections were the most common causes of this prolongation.

Summary/Conclusion: During surgical planning, it should be kept in mind that primary CLP may be accompanied by various syndromes and anomalies, and this will affect follow-up after repair. On the other hand, it should be taken into consideration that a completely healthy baby may be in danger due to respiratory intolerance after the repair. Due to these characteristics of the patient population, cleft surgery will be safe for both the patient and the surgeon at the centers have CLP team, experienced anesthesia, and intensive care facilities.
Management challenges for medically complex children with cleft lip and palate
A Kaye¹, O Jackson², M Tracy³, S Jiang⁴
¹Plastic Surgery, Children's Mercy Kansas City, Kansas City, Missouri, ²Plastic Surgery, Children's Hospital of Philadelphia, Philadelphia, Pennsylvania, ³Plastic Surgery Research, ⁴Plastic Surgery, Children's Mercy Kansas City, Kansas City, Missouri, United States

Background: Congenital cleft conditions frequently occur in conjunction with a syndrome or constellation of other medical abnormalities. Some patients have very few medical issues while some are profoundly affected and require long-term intensive medical care. The infrequency and uniqueness of presentation of some cleft-associated syndromes and conditions provides us with a number of patients for whom highly individualized treatment plans must be devised in order to adequately address their many medical challenges. Factors related to long term prognoses for issues of feeding, speech, hearing, dental development, and cognitive function must be considered when developing cleft care plans. Safety and appropriateness for surgery and the need to prioritize other necessary treatments are factors that may preclude adherence to typical cleft treatment protocols.

Aims: This study aims to assess the challenges posed by a medically complex population and the necessary alterations in their cleft-related care.

Methods: This study was performed as a retrospective review of the demographics and cleft care protocols for patients with a history of multiple medical issues in addition to a diagnosis of cleft lip and/or palate that presented for team-based cleft care over a 15 year period.

Results: 133 patients were identified with a variety of clefting conditions (CL=7, CL/P=51, CP=53, submucous CP=22) co-presenting with 36 different named syndromes, 22 unique chromosomal abnormalities, and 30 unidentified/unconfirmed constellations of anomalies. Diagnoses were made by clinical assessment (31.6%), gene studies (25.6%), high resolution chromosomes (12.8%) or CGH microarray analysis (15.8%). 51.9% have congenital heart disease with 40.6% requiring surgical management. Other common problems include brain abnormalities (36.8%), congenital hearing loss (23.3%), and developmental delays (63.9%). 54.1% have surgical feeding tubes and 15.0% have tracheostomies. 6.7% of patients have died prematurely. 36.2% had delayed cleft-related surgeries or significantly altered cleft care protocols. Average age at primary cleft lip and cleft palate repair was 7.20 months and 23.13 months, respectively, demonstrating delays in cleft-related surgical management. 61.7% of patients have documented speech-language delays, 68.3% of which are severe or profound. Developmental delays are present in 66.2% and growth delays are seen in 35.3%.

Summary/Conclusion: Patients with CL/P and concomitant complex medical conditions present management challenges for coordinated cleft team care due to poor health condition, need for other surgery, and significant speech/developmental/growth delays.
**Background:** Midline cervical clefts are a rare anomaly with less than 200 described cases in scientific literature. Embryopathogenesis is not completely clear up to now, but most authors assume a disturbance in the fusion of the first or second branchial arches.

**Aims:** We report about a 4 month old infant, who was presented with suspected midline cervical cleft in our department.

**Methods:** Clinically the boy showed the typical symptoms of such an abnormality with a reddened medial skin defect in the anterior neck and a skin tag at the upper end. The anomaly started superiorly of the hyoid and reached up to the cranial part of the jugulum. The extension of the neck was slightly impaired. MRI confirmed the diagnosis.

**Results:** Surgery was performed still within the 4th month of life and included also the excision of a strong median muscular cord. In order to create an aesthetic, inconspicuous scar the cleft was closed in broken line technique. No intra- or postoperative problems occurred.

**Summary/Conclusion:** Even though it is a congenital anomaly, midline cervical clefts may be overlooked or wrongly diagnosed due to their rare occurrence. An early surgical correction should be favored to receive best possible aesthetic results and to avoid further impairments. Preoperatively especially the more frequent diagnosis of a thyroglossus cyst should be excluded as it requires a different surgical procedure.
Surgical technical tips for primary nose repair in unilateral clefts

P Knežević1, I Blivajs1, M Tarle1, A Ivoš2

1Department of Maxillofacial surgery, University hospital Dubrava, Av. Gojko Šušak 6, 10000 Zagreb, Croatia, 2School of Dental Medicine, University of Zagreb, Zagreb, Croatia

Background: Nasal abnormalities associated with congenital cleft lip are characteristic, varying only with the extent of the original cleft deformity. Although there have been some controversies in the past, today the generally accepted opinion is that simultaneous early correction of cleft lip and nose is necessary to obtain a satisfactory result that will maintain normal growth. A review by McComb and Coghlan confirmed that early surgery resulted in only minimal interference with nasal development. Salyer et al. published their 33-year experience with unilateral cleft lip-nose repair, further advocating primary repair of the cleft nose.

Aims: Our objective was to describe the management protocol and technique for the repair of associated nasal deformity in unilateral cleft lip as used in the Department of Maxillofacial Surgery, University Hospital Dubrava, Zagreb, Croatia for the last five years.

Methods: The incision line is a modification of rotation - advancement flap without subcolumelar and subalar incision on the cleft side. The technique consists of undermining the skin above the lower lateral cartilage with scissors from the medial approach, elevation of septal mucosa in the base and suturing it later with the nasal vestibulum skin laterally. The incision line in the cleft nose could be extended through the piriform aperture even higher up to the level of lower turbinate. The resorbable stitch between the muscle on the cleft side and the nose on the non-cleft side as the anchorage is very important. Applying this technique the results regarding cleft nasal deformity have been pleasing. Even in the incomplete unilateral clefts, it is beneficial to detach the cleft part of the nose from the piriform aperture. In that way the nose can be better reshaped in three-dimensional view. Any small excess of skin in the floor of the nose can be excised and the nasal vestibulum sutured primarily. The nasal postoperative stents are useful.

Results: Since January of 2014 this technique has been used in all unilateral cleft lip repairs. The technique can be applied to all degrees of unilateral cleft lip.

Summary/Conclusion: In our Department, we have used rotational–advancement flap with minor modifications for cleft lip repair over 35 years. Although this method yields good postoperative aesthetic result regarding lip, results regarding nasal deformity were unsatisfying. Therefore, in the last five years we have been employing certain new tips in the surgical technique for the associated nasal deformity. Whilst muscle repositioning is one of the most important aspects in managing the cleft lip nose, we believe that in addition suturing the skin of the floor of the nose as far posteriorly as possible is particularly important. This technique can be applied both for complete and incomplete clefts to enable better contouring of the nose.

Applying this technique in the last five years we have achieved pleasing aesthetic results in the appearance of the nose. There is no doubt that primary nose repair should be done simultaneously with the lip repair, although long term follow-up is necessary for definitive results.
Corrected cephalometric analysis and hybrid osteogenesis distraction system to control distance and vector with le fort iii osteotomy for syndromic craniosynostosis

S Kobayashi¹, T Fukawa²
¹Department of plastic and reconstructive surgery, Kanagawa Children’s Medical Center, ²Fukawa Orthodontic Office, Yokohama, Kanagawa, Japan

Background: Treatment of syndromic craniosynostosis is critically dependent on the successful advancement of the midface. However, the distance and direction of overcorrection for the midface are difficult to determine accurately in younger patients.

Aims: The purpose of this study was to confirm the utility of corrected cephalometric analysis to facilitate the planning of distraction osteogenesis with Le Fort III osteotomy for syndromic craniosynostosis, and to evaluate the stability and/or resultant change in facial features after at least 1 year of follow-up (range, 1-9 years).

Methods: This prospective study involved 8 cases of Crouzon syndrome and 3 cases of Pfeiffer syndrome (types II and III) (age range, 4 years 6 months to 13 years 2 months) treated with primary Le Fort III maxillary distraction using corrected cephalogram analysis and a hybrid distraction system. Corrected cephalogram analysis involves superimposing a patient's cephalogram onto that of a normal Japanese adult. Both cephalograms are then superimposed using the articulare for guidance to determine the distance and vector of distraction osteogenesis. The hybrid distraction system is composed of both a conventional external distraction device and a newly developed adjustable-angle internal distraction device. Postoperative control of the distraction vector is performed by pulling the midfaces using the external device, while control of distraction distance is done by pushing the midfaces with the internal device.

Results: Midfaces were brought close to the planned position from the corrected cephalometric analysis in Crouzon syndrome. Midfaces were advanced by 19.8±4.0 mm at Or and 29.4±4.1 mm at point A, but accurate alignment to the planned vector was difficult. Meanwhile, midfaces were not brought close to the planned position from the corrected cephalometric analysis in Pfeiffer syndrome. Midfaces were advanced by 32.0±2.1 mm at Or and 34.7±5.0 mm at point A, but accurate alignment to the planned vector was more difficult for Pfeiffer than for Crouzon. Facial features changed little on follow-up, 1-9 years after removal of the distraction device.

Summary/Conclusion: Using corrected cephalometric analysis, the distance and vector of distraction osteogenesis with Le Fort III osteotomy could be determined in patients with syndromic craniosynostosis. This distraction system offers the possibility of bringing facial bones to the planned position using controlling devices. However, final long-term growth remains unclear.
Background: Cherubism is an rare hereditary benign fibroosseous disorder characterized by bilateral enlargement of the jaws and a tendency toward spontaneous remission in the third decade of life. Many authors suggest a wait and see policy and seek to move away from surgical intervention in affected individuals. However, severe jaw malformation can have a heavy impact on physical functions, and social and psychological development in children and adolescents.

Aims: Search for an efficacious and safe treatment method to improve psycho-social rehabilitation and prevent functional dysfunctions.

Methods: Two children 5 and 9 years old with cherubism were treated in the Department of Craniomaxillofacial surgery of the Russian Children's Clinical Hospital, Moscow from 2016 to 2018. Due to progressive growth of the upper and lower jaws remained one child took bisphosphonate therapy, and in another case, due to the unilateral debut of the disease, child was treated with local curettage. Diagnosis was histologically verified in all cases. After the approval of the Ethical Committee, patients received a course of denosumab therapy.

Results: Bisphosphonate therapy has not significant stopped the progressive growth of the upper and lower jaws. Local curettage with implantation hemostatic sponge and triamcinolone has stopped continuing growth of tumor mass, but we observed new tumor foci's in jaws appearance. Denosumab treatment showed a significant response which resulted in decrease tumor mass and disappearance of giant cells. CT before and after the therapy showed an increase in bone density in 5-10 times. The size of tumor nodes reduced, allowing contour resection performed.

Summary/Conclusion: Inoperable forms of cherubism require a comprehensive approach that includes denosumab therapy with subsequent contour resection of excess bone tissue to facilitate medical and social rehabilitation of such patients.
Diffuse sclerosing osteomyelitis of the mandible
A Kugushev¹, A Lopatin², S Yasonov², D Rogozhin³
¹Craniomaxillofacial surgery, Russian Children’s Clinical Hospital, ²Craniomaxillofacial surgery, ³Pathology, Russian Children’s Clinical Hospital, Moscow, Russian Federation

Background: Diffuse sclerosing osteomyelitis (DSO) is a rare jaw disease difficult to treat. None of the proposed various protocols for medical and surgical treatment of DSO guarantees positive outcomes. Recently, many authors have used bisphosphonates (ibandronate, alendronate, pamidronate) to treat DSO during periods of exacerbation. Denosumab, a human monoclonal antibody to the receptor activator of the kappaB ligand of the nuclear factor (RANKL) has shown similar antiresorptive effect in patients with DSO.

Aims: Search for safe and efficacious treatment of patients with diffuse sclerosing osteomyelitis

Methods: 10 children aged 10-16 with DSO were treated in the Department of Craniomaxillofacial surgery of the Russian Children’s Clinical Hospital, Moscow from 2013 to 2018. Treatment included antibacterial monotherapy with or without decortisation and subsequent treatment with antiresorptive drugs (alendronate and denosumab). All patients were diagnosed DSO based on clinical X-ray picture and histological examination

Results: The use of denosumab has allowed reaching long-lasting remission, i.e. disappearance of chronic pain syndrome, reducing inflammatory activity and increasing bone density.

Summary/Conclusion: Antiresorptive drugs should be considered as the main treatment approach for patients with DSO. However, additional studies are needed to determine the mechanisms of action and the required dose of the drug, as well as the intervals of use.
How can we ensure that patients and caregivers are safely discharged from hospital after surgery?
N Lindberg¹, L Bjørnson¹, A Lereggen¹, K Ladegård¹
¹Dept of Plastic and reconstructive surgery, Oslo university hospital, Oslo, Norway

**Background:** Patients born with cleft lip and palate (CLP) usually undergo multiple surgical procedures. A well-planned discharge from hospital may reduce risk, injury and unnecessary readmissions and contribute to positive patient experiences. However, low scores are revealed in a user survey on patients’ experiences of quality of discharge from surgical and medical departments in a Norwegian hospital. The goal for the National Patients safety program in Norway is to avoid injuries. “Safe discharge” - with the patient as an equal party is one of several action areas in the National Patients safety program. We want to improve our routine regarding safe discharge from hospital for patients born with CLP and other diagnosis that are admitted to our department.

**Aims:** To work out and implement a routine that ensures that patients are safely discharged from hospital. The routine must fit into daily work in the department and be in line with “Safe discharge” and other guidelines and strategies we are required to follow.

**Methods:** The action area “Safe discharge” has been studied and parts that address our practice are selected. Written patient information, strategies for communication and a Check-list are developed. A literature review regarding communication and patient information has been conducted. Collaboration with surgeons and coordinators as well as discussions within the nurse staff are performed. Patients have participated in the preparation of written patient information.

**Results:** A routine for safe discharge, embodied in a flow chart, is developed and consists of: Early discharge planning, cooperation with partners in the healthcare system, a structured conversation at discharge and a check list. The routine are implemented in the department.

**Summary/Conclusion:** The routine may facilitate safe discharge from hospital for patients admitted for surgery regarding cleft lip and palate and other diagnoses. We will evaluate the routine and make necessary adjustments.
Otologic and audiologic outcomes in robin sequence patients versus isolated cleft palate patients

B Logies¹, S Coenraad², C Breugem¹
¹Plastic and Reconstructive Surgery, ²Department of Otorhinolaryngology, UMC, Utrecht, Netherlands

Background: Cleft palate (CP) patients have an increased risk of chronic middle ear disease and hearing loss due to the anatomic and functional defect of soft palate muscles that effects Eustachian tube function. Robin sequence (RS) patients also suffer from glossoptosis and pathologic swallowing with frequent aspirations of food. These factors may result in an additional disturbance in the function of the Eustachian tube and ventilation of the middle ear. This abstract presents the preliminary results of our study.

Aims: To review otologic and audiologic outcomes after cleft palate repair in Robin sequence patients and to compare to isolated cleft palate patients (ICP).

Methods: All consecutive RS- or ICP-patients that underwent CP-repair at the Wilhelmina Children’s Hospital were retrospectively reviewed (2003-2016). RS was defined as micrognathia, glossoptosis and upper airway obstruction, and in the ICP-patients there was a clear documentation of no underlying syndromes. All patients were followed by an ENT-surgeon until the minimum age of 2 years and at least one-year post CP-repair, patients with submucous CP were excluded. The cohort was divided into 3 different groups based on age in follow-up: 0-2 years, 4-6 years, and ≥ 7 years. Variables included sex, age at CP repair, CP characteristics, type of CP repair, and underlying syndromes in RS. Indication for tube placements included clinical signs otitis media with effusion (OME) during examination combined with failure of otoacoustic emissions testing. Outcomes were primary tube placements within the age of two years, total tube placements in follow-up, type of hearing loss (conductive, sensorineural or mixed), in patients aged 4-6 years the worst audiogram pre tube placement was used to evaluate hearing thresholds, and in patients aged ≥ 7 years the most recent audiogram was used. In addition, otologic outcomes of all CP-patients were noted.

Results: A total of 118 CP-patients (57 RS vs. 61 ICP) underwent CP-repair (median age RS: 12.4 months vs. ICP: 9.1 months, p = 0.001) who met the inclusion criteria. The total CP-group consisted of 51 males and 67 females, and RS associated syndromes were identified in 56%. All CP-patients had Von Langenbeck repair with intravelar veloplasty, except for 3 RS-patients (1 Furlow and 2 Malek repair). Mean width of CP was 10.1mm (SD2.7) in RS vs. 8.7mm (SD2.6) in ICP (p = 0.036), and CP involvement into the hard palate was 77% in RS vs. 51% in ICP (p = 0.021). Median follow-up post CP-repair was 5.2 years (range:1.0-14.9 years) and median age at last ENT follow-up was 6.0 years (range:1.9-15.7 years). Primary tube placement within the age of two years was performed in 77 ears (71%) in the RS-group vs. 64 ears (54%) in the ICP-group (p = 0.014). Mean number of tube placements in total follow-up was 2.5 (SD1.7) in the RS-group vs. 1.7 (SD 1.6) in the ICP-group.

Summary/Conclusion: Our data demonstrates that within the age of 2 years RS-patients needed more tube placements for treatment of OME. Further conclusions/results regarding otologic and audiologic outcomes at a later age will be presented in June.
P-059
Ten years of p.I.S.A. Technique: periosteoplasty improves symmetry and aesthetic. An analysis of 642 primary surgeries
A Giacomina¹, G Gatti², B Kuppers³, B De Lorenzo³, L Rosato⁴
¹Hospital of Pisa - Dept. of Plastic Surgery, ²Plastic Surgery, ³Anesthesia, Hospital of Pisa, Pisa, ⁴Plastic Surgery, Hospital of Piacenza, Piacenza, Italy

Background: Surgical treatment of cleft lip and palate is a great struggle between function, growth and aesthetic. A well-defined protocol, for both unilateral and bilateral cases, is followed in Pisa and everything starts in early ’60. A dedicated ward with 10 beds is fully working for cleft babies since 2016.

Aims: The keystone of surgical treatment of the lip is the periostioplasty, a special modification of Skoog’s one by Massei, that helps bone formation and aesthetics improvements. Other two main points are surgical timing and nose treatment. The cheiloplasty is a Tennison modification, similar to Fisher’s technique, for unilateral cases while Mulliken’s technique is used for bilateral ones.

Methods: In unilateral and bilateral cases, periosteoplasty procedure is always performed during the cheiloplasty at 2,5-3 months of age; a previous lip adhesion is performed in the severe maxillary disorders at 40 days of age; the palatal surgery is performed at 6 months of age. Orthopaedic pre-surgical devices are not commonly used in our practice. The multidisciplinary follow-up takes place in our office since pregnancy.

Results: From 2009 to 2018 a number of 642 primary cheiloplasties were performed. A number of 387 complete CLP were treated, 128 CL with maxillary cleft and 127 CL with minimal or no affection of the bone. Left clefts were the most represented (above 60%), while right and bilateral cases were on approximately 20% each. Periosteoplasty procedure, in our hands, has decreased the need of bone graft in almost 70% of cases. The III class skeletal malocclusion is comparable with major studies (15%>20%)

Summary/Conclusion: We do believe that an earlier surgical approach represent the best way to achieve better result and periosteoplasty procedure is our milestone to decrease the need of bone graft.
P-060

Use of the turbinate flap in primary cleft nose correction in wide uclp

F Maggiulli¹, A Sadri¹, B Rafique¹, P Morris¹
¹Plastic Surgery, Great Ormond Street Hospital, London, United Kingdom

Background: The nasal deformity in UCLP is a challenge because the maxillary hypoplasia on the cleft side represents an insufficient platform in the antero-posterior (AP) plane for the ala to rest upon, above all in wide cleft deformity where the AP gap is >10mm. Nasal dissection in the pediatric population is associated with scar, growth restriction and nostril stenosis.

Aims: The aim if this paper is to describe our technique of using the inferior turbinate flap at the time of the nasal correction to achieve a symmetrical result without any further nasal dissection.

Methods: In 5 years, from 2014 to 2018, 100 (70 males and 30 female) consecutive complete UCLP patients were treated at 3 months by the senior author. In 70 cases (45 males and 25 females) the AP gap was >10mm and needed the turbinate flap. No pre-surgical orthopedics has been used. The flap, raised before vomerine flap under the microscope, is inserted in the opening of the nasal floor as a continuum of the vomerine flap without any further nasal dissection.

Results: The early outcomes consisted in:
- on table symmetry in all 70 cases
- no soft tissue secondary healing
- no peri-alveolar fistula
- reduced early nasal distortion and stenosis
- no Mc Comb dissection
- no nasal regurgitation (100% vomerine flap integrity)
- early speech improvement.

The late outcomes will be assessed at 5 and 10 years of age to look at the impact on the alveolar bone graft and at the nasal symmetry as we think that the full superior soft tissue availability maintains vascularity and position of the bone graft.

Summary/Conclusion: The inferior turbinate flap is a reliable vascularized flap to achieve a symmetrical result without any further nasal dissection, although long-term outcomes are not available yet.
The functional palate suspension: how to expand the versatility of the double opposing buccal flap palatal lengthening procedure

Robert Mann

1Pediatric Plastic Surgery, Helen DeVos Childrens Hospital, Grand Rapids, Michigan, United States

Background: The Double Opposing Buccal Flap Procedure for Palate Lengthening (DOBFP) has become a proven alternative to the non-physiologic pharyngeal flap (PF) and sphincteroplasty (SP). The traditional PF and SP tether the velum rendering it essentially immobile, and rely on the lateral and posterior wall muscles to achieve nasal pharyngeal closure. Both the PF and SP dramatically reduce the standing nasal pharyngeal aperture and contribute to lifelong risks of obstructive sleep apnea. The DOBFP replaces missing embryonic palate tissue and lengthens the palate and allows the natural velar movement to be the closing mechanism for the nasal pharyngeal space. The DOBFP measurably lengthens the anterior portion of the velum, but as the palate is lengthened in patients with wider velar gaps, the palate invariably falls more inferiorly with every millimeter gained posteriorly. As this progression continues the natural point of velar contact on the posterior pharyngeal wall also falls progressively downward to a point where the closure may be rendered incompetent. This is one reason why some patients with wide nasal pharyngeal gaps fail to achieve effective closure with the DOBFP alone. The purpose of the Functional Palate Suspension (FPS) is to expand the gap range of the DOBFP to treat wider gaps and shorter palates. The Functional Palate Suspension is a new way to use PF tissue that does not tether or immobilize the velar muscles. By connecting a narrow superiorly based unlined flap of superior constrictor muscle and mucosa from the high posterior nasopharyngeal wall to the buccal flap space in the anterior velum, the whole soft palate can be elevated and the velar muscles remain undamaged.

Aims: To demonstrate how the effectiveness of the DOBFP can be expanded by adding the FPS for those patients who fail to achieve normal speech with the DOBFP alone.

Methods: A qualitative review of 13 patients born with cleft palate whose secondary Velopharyngeal Dysfunction (VPD) was initially treated with a DOBFP operation and subsequently with a FPS.

Results: Group 1: All patients achieved normal speech without developing any post-operative signs of obstructive sleep apnea.

Summary/Conclusion: The Functional Palate Suspension can effectively expand the success of the Double Opposing Buccal Flap as a treatment for the vast majority of secondary VPD Cleft palate cases. In our practice by using the DOBFP and FPS (as needed) as the first line of treatment for VPD has essentially eliminated the need for classic PF and SP operations. The DOBFP +/- FPS are more physiologic reconstructions which do not incorporate the dramatic obstructive nasopharyngeal aperture reduction of the classic PF and SP.
A new primary cleft palate repair based on the buccal flap approach: the no touch cleft palate repair: the palatoplasty for patients with small velar muscles, a 31 year review

R Mann

Pediatric Plastic Surgery, Helen DeVos Childrens Hospital, Grand Rapids, Michigan, United States

Background: Every cleft lip and palate patient is different anatomically. This vast diversity makes it difficult to achieve consistent results with traditional palate repairs. Those children born with very small velar muscle, and deficiency of the tensor villi palatini aponeurosis, are at a high risk for significant velar muscle scarring when treated with the commonly used standard or radical Intervelar velar plasty or Z plasty. All too frequently these children progress to Velopharyngeal Dysfunction and are frequently treated with non-physiologic pharyngeal flaps or sphincteroplastys. Children born with Pierre robin sequence frequently have very anteriorly displaced muscles also placing them in a high risk group. While traditional repairs pull the available tissue together and attempt to rearrange the velum, the No Touch repair developed in 1987 positions the velar tissue as a unit to its natural position, reducing scarring around the muscles and replaces the embryologic missing tissue with Buccinator Myomucosal flaps (Buccal Flaps) to complete a more anatomic reconstruction.

Aims: To give patients born with small velar muscles improved speech outcomes with low complications.

Methods: 1. A 31 year quality assessment review of the results of 20 consecutive patients born with small velar muscles treated with the No Touch technique. A qualitative review of results was done with specific analysis of the speech results and complications. 2. The No Touch procedure will be demonstrated.

Results: This specific anatomic subgroup of patients with small velar muscles normally presents the reconstructive surgeons with some of the most difficult challenges. Treating this patient group with the No Touch repair achieved excellent speech results, equivalent to the results obtained in the Double Z Plasty +/- Buccal Flap Repair series. Post-surgical complication rates are even lower than those reported in the Double Z Plasty +/- Buccal Flap Repair series, demonstrated over a 31 year period.

Summary/Conclusion: The No Touch Palate Repair achieves successful velar function with less radical velar muscle dissection than traditional repairs. All traditional repairs dissect either partially or completely around the velar muscles. Children with abundant muscles can tolerate this scarring, but for children born with small muscles, this degree of scarring can lead to reduced velar movement and permanent vocal compromise. The No Touch Repair moves the velum more as a unit, there is very little mucosal dissection which reduces scarring around the delicate muscles. The second advantage of the No Touch repair incorporates the most fundamental principle of the Buccal Flap Approach which is the replacement of the missing embryonic tissue. The tissue replacement effectively reconstructs the missing elements of the Tensor Villi Palatini Aponeurosis as well as the deficiency of the mucosal tissue and allows the velar muscle to easily assume its natural velar position.
Background: Treatments for the bilateral cleft lip and nasal deformity used to normalize the face in infancy and childhood can often leave disfiguring scarring that can compromise the opportunity to achieve the best result when the child reaches skeletal maturity. The Bilateral cleft nasal deformity with its displaced nasal cartilages, drooping dorsum, short columella and broad nasal tip creates a visual deformity consistently identified with the patient born with a facial cleft. Children in pre-teen and teenage age years are faced with ever increasing social pressures. The Step Rhinoplasty allows the surgeon to have an option for a treatment where both the nose and the lip can be brought into a more normal balance in the pre-adolescent age group without having to use cartilage grafting from the septum, leaving the septum cartilage available to be used at the definitive rhinoplasty. The Step Rhinoplasty achieves markedly improved pre-adolescent nasal-lip balance and allows for the young teen to mix well with their peers, without compromising the result achievable at the definitive rhinoplasty at full facial growth.

Aims: 1. To illustrate an alternative to some of the common treatments that often leave our adult patients with noses that still do not look normal. 2. To demonstrate how to perform the step Rhinoplasty.

Methods: 1. The surgical technique will be described in detail. 2. A qualitative review of the results of 7 patients with 1-5 year follow up will be demonstrated.

Results: Early results were most effective in the Asian population. The more prominent Caucasian nose required some adjustments to achieve similar excellent results. The treatments were well tolerated without complication.

Summary/Conclusion: The bilateral cleft nasal deformity is one of the most difficult to normalize. The Step Rhinoplasty allows for improved self-esteem at the critical pre-adolescent age but does not compromise the definitive rhinoplasty performed at skeletal maturity. All too often the patients we treat as infants are left with unsatisfactory results when they reach adulthood. They often spend adulthood quietly unhappy with their appearance, and most often will not seek more treatments. Many times the treatments that can be offered to the adult patient are less effective due to the scarring from the multitude of childhood treatments. The Step Rhinoplasty addresses the psychologic need to maintain a normal face during the developing years yet respects the desire of the adult patient to have the very best long term outcome.
Impact on parents with the birth of a child with cleft lip and/or palate

R. Aleman¹, L. Aparicio², S. Guzman³, A. Romo³, J. Aparicio⁴


Background: Reviewing the current scientific literature, the authors found there are very few studies related to the impact on parents related to the birth of a child with cleft lip and/or palate, most of the studies are related to the impact on the child related to his or her facial difference. Parents are and will be of paramount importance in successful holistic management of the cleft lip and/or palate patient, most of them experience feelings of guilt, extreme sadness, and denial. According to the literature there are some differences among different cultures in the way parents face the challenges of taking care of the new-born, Japanese and Chinese moms describe more intense reactions to this event than moms in the USA, in Nigeria many believe that evil spirits are the main cause of CLP, and moms are usually more protective, Thai and Colombian mothers are more positive towards their children born with CLP probably related to their religion. All these led the authors to our research question: Which is the impact on parents with the birth of a child with CLP in El Salvador, Central America?

Aims: The purpose of this study was to explore the impact on parents with the birth of a child with cleft lip and/or palate.

Methods: A descriptive qualitative study was conducted from April to August 2018 at a cleft lip and palate center in San Salvador, El Salvador, in Central America. A convenience sampling was used, the sample consisted of 11 parents which were interviewed. A thematic content analysis was used to code the interviews. The study was approved by the ethics committee of the Evangelical University of El Salvador. Informed consent was obtained from each participant.

Results: The following 4 major themes emerged: (a) information given by the healthcare professionals at the moment of birth, (b) emotions related to the birth of a child with cleft lip and/or palate, (c) challenges and responsibility, (d) sources of support.

Summary/Conclusion: Parents felt that the information received from healthcare professionals at the moment of birth was poor. The emotions related were: guilt, depression, denial, sadness, suicide thoughts. Among the sources of support, the following were found: (a) support from the couple, family and friends, which brings family into a union, fighting together with the adversity, (b) religion, faith in a supreme power that will help them to solve their problems.
The incidence of velopharyngeal insufficiency and oronasal fistula after primary palatal surgery with Sommerlad intravelar veloplasty: a retrospective study in Isfahan cleft care team

D Mapar1, F Derakhshandeh1, F Khanlar1, S Sadeghi1, H Abdali1, M Memarzadeh1, H Davari1

1Isfahan University of medical sciences. Isfahan. Iran, Craniofacial & Cleft Research Center, Isfahan, Iran, Islamic Republic Of

Background: Primary palatal surgery is the first physical intervention to repair the cleft palate and has a determining role in the quality of speech outcomes. The main objective of this surgery is to create a healthy structure for normal speech production, direct oral airflow, and a balance in resonance with minimal effects on the facial growth.

Aims: Main purposes of this paper are studying the incidence of different types of hypernasality severity and oronasal fistula development in people with cleft palate after primary surgery with Sommerlad intravelar veloplasty (SIVV) technique that is performed by Isfahan cleft lip and palate team after 2011. Furthermore the association between sex, type of cleft palate and age at primary surgery with incidence of them is determined.

Methods: The present study is retrospective study that investigate the speech evaluation results and oral examination videos of 40 patients who have been operated with SIVV. Two speech therapists listened to the recorded sounds and rated them according to CAPS-A (Kappa = 82.4). Deciding whether or not to have a fistula was based on the videos.

Results: Severe and moderate hypernasality was observed in 42.5% of patients. Normal resonance and mild hypernasality was observed in 37.5% and 20% of patients, respectively. The frequency of fistulas was 7.5%. There was a significant association between hypernasality with type of cleft (p < 0.05). The frequency of hypernasality increased significantly with an increase in age at primary surgery (p = 0.04). Although the frequency of hypernasality was higher in boys versus girls (75% vs. 54.2%), this difference was not statistically significant. 12.5% of boys (2 out of 16) and 4.1% of the girls (1 out of 24) had oronasal fistula but the differences were not significant (p = 0.38). Incidence of oronasal fistula was more common in bilateral cleft lip and palate compared to other types of cleft, but was not statistically significant (p = 0.41). There was no significant difference between age groups at primary surgery in this regard (p = 0.36).

Summary/Conclusion: Significant progress has been made in the outcomes of the primary palate surgeries with the SIVV technique compared to the previous study that used other surgical procedures in the Isfahan cleft lip and palate team.
Furlow palate repair – a new day case pathway introduced in the regional cleft unit

S Martin¹, T O’neill², J Quinn², C Hill³
¹Cleft unit, Royal Belfast Hospital for Sick Children, ²Cleft unit, ³Cleft uni, RBHSC, Belfast, United Kingdom

Background: Furlow palate repair is a reliable method of palate repair and can be used for both primary palate repair and as a secondary procedure.

Aims: Our aim was to determine the feasibility of performing Furlow palate repairs as a day case procedure in the Regional cleft centre. Following the initial feasibility study, our aim was to optimise the day case pathway for patients undergoing Furlow palate repair based on both parent and staff feedback.

Methods: A retrospective review of the Nurse Controlled Analgesia (NCA) volumes and durations for 50 patients who had undergone Furlow Palatoplasty for submucous clefts and clefts of the soft palate were retrieved and examined. Co-efficient analysis was performed. The maximum duration and volume of NCA for both syndromic and non-syndromic patients was retrieved. The tonsillectomy pain pathway was adapted using the information retrieved from the NCA administration audit. The regional Cleft unit used this adapted pain pathway to initiate day case Furlow palate repairs in non-syndromic patients. A review of the first year of day–case Furlow palate repairs is presented.

Results: The maximum duration of NCA administration in non-syndromic patients was 24 hours. Syndromic patients tended towards longer NCA durations and for this reason syndromic patients are not treated according to the day case protocol. We have successfully been performing Furlow palate repairs in non-syndromic patients for one year. Parental feedback has all been positive and has led to the introduction of more informative booklets on post-operative care which are now given home to parents on discharge.

Summary/Conclusion: Furlow palatoplasty is a safe and reliable method to repair sub-mucous clefts and clefts of the soft palate. It produces excellent speech outcomes and has been shown to be appropriate to undertake as a day case procedure in certain circumstances and patient sub-groups. This not only represents an economical saving to the hospital in terms of bed days saved, but is also an acceptable and preferable approach from a parent perspective. Early hurdles have been overcome to ensure this new day case pathway can continue in the regional cleft unit.
Background: Secondary surgery for patients with cleft lip and palate (CLP) is common. The timing and nature of surgery required will depend on several factors. Revision surgery may be needed to improve cosmesis and/or function of each area; including the lip, palate or the nose.

Aims: To determine the type and amount of secondary surgery required up to 21 years in both unilateral and bilateral cleft lip and palate patients.

Methods: A retrospective review of the regional cleft lip and palate database was used. To incorporate all secondary surgery required up to 21-years, patients born between 1988-1995 were included. Cleft palate only, isolated cleft lip and cleft lip and alveolus were excluded. Information on both primary and secondary surgery performed was reviewed; including lip revision, fistula repair, speech surgery and rhinoplasty.

Results: Over the 8 year period 28 patients were born with CLP; 22 unilateral and 6 bilateral.

Unilateral – 68% required secondary surgery. The commonest area requiring secondary surgery was the palate (41% 9/22), followed by the nose (36% 8/22) and the lip (9% 2/22). Two patients required revision surgery for all 3 areas. Secondary palate surgery included; fistula closure (33%) and speech in (66%). Average age at secondary palate surgery was 8-years and 17-years for nasal surgery.

Bilateral – 50% required revision surgery. Secondary palate surgery and rhinoplasty were the commonest. All secondary palate surgery was for speech.

Summary/Conclusion: Unsurprisingly, the rate of secondary surgery is high. In this review speech surgery was the main burden. We continually strive to improve outcomes from primary surgery but some factors are beyond our control as surgeons, including the impact of facial growth on changing aesthetics as the child becomes an adult.
Posterior pharyngeal wall augmentation with a superiorly based midline myomucosal flap: a novel technique in the surgical management of both cleft and non-cleft velopharyngeal dysfunction.

P McAllister1, T O'Neill1, M Devlin1, D McAuley2, C Russell1, L Campbell3, L Crampin3
1Cleft Lip and Palate, 2Oral and Maxillofacial Surgery, 3Speech and Language Therapy, NHS Greater Glasgow and Clyde, Glasgow, United Kingdom

Background: Velopharyngeal dysfunction (VPD), an inability for co-ordinated closure of the velopharyngeal sphincter, can result from either anatomical or physiological aberrations in the soft palate, lateral or posterior pharyngeal walls, or be due to a combination of such issues. The result of such issues includes alterations in speech resonance, compensatory articulation errors and nasal regurgitation of food with consequent social embarrassment. The surgical management of VPD across the UK and Ireland has been shown to be varied.

Aims: The aim of this technical note is to describe a simple technique of posterior wall augmentation using a superiorly based midline pharyngeal flap that is technically straightforward to execute, has minimal complications and in our experience, delivers consistently good outcomes.

Methods: A superiorly based myomucosal flap is designed originating just caudal to the adenoidal bed. The lateral dimensions of the flap are conceived on a case by case basis and are as generally wide as possible but still permitting primary closure of the flap donor site. A chevron is incorporated into the most distal aspect of the flap both to facilitate closure without a dog-ear caudally and also to allow inset of the flap. Hydrodissection with 1% xylocaine with 1:200 000 adrenaline is performed. Seven minutes are allowed to elapse in order to maximise the effect of the adrenaline. The flap is raised just superficial to the prevertebral fascia incorporating both the inferior and middle constrictors. Meticulous haemostasis is performed. The donor site is closed with a 4/0 Vicryl™ suture leaving a small gap inferiorly to allow any collection to drain easily under low pressure. A horizontal mattress suture is placed in the middle of the flap to create the point of flexion. Care is taken not to over tighten this suture and strangulate blood flow through the flap (Fig 2). The edges of the flap are opposed to reduce the amount of secondary intention healing as much as possible. The chevron at the distal end of the flap is inset. A Sommerlad suction test is performed, and the result recorded in the operation note.

Results: The on-table Sommerlad Tests results are satisfactory. Considerable more pharyngeal augmentation is felt to be achieved than that delivered by a Hynes pharyngoplasty. The longer the flap raised, the greater the AP projection of the augmentation. Furthermore insert of the flap is technically easier that that for Hynes or Orticochoea pharyngoplasty. We have not experienced any difficulty with peri- or post-operative bleeding and recovery is similar to that of a Hynes pharyngoplasty or tonsillectomy. Anecdotally, the early speech outcomes are favourable however we plan to objectively assess this in the future when a suitable volume of patients and post-operative speech assessments have been undertaken.

Summary/Conclusion: The superiorly based pharyngeal flap is a subjectively reliable, reproducible and safe pharyngeal augmentation technique that appears to improve VPD speech characteristics in cleft and non-cleft patients. We are currently accruing an adequate number of patients to enable objective evaluation of this under-reported technique.
Unilateral soft palate palsy secondary to blunt neck trauma: a case report

P Mcallister¹, T O'Neil¹, G Baniulyte², M Devlin¹, C Russell¹
¹Cleft Lip and Palate, ²Oral and Maxillofacial Surgery, NHS Greater Glasgow and Clyde, Glasgow, United Kingdom

Background: Unilateral acquired soft palate paralysis is a rare but recognised condition. Idiopathic cases, most commonly presenting in the paediatric population have been documented, with infective or vascular theories to explain their occurrence. Penetrating neck trauma has previously been identified as a cause of unilateral palatal paresis.

Aims: The aim of this report is to present a case of unilateral soft palate paralysis with associated velopharyngeal insufficiency (VPI) secondary to minor blunt neck trauma. No similar presentations have been reported in the medical literature to date.

Methods: A 16-year old male was referred to the palatal function clinic with speech characteristics of velopharyngeal insufficiency (VPI) following an episode of trauma to the neck after a collision with an opponent playing soccer. Immediately following the clash he was aware of changes to his voice strength and quality. The patient was seen locally by a Speech and Language Therapist and referred to the palatal function clinic provided by the National Cleft Service.

Results: On examination there was speech hypernasality, asymmetric palatal elevation and uvular deviation to the non-affected side. There was no evidence of muscle wasting, no abnormalities in swallowing or nasal regurgitation of food or fluids and no other focal neurology was identified. The gag reflex was intact. Nasoendoscopy identified normal vocal cord position and movement. At last review, four months following injury, speech had improved in the absence of identifiable palatal recovery presumably due to compensation from the tongue and unaffected contralateral side.

Summary/Conclusion: Levator veli palatini, innervated by the pharyngeal branch of the Vagus nerve (X), is the principal elevator of the soft palate during function. A traumatically induced focal neuropaxia or axonotmesis of the pharyngeal branch of the vagus nerve was suspected. A sphincter pharyngoplasty for unilateral VPI following resection of skull base tumours has been described. A similar dynamic corrective procedure like the modified sphincteroplasty detailed could be considered in our patient if problematic VPI persists at a time beyond which spontaneous nerve recovery is deemed likely. Peripheral nerve regeneration is estimated at 1mm/day. It has been suggested a period of observation for 6–9 months is a reasonable timeframe to monitor for spontaneous nerve recovery in high vagal injuries. At present the patient has no concerns regarding his speech and with the possibility of complete spontaneous nerve recovery, no surgical intervention is currently planned. This case demonstrates the previously unreported case of an isolated high vagal injury affecting only the pharyngeal branch of the vagus nerve with associated VPI following blunt neck trauma. It highlights infrequent sequelae of neck injuries and the role of the national cleft service in rare cases of acquired non-cleft velopharyngeal insufficiency.
Background: The aim of primary palatoplasty is to achieve speech within the normal range without disrupting breathing/future growth. Symptomatic fistulae are well-recognised complications of palatoplasty and may require additional surgical intervention increasing burden of care.

Aims: We aimed to better understand fistula experience in our unit and compare to published standards.

Methods: Post-operative fistulas were prospectively and independently recorded by cleft nurse specialists as part of routine six week post-operative reviews. Cleft type and intra-operative hard–soft palate junction (HSJ) width were prospectively recorded by operating surgeons. Data was collated and analysed using Microsoft Excel.

Results: Between Jan 1\textsuperscript{st} 2014 and Dec 31\textsuperscript{st} 2018 \(X\) number of primary palatoplasties were performed. The overall fistula rate was 8\%. (0\% SMCP, ICP 7\%, UCLP 8\%, BCLP 19\%). Fistulae clustered in clefts with a mid-range HSJ width of 12 to 16mm. Numerically fistula rates remains similar over time despite increased unit activity (Doubling of primary surgery 2017-18). There was no significant difference in fistulae rates between surgeons (\(P > 0.05\)).

Summary/Conclusion: Overall fistulae rate compared favourably with published data. These results indicate that TIG training has the potential to minimise effects of new consultant learning curve in cleft surgery, (results from a newly appointed TIG trained surgeon comparing well with that of an established TIG trained surgeon). Data suggest surgeons should be aware of the risk of fistulae in the mid-range palatal defect with fistulae more common in HSJ widths of 12-6mm.
Treatment of maxillary deficiency with increased lip angle and close nasolabial angle in a CLP patient. Case report.

A Medrano Gutierrez1, R Yassutaka Faria Yaedú1, M Barbosa Mello1
1Postgraduate Program, Hospital for Rehabilitation of Craniofacial Anomalies, Bauru, São Paulo, Brazil., Bauru, Brazil

Background: This paper presents the planning for a double-jaw orthognathic surgery. The patient presented skeletal Class III discrepancy, CLP, maxillary deficiency and a closed nasolabial angle. The lip angle and nasolabial angle are measurements that indicate a correct maxillary position on the sagittal anteroposterior plane.

Aims: This report discusses possible reasons for the increased lip angle, despite the maxillary deficiency, as well as the reason that allowed Le Fort 1 advancement, improving the facial balance and harmony after orthognathic surgery.

Methods: The patient was an adult male with non-syndromic bilateral cleft lip and palate (BCLP) and skeletal Class III malocclusion due to a maxillary deficiency and mandibular prognathism. The patient's main complaint was mandibular prognathism and lack of nose support.

Evaluation of facial profile revealed a deficit in the middle third, as well as underdevelopment of maxillary growth; however, the patient presented a close nasolabial angle, closed lip angle and upper lip with normal projection and overjet of 8mm.

Results: The surgical planning comprised double-jaw orthognathic surgery with maxillary Le Fort 1 osteotomy and mandibular sagittal osteotomy with clockwise rotation of the occlusal plane. Thus, it was possible to correct the maxillomandibular skeletal Class III discrepancy, improving the facial balance and harmony and achieving normal occlusal relationship, despite the upper lip projection and closed nasolabial angle.

Summary/Conclusion: The upper lip projection, closed lip angle and closed nasolabial angle are measurements that indicated incorrect maxillary positioning on the sagittal anteroposterior plane. However, an accurate three-dimensional planification for a double-jaw orthognathic surgery may improve the maxillomandibular skeletal Class III discrepancy.
Background: The post-surgical treatment of cleft patients requires time and is important for the overall success with the facial growth and appearance of the patient. During the first four months after the operation, it is very important to massage over the scar in the correct fashion and management nasal alignment. Keep straight the nasal septum and size of the nostrils. In our treatment protocol, an initial appointment is made with the parents 3 weeks after the operation to train the parents on the correct way to perform the massages and evaluated if it is necessary a nasal appliance.

Aims: Provide tools to managment cleft palate patients after surgery. Maintaining the patency of a round stucture against the forces of scar contracture is difficult matter in many areas of the human body, including stenotic nostrils. We present nasal appliances to shape and improve the form of the nasal nostrils and elongate the nasal columnella in post-surgical treatment of cleft lip and palate.

Methods: During the first period of healing, the nasal appliance is used 24 hours a day. Preferably, the appliance is used 24 hours a day for about two to three months. During this period, intermittent visits can be conducted with a clinician for any revisions or adjustments to the rhinoplasty appliance. Following this period, the rhinoplasty appliance is worn only at night. Preferably, the rhinoplasty appliance is worn at night for about four to six months. Because recurrence of nostril stenosis and other nostril shape abnormalities may occur, overcorrection is recommended.

Results: Porex stent is placed at the time of surgery and maintained in place for the first week or two; an expansile stent of the same dimensions of the silicone stent is fabricated in the meantime and inserted at the time of removal of the silicone stent. Further expansion, if necessary, may be started 2 weeks postoperatively. No complications have been encountered with the use of this appliance in 31 patients.

Summary/Conclusion: The collaboration between a dentofacial orthopedics specialist accustomed to performing post-surgical nasal molding and a surgeon has provided an “Rhinoplastic” approach to the correction— and maintenance of correction—of nostril stenosis and other nostril shape abnormalities. Lesser degrees of deformity can be corrected nonsurgically by the use of this device alone.
**P-073**

**Analysis of urogenital malformations in cleft lip and palate patients; single craniofacial center report**

D Menku¹, M Calis¹, F Ozgur¹

¹Plastic Reconstructive and Aesthetic Surgery Department, Hacettepe University, Ankara, Turkey

**Background:** Cleft lip and palate are common congenital anomalies. It occurs approximately 500 to 700 births. The most common anomalies associated with anatomical localization are facial anomalies, skeletal anomalies, central nervous system, cardiovascular system, urogenital system, ear and eye anomalies. The coincidence of these anomalies is different only in the palate cleft, or in the cleft lip and palate. Although the incidence of urogenital anomalies varies between 2% and 10%, this rate can be increased according to the region studied and the ethnic group. Urogenital anomalies most commonly associated with palate clefts are mostly found in patients with both cleft lip and palate. These anomalies can be listed as cryptorchidism, micropenis, congenital pelvic uterine separation, polycystic kidney, horseshoe kidney, dysplastic kidney, ectopic kidney, hypoplastic kidney, renal agenesis, hydronephrosis, vesicoureteric reflux, hydrocele, hypospadias, ambiguous genitalia.

**Aims:** We aimed to determine retrospectively the analysis of the urogenital anomalies which may accompany the patients with cleft lip and palate.

**Methods:** In our study, 3304 patients who were treated with the cleft lip of the palate at our clinic between June 2014 and June 2018 were examined retrospectively for many variables such as age, gender, cleft type, congenital urogenital anomaly, the type of operation. In the database of hospital information systems and palate lip cleft council records, surgery notes and patient epicrisis were examined.

**Results:** Thirty patients (21 male, 9 female) had concomitant urogenital disease. Two patients were diagnosed as focal segmental glomerulosclerosis and central diabetes insipidus. Accompanied anomalies were evaluated separately in the cleft of the lip, unilateral lip and palate, bilateral cleft palate, and isolated palate. The most frequently detected anomaly was hypospadias in 9 patients, followed by hydronephrosis in 5 patients. Multiple anomalies were found in patients. Other detected anomalies include renal agenesis, renal ectopia, cryptorchidism, renal hypoplasia, multicystic dysplastic kidney, nephrolithiasis, hydrocele, horseshoe kidney, double collecting system. In 6 patients, no other accompanying anomaly was found, while the most common anomaly in other patients was cardiovascular system diseases. Atrial septal defect is the most common. 7 patients with syndrome were DiGeorge syndrome, Trizomy 13, Cornelia de Lange syndrome, Micheals syndrome, Orofascialdigital Syndrome, Down Syndrome and a patient who was diagnosed. When compared with the literature, it is seen that the ratio ranging from 2% to 10%. Our ratio was 0.9% which was lower compared with the literature.

**Summary/Conclusion:** In this study, it was possible to obtain information about the anomalies that may be encountered in the geography we live in as a result of examination of the patients with cleft lip and palate.
Contemporary unilateral cleft lip and palate repair in one surgery with pure primary healing
A Mueller¹, B Benitez¹, A Brudnicki², R Kant Singh³, P Nalabothu⁴, Z Surowiec⁶, D Schumann⁷
¹Cranio-Maxillofacial Surgery, University and University Hospital Basel, Basel, Switzerland, ²Clinical Department of Child and Adolescent Surgery, Institute of Mother and Child, Warsaw, Poland, ³Maxillofacial Surgery, Peace Point Hospital, Bhelupur Varanasi, India, ⁴Department of Orthodontics and Pediatric Dentistry, University Centre for Dental Medicine, ⁵Department of Biomedical Engineering, University Basel, Basel, ⁶Clinical Department of Child and Adolescent Surgery, Institute of Mother and Child, Warsaw, ⁷Maxillofacial Surgery, University and University Hospital Basel, Basel, Switzerland

Background: Common practice uses multiple surgeries for unilateral cleft lip and palate repair. However, a stepwise approach has two inevitable drawbacks. First, it brings repeated burden. Second, it leaves an area of secondary wound healing at the junction between the repaired and un repaired cleft area in the first operation. In the following operation, the same area needs re-exposure to allow gapless suturing.

Aims: To reduce the burden of care to patient and tissue we developed a single stage cleft repair with healing by pure primary intention all along the oral and nasal surface.

Methods: A traditional infant orthopaedic plate with nasal stent is fabricated at birth, which is renewed only once after 3-4 months. No preoperative active tissue molding is performed, neither on the alveolar segments, by thus frequent consultations are avoided. The plate is renewed at maximum one-time before surgery. Surgery takes place at 8 months of age (mean weight 8.3kg) and comprises: hard-palate repair in two layers comprising: (1) bilateral bipedicle flaps with allocation of the oblique vomer for nasal and oral layer closure (2) soft-palate repair by a combination of subperiosteal medial pterygoid detachment and velar muscle release from the nasal layer for muscle sling backward positioning. The lip repair comprises primary rhinoplasty. No gingivoperiostplasty is performed but the two-layer hard palatal closure extends gapless across the alveolar cleft region. The surgical technical keypoints are highlighted by intraoperative photographs and video (n=11).

Results: The infant orthopaedics leads to significant spontaneous narrowing (Mean, Range) of the anterior palatal cleft width from birth (12.9mm, 17.0-10mm) to 8 month (5.6mm, 10.0mm-2.5mm). The surgery consistently allowed for tensionless soft tissue closure leading to complete primary wound healing in two layers all along the oral and nasal surface. The nasopalatine and greater palatine neurovascular bundles were maintained to assure full vascularisation and sensation on the hard palate. Hemoglobin level at the end of operation was 98.2 g/L (SD 10.1). There was no need for blood transfusion. Postoperative spontaneous respiration was uneventful in all patients and oral feeding promptly started in all patients with no need for nasogastric feeding tubes. Patient were discharged from hospital in the mean of 5 days (SD 1.5) after the operation.

Summary/Conclusion: Although we cannot report any long-term growth results of the presented technique yet, both co-authoring centers have published their long-term growth results with other single-stage techniques. However, these other techniques lead to areas of raw surface healing on the oral- or nasal side respectively. Raw surface healing is accused to inhibit growth and tissue mobility. The presented surgical evolution overcomes this shortcoming and allows for a raw-surface free healing all along the oral- and nasal surface. Cleft repair in one surgery with pure primary-intention wound healing is feasible if the traditional concepts of early lip repair, unipedicled palatal flaps or single layer palatal closure is abandoned.
Anthropometric and aesthetic outcomes for the nasolabial region in 101 consecutive African children with unilateral cleft lip one year after repair using anatomical subunit approximation technique.

M Sébastien¹, H Reychler², K Devriendt³, P Kalenga⁴, P Lukusa⁵, F Tshilombo¹
¹Pediatric and maxillofacial surgery, University of Lubumbashi, Lubumbashi, Congo, The Democratic Republic of the, ²Maxillofacial surgery, Université catholique de Louvain, Brussels, ³Centre of human genetics, KU Leuven, Leuven, Belgium, ⁴Gynecology, University of Lubumbashi, Lubumbashi, Congo, The Democratic Republic of the, ⁵Genetics, KU Leuven, Leuven, Belgium

Background: Cleft lip and palate are the most common malformation of the head and neck region. Its incidence is 1/700 newborn worldwide. Adequate surgical repair of CL/P is key to improving the anatomical and functional aspects of these structures.

Aims: The objective of this study was to determine the outcomes for the nasolabial area through anthropometric measurements and assessment of the Asher-McDade Aesthetic Index and Steffensen's criteria at 1 year after surgery.

Methods: We conducted a descriptive longitudinal study in the health district of Lubumbashi, DR Congo, from February 2012 to July 201. One hundred and one patients with complete or incomplete cleft lip underwent the anatomical subunit approximation technique for repair. The patients were followed up prospectively for 1 year. Six assessors (three cleft surgeons and three non-surgeon medical professionals) examined cropped images; reliability was assessed using Cronbach's alpha.

Results: The difference in lip length between the healthy and operated sides was 0.61mm and the difference in nostril diameter was 0.37mm (differences not significant). The average scar width was 2.78±1.35mm. Hypertrophic scars were observed in 9.9% of cases. The average Asher-McDade Aesthetic Index rating varied between 1.35 and 1.98 for all parameters. Cronbach's alpha coefficient was 0.83, 0.89, 0.98, and 0.89 for nasal form, nasal symmetry, vermilion border, and nasolabial profile, respectively. Steffensen's criteria rated appearance as 'good' in 69.3% to 91.1% of cases.

Summary/Conclusion: The anatomical subunit approximation technique can be performed in Sub-Saharan Africans for all types of unilateral cleft lip. It significantly improves the length of the medial and lateral lips, leaving an acceptable scar. A study with a larger sample size and longer follow-up is warranted.
Asymmetric lift of the soft palate: a sign of something more.
A Naparus,1,2 A Soueid2, G Phippen2, M Cadier2
1Surgical Science, University College London, London, 2The Spires Cleft Centre, Salisbury, 3Craniofacial and Cleft Surgery, Oxford University Hospitals, Oxford, United Kingdom

Background: Children, and rarely adults, present to speech and language therapists or cleft surgeons with speech problems that are sometimes related to the movement of their soft palate. This could be due to a primary disorder or secondary to a cleft palate. Depending on the findings of the clinical examination, speech assessment and special investigations, a subsequent management plan is formulated.

We describe asymmetric lift of the soft palate (AL). This clinical finding is significant as it may require adjustment of surgical technique and could be an indicator of underlying pathology.

Aims: The aim of this study was to assess whether AL, observed in a small number of our patients, has implications on outcome, surgical technique or is associated with more serious underlying pathology.

Methods: The case notes of all patients identified with AL were analysed for demographics, presentation, presence of a cleft palate, related pathologies and syndromes, speech therapy investigations, surgeries and final outcomes.

Results: 12 cases of AL were identified over 11 years (incidence 2.4%; M:F distribution 1:1). Age ranged from 0.5 - 12 years. All were referred with velopharyngeal dysfunction. 7 were idiopathic, 2 were associated with a submucous cleft palate, 2 were post CP repair and 1 was associated with forme fruste. 4 had a diagnosis of 22q11 syndrome (33%) and one 1p deletion. All had AL when asked to phonate during oral inspection. Nasometry ranged from 38 to 74% nasalance (normal< 20%). AL was noted in only 2 of 5 cases on nasoendoscopy. Video fluoroscopy showed incomplete closure of the velopharynx in 10. 75% were operated. Follow up ranged from 0.5-11 years. 5 had significant improvement post surgery, 1 moderate, and 1 poor outcome. 1 case of subtle hypoglossal nerve palsy later was shown to be caused by a space occupying lesion requiring neurosurgery.

Summary/Conclusion: Asymmetrical lift is a phenomenon not previously described in the published literature, which may be caused by differential muscle development or neurological impairment. We emphasise the importance of oral inspection of the uvula to observe this effect. Apart from presenting technical limitations in terms of surgical repair, it can be a sign of an undiagnosed syndrome, or even a more serious condition as described in our series.
Large bilateral cleft series demonstrates the utility of pre-surgical orthopaedics in minimising revision of primary definitive lip repair & the safety of bilateral vomerine flaps at primary surgery

R Nicholas¹, L Giwa¹, S Chummun¹, N Timoney¹
¹Evelina London Children's Hospital, London, United Kingdom

Background: Following of centralisation of UK cleft care in 1998, the impact of this change in cleft care delivery was audited at a national level by the subsequent ‘Cleft Care UK’ cross sectional survey in 2015. This demonstrated that it had resulted in improved outcomes for non-syndromic unilateral cleft patients. Outcomes in the bilateral cleft group justify separate consideration.

Aims: To review the outcomes of bilateral cleft patients who underwent pre-surgical orthopaedics, followed by two stage closure: Definitive lip repair was accompanied by bilateral vomerine flaps and intravelar veloplasty type completion of palate repair was performed.

Methods: A retrospective case note review of all bilateral cleft patients operated on by a single surgeon in a designated cleft centre.

Results: 72 consecutive cases were treated over an 11-year period. 45 were male and 27 were female. Median age was 6 years 1 month at time of review. 66 of these patients had bilateral cleft lip and palate (CLP) and 6 had bilateral cleft lip only (CL). 11% of cases were associated with a syndrome and 24% had significant co-morbidities. Median follow-up was 5 years 2 months. 71% of complete CLP patients underwent pre-surgical orthopaedics, which consisted of a static plate with extraoral taping (without naso-alveolar moulding). Median age at time of primary lip and anterior hard palate repair was 4 months (range 3-10 months) and was 10 months (range 6-15 months) at time of completion palate repair. The vast majority (88%) CLP cases underwent bilateral vomerine flaps (leaving a central strip of mucosa) at time of primary surgery.

At 5-year audit, 42% had good speech articulation and velopharyngeal function using CAPS A speech assessment. 74% had Class I & II occlusion. Infection rate was 2.8%, reoperation rate 1.4%, and 31% had type I-IV palatal fistulae. There was no loss of prolabium and no primary definitive lip repairs required revision.

Summary/Conclusion: This is a larger than National average single-surgeon series. Despite a relatively long follow up period, the majority had not reached the ages necessary for completion of validated skeletal growth and speech assessment. Although lower than the published target for unilateral cases, speech outcomes that were available are comparable to a recent 11 centre audit of bilateral cleft cases.

Pre-surgical orthopaedics is not in widespread use in the UK. However, it was found to facilitate primary definitive lip repair. No definitive lip repairs required revision. Bilateral vomerine flaps at time of primary definitive lip repair were demonstrated to be safe.
Pre-surgical functional maxillary orthopedic treatment

Background: Some of the defects described in cleft patients such as the width of the fissure, the smaller segment size, the deviation and inclination of the premaxilla can be modified and improved before lip surgery applying pre-surgical Functional Maxillary Orthopedic treatment (FMO).

Aims: There are different strategies to improve dental results for cleft patients. The goal of these strategies is to allow for minimally invasive surgeries that do not compromise the growth and shape of the maxillary arch.

Methods: The FMO technique for cleft patients is based on bone tissue properties and orthopedic plasticity in order to program stimulation of the maxillary bone through the intermittent mechanical action caused by the movements of the tongue on the surface of the FMO intraoral Plate.

On the surface of the plate we make transverse scratches to give the plate a different texture to stimulate the continuous touch of the tongue through proprioception; this technique calls ¨Pannaci’s Modification¨ was published in 1997 and has been applied in 7 countries from Operation Smile for 20 years, and also applied in Spain since 2016.

Results: Due to the constant and rapid growth of the baby occurring during the first year of age the FMO technique can modify the direction of maxillary bone growth and the same time can induce the volume increase of each maxillary segment in cleft patient resulting after 4-5 months of continues treatment a well-shaped maxillary arch while at the same time the alveolar and palatal gap is achieved by almost 70%.

This technique has functional in its name because it also rehabilitates the altered functions of breathing, suction and swallowing reducing the risk of aspiration pneumonia and allowing for the progressive increase in size and weight of the cleft baby before surgery.

Summary/Conclusion: By applying FMO technique, we can reduce the severity of the oral defects before surgery thus increase the possibilities of excellent aesthetic and functional results in cleft patients.
**P-079**

**Measuring system for maxillary lingual forces in cleft lip and cleft palate patients**

A Pares¹, T Pannaci², O Pelliccioni⁵

¹Technical sales and consulting, Ineltk Nord GmbH, Hamburg, Germany, ²Electrical Engineering, Simon Bolivar University, Caracas, Venezuela, Bolivarian Republic Of, ³Maxillofacial Department, Vall d’ Hebron Hospital, Barcelona, Spain, ⁴Dentist, Operation Smile, Norfolk, United States, ⁵Biomechanical Group, Simon Bolivar University, Caracas, Venezuela, Bolivarian Republic Of

**Background:** The FMO technique for cleft patients stimulates the maxillary bone through the intermittent mechanical action caused by the movements of the tongue on the surface of the FMO intraoral Plate. Knowing the pressures exerted by the tongue we can develop numerical methods that allow the reconstruction of the malformation in order to provide complementary information for the programming of the stimulus that the plaque gives to the maxillary bone.

**Aims:** Designing and manufacturing of a device capable of recording tongue pressures in patients with cleft lip and cleft palate in order to develop new treatments and improve the of Functional Maxillary Orthopedics technique, as well as supporting them. It also allows making better decisions regarding the treatment of the patient.

**Methods:** We developed a system for the acquisition and recording of loads (pressure) transmitted by functional maxillary orthopedic intrabucal plates, intended for patients between 0 and 5 months of age who present cleft lip and cleft palate. The age range was chosen because during the first four months of life the highest growth peak occurs in which a baby doubles its weight and the analysis of the data is more accurate. The device consists of two sensors attached to a functional maxillary orthopedic (FMO) intraoral plate and connected to a data acquisition system that by serial port, using a microcontroller, transforms and stores the data in digital files for its further analysis and use.

**Results:** After building the device and testing it in six different patients between 0-5 years old with lip cleft palate, three hypotheses were proven: major segments present greater pressures, the pressures in the maxillary segments are different from the pressure measured in the center of the FMO plate and the greater the applied pressure, the greater the volume of the bone.

All of these help to improve the design of the FMO plate and the correction of the condition in the early years of life.

**Summary/Conclusion:** The literature related to recording loads in the mouth of newborns is scarce because an infant does not follow instructions and the work space is small and complex. The actual solution is to do surgery, losing the space where the bone could still grow, obtaining an asymmetrical result that needs more surgeries in the future and subjecting the patient to painful treatments.

The pressures obtained are associated with the bone growth response. The recording of pressures in the patients allows checking and improving the FMO techniques. By detecting in time the magnitude of the patient’s tongue strength, the presurgical treatment of the cleft patient can be better programmed contributing to minimally invasive surgeries.
Background: Orthognathic surgery is often necessary in cleft lip and palate patients after their skeletal growth is finished in order to achieve good facial aesthetics and functional occlusion because of maxillary hypoplasia. Maxillary morphology is very specific in cleft lip and palate patients and therefore it is necessary to follow up the surgery results not only in short-term but in the long-term in particular to verify stability of the surgically achieved maxillary position.

Aims: Aim of this preliminary study was to determine extent of relapse after maxillary advancement surgery in adult cleft lip and palate patients.

Methods: In the study four patients were followed up, 20-24 year old males with unilateral cleft lip and palate. All patients underwent LeFort I. osteotomy. The cephalograms were taken before, right after the surgery and 2-3 years after the procedure. On the cephalograms the position of maxilla was determined by measuring the SNA angle and horizontal and vertical position of the A point. Frankfurkt horizontal (FH) and perpendicular to the FH from Porion point were drawn on each cephalogram. Vertical dimension was measured as distance between A point and FH and horizontal dimension as distance between A point and the perpendicular to FH. All values were measured twice in four weeks distance by one evaluator and average values were used to eliminate measurement errors. In the study four patients were followed up, 20-24 year old males with unilateral cleft lip and palate. All patients underwent LeFort I. osteotomy. The cephalograms were taken before, right after the surgery and 2-3 years after the procedure. On the cephalograms the position of maxilla was determined by measuring the SNA angle and horizontal and vertical position of the A point. Frankfurkt horizontal (FH) and perpendicular to the FH from Porion point were drawn on each cephalogram. Vertical dimension was measured as distance between A point and FH and horizontal dimension as distance between A point and the perpendicular to FH. All values were measured twice in four weeks distance by one evaluator and average values were used to eliminate measurement errors.

Results: Directly after LeFort I. osteotomy advancement in all UCLP patients SNA angle increased by 6.4° in average, mean horizontal displacement was 10.45% and mean vertical displacement was 0.95%. 2-3 years after surgery in all patients partial relapse was detected, mostly in vertical dimension – the resulting position of the A point in comparison to the position before surgery decreased by 4.68%. SNA angle enlarged by 1.8° in average, horizontal dimension in average remained 4.43% bigger than before the surgery.

Summary/Conclusion: LeFort I. osteotomy advancement in adult UCLP patients showed satisfactory stability in horizontal dimension. Reduction in vertical distance of the A point to Frankfurt horizontal could be caused by tissue retraction in scar area. Further research is necessary to verify extend of relapse of the LeFort I. advancement surgery in cleft lip and palate patients. LeFort I. osteotomy advancement in adult UCLP patients showed satisfactory stability in horizontal dimension. Reduction in vertical distance of the A point to Frankfurt horizontal could be caused by tissue retraction in scar area. Further research is necessary to verify extend of relapse of the LeFort I. advancement surgery in cleft lip and palate patients.
P-081
treatment outcome of 4 - 7 years-old patients with cleft lip and cleft palate in surgery examination room srinagarind hospital
S Pongpagatip¹, S Pradubwong², K Winaikosol³, K Jenwitheesuk³, P Surakunprapha³, B Chowchuen³
¹Division of Nursing, Khon Kaen University, ²Division of Nursing, Srinagarind Hospital Khon Kaen University, ³Department of Surgery, Khon Kaen University, Khon Kaen, Thailand

Background: Cleft palates patients need a complete treatment from a multidisciplinary team including surgeries, follow-up treatments, and a recovery protocol of Tawanchai Center in order to have good results of the treatment.
Aims: To study the follow-up treatment and recovery after surgeries of cleft palates patients in the surgery examination room at Srinagarind Hospital.
Methods: This descriptive study used history and background of 93 patients who have spent 4-7 years in the surgery examination room at Srinagarind Hospital and took 3 months to collect the data. The research tool was a general questionnaire on the follow-up treatment and recovery after the patients’ surgeries at the surgery examination room and applied statistics and a content analysis conclusion to analyze the data.
Results: 68 out of 93 patients suffer unilateral cleft palates (73.12%): 82 patients came to receive a follow-up treatment and recovery after the surgery within a two-week period(87.17%); 78 patients received follow-up treatments after the surgery and recovered from cleft palates conditions within the first two weeks(86.67%). They were evaluated, consulted, given treatments from surgeons and nurses in the surgery examination room and Tawanchai Center, and transferred to the multidisciplinary team for further treatment in accordance with the protocol.
Summary/Conclusion: 93 patients aged around 4-7 years old suffer from unilateral cleft palates the most; most of them came to receive follow-up treatments two weeks after the surgery as well as recovered in accordance to the protocol of Tawanchai Center. They were also sent for further inclusive and suitable treatment from multidisciplinary team according to their ages.
Perioperative management of clp patients – an international survey
R Preidl¹, M Kesting¹, A Rau¹
¹Department of Oral and Maxillofacial Surgery, University Hospital Erlangen, Erlangen, Germany

Background: In cleft lip palate surgery, perioperative treatment strategies like antibiotic prophylaxis, postoperative feeding and duration of hospital stay are non-standardized and vary between different cleft centres worldwide. Likewise, intraoperative choice of suture materials, timepoint of suture removal as well as preoperative ENT-consultation and the application of presurgical orthopedics are inconsistently performed.

Aims: We wanted to collect information on protocols focussing these perioperative topics in order to summarize and subsume currently approved treatment strategies from different departments and healthcare centres.

Methods: A designed questionnaire was distributed asking for the respective treatment strategies. 70 centres returned the questionnaires and descriptive statistics were performed.

Results: During cleft lip repair absorbable suture material is used for closure of the outer lip skin in 20 centers whereas 50 centers prefer non-absorbable materials. Suture removal is conducted after 7,0 ± 1,5 days at the skin. Reconstruction of the orbicularis oris muscle, suturing of the enoral as well as the nasal mucosa and the palatal musculature is predominantly performed with absorbable suture materials at a suture size of majorly 4-0 or 5-0. Intraoperative antibiotic prophylaxis is applied in 82,9 % of the participating centres primarily by using penicillins and cephalosporins. In contrast, 31,9% of the departments do not apply any antibiotic postoperatively. Postoperative nutrition is performed in 27 centres via a nasogastric tube for 4,6 ± 2,3 days in average. Mean length of postoperative inpatient stay is 4,1 ± 2,6 days in children after cleft lip surgery and 4,5 ± 2,7 days after cleft palate repair. Preoperative NAM is conducted in 37,1% of the centres. ENT-consultation prior to surgery is routinely conducted in 52,8% and in 82,9% hospitals ENT-colleagues investigate middle ear pathologies in the same operation as the cleft surgery is performed.

Summary/Conclusion: Beside the great variations of treatments concepts between the participating departments, intraoperative antibiotic prophylaxis as well as an inpatient hospital stay of less than one week in combination with a preoperative consultation and intraoperative cooperation with the ENT- department seem to be well-proven concepts in CLP-patient care. Further investigations - favourably in the context of multicentre studies - will be needed to verify perioperative management concepts in cleft surgery.
Mandibular asymmetry reconstruction after free fibular flap using unilateral sagittal split osteotomy
K Bangun¹, N Putri¹, P Atmodiwirjo¹, J Pancawati¹
¹Plastic Reconstructive and Aesthetic Surgery Division, Department of Surgery, Cleft and Craniofacial Center Cipto Mangunkusumo Hospital, Plastic Reconstructive and Aesthetic Surgery Division, Department of Surgery, Medical Faculty, Universitas Indonesia, Jakarta, Indonesia

Background: Mandibular asymmetry after free fibular flap reconstruction in a patient following mandible tumor resection is one of the late complication. Despite the accuracy of the presoperative planning and surgical execution, mandibular reconstruction can sometimes result in a facial deformity or asymmetry. Orthognathic surgery is aimed to restore the proper anatomical and functional relationship in patients with dentofacial skeletal abnormalities. Asymmetry of the mandible cases usually corrected with BSSO. From the previous literature, one of the sides of the ramus osteotomy does not change or minimal changes in 3D planning. Based on this phenomenon, the unilateral sagittal split ramus osteotomy (USSRO) was used to correct the asymmetry.

Aims: To present the evaluation and the outcome of surgical reconstruction of mandibular asymmetry after free fibular flap reconstruction

Methods: A case report based on the medical and surgical records was done. We did pre-surgical assessment by the orthodontist by using wafer as guidance to get the optimal occlusion. USSO was done to correct the asymmetry of the mandible on patient who had a history of hemimandibulectomy and mandible reconstruction with free fibular flap.

Results: The mandibular asymmetry was corrected with USSO. NSAID was given post operatively to reduce the edema. Edema and hematoma post-operative was found. Patient was stay in hospital for 5 days. Better occlusion and aesthetic outcome was achieved.

Summary/Conclusion: The mandibular asymmetry requires multidisciplinary approach including presurgical assessment by the orthodontist to get the optimal result of the occlusion and aesthetic outcome. USSO can be used to correct the mandibular asymmetry in patient with the history of mandible reconstruction with free fibular flap. The edema and hematome post operative must be considered as an early complication.
**Versatility of the vomerine flap in hard palate closure for complete unilateral and bilateral cleft lip and palates.**
B Rafique¹, F Maggiulli¹, A Sadri¹, M Pinkstone², P Morris¹
¹Cleft Lip and Palate Surgery, ²Speech and Language Therapy, Great Ormond Street Hospital, London, United Kingdom

**Background:** In the UK, the vomerine flap is a widely used method of reconstructing the hard palate at the time of primary lip repair. Despite evidence which suggests there is a high failure rate and subsequently higher risk of fistula and complications¹,², the senior author has found this technique to be routinely reliable with excellent results. These are in cases of both complete unilateral and bilateral cleft lip and palate.

**Aims:** We present a retrospective cohort study of 100 consecutive cases of UCLP and 31 BCLP between 2012 and 2018. The aim was to examine flap survival rate, fistula rate, early speech outcomes and demonstrate the important relevant technical aspects of the procedure with a short video.

**Methods:** Using an institutional electronic database we retrospectively reviewed 131 cases which were performed over five years by the senior author. At the time of soft palate repair (intra-velar veloplasty) the integrity of the hard palate closure was confirmed and documented in all cases.

**Results:** In all 131 cases there was no loss of vomer flaps and no hard palate fistula present. We present speech outcomes, where appropriate, including articulation and perceptual features of velopharyngeal dysfunction using the CAPS-A protocol.

**Summary/Conclusion:** In our experience, we have found the vomerine flap an excellent technique for reconstructing the hard palate. From a technical point of view one must always be mindful to plan a large broad based flap with minimal tension at inset and careful dissection on the oral layer enough to allow sufficient double breasting of the raw surface of the flap deep to the oral layer.

L-085
Lip adhesion: a solid keystone for departments with no presurgical nasoalveolar moulding
L Rosato1, A Giacomina2, G Gatti2
1Plastic Surgery, Hospital of Piacenza, Piacenza, 2Plastic Surgery, Hospital of Pisa, Pisa, Italy

**Background:** Patients with a wide bone cleft represent a huge challenge for surgeons. Moreover in Plastic surgery unit with no possibilities of pre surgical nasoalveolar moulding.

**Aims:** The lack of the bone, the asymmetry of the mandibular arches and the nostrils, the tension on the soft tissue are the most common surgical challenge. Lip Adhesion is a surgical procedure with almost 50 years of history and represent an open field of discussion regard pros and cons. Different techniques has been described over the years, we combined some tips and tricks from Randall-Graham’s and Seibert’s techniques, in order to achieve our best.

**Methods:** Lip Adhesion is a procedure used in our department since the ‘70. We analyzed 214 patients from 2009 to 2018. We used to perform the procedure in unilateral and bilateral cases with a bone gap ≥7 mm. The average age was of 6 weeks and after 3 weeks we performed the lip repair. We had 6 cases with dehiscence. Preoperative and postoperative photographs were taken for all the patients.

**Results:** We’ve achieved good results even with extra rotated pre maxilla. The precocious restore of the orbicularis oris muscle and the equal distribution of the forces, helps us to reduce the extra-rotation of the pre maxilla, restore its central position and reduce the bone gap (with an average of 50%). It also helps to perform an easier lip repair with less soft tissue tension and a better symmetry.

**Summary/Conclusion:** Lip Adhesion procedure is a solid and useful tool for the cleft surgeons. Even more in departments where there’s no possibility presurgical nasoalveolar moulding, it helps to achieve good results in few weeks.
Long-term follow-up of maxillary advancements in cleft lip and palate patients
G Rossetti¹, L Pellegrinelli¹, M Ferrari¹, V Battista¹, M Meazzini¹, L Autelitano¹
¹Smile House Maxillo Facial Surgery, ASST Santi Paolo e Carlo, Milano, Italy

Background: Maxillary hypoplasia is common among cleft children and often requires Le Fort I advancement. Conventional orthognathic surgery is often performed for the correction of this deformity. This treatment allows an immediate improvement of the morphology and occlusion of the patients. In cleft patients the presence of scar tissue can have an effect on relapse, therefore a long term evaluation is important to establish the stability of the results.

Aims: The objective of this study was to evaluate long-term results of maxillary advancements in patients with cleft in terms of skeletal position and occlusion.

Methods: The records of 30 consecutive patients affected by cleft lip and palate, who underwent Le Fort I osteotomy were reviewed. All patients had cephalometric records pre-operatory, immediately post-operatory, 12 months post-operatory and long-term records with a long-term follow-up of >3 years (range 3-13 years). Vertical an horizontal changes were recorded.

Results: Le Fort osteotomy allows to normalize occlusion and facial profile in cleft patients. Post-surgical stability of the occlusion was recorded with some skeletal relapse. Total skeletal movement, dental post surgical movement and skeletal relapse will be reported.

Summary/Conclusion: Conventional Le Fort I advancement can improve facial profile and occlusion in cleft patient with maxillary hypoplasia and is a relatively stable and reliable procedure.
Cleft rhinoplasty: correction of the alar scroll to achieve aesthetic and functional improvement
A Sadri¹, L Kangesu¹, H Hawys Lloyd-Hughes²
¹Plastic Surgery, Great Ormond Street Hospital, ²London Deanery, London, United Kingdom

Background: Disruption of the alar scroll region on the cleft side in patients with unilateral cleft lip nasal deformity leads to aesthetic asymmetry and blockage of the nasal airway.
Aims: The aim of our study was to analyse how correction of the alar scroll affected aesthetic and functional outcomes in patients undergoing cleft rhinoplasty.
Methods: Patients undergoing cleft rhinoplasty prospectively had pre- and postoperative (6 weeks) patient reported nasal obstruction scores assessed using a validated questionnaire (NOSE [Nasal Obstruction Symptom Evaluation] score). Pre- and postoperative standardised rhinoplasty photographs were also taken for analysis of aesthetic improvement.
Results: Thirteen patients underwent open cleft rhinoplasty with our technique. All patients had unilateral cleft lip nasal deformity, 92% of patients had complete UCLP. Mean age at surgery was 22.8 yrs. No patients underwent septoplasty. Mean pre-operative NOSE score was 55, mean post-operative score at 6 weeks was 16.7. All patients reported good satisfaction with aesthetic outcome post surgery.
Summary/Conclusion: Restoration of the normal anatomical association between the alar and upper lateral cartilage leads to improvement in nasal appearance and airway. Our technique avoids the need for septoplasty in patients undergoing cleft rhinoplasty.
Surgical treatment for internationally adopted children compared to Swedish born children with cleft lip and palate, using the Swedish cleft lip and palate (CLP) registry.

J. Sahlsten Schölin¹, A. Paganini¹, S. Rizell², M. Becker³, H. Mark¹
¹Dept of Plastic Surgery, Sahlgrenska University Hospital, ²University Clinics of Odontology and University of Gothenburg, Orthodontic clinics, Gothenburg, ³Dept of Plastic Surgery, Malmö University Hospital, Malmö, Sweden

Background: During 2008-2011 a peak in increase was seen of internationally adopted children (IAC) with CLP from China to Europe and the US, and among them, Sweden was a top receiving country. IAC tend to arrive with un-operated clefts at an age when Swedish born peers have completed their primary palate surgery. Studies on palatal surgery, comparing the treatment and outcome between IAC and native cohorts have shown higher fistula rates, velopharyngeal insufficiency and higher need for surgical revisions, but the reason for this still remains unclear. To ensure a good quality of treatment for the patient, the Swedish cleft lip and palate registry allow comparison and open reporting of results. Average coverage ratio is reported to be 91.3% in the last annual report. All Swedish CLP centers are involved.

Aims:
1: To compare all registered IAC with Swedish born children with CLP regarding demography (diagnosis, date of birth, sex, other malformations/comorbidities and age at first visit at the cleft clinic), in the Swedish CLP registry.
2: To compare all registered IAC with Swedish born children with UCLP and BCLP in their respective diagnosis groups, regarding surgical treatment.

Methods: Baseline data was obtained from the Swedish National registry for all children with any cleft diagnosis, born between 2000-2014. Data on surgical treatment was obtained from the Swedish National registry for children with uni- or bilateral CLP who were born 2000-2014.

Results: The study is ongoing and results will be presented at ECPCA.

Summary/Conclusion: Data from a national health care registry with good coverage ratio such as the Swedish CLP registry, is valuable when studies are to be performed on heterogeneous patient cohorts like IAC with CLP.
A less invasive approach to infants with pierre robin sequence and cleft palate: could that be efficient?

M Schettino¹, O Senese¹, A De Mey¹, D Franck¹
¹Pediatric Plastic Surgery Department, Queen Fabiola University Children’s Hospital, Brussels, Belgium

Background: The standard surgical care of cleft palate patients presenting a Pierre Robin sequence is still controversial. Four treatment modalities have historically been used to address the airway: tracheostomy, conservative management, tongue-lip adhesion, and mandibular distraction osteogenesis. Tracheostomy achieves control of the airway but is associated with significant morbidity and is now considered a treatment of last resort. Tongue-lip adhesion is a procedure which addresses glossoptosis by fashioning a temporary adhesion of the tongue to the lower lip. Mandibular distraction osteogenesis is a newer procedure that addresses mandibular hypoplasia, thereby allowing the tongue to move anteriorly. Conservative management is another option for newborns with Pierre Robin sequence who demonstrate adequate growth and mild airway obstruction relieved with side or prone positioning. Conservative management was the standard of care for our patients.

Aims: We report our experience at Brussels Queen Fabiola Children’s Hospital, regarding the clinical management and surgical planning of this entity, over 28 years.

Methods: Between 1988 and 2017, we collected a series of 600 consecutive patients operated for cleft palate closure. Among them, 47 presented Pierre Robin sequence. A single surgical procedure has been performed by the same team overall these period. Pre- and postoperative data were retrospectively analysed, with the approval of the ethical committee of our institution (CEH n° 101/18).

Results: Median age and weight are respectively 171 days (between 5 and 6 months of age) and 6.3 kilograms at time of surgery. No one needed major surgical procedure or invasive respiratory assistance care before closing the palate, except for one patient who needed endotracheal intubation. An associated malformation was found in 31% of the patients and among them, 4% resulted in cardio respiratory pathology. Polysomnography studies have been performed for all patients. The average of cardio respiratory monitoring was 80 days. The neonatal intensive care average permanence was 41 days. Palate plate was used before surgery in 40% of our patients. Nasogastric feeding was registered in 29% of the sample, with an average time of 162 days. No major complication of the surgery have been encountered excepted two local haemorrhages, one of which needed surgical exploration to control bleeding. The median length of hospitalisation was 5.6 days. Post-operative oronasal fistula occurred in 6 patients (12%) whose require further surgery.

Summary/Conclusion: Infants with Pierre Robin sequence are an heterogenic group with a wide spectrum of associated anomalies. The decisional approach is challenging and must be multidisciplinary. We did not observe more postoperative major complications on our patients group in comparison with the results found in the literature and with the non-syndromic palate patients operated during the same period in our department with the same gentle approach. We believe that an adequate medical care during the neonatal period allow to avoid invasive surgical procedures without extending the length of neonatal intensive care.
Surgical management of a rare variant of submucous cleft palate

P Sharma¹, S Krishnan¹, Z Chaudhary¹, R Sharma¹
¹Oral and maxillofacial surgery, Maulana azad institute of dental sciences, New delhi, India

Background: A submucous cleft is a palatal defect that is bridged over by mucosa; such a defect has been recognized for many years. A small portion of all cleft palate defects shows this phenomenon, but the cryptic nature of the lesion and the frequent failure to include it in the differential diagnosis of speech problems may make the defect’s discovery a belated one.

Aims: This report is a case of incomplete submucous cleft palate that resembled as postoperative palatal fistula initially,and its surgical management is also described.

Methods: An 18-year-old female patient reported to us with complaints of nasal regurgitation of both solids and liquids on eating along with difficulty in speech since birth. Her history suggested no surgeries.

Intraorally the defect strikingly resembled a postoperative palatal fistula,. Apart from the history, there was no discernable surgical scar over the palate . The needle probing indicated a V-shaped bony defect starting from possible posterior border of the hard palate until just 8 mm posterior to the nasopalatine foramen.Considering the situation, a plan of a modified Bardach 2-flap technique,was decided as the surgical plan for closure.

Results: The most important aspect of cleft palate surgery is complete tension-free closure. This reduces the chances of occurrence of oronasal fistulas, and a proper repair also ensures a good function of the soft palate aiding in speech. A properly performed palatal closure often avoids a secondary surgical procedure of pharyngoplasty. The oronasal fistula was of a large size and surgical procedure provided good result,

Summary/Conclusion: Submucous cleft palate is a cryptic condition in which identification of the condition is difficult, and a large number of these patients have a normal speech and often minimal muscle defect. The true incidence of submucous cleft palate is hard to pinpoint because of its difficulty in identification. The early recognition of submucous cleft palate is important even that of a mild nature. Such patients should be followed up carefully by a physician or a group involved with cleft care.
Background: The submucous cleft palate (SMCP) is less prevalent and can be considered the most subtle type of cleft palate. Because SMCP is less visible it could be missed during the initial after birth screening and is often diagnosed late. Early detection is necessary to initiate possible speech therapy and to make it possible to operate before the patients develop compensatory speech mechanisms.

Aims: The aim of this study was to investigate at what time patients with a SMCP present at our unit, determine what and when patients are operated and analyze the postoperative speech outcomes in SMCP patients after different operation techniques.

Methods: Patient records from 766 individuals registered in the cleft registry from 1997 to 2014 in the Wilhelmina’s Children’s Hospital, Utrecht, were retrospectively reviewed. Inclusion criteria were isolated SMCP. Data were collected for age at diagnosis, inspection upon diagnosis, age at surgery, surgical technique, speech therapy pre- and post-surgery, Otitis media, secondary cleft surgery, family history and anomalies.

Results: In total, 53 SMCP patients were identified. The mean age of diagnosis was 3.8 years (SD = 3.0). In 47 patients (89%), surgical intervention was performed (Furlow plasty, intravelar veloplasty, pharyngoplasty or a combined operation). The mean age at time of operation for all patients was 5.7 years (SD 2.8). There were no significant differences in operating outcomes between surgical techniques.

Summary/Conclusion: This study reconfirms that SMCP often present late age and that almost 90% patients ultimately need palate surgery. No conclusions could be made about operative success rate between surgical techniques due to small patients groups for different types of surgery.
Primary rhinoplasty in the treatment of children with congenital cleft lips
Y Stepanova¹, M Tsyplakova ¹, A Usoltseva¹, A Baindurashvili¹
¹The Turner Scientific Research Institute for Children’s Orthopedics, The Turner Scientific Research Institute for Children’s Orthopedics, Saint-Petersburg, Pushkin, Russian Federation

Background: Treatment of children with congenital clefts of the upper lip and palate is an actual problem of maxillofacial surgery. Congenital deformity of the nose in varying degrees is accompanied by any form of cleft lip. As the child grows, the deformity of the nose progresses and becomes the cause of psychological dissatisfaction.

Aims: The objective of the study was to evaluate the benefits of using primary rhinoplasty in the treatment of children with congenital unilateral and bilateral lip clefts.

Methods: Correction of the deformity of the nose for children with congenital cleft lip was performed in 328 children aged 2–11 months old (272 patients with unilateral and 56 patients with bilateral cleft lip) during the primary operation with the technique advanced at the clinic. In the preoperative period, children with complete clefts of the upper lip underwent early orthodontic treatment in order to reduce congenital deformity of the upper jaw. In the postoperative period, all children used individual nasal tubes in the nose, and also they received a massage of postoperative scars.

Results: When assessing immediate and long-term results of treatment with primary rhinoplasty, the results of treatment of children in both patients’ groups with one-sided and bilateral lip clefts were improved. In all cases, good treatment results were obtained. The use of the technique of primary rhinoplasty does not have a significant effect on the growth of cartilaginous structures of the nose, which leads to a decrease in the number of repeated corrective operations.

Summary/Conclusion: Carrying out early radical rhinoplasty in the primary operation in combination with orthodontic treatment and constant dynamic follow-up in the conditions of the dispensary center allows to improve the results of treatment significantly, to create conditions for normal growth of tissues of the middle zone of the face.
Influence of repaired cleft lip and palate on layperson perception following orthognathic surgery
L Lin¹, R Zhang¹, I Hoppe¹, J Swanson¹, S Bartlett¹, J Taylor¹
¹Children's Hospital of Philadelphia, Philadelphia, United States

Background: Facial scarring and disharmony caused by clefting are associated with psychosocial stress, which may be improved by orthognathic surgery.

Aims: The authors examine how clefting influences change in layperson perception of a patient following orthognathic surgery.

Methods: One thousand laypersons were recruited through Mechanical Turk to evaluate patient photographs before and after orthognathic surgery. Nineteen patients-five with unilateral and five with bilateral clefting-were included. Respondents assessed six personality traits, six emotional expressions, and likelihood of seven interpersonal experiences on a scale from 1 to 7.

Results: Changes in all aspects of social perception after the procedure differed significantly between cleft versus noncleft cohorts (p < 0.01 for all). Respondents evaluated the change for the cleft cohort compared with the noncleft cohort as more trustworthy, friendly, sad, and afraid; more likely to feel lonely, be teased or bullied by others, or feel anxious around others; less angry, disgusted, threatening, dominant, intelligent, happy, and attractive; and less likely to have romantic relationships, friends, or be praised by others. For unilateral versus bilateral cleft cohorts, change in social perception was significantly different in four of the 19 items (p < 0.05 for all). Social perception change for the unilateral cohort was less surprised, sad, dominant, or happy compared with the bilateral cohort (p < 0.05 for all).

Summary/Conclusion: Despite significant improvements in social perception following orthognathic surgery, cleft patients benefit less than noncleft patients. These findings may be useful to counsel postsurgical expectations for cleft patients undergoing orthognathic surgery.
Modified buccal myomucosal flap closure of large anterior palatal oronasal fistula
J Swanson, A Siu
Operation Smile Nicaragua, Operation Smile, Managua, Nicaragua

Background: Postoperative palatal fistulas may occur in 25-57% of patients with cleft palate who are treated in low-resource settings. An ideal corrective procedure would be performed in a single-stage, for a wide age range, with low airway/anesthetic risk, with reliable outcome, low comorbidity, and be teachable. Either a tongue flap or conventional facial artery myomucosal (FAMM) flap would meet only several of these criteria; FAMM flaps also often necessitate a dental splint between stages.

Aims: We present a modified buccal myomucosal flap with the goal of achieving each of the six objectives, and review medium-term outcomes.

Methods: In this technique, large anterior palatal fistulas are closed in two layers: first, with apposed nasal turnover flaps of vomer mucosa medially and nasal wall mucosa laterally; second, utilizing a posteriorly-based buccal flap incorporating full-thickness buccinator muscle and overlying mucosa, with interposition of the flap in the retromolar trigone and lateral palate to preserve dental occlusion. This technique was employed in pediatric and young adult patients in low resource settings. Follow-up was attempted at 1 week and 2 months postoperatively, with evaluation of palatal patency and any donor-site comorbidity.

Results: Among 8 consecutive patients treated in Nicaragua, Guatemala, the Phillipines, and Jordan, 7 completed full postoperative followup. Patients ranged in age 3-22 years, with mean anterior palatal fistula size of 2.5 cm² (range 0.8-3.5 cm²), and mean followup of 5 months (range 2-13 months). All 7 patients showed decreased fistula symptoms; 5 healed expediently, 1 flap exhibited delayed tip mucosalization, and 1 flap experienced partial anterior flap dehiscence that required revision in the operating room. 5 of 7 flaps were successfully performed as a single stage; in addition to the above revision, one flap required delayed flap division to achieve comfortable occlusion. Anesthesia was straightforward in all 8 cases. No patients exhibited facial asymmetry dynamically or in repose, limitation in lip purse or pucker, or had limited mouth opening. This technique is being taught and increasingly performed by trainees in these settings.

Summary/Conclusion: Palatal fistulas in low-resource settings are a prevalent problem. Current methods have limited applicability due to lack of surgeon familiarity and constrained resources, such as skilled anesthesia; regardless of technique, outcomes are often unknown due to patient barriers to follow-up. The modified buccal myomucosal flap has shown promising medium-term results with regard to being performed in a single-stage, for a wide age range, with low airway/anesthetic risk, with reliable outcome, low comorbidity, and teachability.
**Background:** Furlow double opposing z plasties has become a popular choice of surgical repair for cleft palate in the recent years. It was hypothesized that Furlow palatoplasty would result in velar elongation and retro-positioning of the levator sling, resulting in clinical speech improvement. However, objective speech outcome data is lacking, especially in the Chinese population.

**Aims:** Retrospective analysis of the speech data in patients receiving Furlow palatoplasty using the Pittsburgh Weighted Speech Scale (PWMSS) in a single center in Hong Kong.

**Methods:** All patients who had received Furlow palatoplasty by a single surgeon since January 2016 with cleft palate with and without cleft lip were recruited in the study.

**Results:** 23 patients' data were reviewed (13F: 10M), age at Furlow palatoplasty ranged from 10-month-old to 20-yaer-old. 21 palatoplasties (6 cleft lip & palate, 15 cleft palate without cleft lip) were performed as part of the primary repair of the cleft palate, 2 palatoplasties were performed as "revision" operation for significant velopharyngeal insufficiency after primary palate repair. Formal speech assessment was performed by a speech therapist. Only 3 patients (13%) received a PWMSS of >= 7, indicating that moderate hypernasality.

**Summary/Conclusion:** Objective speech assessment in post palatoplasty patients remained difficult, and a comprehensive assessment would often requires multiple assessments at different time intervals. While there is currently no validated Cantonese assessment tool for cleft speech in Hong Kong, we could only rely on the PWMSS in order to benchmark our current surgical outcome, in order to provide a reference point and to facilitate further technical advancement. Herewith we presented our short term speech outcome for a heterogenous group of cleft palate patients using the PWMSS.
Revision furlow palatoplasty: does it work?
P. Tanq, K. Liu

Background: The Furlow double opposing z-plasty had gained popularity as the technique of choice for the primary repair of cleft palate in the recent years. It utilizes the principles of transpositioning the palatal muscles and lengthening of the soft palate to achieve adequate velo-pharyngeal portal closure while minimizing dissection onto the palate, theoretically reducing the risk of subsequent maxillary growth inhibition. However, data on the use of revision Furlow palatoplasty in the management of post primary palatoplasty complication is scanty.

Aims: We review the short term outcome of patients who underwent revision Furlow palatoplasty in our center by a single surgeon.

Methods: Medical records of patients who had undergone revision Furlow palatoplasty in our center were reviewed retrospectively. Indications for surgery, complications and the short term speech outcomes were reviewed.

Results: From June 2016 to October 20218, ten patients received revision Furlow palatoplasty in our center. There were 8F:2M with a mean age of 13.3 year-old (age at revision Furlow surgery) (range: 2 year-old to 24 year-old). Four patients had received primary palatoplasty in another institution while size patients had received the von Langenbeck primary repair at our center previously. For the indication for revision Furlow: two patients had symptomatic palatal fistula, one patient had significant soft palate dehisence post primary palatoplasty and seven patients had hypernasal speech secondary to velo-pharyngeal insufficiency. For the surgical outcomes of the revision Furlow: there were no palatal fistula or dehisence, and all but two patients reported improvements in their hypernasality by perceptive speech assessment.

Summary/Conclusion: Revision Furlow palatoplasty is a safe and feasible surgical option in the management of post palatoplasty complications. Appropriate patient selection is important in optimizing outcome.
207 primary cheiloplasty in a single center in Hong Kong
P. Tang, L. Kelvin

**Background:** Various surgical techniques have been developed over the years for the primary surgical correction of cleft lip deformity. Understandably, objective and quantitative outcome measures is almost impossible. Surgical revision rate would provide a relatively hard outcome assessment.

**Aims:** To review the revision rate of patients who received primary cheiloplasty operation in a single center in the past ten years.

**Methods:** Medical records of patients who had received primary cheiloplasty in our center since 2009 to 2018 were retrospectively reviewed.

**Results:** 207 patients’ data was retrieved. Out of the 207 patients, 38 were bilateral cleft lip and 4 of them were lateral cleft lip (macrostomia). For the remaining 165 patients with unilateral cleft lip: 106 (64%) underwent rotation advancement repair with Mohler incision.; 36 (22%) underwent modified Tennison’s repair, and 20 (12%) underwent modified Fisher’s repair. Three patients’ unilateral cleft lip were repaired with other surgical techniques. Lip revision rates for the different primary surgical techniques were as followed:
- Rotation advancement with Mohler incision : 30 (28%)
- Modified Tennison : 13 (35%)
- Bilateral cleft lip repair : 2(5%)
- repair of lateral cleft lip : 1(25%)

**Summary/Conclusion:** From our review, primary modified Tennison repair of unilateral cleft lip produced the highest lip revision rate. However, as unilateral cleft lip is a highly heterogenous conditions and there are various different parameters influencing the decision for lip revision surgery, lip revision rate would not provide a reliable indication for the appropriateness of the selection of primary cheiloplasty technique. Our review should only serve as a benchmark for future reference.
An uncommon complication of buccal flap repair for palatal fistula

P. Tang¹, L. Kelvin²
¹Kosovo, ²Hong Kong, Hong Kong, Hong Kong

Background: The use of buccal fat pad flaps for reconstruction of oral defects was first introduced by Egeydi and has then been used by many surgeons. However, its use is often restricted in the paediatric population for the repair for post palatoplasty palatal fistula. Herewith we present an uncommon complication of buccal flap repair in an adult patient with an anterior palatal fistula.

Aims: We described the surgical techniques for anterior palatal fistula closure with buccal flap and presented the complication. Preventive measures to avoid similar complications are discussed.

Methods: A 24-year-old boy complained of diplopia after blunt soccer injury to the right eye. CT confirmed fractured right orbital floor with herniation of the orbital contents, tarsal plate repair with hard palatal graft was performed. The hard palate donor site failed to heal completely after 6 months, giving rise to a 6 x 10 mm anterior palatal fistula.

The anterior palatal fistula remained symptomatic, resulting in significant hypernasal speech and persistent nasal regurgitation of liquid. In view of the size and location of the fistula, double layer closure was decided. The nasal layer was closed by local flap in a flip-over manner, the oral closure was reinforced by a buccal fat pad flap from the left buccal cavity. A 1 cm mucosal incision was made at the gingivobuccal groove above the left second molar tooth, and the parotid duct opening was safeguarded. The flap was advanced to the anterior palatal fistula from behind the molar and fixed with 5-O vicryl sutures. The patient was hospitalized for 3 days and advised to have liquid diet and oral prophylactic antibiotic for one week.

Results: At clinic follow up on day 10 post fistula repair, there was partial necrosis of the buccal flap, which was debrided surgically. Clinic follow up at on month post op showed residual 2 mm anterior palatal fistula, which healed completely at two months post op. Both the hypernasal speech and nasal regurgitation had resolved completely.

Summary/Conclusion: While the use of buccal flap in the repair of palatal fistula in post palatoplasty patient has been well documented, it is most frequently used in the paediatric population. We believe the partial necrosis of the buccal flap in our case could be due to our poor understanding of the adult dentition, especially the anatomical relationship of the buccal flap and the wisdom tooth, resulting in the inadvertent injury to the pedicle and the subsequent partial flap necrosis. To avoid similar complication, we adopted three preventive measures for patients receiving buccal flap operation:

1. careful siting of the buccal flap: to avoid passing the flap over the molars
2. post op mouth guard / bite blocking device: to prevent dental trauma to the pedicle
3. removal of wisdom tooth prior to the buccal flap operation if patient is of the adult age group

We hope that with these measures in place, similar complication of flap necrosis secondary to dental trauma can be avoided.
Incidence and factors of palatal fistula after primary palatoplasty as the medical cooperation in Madagascar for eight years
Y Tosa

1Plastic and Reconstructive Surgery, Showa University School of Medicine, Yokohama, Kanagawa, 2Plastic and Reconstructive Surgery, Showa University School of Medicine, Tokyo, Japan

**Background:** Cleft lip and palate are the most frequent anomalies among congenital malformations of the face. We have been conducting medical cooperation for cleft lip and palate in the Republic of Madagascar for eight years, from 2011 to 2018.

**Aims:** The purpose of this study is to investigate the incidence of fistula after primary palatoplasty in the Republic of Madagascar, which is a developing country. A further objective is to compare the group of cases in Madagascar with the group of cases in Japan to examine the presence or absence of increased incidence of fistula, and the factors that might be involved in the increase.

**Methods:** We conducted a survey of the age at time of surgery, cleft type and fistula incidence in 48 cases (30 males and 18 females). Age at the time of surgery was 11 months to 29 years (average: 7 years and 5 months), the cleft type was by the Veau classification (1 of class I, 15 of class II, 19 of class III, 13 of class IV). We used the modified two-flap palatoplasty for palatal closure. Besides the selection of the surgical technique, factors suggested to influence the fistula incidence include the experience of the surgeon, the extent of cleft.

**Results:** None of the 48 patients had complications resulting in fistula formation, and the incidence of postoperative fistula complications was 0%.

**Summary/Conclusion:** In medical cooperation in developing countries, there are factors in delayed wound healing, such as poor oral hygiene, low nutrition, and instability of the flap blood flow. Understanding such factors in surgery is important in order to avoid palatal fistula. We report that a palatal fistula incidence rate of 0% could be realized by avoiding those factors which contribute to fistula formation.
Nasal and upper lip harmony in cleft patients - a combined surgical approach
A Ujam¹, N Vig¹, N Nasser¹
¹Maxillofacial surgery, BARTS & THE ROYAL LONDON HOSPITAL, london, United Kingdom

Background: Secondary nasal deformity in bilateral cleft lip is one of the great surgical challenges. The problems are an under projected tip, an infra-tip lobule merging with the pro-labium and a short columella. Upper lip vermilion border deformity is a further significant problem.

Aims: A combined surgical approach that addresses both the lip and nasal deformity in this group of patients will be advantageous and is described.

Methods: We illustrate the combined use of the Abbe flap to reconstruct upper lip deformity with the concomitant use of pro-labial skin flap to correct the short columella and the under-projected nasal tip, commonly encountered in cleft lip patients. Critically, we demonstrate how complete release of the pro-labium from the lip improves nasal tip projection without inadvertently elevating the upper lip. We also describe the simultaneous use of 10th rib costochondral grafts to provide further enhancement of nasal tip projection.

Results: Our cases demonstrate that cleft patients with nasal deformity and poor upper lip aesthetics can be managed with a single operation. Lengthening of the columella by releasing the pro-labium and insertion of a 10th rib columellar strut graft can result in excellent nasal tip projection, while simultaneous improvement of upper lip deformity can be achieved by utilising either the Abbe Flap or a full thickness post-auricular Wolfe graft.

Summary/Conclusion: Re-establishing upper lip thickness, concavity, philtral lines, a smooth vermilion border and an aesthetically pleasing nasal profile are all crucial in achieving excellent outcomes in bilateral cleft patients. Our surgical techniques have produced predictable and most satisfying results.
Does microbial colonization of the cleft and nose affect alveolar bone grafting?

W Urbanova¹, M Kotova¹, E Leamerová², J Dvorakova³, F Prusik⁴, P Waldauf⁵
¹Department of orthodontics and cleft anomalies, Dental Clinic 3rd Medical Faculty Charles University Faculty Hospital Královské Vinohrady, ²Plastic Surgery Clinic, 3rd Medical Faculty Charles University Faculty Hospital Královské Vinohrady, ³Plastic Surgery Clinic, 3rd Medical Faculty Charles University Faculty Hospital Královské Vinohrady, ⁴Department of Laboratory Diagnostics, ⁵Department of Anaesthesiology and Resuscitation, 3rd Medical Faculty Charles University Faculty Hospital Královské Vinohrady, Prague, Czech Republic

Background: Treatment protocol of UCLP patients includes swabs before each surgery as standard. However the exact approach when and which swabs are collected before alveolar bone grafting (ABG) differs among the Cleft centres and their clinical impact in the scientific literature remains uncertain.

Aims: To determine aerob microbial colonization of the alveolar cleft and nose in patients before ABG and its impact on the preoperative and postoperative C-reactive protein levels (CRP), white blood cells count (WBC) and subsequent graft healing.

Methods: In 43 consecutive UCLP patients (♂29, ♀14, age 9.7 years +/-0.9) scheduled for ABG; swabs from the alveolar cleft and nose were taken before ABG and standard microbiological assessment was done. The CRP level and WBC count from blood tests were monitored before and twice in a first week after the surgery. Informed consent was obtained before the procedure. Bone graft healing was followed up for two years during clinical checkups. The linkage between the swab results, the inflammatory markers levels and subsequent graft healing was evaluated using Exact logistic regression and Linear mixed effect model analysis.

Results: In 23 (54%) patients in at least one of the swabs pathogenic microorganisms were detected, inside the alveolar cleft pathogens were found in 11 cases. Among the pathogens detected in the cleft were Staphylococcus aureus (no MRSA was detected) and Streptococcus pyogenes, and opportunistic pathogens as Proteus mirabilis, Enterobacter Cloacae, Pseudomonas aeruginosa. Candida albicans was identified in 13 cleft swabs, in none of the nose swabs. Complications in graft healing (e. g. sequestration or wound dehiscence) occurred in 11 patients (26%), repeated ABG was necessary in 6 out of them (14%). The CRP and WBC increased in the first days after surgery in all patients, however no statistically significant difference of the inflammatory markers levels before and in the first week after surgery and subsequent graft healing was found between patients with pathogens or opportunistic pathogens in the cleft/nose swabs. Presence of the Candida in the cleft had no influence on the inflammatory markers and graft healing.

Summary/Conclusion: Swabs are useful to monitor general health of patients before surgery, however direct impact of the cleft/nose swabs results on the bone graft healing was not proved. Inflammatory markers levels increased after surgery as expected, but the increase did not differ between patients with and without pathogens/candida in cleft/nose swabs. Relatively high complications rate of the ABG was found in UCLP patients and further research is necessary to identify the reason for poor graft healing in some patients (e.g. anaerob microbiota in the cleft, cleft size, soft tissues distress, ABG technique and others).
Background: Bilateral cleft lip and palate /BCLP/ is one of the most complicated oronasal defects. The methods and timing of consecutive operations are discussed worldwide. The number one task for the surgeon is anterior and upper position of a premaxilla, which should be overlayed by the orbicularis oris muscle during the lip closure. We had a group of 5 children with BCLP with an excessive vertical growth of premaxilla. The lip closure was provided at the age 3 to 8 months with no preoperative alveolar molding and the palatal closure at the age 11-12 months. The average number of operations until the premaxilla reposition was 4, including lip and nose corrections, vestibulum correction and insertion of gromets. The lip closure was done using Veau technique, where the orbicularis oris muscle was sutured on both sides and it covered from the front the whole volume of premaxilla as a molding system. The pressure of muscle induced premaxilla reposition, but we also observed the excessive vertical development with a large overbite.

Aims: In the study we evaluated two approaches for a premaxilla stabilization. The evaluation was done preoperatively and one year after the operation using wax bite register, models and photographic documentation.

Methods: Premaxilla reposition was performed at the age 9-13 years depending on the eruption of upper permanent canine root. Preoperatively we indicated PAN, intraoral CT and presurgical setup on study models. The evaluation of the intermaxillary relationship was performed using wax bite register. The operation included transversal osteotomy of vomer in the place where it links to the premaxilla. It also included a reposition of the premaxilla upwards. We divided patients into two groups according to the type of fixation. In the first group the fixation was done only by an orthodontic appliance and a simultaneous bone graft harvested from the iliac crest. In the second group we provided ORIF /open reduced internal fixation/ with titan screws and miniplates together with a xenogenic augmentation material /bone and membrane/. We combined it with an orthodontic appliance for 6-8 weeks. Both groups were followed by orthodontic treatment with fixed devices.

Results: We observed a better stabilization and a smaller percentage of relapse in the second group. We explain it by using permanent internal fixation which remains in situ throughout the life.

Summary/Conclusion: In BCLP patients, the lip closure with thorough suture of orbicularis oris muscle works as a molding system for the premaxilla reposition. In some patients the premaxilla grows excessively in a vertical plane. These patients are indicated for a premaxilla reposition with ORIF in combination with xenogenic augmentation materials completed by an orthodontic appliance. This method represents better stabilization of the premaxilla and prevents from the relapse.
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*Surgical part of an interdisciplinary evaluation of 30 consecutively treated patients with unilateral cleft lip (UCLP) at age 40 years. Surgical procedures and results are presented*

H Vindenes¹, Å Sivertsen¹, H Sylvester-Jensen¹, P Sæle¹, N Pedersen¹

¹Cleft Lip and palate team Bergen, Haukeland University Hospital, Bergen, Norway

**Background:** This is a quality assurance project carried out by the Bergen Cleft team in Norway. The surgical procedure were carried out by the same surgeon at 3 months and 2 years. The Pushback procedure was applied for palate closure.

**Aims:** The aim is to meet the patients born between 1973 and 1979 at the age of 40 years.

**Methods:** 30 consecutively treated patients with UCLP were invited. 21 patients showed up to the examination. Records from birth to the examination at 40 years of age were investigated. All forms of surgery to the face and CLP area were registered.

**Results:** Two patients did not need bonegrafting, however 1 patient had to have 2 bonegrafting procedures. Seven patients had LeFort I and three of these were in combination with mandibular set back. Four patients had pharyngoplasty. 3 patients had no need for secondary nose/lip correction. However, 11 had one-, 4 had two-, 2 had three- and 1 had four secondary nose/lip operations.

**Summary/Conclusion:** The results show us the importance of evaluating adults and looking back at earlier treatment procedures.
Treacher collins syndrome with unilateral incomplete cleft lip: an evidence based case report
D Wicaksono¹, P Kreshanti¹, S Handayani¹
¹Plastic Reconstructive and Aesthetic Surgery, Ciptomangunkusumo Hospital, Faculty of Medicine Universitas Indonesia, Jakarta, Indonesia

Background: Treacher Collins Syndrome (TCS) is an autosomal dominant disorder of craniofacial development with variable clinical forms, occurs with an incidence of 1 in 50,000 live births. TCS occurs as an autosomal dominant disorder, 60% of cases arise as sporadic mutations and 40% of cases may be inherited from the parents. TCS has a full clinical presentation, characterized by hypoplasia/aplasia of the body and arch of the zygoma, a significantly increased facial convexity, mandibular hypoplasia, a retrusive chin with increased vertical height, cleft palate, and external ear anomalies.

Aims: To present a rare case of patient with TCS and unilateral incomplete cleft lip.

Methods: This is an Evidence Based Case Report. Literature review and searching were performed. The Cochrane Library, PubMed, Scopus and EBSCO databases were searched for English language publications, including abstracts. The search was performed using the following terms: “Treacher Collins Syndrome” AND “cleft lip”. We also manually searched for relevant articles from the reference lists of the retrieved articles.

Results: Based on literature review and searching, we reviewed 7 varied cases of TCS. There was no reported TCS with cleft lip case. The present case has shown the positive family history with variable clinical form of TCS from mother’s side and cleft lip from father’s side. His mother, aunt and grandmother had variable clinical forms of TCS without cleft lip, but his father had incomplete cleft lip. The patient was undergone labioplasty as the first stage of definitive treatment and planned to have another surgery in the future.

Summary/Conclusion: TCS with cleft lip is a very rare case. Other clinical features are unilateral cleft palate, downward slanting of palpebral fissures, type 3 microtia on right ear and type 2 microtia on left ear and micrognathia. The patient has shown the positive family history with variable clinical form of TCS from mother’s side and cleft lip from father’s side.
Development nursing practice guideline for care of patients with cleft lip/palate in Srinagarind hospital: antenatal care unit

J. Wongkham 1, S. Pradubwong 1, P. Chatvised 1, T. Ratanasiri 2
1Nursing Division, 2Department of Obstetrics and Gynecology, Khon KAEN University, Khon KAEN, Thailand

Background: Medical knowledge and new technology can be used to detect abnormal condition of infants who have cleft lip and cleft palate since 4-5 months.

Aims: To develop nursing practice guideline for care of pregnant women with fetal cleft lip and palate.

Methods: The study design by evidence-based practice. Researchers use empirical evidence of South nursing practice method as a guideline for develop the plan which divided into 4 phases; 1) discovering the problem, 2) searching for empirical evidence, 3) develop nursing practice and trial applying, and 4) apply the adjusted plan to organization. This study starts from the second phase to third phase. A total of 10 researches including 2 systematic review, 3 prospective studies, 2 retrospective studies, and 3 qualitative studies include were used to develop this nursing practice guideline.

Results: The result of this study consists of 3 main topics; 1) the evaluation of psychological and support requirements, 2) giving consulting and information, and 3) mental nursing care and use the result of the study to develop a Development Nursing Practice Guideline for care of Pregnant women with Fetal cleft lip and Palate Srinagarind Hospital after 5 experts examined and adjusted the content.

Summary/Conclusion: Nursing practice using for take care of pregnant with fetal cleft lip/palate consists of 1) knowledge evaluation of pregnancy women 2) evaluation of information need 3) providing knowledge 4) take care with understanding of their feeling. Before apply this Nursing Practice Guideline, it should be evaluated both process and outcome. The implementation of the CNPG should be adjusted for the appropriateness for using in Unit. In addition, the evaluation of the anxiety of pregnant women before and after the implementation of this nursing practice Guideline has been continuously improved.
Background: Bleeding after palatoplasty is a surgical complication which may require return to theatre in the first few postoperative hours. Its most common causes are vessel injury or suture breakdown.

Aims: To focus attention on unusual causes of immediate postoperative bleeding after palatoplasty requiring return to theatre and discuss the management of such complications.

Methods: We report a case of immediate postoperative bleeding which mandated return to theatre and examination under general anaesthesia. The Patient was a six months old boy, weighing 7 kilos, with unilateral cleft lip and alveolus and cleft of the soft palate with intact hard palate. Blood tests, including clotting, were unremarkable. Tranexamic acid was routinely given at the induction at a dosage of 10mg/kg. The Patient underwent a Millard-Tennison lip repair and a Sommerlad palate repair in one surgery. No intraoperative bleeding was observed. After the operation, throat pack was removed and trans nasal suction was performed before cuffed endo tracheal tube removal.

Results: In the recovery room the Patient presented persistent bleeding from the mouth. After a second dose of Tranexamic acid 10mg/kg, the Patient was put under general anaesthesia. The lip and the palate repair appeared intact. Blood was coming from the rhynoparinx. On examination, a mass protruding from the nasal side of the reconstructed soft palate was observed. A tissue sample was sent to histopathology. The bleeding eventually stopped, but haemoglobin dropped to 6g/dl requiring haemotranfusion. The Patient was sent to Intensive Care and a contrast CT scan showed a mass in the rhinopharynx which was then removed by the ENT doctors. Histopathology showed an angiomatoid polyp.

Summary/Conclusion: Polyps of the rhinopharynx have been described in association with cleft lip and palate in some syndromes. The Patient described here has no demonstrated syndrome to date. We believe that the pedunculated angiomatoid polypoid mass was sitting in the rhinopharynx of the baby and pushed down by the suction tube at the end of the operation. Although postoperative routine trans nasal suction is advocated to prevent aspiration, it may push down pre existent rhinopharyngeal masses which can cause bleeding and obstruction. We then advice a routine thorough examination after postop trans nasal suction and before extubation to prevent life threatening postop complications.
2. Airway and Feeding Management

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The importance of feeding specialists in the treatment of pierre robin patients with the pre epiglottic baton plate (tuebingen plate)
K Micheletti\textsuperscript{1}, M Meazzini\textsuperscript{1}, L Autelitano\textsuperscript{1}, F Mazzoleni\textsuperscript{1}, S Colombini\textsuperscript{1}
\textsuperscript{1}Smile House, ASST Santi Paolo e Carlo, Milan, Italy

Background: Background: Pierre Robin Sequence involves micrognatia, cleft palate and glossoptosis, together with breathing disorders. In this study we present the different solutions we used to feed children affected by PRS after the positioning of a pre epiglottic baton plate or Tuebingen Plate (TP). Eight PRS children where treated with a TP in our unit.

Aims: Understand the importance of feeding guidance in patients undergoing TP (Tuebingen Plate) treatment

Methods: We present our feeding approach through a case report of an infant. The patient presented at the Smile House at two days. Birth weight was 3350 gr at birth, 3105 gr at discharge. The parents where trained to use a Medela Special Needs bottle in a semi prone position to ensure that the tongue was kept away from the airways and the infant could suck, swallow and breathe correctly. The patient gained 50g in 4 days, exclusively with breastmilk, 60/70ml, 7 times a day. After two episodes of respiratory distress a TP was prepared. After plate positioning, breathing improved, sternal and jugular introflections were eliminated and the tongue position was advanced, but feeding was significantly worsened. As the space between the plate and tongue was narrow, sucking was rendered more difficult and vomit reflexes were stimulated. Initial acceptance of the plate by the infant was difficult. The patient was admitted in the pediatric ward to monitor his feeding and breathing.

The first day we successfully fed the patient with 30/40ml per feeding, using the finger feeder directly in his mouth. At the very beginning, this was the only device accepted. In the following days, different devices were attempted, the mother was trained to exercise the tongue with pressures on the tongue itself and letting the child suck her finger and immediately after putting the teat in his mouth. This training turned out to be really useful and preparatory to teat suction. We then adopted Dr. Brown's specialty feeding and the hospital teat, which is softer and has a bigger hole. This way, the milk flow was faster and the baby began to drink more, reaching 400ml in 24hrs. IV Fluid was interrupted. After one week from admittance, the plate was activated to advance the positioning of the tongue during sucking. A fiberoptic exam confirmed that the positioning was correct.

Results: During the subsequent days, feeding improved significantly. At the time of discharge, reaching 500ml in 24 hours. In the meantime, the mother went on with pumping and maintaining exclusive breastmilk production.

Summary/Conclusion: TP are useful tools to improve breathing in PRS, but careful follow up by trained feeding specialists is fundamental for adaptation.
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**Postoperative complications and nutrition management after neonatal cleft lip repair**  
M Fiala¹²³, O Koskova¹²³, J Vokurkova¹²³, L Elstnerova⁴, P Rotschein¹²³  
¹Department of Pediatric Plastic Surgery - Department of Pediatric Surgery, Orthopedics and Traumatology, University Hospital Brno, ²Faculty of Medicine, Masaryk University, ³Department of Burns and Plastic Surgery, ⁴Department of Pediatrics, University Hospital Brno, Brno, Czech Republic

**Background:** In recent decade there has been a diversion from cleft lip repair in 3rd month of age and conversely, neonatal cleft lip surgery has become a routine procedure in the Czech Republic. Early cleft lip closure is considered controversial but sometimes criticized unsubstantially.

**Aims:** The aim of the study was to assess postoperative complications and nutrition management in newborns after primary cleft lip surgery at the University Hospital Brno.

**Methods:** Out of 210 patients who underwent neonatal cleft lip surgery (both unilateral and bilateral cleft lip/cleft lip and palate) between 2010 and 2015, 202 were included in the study. The remaining ones were excluded because of diagnosis of syndromes associated with orofacial clefts. Postoperative complications, as a wound dehiscence and infectious complications, and enteral feedings following surgery (including possibility of breast-feeding and need of nasogastric tube) were evaluated.

**Results:** A total of 93.6% of patients had no complications following neonatal cleft lip surgery. A wound dehiscence was observed in 2 patients (1% of patients), both with a wide bilateral cleft lip and palate. An incidence of neonatal infection requiring antibiotics was 2.5% (4 patients). Seven patients (3.5%) had a respiratory insufficiency rectified by applying oxygen mask (1.5%) or performing unplanned reintubation (2%). Severity of cleft was not directly related to respiratory and infectious complications. Enteral feeding was able to resume in 7.9 hours in average after the surgery and only 7.4% of children needed short-term nasogastric tube placement. While being released from hospitalization, 66% of children with isolated cleft lip were breastfed. The mean duration of hospital stay was 7.1 days.

**Summary/Conclusion:** This study shows that the neonatal cleft lip repair is a safe surgical method based on a rare occurrence of wound healing problems. A high percentage of breastfed children with cleft lip and, on the contrary, a low percentage of nasogastric tube usage indicates minimal impact on enteral feeding after neonatal surgery.
Car seats: a challenge for babies with robin sequence
H Robson¹, H McClements¹, D phare¹, L hall¹
¹Cleft Lip and Palate, Alder Hey Childrens Hospital, LIVERPOOL, United Kingdom

Background: In 1985 European law stipulated all children under 3 must travel in a secured child seat. For babies with Robin sequence this creates a difficulty as they may require lateral positioning to stabilize their airway. The majority of standard car seats do not accommodate this.

The development of our multi disciplinary algorithm for management of a robin baby stipulates a car seat challenge. This is to ensure Robin babies are safe during car journeys.

Aims: This has been challenged by maternity units, as there are no specific guidelines with regard to the safe transport of babies in car seats with Robin sequence.

Therefore this demonstrated a need for evidence based criteria for a robust policy for ensuring safe transportation of Robin babies.

Methods: A literature search to identify Robin sequence and car challenge test was performed.

Results: Evidence in the United Kingdom (UK) reports that babies are at risk of adverse cardiorespiratory events when placed in semi upright position. For babies with Robin sequence the risk is higher. There is a variance and no consensus to what is a car seat challenge and what the expected outcome would favour for safe transportation by car. The units have raised concerns that there is no strategy if the baby fails the car seat challenge, as it is not possible to adopt alternative positions which would be the advice for pre term babies.

Summary/Conclusion: Parents have sought alternative car seats which offer alternative positioning however this creates financial burden as they had invariably purchased a standard car seat.

This creates a paradigm of safe discharge or an increased length of stay in hospital until the baby can be nursed in an upright position. It is vital care givers provide with Robin sequence counsel parents about car seat safety. This presents a challenge to provide policy guidelines for the safe transport of Robin babies for both parents and health care providers. The lack of guidelines needs to be addressed and a safe policy needs to be drafted that would be accepted nationally.
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**Palatal morphology and upper airway structures in patients with cleft lip and palate**

V Nguyen\(^1\), T Jagomägi\(^2\)

\(^1\)Faculty of Odonto-Stomatology, Hue University of Medicine and Pharmacy, Hue, Viet Nam, \(^2\)Institute of Dentistry, University of Tartu, Tartu, Estonia

**Background:** Patients with cleft lip and palate usually have a smaller palate in different dimensions due to scarring of cleft surgery. They also had a smaller upper airway due to changes in the craniofacial morphology as well as cleft surgery.

**Aims:** This study aimed to evaluate palatal morphology and upper airway structures in patients with cleft lip and palate.

**Methods:** The research was approved by the ethics committee of Hue University of Medicine and Pharmacy. Informed consent was obtained from the patients or their parents if the patients < 18 years. Patients with cleft palate with or without cleft lip, no prior orthodontic and/or prosthodontic treatment were included. The cleft group included 17 patients (12 children and 5 adults). They were operated by a Korean charity operation team. The lip surgery was performed at age of 6–12 months using modified Millard or Tennison technique. The palate surgery was performed at age of 12–24 months using V-Y pushback technique. The control group included 34 participants (24 children and 10 adults). Digital dental models were obtained from the participants using the intraoral scanner TRIOS 3 Color Pod (3Shape, Denmark). Lateral cephalograms were obtained using Galileos (Dentsply Sirona, Germany). We investigated four features of the palatal morphology: palatal widths, palatal lengths, palatal depths, and palatal angles. We also studied four features of the upper airway structures: the pharynx, the soft palate, the tongue, and the hyoid bone.

**Results:** Children with clefts had significantly narrower palatal widths, shorter palatal lengths, and shallower palate. Adults with clefts had significantly narrower palatal widths, shorter palatal lengths, but not palatal depths and angles. Children with clefts had more anteriorly positioned hyoid bone, smaller inferior oropharyngeal airway space and retroglossal airway dimension, and shorter soft palate. Adults with clefts had more posteriorly positioned hyoid bone, and shorter soft palate.

**Summary/Conclusion:** The palatal dimensions were reduced in children with clefts in three dimensions. Adults with clefts had a similar size of the palate compared with the controls. The airway space was reduced in children with clefts but not in adults with clefts.
Clefts and interaction during feeding (clinng) early observation of feeding in children with a cleft lip and/or palate
B Van Gessel1, S Haverkamp2, C Guillaume1, I de Vries1, C Breugem3, S Duijff4
1Department of Pediatric Plastic Surgery, Wilhelmina Children’s Hospital, University Medical Center Utrecht, 2Department of Speech Therapy, Wilhelmina Children’s Hospital, University Medical Center Utrecht, 3Department of Pediatric Plastic Surgery, Wilhelmina Children's Hospital, University Medical Center Utrecht and Meander Medical Center, Amersfoort, 4Department of Pediatric Psychology & Social Work, Wilhelmina Children's Hospital, University Medical Center Utrecht, Utrecht, Netherlands

Background: Different studies indicate that children with a cleft lip and/or palate (CL/P) are at higher risk of developing feeding disorders. The feeding moment is one of the most essential moments of the day for a newborn baby, furthermore it is one of the first interactions between a child and its parents. If acquired appropriately it can be a pleasant and enjoyable experience for both the child and parent and strengthen the parent-child relationship. Feeding difficulties on the other hand can lead to stress, dysregulation and strain on the parent-child interaction.

Aims: The aim of this study is to increase understanding of potential difficulties with feeding that parents may encounter when feeding a 4-week old child with CL/P, hence enhancing insight in children at risk of developing feeding difficulties and signalling these possible difficulties at an earlier stage. Ultimately we aim to help clinicians involved in the care of cleft patients, in providing proper information and to the determination of adequate therapeutic strategies and carefully adjusted support.

Methods: This cross-sectional study was part of the longitudinal CLEFED study. We observed parent-child interaction during bottle feeding at the age of 4 weeks. During a home visit we videotaped one complete feeding moment. Also, parents were asked to fill in a questionnaire. We consecutively included 49 children with a CL/P who visited the Wilhelmina Children Hospital in Utrecht, the Netherlands. We registered growth, parent-child interaction and feeding problems, such as choking, gagging, nasal regurgitation and prolonged feeds, as well as subjective feeding problems. The interaction was scored by use of a specific tool, the Observatielijst van de Ouder/Kind Interactie(OOKI). Correlations were assessed by the linear-by-linear association and the Pearson chi-square test.

Results: Feeding difficulties, type of facial expression, and birth order correlated with specific components of the interaction between parent and child during feeding. The total score of the OOKI and the mean OOKI score per item, higher scores mean more positive interaction, showed significant positive correlations with the occurrence of feeding difficulties (Pearson = -.337, p = .018 and Pearson = -.328, p = .021).

Summary/Conclusion: More positive interaction was established when the child had more feeding difficulties and if the child was the first- or second born child in the family. Facial expression of the parent provided us with information about the interaction and could help us signal possible psychological difficulties of the parent. No differences in interaction were found across different types of clefts. Early observation of a feeding moment by a specialised team benefits signalling (potential) difficulties at an early stage to prevent further complications such as pressure ulcers and malnutrition. Our practical advice would be to observe one feeding moment within one week after birth, preferably in a home setting. Regular follow-up is necessary to monitor the child’s nutrition and growth.
3. Diagnostics and Imaging

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Palatal morphology in unilateral cleft lip and palate patients: association with infant cleft dimensions and timing of hard palate repair

S Botticelli12, A Kuseler12, K Mølsted3, S Nørholt45, P Cattaneo2, M Ovsenik6, T Pedersen24

1Cleft Lip and Palate Department, Institut for Kommunikation og Handicap, 2Section of Orthodontics-Department of Dentistry and Oral Health, University of Aarhus, Aarhus, 3Cleft Lip and Palate Center, Copenhagen University Hospital, Copenhagen, 4Department of Oral and Maxillofacial Surgery, Aarhus University Hospital, 5Section of Oral Surgery and Oral Pathology-Department of Dentistry and Oral Health, University of Aarhus, Aarhus, Denmark, 6Department of Orthodontics and Dentofacial Orthopedics, University of Ljubljana, Ljubljana, Slovenia

Background: Size and shape of the palatal vault differ in subjects born with cleft lip and palate when compared to non-cleft individuals, due to the initial palatal defect and the scars following the surgical repair. The interest of the cleft team in the anatomy and morphology of the palatal vault refers to the hypothesis that it may have an impact on articulation in speech, oral function in general and biomechanics and stability of the orthodontic expansion.

Aims: 1) to describe palatal dimensions and morphology in children born with UCLP, randomized for early versus delayed hard palate closure, in relation to non-cleft controls; 2) to introduce a novel 3D morphometric analysis of palatal size and shape; 3) to establish if cleft size at infancy has an influence on palatal dimensions.

Setting: Tertiary healthcare. One surgical center. Age, gender, ethnicity matched controls.

Design: single center-controlled subgroup analysis within an RCT of primary surgery (Scandcleft)

Methods: A total of 122 infants born with unilateral cleft lip and palate, operated in one surgical center, were included in multicenter RCT of primary palatoplasty and were randomized for early (at 12 months of age) versus delayed (at 36 months of age) hard palate closure. Linear measurements of palatal height and width, in a reference coordinate system, were performed on 116 digital dental models (mean age 8.2 years) and compared to a control group of non-cleft individuals (28 subjects). Cleft dimensions at infancy (mean age 1.1 months) were considered in the analysis in a linear regression model. Thereafter, a novel 3D method for analysis of morphological differences was applied and the mesh to mesh distances between 3D cleft palatal objects and control objects visualized with color mapping. Differences between groups were assessed.

Results: UCLP subjects presented with a palate higher than the controls at the level of the anterior scar (P=.002), but generally lower in the middle region (P<.001). The group who received delayed hard palate closure presented a flatter palate posteriorly (P=.048) in relation to the group where the hard palate was closed earlier. Reduced transversal dimensions were evident in both UCLP groups but more in the group who received earlier hard palate closure (P= .003 and .031). The morphological analysis in 3D better depicted the anatomical complexity, revealing that, after delayed hard palate closure, the central palate is shallower in the middle and posteriorly.

A significant correlation was found between anterior cleft size at infancy and palatal height at different levels (P=.029; .010).

Summary/Conclusion: a UCLP palate significantly differs from a non-cleft control in width and height. Delayed hard palate closure may represent an advantage for the transversal dimension, but a disadvantage for palatal height. Infant cleft dimensions at birth partially explain differences in palatal height.
Three-dimensional facial development in infants with unilateral cleft lip and palate

S Brons¹, J Meulstee², T Loonen², R Nada³, M Kuijpers¹, E Bronkhorst⁴, S Bergé⁵, T Maal², A Kuijpers-Jagtman¹

¹Orthodontics and Craniofacial Biology, ²Radboudumc 3D Lab, Radboud University Medical Centre, Nijmegen, Netherlands, ³Faculty of Dentistry, Kuwait University, Kuwait City, Kuwait, ⁴Preventive and Curative Dentistry, ⁵Oral and Maxillofacial Surgery, Radboud University Medical Centre, Nijmegen, Netherlands

Background: Stereophotogrammetry has the potential to serve as a method for early detection of facial growth disturbances related to certain surgical treatments as well as soft-tissue changes related to presurgical nasoalveolar molding. Comparison of the facial morphology of individuals with craniofacial malformations and normal controls can be achieved by registration of average faces of each group. This enables early identification of treatment protocols that are potentially detrimental for the development of normal facial morphology and can be subsequently abandoned earlier. To our knowledge, there is no published report on facial growth and treatment outcome in infants with orofacial clefts and controls matched for age during the first year of life using stereophotogrammetry and superimposition with the use of a generic mesh and average faces.

Aims: This study aimed to compare the three-dimensional facial morphology of infants born with unilateral cleft lip and palate with an age-matched normative three-dimensional average face before and after primary closure of the lip and soft palate.

Methods: Thirty infants with a non-syndromic complete unilateral cleft lip, alveolus, and palate participated in the study. Three-dimensional images were acquired at 3, 6, 9, and 12 months of age. All subjects were treated according to the primary surgical protocol consisting of surgical closure of the lip and the soft palate at 6 months of age. Three-dimensional images of patients with unilateral cleft lip and palate at 3, 6 (pre-treatment), 9, and 12 months of age were superimposed on normative datasets of average facial morphology using the children's reference frame. Distance maps of the complete three-dimensional facial surface and the nose, upper lip, chin, forehead, and cheek regions were developed.

Results: Assessments of the facial morphology of patients with unilateral cleft lip and palate and control subjects by using color-distance maps showed large differences in the upper lip region at the location of the cleft defect and an asymmetry at the nostrils at 3 and 6 months of age. At 9 months of age, the labial symmetry was completely restored although the tip of the nose towards the unaffected side showed some remnant asymmetry. At 12 months of age, the symmetry of the nose improved, with only some remnant asymmetry noted on both sides of the nasal ridge. At all ages, the mandibular and chin regions of patients with unilateral cleft lip and palate were 2.5-5 mm posterior to those in the average controls.

Summary/Conclusion: In patients with unilateral cleft lip and palate deviations from the normative average 3D facial morphology of age-matched control subjects existed for the upper lip, nose, and even the forehead before lip and soft palate closure was performed. Compared to the controls symmetry in the upper lip was restored, and the shape of the upper lip showed less variation after primary lip and soft palate closure. At this early age, retrusion of the soft-tissue mandible and chin, however, seems to be developing already.
Cone beam computed tomography comparative assessment of maxillofacial morphology in patients with bilateral cleft lip and palate

M Elkassaby¹, Y Elhoseiny², R Mostafa³, M Aboelfotouh³
¹Oral and Maxillofacial Surgery, ²Oral and Maxillofacial Radiology, Faculty of Dentistry, Ainshams University, ³Oral and Maxillofacial Radiology, Faculty of Dentistry, Ainshams University, Cairo, Egypt

Background: Bilateral cleft lip and palate (BCLP) patients have been described to have variable maxillofacial features as well as different response to presurgical infant orthopedics and lip repair. Difference in features suggest different underlying etiologic factors related to the clefting process. Validation of this hypothesis on the radiographic level is required for further elaboration of the defect.

Aims: The purpose of this study is to radiographically compare the maxillofacial morphology in the two previously suggested subtypes of BCLP patients using CBCT images from the cleft center archives.

Methods: CBCT scans of 22 BCLP (10 males and 12 females with mean age 9.6) were divided into two groups according to the SNA. Group P, showing well developed prominent premaxilla and SNA >80±2. Group R, showing ill developed rudimentary premaxilla and SNA <80±2. The relationship between the maxilla and the mandible (ANB), angle of septal deviation (ASD), and anterior upper facial height (AUFH) were assessed using Ondemand 3D software and compared between the two groups.

Results: There was a significant difference in ANB and ASD between the two groups, while the AUFH showed no statistically significant results.

Summary/Conclusion: The two subtypes of BCLP investigated in this study showed different maxillofacial features that can later affect treatment objectives and outcomes.
Comparison of three-dimensional and two-dimensional soft tissue measurements of patients with cleft lip and palate

S Alpagan Ozdemir¹, E Esenlik²
¹Orthodontics, Private Practice, ²Orthodontics, Akdeniz University, Antalya, Turkey

Background: Lateral cephalometric films have been reported to have certain limitations such as magnification or identification of the landmark problems especially in patients with facial asymmetries. Therefore non invasive three dimensional (3D) photographic method has become popular for better facial evaluation.

Aims: The aim of this study was to analyze the differences in facial soft tissue measurements between two-dimensional (2D) lateral cephalograms and 3D stereophotographs of patients with unilateral cleft lip and palate (UCLP), bilateral cleft lip and palate (BCLP), skeletal Class III and skeletal Class I malocclusions.

Methods: 3D stereophotogrammetric and 2D lateral cephalometric soft tissue measurements of facial height, lip lengths and convexity, mentolabial, and nasolabial angles of 158 patients (UCLP, n = 29; BCLP, n = 22; skeletal Class III malocclusion, n = 54; skeletal Class I malocclusion, n = 53) aged 8–32 years, were compared.

Results: The differences between 2D and 3D data for upper and lower facial heights, lip lengths, soft tissue convexity angle, nasolabial angle, and proportional measurements were significant (p<0.05). Upper facial height was lower in the 3D evaluation of all groups, whereas, the lower facial height values were higher in the 3D analysis of Class I malocclusion, UCLP, and BCLP groups. While most of the angular measurements were highly correlated in both techniques, the nasolabial angle was smaller in the 2D assessment of all groups.

Summary/Conclusion: Significant differences were found between 2D and 3D analysis of patients with cleft lip and palate, as well as patients with Class I and Class III malocclusions. Almost all linear measurements exhibited different results in 2D versus 3D analyzes while angular measurements were more similar between the two methods.
Differences in infraorbital foramen position in unilateral cleft lip and palate
A Kaye¹, S Tomur², M Tracy³
¹Plastic Surgery, Children’s Mercy Kansas City, ²UMKC School of Medicine, ³Plastic Surgery Research, Children’s Mercy Kansas City, Kansas City, Missouri, United States

Background: The maxillary division of the trigeminal nerve is directed along the floor of the orbital through a bony canal and exits through the infraorbital foramen (IOF). As the nerve exits it becomes the infraorbital nerve and supplies sensory innervation to the ipsilateral lower eyelid, cheek, nasal sidewall, upper lip, and oral mucosa. Knowledge of the presence and position of the infraorbital nerve is relevant for regional anesthesia of the face and surgical interventions on the face, jaw, and orbit. Children with cleft lip/palate have intrinsic maxillary differences on the cleft side that could potentially alter the anticipated location of the IOF. There is minimal research describing these differences in children with cleft lip and palate.

Aims: This study aims to investigate anatomic variations in infraorbital nerve positioning in children with unilateral cleft lip and palate.

Methods: A retrospective chart review identified a cohort of patients with unilateral complete cleft lip and palate who have undergone pre-operative work-up for alveolar bone grafting with 3D maxillofacial CT scans used for digital measurements.

Results: 17 patients were identified with adequate pre-operative 3D maxillofacial CT scans. Vertical and horizontal position measurements of the infraorbital foramina on the cleft and non-cleft side for each patient. Paired student t-test was used for statistical analysis. Mean value of the vertical height from the infraorbital rim to the IOF in the sagittal plane was 8.34±1.49mm on the cleft side and 9.28±1.62mm on the non-cleft side (p=.0001). An orthogonal horizontal measurement between the IOF and the vertical midline in the coronal plane revealed mean values of 24.3±1.53mm on the cleft side versus 26.13±1.48mm on the non-cleft side (p=.001). Differences in vertical maxillary height were seen with mean height of 35.30±4.19mm on the cleft side versus 36.00±4.27mm on the non-cleft side (p=.008).

Summary/Conclusion: Statistically significant differences were seen between the vertical and horizontal position of the IOF and overall maxillary height between cleft and non-cleft sides.
Prevalence of submucous cleft palate in children with initial diagnosis of isolated cleft lip

O Koskova\textsuperscript{1,2,3}, J Vokurkova\textsuperscript{1,2}, M Fiala\textsuperscript{1,2}, P Rotschein\textsuperscript{1}

\textsuperscript{1}Department of Pediatric Plastic Surgery - Department of Pediatric Surgery, Orthopedics and Traumatology, The University Hospital Brno, \textsuperscript{2}Faculty of Medicine, Masaryk University, \textsuperscript{3}Department of Burns and Plastic Surgery, The University Hospital Brno, Brno, Czech Republic

Background: Cleft lip with or without cleft palate (CL±P) is one of the most frequent types of orofacial congenital malformations, ranging from 3.4-22.9 per 10,000 births. On the other hand, submucous cleft palate (SMCP), in overt or occult form, belongs to one of the rarest forms of facial clefts. The incidence of CL +SMCP is not exactly known, however, the rate of SMCP is higher when a cleft of the alveolar ridge occurred with the cleft lip (CLA – cleft lip and alveolus).

Aims: A retrospective study was designed to determine the prevalence of overt and occult form of SMCP in patients with initial diagnosis cleft lip only who underwent cleft surgery at Department of Pediatric Plastic Surgery University Hospital Brno.

Methods: Out of 432 patients, registered for cleft lip ± palate with the cleft team of the University Hospital Brno between 2006 and 2018, 198 were diagnosed with isolated cleft lip. Twenty five children (12.6%) were included in the study because of associated submucous cleft palate. Following parameters were recorded and evaluated: patients’ age at the time of surgery, presence of classic submucous cleft palate triad (bifid uvula, a bony notch in the posterior hard palate, and a furrow along the midline of the soft palate – zona pellucida) and its correlations with Smyth’s grading system.

Results: 18 boys and 7 girls with submucous cleft palate and cleft lip were included in the study. The mean age of the patients at the time of palatal surgery was 3.3 years (min-max 0.5-6.8yr). Fifteen patients (60.0%) have been diagnosed as overt form of SMCP, the remaining ones as occult form of SMCP. All patients diagnosed as occult form of SMCP correspond to clinical grade I of Smyth’s grading system. 68.0 % of patients has alveolar ridge affected by cleft and the finding of CLA wasn’t in correlation with overt or occult form of SMCP (p=0.14). The patients with occult form of SMCP underwent a surgery in later age then patients with overt form of SMCP, respectively, at age of median value 2.3 years for overt form and 5.5 for occult form (p <0.05).

Summary/Conclusion: Cleft lip associated with submucous cleft palate (in overt or occult form) occurs in the prevalence of 12.6%. All patients with cleft lip ± alveolus should have been examined for soft palate on a regular basis.
Attempt of intraoperative simulation for le fort i osteotomy with hololens
D Mitsuno¹, K Ueda¹, Y Hirota¹, H Kino¹, T Okamoto¹, C Umeda¹
¹Dept. of Plastic and Reconstructive Surgery, Osaka Medical College, Takatsuki city, Osaka, Japan

Background: The technology for adding various information to real visual field is defined as Augmented Reality (AR) technology, and application studies have flourished in recent years also in the medical field. By superimposing the hologram created from inspection images or simulation image created by processing them on the surgical field, intraoperative judgment becomes easy. Various AR devices are already available on the market because of technological innovation and low pricing.

Aims: In this study, toward the practical application of intraoperative bone / soft tissue movement simulation by AR technology, we attempted intraoperative simulation for Le Fort I osteotomy with AR device HoloLens (Microsoft Corp., Redmond, Wash.).

Methods: Inspection images were obtained with CT and VECTRA H1 (a hand-held 3D imaging system). 3D images of facial bone / skin surface were created using 3D slicer (a free and open-source software package for image analysis and scientific visualization). The 3D images were further exported to Blender (a professional free and open-source 3D computer graphics software), and data processing such as bone division / setting of bone fragment moving direction etc. was performed. Furthermore, the data was installed on HoloLens after setting the moving amount of the bone fragment arbitrarily adjustable. When performing a surgical procedure, we mounted HoloLens, and started the AR application. The bone movement simulation was performed with the 3D image aligned to the body surface.

Results: Case 1: male, 19 years of age, maxillary dysplasia (bilateral cleft lip and palate)
The image of Le Fort I osteotomy was superimposed onto the surgical field. It was useful for checking the planned amount of bone movement.
Case 2: male, 18 years of age, maxillary dysplasia (bilateral cleft lip and palate)
The Simulation image in which skin also moves in conjunction with bone was created and then superimposed onto the surgical field. It was useful for checking the planned amount of bone movement and facial change.

Summary/Conclusion: Ming et al. reported that they have displayed the osteotomy data simulated preoperatively onto the surgical field by AR-based navigation system (Ming Z, Gang C, Li L, et al. Effectiveness of a novel augmented reality-based navigation system in treatment of orbital hypertelorism. Ann Plast Surg. 2015;77:662–668.). On the other hand, we made it possible to adjust the moving amount of bone fragment even during the operation, and furthermore the image of the skin can be deformed in conjunction with bone movement. It is significant that those were possible with existing AR device and free software.

In deformation simulation of soft body such as skin, in general, it is necessary to use a complicated calculation method (such as finite element method) that takes into consideration the interaction of adjacent tissues. In this study, the movement of skin was made parallel to the movement of bone fragment, and the coefficients of the movement amount at each point were adjusted. Thereby it was possible to perform skin deform simulation, which is makeshift method but has less discomfort in appearance. The future task is to measure and analyze the actual "amount of skin movement per part with respect to the amount of bone movement" based on the CT / VECTRA postoperative follow-up, and feedback it to the "more natural skin movement coefficient" for Le Fort I osteotomy.
Three-dimensional analysis of rare facial cleft and association with hard tissue and soft tissue

P Moura 1, R Zechi-Ceide 1, A Porto Peixoto 2, C Tonello 3, A Richieri-Costa 1, D Garib 2

1 Clinical Genetics, 2 department of orthodontics, 3 department of craniofacial surgery, Hospital for Rehabilitation of Craniofacial Anomalies, University of Sao Paulo, Bauru, Sao Paulo, Brazil, Bauru, Brazil

Background: Rare facial clefts affect different structures of the face and can extend to the skull. They are classified numerically from the location of the cleft in relation to the sagittal midline, in 16 distinct regions, and may refer to soft tissue and bone tissue or hard tissue (Tessier’s classification).

Aims: The aim of this study was to evaluate morphologically the craniofacial structures involved in individuals with complex facial clefts to better understand the pathogenetic mechanisms involved in the occurrence of these clefts, as well as to aid in the syndromic diagnosis, management and craniofacial reconstruction.

Methods: We included in this study radiological data of 15 patients from Hospital for Rehabilitation of Craniofacial Anomalies, University of Sao Paulo (HRCA/USP), Bauru, Sao Paulo, Brazil. The data were assessed by 3D craniofacial computed tomography (helical or cone beam) in the Mimics software program and the images were segmentation for morphological description of craniofacial alterations and comparison with cleft in hard tissue and soft tissue.

Results: All patients had more than one type cleft (facial/orofacial or cranial). Tessier clefts in six cases were not associated with other anomalies. Other 9 cases had a known condition associated: three cases presente amniotic band sequence, three oculoauriculovertebral spectrum, one frontonasal dysplasia, one pai syndrome and one Treacher Collins syndrome. Different combinations of rare facial clefts was observed in all cases studied, but none pattern can be established, reflecting the diagnostic heterogeneity of this group. The complex cleft was observed in both soft and hard tissue, but they did not show the same severity.

Summary/Conclusion: The morphological alterations observed of soft tissue and bone tissue are fundamental importance for the planning of craniofacial rehabilitation and treatment and due to the complexity of the cases and absence of a pattern of malformation in patients evaluated in this study suggested that 3D images of hard tissue and soft tissue should be evaluated to contribute in diagnosis of complex rare clefts and associated of syndromes. The better characterization of type by Tessier clefts might to aid in understanding the pathogenesis and etiology of rare facial clefts.
On the role of the frontal projection in videoradiography of velopharynx in decision-making for a velopharyngeal flap plasty in patients with cleft palate

F Appelros¹, M Stiernman¹, M Becker¹, H Salé², H Svensson¹
¹Department of Plastic and Reconstructive Surgery in Malmö, Sweden, ²Department of Diagnostic Radiology, Clinical Sciences in Malmö, Malmö, Sweden

Background: Despite successful primary surgery, patients with cleft palate (CP) and cleft lip and palate (CLP) may experience velopharyngeal insufficiency and hypernasal speech. To assess this, a perceptual speech analysis by a speech and language pathologist is a first step. The next step is a videoradiographic investigation with a lateral and a frontal projection. In selected cases nasoendoscopy may be performed. Depending on the findings, a velopharyngeal flap may be performed with the aim of reducing the hypernasality.

Aims: The aim of this study was to investigate whether the frontal projection on videoradiography plays a role in the decision-making about velopharyngeal flap surgery, or the width and orientation of the flap. A secondary aim was to evaluate the effect of the flap in reducing hypernasality.

Methods: Between 2007 and 2016, 75 patients had received a flap at our department. During the same period of time, 41 patients who had undergone videoradiography did not receive a flap. Medical records, particularly regarding speech assessments, videoradiography statements and operating records, were scrutinised to seek information about the factors leading up to the decision about whether or not to performing a flap.

Results: The median age at surgery was six years. Only a slight agreement was found between pre-operative speech assessment and videoradiography. Regarding the risk of hypernasality and the risk for getting a flap, no significant differences were found between CP and CLP, nor between girls and boys. Hypernasality was reduced by flap surgery in 97% of the patients. For all patients the information from speech analysis and the lateral projection were essential for the decision on whether to perform flap surgery or not.

Summary/Conclusion: The frontal projection on videoradiography does not play a substantial role in the decision-making regarding performing a flap or the design of the flap. The flap reduces hypernasality efficiently.
Upper airway dimensions and respiratory flow simulation in individuals with cleft palate and sleep apnea.

Background: Individuals with cleft lip and palate (CLP) have an increased risk of developing obstructive sleep apnea (OSA) and one of the potential contributing factors is the reduction of the pharyngeal dimensions, commonly observed in this population.

Aims: This study aimed at investigating the occurrence of OSA in individuals with CLP by means of polysomnography and to correlate these findings with the upper airway dimensions and respiratory flow simulation data, using cone-beam computed tomography (CBCT) and computational fluid dynamics (CFD).

Methods: Twenty-seven young adults with non-syndromic unilateral CLP (20-29y) were prospectively evaluated by means of nocturnal polysomnography (OSA = apnea-hypopnea index ≥5 events/h). Upper airway dimensions (volume [V] and minimum pharyngeal cross-sectional area [mCSA]) were assessed on CBCT-generated 3D models (Mimics Research 17.0, Materialise Medical, Leuven, Belgium). Inspiratory airflow simulations were done by the CFD technique (ICEM-CFD 14.5.7, Fluent 14.5, FieldView, ANSYS, Canonsburg, PA and Intelligent Light, Lyndhurst, NJ) and the mean and maximum inspiratory pressures and the resistance to respiratory flow were determined.

Results: Polysomnographic data showed that 26% of the subjects had OSA (AIH: 12±8, min:6/max:27). Subjects with OSA had significantly smaller upper airway volume (36±5cm³) than subjects without OSA (43±9cm³). Although not statistically significant, mCSA of OSA individuals were numerically reduced (94±19mm²) when compared to non-OSA subjects (145±84mm²). Even though variables evaluated by the CFD technique did not differ between subjects with and without OSA, a tendency for more negative inspiratory pressures (OSA: 24±8Pa(ml/s), Non-OSA: 19±11Pa(ml/s)) and higher resistance (OSA: 0,133±0,043Pa(ml/s), Non-OSA: 0,111±0,052Pa(ml/s)) was observed in subjects with OSA.

Summary/Conclusion: The results have shown that young adults with repaired cleft lip and palate are at risk for sleep apnea. The reduced airway volumes observed may lead to more negative inspiratory pressures and higher upper airway resistance to airflow, increasing the chance for pharyngeal collapse and predisposing these individuals to obstructive sleep apnea.
Background: The surgical reconstruction of the lip and palate, which aims at restoring the shape and function of the structures, paradoxically impairs maxillary and nasal growth, leading to a class III skeletal discrepancy and reduced nasal patency.

Aims: The objective of this study was to three-dimensionally evaluate the internal nasal dimensions of young adults with cleft lip and palate (CLP) by means of cone-beam computed tomography (CBCT).

Methods: This was a cross-sectional prospective study. Fourty-five subjects were enrolled on this study and divided into 3 groups, as follows: 1) Control group (CON): 15 CBCT scans of individuals without CLP, 2) Unilateral Cleft group (UCLP): 15 CBCT scans of subjects with complete unilateral FLp, 3) Bilateral Cleft Group (BCLP): 15 CBCT scans of individuals with complete bilateral CLP. CBCT-generated nasal 3D models were obtained using Dolphin Imaging 11.8 software. Two trained examiners assessed the images. Significant differences among groups were evaluated using ANOVA and Tukey’s test. (p<0.05).

Results: The mean nasal volumes (± SD) of the CON group corresponded to 18.1 ± 3.8cm$^3$. In the UCLP and BCLP groups, the values were smaller and corresponded to 14.7 ± 2.2 cm$^3$ and 17.1 ± 2.2 cm$^3$, respectively. A significantly smaller volume was observed for the UCLP group in relation to CON group (p = 0.006). No significant differences were observed between BCLP and CON or between UCLP and BCLP.

Summary/Conclusion: The nasal volumes of individuals with complete cleft lip and palate, especially those with unilateral clefts, are dimensionally smaller than that of individuals without CLP. This fact indicates the impact of the cleft per se on the internal nasal geometry, probably leading to a reduced nasal patency in this population.
4. Genetics

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Prevalence of consanguineous marriages between parents of individuals with cleft lip and/or palate
C Silva¹, T Queiroz¹, M Pereira¹, J Mateo-Castillo¹, A Gonçales¹, L Neves¹
¹Department of Biological Sciences, Hospital for Rehabilitation of Craniofacial Anomalies (HRAC), University of São Paulo, Bauru, São Paulo, Brazil

Background: Cleft lip and palate are the most common congenital craniofacial deformities seen in children and present a complex etiology involving genetic and environmental factors. The occurrence of orofacial clefts in the world varies with frequency of 1-2.2/1000 live births, and in Brazil it is around 1 for 650 live births. Environmental factors related to orofacial cleft are associated with maternal health and habits during pregnancy. Regarding genetic/hereditary factors, the issue of familial recurrence and consanguineous marriages are highlighted. Consanguineous marriage is traditional and respected in same communities of North Africa and West Asia. In Brazil, the consanguineous union are heterogeneous and there are few studies verifying the relation between intra-familial union and the occurrence of the orofacial clefts.

Aims: The objective of this study was to verify the prevalence of consanguineous marriages between parents of subjects with non-syndromic cleft lip and/or palate and to investigate whether there could be an association between consanguineous unions and the severity of the cleft.

Methods: This study was funded by retrospective study analyzed 1179 medical records of patients with confirmed diagnosis of non-syndromic cleft lip and/or palate, treated in a Brazilian reference hospital. The information about consanguineous union between the parents and the type of cleft were obtained by analyzing the medical records.

Results: A total of 1179 medical records were evaluated and all contained the information regarding consanguineous marriages among parents. Of these, 30 cases of consanguinity were found between parents being that 19 patients were male and 11 were female. The degree of kinship that prevailed was consanguineous union between first degree cousins, with fourteen cases; followed by cousins of second degree, with nine cases; three cases between cousins of third degree; three cases where the degree of kinship was not informed; and a case between uncle with niece. The most frequent type of cleft was isolated cleft palate with 12 cases (40%); in second, cleft lip and palate with 10 cases; in third, isolated cleft lip with 8 cases.

Summary/Conclusion: It was observed a prevalence of 2.54% of consanguineous union among parents of subjects with non-syndromic cleft lip and palate. The kind of cleft that prevailed was isolated cleft palate and the male was the most affected with these orofacial anomalies. We didn't find a positive association among intra-familial union and the severity of the cleft. These results are within the expected rate of consanguineous marriages in this population studied. Financial Support: FAPESP.
Robin sequence, atypical face and intellectual disability in brazilian siblings
R Souza Sandri¹, M Rafacho¹, L Maximino², A Richieri da Costa¹, R Zechi Ceide¹
¹Clinical Genetics and Molecular Biology Section, Rehabilitation Hospital Craniofacial Anomalies, University of Sao Paulo, Bauru, Brazil, ²Department of Speech-Language Pathology and Audiology, Faculty of Dentistry, University of Sao Paulo, Bauru, Brazil

Background: The Pierre Robin sequence (RS) is a rare heterogeneous condition, characterized by the combination of micrognathia and glossoptosis, which can cause respiratory obstruction and severe eating difficulties. Palate cleft, variable feature and has often been incorporated into the definition. It occurs in isolation or associated with other anomalies, characterizing several syndromic conditions or not in a recognized standard such as a known syndrome or genetic condition, termed Robin Plus sequence (RS-plus). Here, we reported two Brazilian siblings, both male, with RS-plus with moderated intellectual disability and atypical face consisting of frontal hirsutism, low hair implantation, prominent thick eyebrows, narrow eyelid fissures, middle face hypoplastic, long and prominent filter, broad nasal bridge and base, hypoplastic nostrils and collumela, broad lobe ear, cleft palate. They also presented small hands, short digits, plantar hyperkeratosis, developmental delay and learning difficulties. Karyotype was normal. The evaluation of language showed an important oral and written language disorder, indicating cognitive deficit. The parents was phenotypically normal.

Aims: To investigate possible chromosome microdeletion or microduplication related to the phenotype of the siblings.

Methods: Microrearrangement analysis by MLPA in subtelomeric region (P036) and multiple microdeletion syndromes (P064).

Results: No change in the regions specific to syndrome with intellectual disability or subtelomeric region were found.

Summary/Conclusion: The same pattern of clinical findings observed in siblings with RS-plus suggest that this phenotype represent a new syndrome. The absence of changes in the subtelomeric regions, as well as in regions of known microdeletion/duplication syndromes, points to a probable genetic etiology. Additionally, the recurrence in male siblings and the presence of intellectual disability suggest that this is an autosomal recessive or X-linked condition. Additional molecular studies in collaboration with European group have being planned to clarify the etiology of this rare RS-plus condition.
Background: Epidermolysis Bullosa Congenita (EBC) is a rare hereditary disease which is characterized by fragility of the skin and mucous membranes. It has wide range of clinical manifestation due to the malfunction of different proteins of the skin caused by genetic mutations. The most severe form is called Epidermolysis Bullosa Dystrophica (EBD), which is characterized by blisters spontaneously occurring on the skin and mucous tissue. EBD is caused by mutations in the COL7A1 gene encoding collagen VII and it is inherited in autosomal recessive manner. Isolated cleft palate (ICP) may be caused by genetic or environmental factors and in some cases it is linked to certain syndromes. EBD linked to orofacial clefts is extremely rare.

Aims: Our aim was to provide an analysis of a family consisting of both parents and two children with COL7A1 gene mutation. The parents were carriers of EBD with no clinical manifestation of the disease. Their first born child was a daughter with postnatal clinical manifestation of EBD due to inherited recessive allele of the COL7A1 gene from both parents. Their second born child was a son with ICP with no clinical manifestation of EBD. He was carrier of EBD just like his parents.

Methods: Members of the family underwent genetic examination with DNA sequence analysis after the daughter was born. The method of DNA analysis was Sanger sequencing of the COL7A1 gene, which is now obsolete, but was considered as a gold standard at the time of testing. Prenatal diagnose of the son was executed by amniocentesis.

Results: The daughter was diagnosed with recessive type of EBD postnatally due to the clinical manifestation of the disease. The parents underwent genetic examination prior the next gravidity. They both were carriers of EBD with 25% chance of having a child with the disease. During the next pregnancy amniocentesis was performed and only paternal recessive allele was found, meaning that their son is a carrier of the disease. There was found no other prenatal pathology and the son was born with no clinical manifestation of EBD. However, after birth he was diagnosed with ICP. No correlation was found between the genetic mutation in COL7A1 and ICP. The surgery of the ICP was performed at 6 months of age at our department using one stage closure of hard and soft palate. A follow up with regular check-ups was established.

Summary/Conclusion: There was found no connection between EBD and ICP of the son. The cause of ICP was random with no familiality. Due to exhausting care of the daughter with EBD parents didn’t consider the ICP of the son as a “true disease.” From their point of view the ICP was just an obstacle they had to overcome.
Clinical and genealogical analysis of families with cleft lip and palate
T Sorokman

Pediatrics and medical genetics, BSMU, Tshernivtsi, Ukraine

Background: Cleft lip and palate (CLP) are birth defects that have multifactorial etiology, including genetic factors and environmental factors. Several genes (22 (TBX22), 1 (PVRL1), 6 (IRF6)), which causes syndromic CLP, have been described. The etiology of non-syndromic CLP is still largely unknown, mutations in candidate genes have been detected in a small proportion of cases. The determination of relative risk on the basis of genealogical analysis and environmental impact is relevant.

Aims: To determine the relative risk of the occurrence of CLP on the basis of genealogical analysis and the influences of environmental factors.

Methods: Clinical and genealogical analysis of the pregnancy course among families of 76 proband births with CLP were conducted. The following factors have been analyzed: mother's smoking, drinking alcohol, taking folic acid, steroids and anticonvulsants during pregnancy.

Results: The reproductive history of mothers who gave birth to a child with CLP showed that 42.1% of mothers had different pathological conditions of their pregnancy. In 18 cases (23.6%), CLP was detected. Such defects were only in native sibs. We can assume that in this situation there was an autosomal-recessive type of inheritance of this pathology observed in these cases. Among the relatives of the I-III degree of affinity, the CLP was transmitted by the parent in 12 cases, on the mother's line - in 6 cases. In the proband, who inherited CLP on the maternal line, CLP is registered with grandparents, mothers and daughters. We can assume that in this case there is an autosomal-dominant type of inheritance. The relative risk of developing CLP in a future baby in mothers who smoked during pregnancy was 1.4-1.6. Alcohol consumption by the mother (more than 5 servings a day) increased the risk of CLP occurrence by 3.4-4.7 times. In women who did not use folic acid and cobalamin in pregnancy, the risk of developing CLP increased threefold. The use of corticosteroids in the periconceptual period increased the risk of developing CLP by 3.1 times. According to the literature, anticonvulsants (phenytoin / hydantoin, diazepam, valproic acid) are clearly associated with an increased risk of birth defects. In our observations, only in cases of drugs valproic acid, the risk of CLP increased 3.4 times.

Summary/Conclusion: Women who have CLP in their families should undergo medical genetic counseling to determine the nature of the disease (hereditary, non-inherited, teratogenic). Research is promising that studies the relative risk associated with candidate genes and the interaction of the gene and the environment.
5. Psychology and Neurological Development

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Is cleft still a taboo? Beliefs and expectations of parents of cleft lip and palate patients: a study from an institute in north india
D Chattopadhyay¹, A Das², R Deol³
¹Burns and Plastic Surgery, ²Psychiatry, ³Nursing, AIIMS Rishikesh, Rishikesh, India

Background: Cleft lip and palate are a common but significantly disfiguring congenital anomaly affecting children. Besides the multiple problems they pose regarding the child’s feeding, speech and hearing, being visible they cause a major social stigma. Different degrees of parental guilt and shame are frequently encountered, primarily due to the perceived cause of the birth defect.

Aims: This study aims to define the parental perceptions associated with a child of cleft lip or palate and their social significance.

Methods: A detailed questionnaire was structured based on a previous study by Weatherly White (2000). The questions were focused to identify parental perceptions concerning the causation of clefts and belief systems that might be responsible for these perceptions. They were also asked about the degree of social interaction the child was permitted with schoolmates and other peers. A second set of questions were used to know about the parental expectation from the surgery.

Results: Parents of 63 patients were interviewed and the results tabulated.

Summary/Conclusion: The study identified many cultural and societal attitudes that deeply affect the way that communities treat children with clefts and other facial deformities. The results interestingly almost mirrors the last study similar to this, almost 20 years ago. It is surprising to note the beliefs and perceptions about clefts have remained largely unchanged.
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**Self-esteem of patients with cleft-lip cleft-palate attending the self-esteem enhancement program camp activities**

D Rod-Ong¹, H Samretdee¹, J Singkhornard¹, S Maneeganondh¹, A Theeyoung¹, N Patjanasootorn¹
¹Faculty of Medicine Khon Kaen University, Srinagarind Hospital, Khon Kaen, Thailand

**Background:** The patients with cleft lip and cleft palate [CLP] have physical problems in chewing and swallowing food as well as problems related to the intellectual level, living with others in the society, and low self-esteem. Tawanchai Cleft Center, therefore, organized a camp to improve the self-esteem of patients.

**Aims:** To study the result of the Self-Esteem Enhancement camp activities.

**Methods:** This is a descriptive study to evaluate the effects of the Self-Esteem Enhancement Program camp, which was scheduled on May 5, 2017. The subjects were divided into a group of 39 patients with cleft lip and palate and a group of 31 normal volunteers without CLP. The instrument included the questionnaire for general baseline data and the 10-item pre-test and post-test for self-esteem using the Thai version of Rosenberg Self-Esteem Scale [SES]. Data Analysis: The Paired t-test was used to compare the pre-test and post-test scores of the self-esteem scores. The correlation analysis was based on ANOVA.

**Results:** The patients with cleft lip and palate had self-esteem score not different from volunteer in pretest, but after joined the camp showed a significant increase of self-esteem scores in patients with cleft lip and palate compare to volunteers.

**Summary/Conclusion:** The sense of self-esteem of the patients with cleft lip and cleft palate did not differ from normal people. The camp activities could increase the level of self-esteem in both groups, and significantly in the patients with cleft lip and cleft palate.
Neuropsychomotor development of children aged 3 to 6 years with isolated pierre robin sequence
M Cavalheiro¹, R Zechi-Ceide¹, L Maximino²
¹Clinical Genetics and Molecular Biology, ²Faculdade de Odontologia de Bauru, Hospital for Rehabilitation in Craniofacial Anomalies (HRAC-USP), Bauru, Brazil

Background: Pierre Robin sequence is characterized by micrognathia, glossoptosis with or without, palate fissure, alone or in combination with other congenital anomalies or genetic syndromes. When it occurs alone it is called isolated Pierre Robin Sequence. The clinical phenotype is varied, but it is expressed mainly by airway obstruction and eating difficulties, which are more serious and frequent in the neonatal period. The risk factors for neuropsychomotor and language development are attributed to respiratory and alimentary difficulties in the first months of life, the time and frequency of hospitalization and the presence of cleft palate.


Methods: The study was approved by the ethics committee on human research of the institution and the consent and assent term was obtained. The sample consisted of 15 children attended at the Hospital for Rehabilitation in Craniofacial Anomalies (HRAC-USP), aged between 3 and 6 years, 10 girls and 5 boys, with genetic-clinical diagnosis of isolated Pierre Robin Sequence performed by the Clinical Genetics and Molecular Biology of quoted Hospital. The diagnostic criteria were to present the triad: micrognathia, glossoptosis and cleft palate, not associated with other congenital anomalies that may constitute syndromes, other sequences or associations. The children were evaluated through the Denver Developmental Screening Test II, which allows to verify the development in the Personal-social, Gross Motor, Language and Fine Motor-Adaptive areas. Children who presented otitis during the data collection period and who had a diagnosis of genetic syndromes or other malformations associated were excluded.

Results: Of the children evaluated, 60% (n = 9) failed the test and were considered at risk for child development and 40% (n = 6) passed the test. The most impaired ability was Language with 60% of children with delay, followed by 18.75% in Fine Motor-Adaptive. It is important to be noted that 12.5% (n = 2) of the children presented a delay in all the abilities contemplated by the instrument and one child presented impairments in language ability, however, it was not characterized as delayed child development according to the norms of analysis of the test manual.

Summary/Conclusion: Children with isolated Pierre Robin Sequence presented a risk for delayed neuropsychomotor development, being the language ability most impaired.
The effect of le fort i osteotomy on dental consonant in cleft patients

S Alaluusua, L Turunen, J Leikola, A Heliövaara

1Cleft palate and craniofacial, Department of plastic surgery, Helsinki University hospital, Helsinki, Finland

Background: Maxillary advancement may affect speech in cleft patients.

Aims: The aim of this study was to evaluate the effect of maxillary advancement on Finnish dental consonants /r/, /s/ and /l/ in patients with cleft lip and palate (CLP).

Methods: Fifty-nine Finnish-speaking nonsyndromic cleft patients (35 females, 24 males) with CP (n=12), UCLP (n=31) and BCLP (n=16) who underwent Le Fort I or bimaxillary osteotomies were evaluated retrospectively. The production of Finnish dental consonants /r/, /s/ and /l/ was assessed from pre- and postoperative standardized video recordings by two experienced speech pathologists. McNemar's test was used in statistical analyses. Kappa statistics were calculated to assess reliability.

Results: There was a significant improvement in articulation of Finnish /s/ and /l/ sounds after maxillary advancement. The preoperative mean percentage of /s/ errors was 34% and the postoperative 22%. /L/ was misarticulated preoperatively by 34% and postoperatively by 19% of the patients. R/ was misarticulated preoperatively by 47% and postoperatively by 42% of patients. Four patients had mild misarticulations in 1-3 sounds preoperatively, and after osteotomy the articulation was evaluated as being normal. The amount of mild articulation errors rose from 25% to 31% whilst severe articulation errors decreased from 37% to 25%. The reliabilities were good.

Summary/Conclusion: Maxillary advancement improved dental articulation of Finnish consonants /s/ and /l/ in cleft patients.
Background: In resource-poor countries, traditional models for delivery of speech therapy (i.e. intervention with a frequency of 2 sessions/week and an intensity of 21-30 minutes) are not adequate to reach all patients in need. In those countries, intensive speech therapy might be a solution.

Aims: Present study aimed to assess both short-term and long-term effectiveness of short and intensive speech therapy provided to patients born with cleft (lip and) palate (CL/P) in terms of articulation, resonance and satisfaction with cleft-related features.

Methods: Patients (n=5) who received individualized intensive speech therapy (i.e. 6 hours of speech therapy) were contacted in order to obtain long-term data. Finally, three patients agreed to participate in this study. Perceptual and instrumental speech evaluations were performed. Furthermore, a questionnaire (the Cleft Evaluation Profile), investigating patients' satisfaction with cleft-related features before and after speech therapy, was included in the assessments.

Results: Short-term post-treatment results for patient 1 showed mildly impaired speech understandability and acceptability, combined with resonance disorders. A decrease in active nasal fricatives was seen after speech therapy. On the long term, this patient presented with an increase in active nasal fricatives, which was reflected by decreased PCC scores. For patient 2, both short-term and long-term findings showed no resonance disorders resulting in normal speech intelligibility. Immediately after speech therapy, patient 3 still presented with severely impaired speech understandability and speech acceptability, combined with compensatory articulation errors (i.e. glottal stops, active nasal fricatives and pharyngeal articulation). On the long term, presence of glottal articulation (19.47%) and pharyngeal articulation (25.66%) was observed. Regarding self-perceived satisfaction, all patients showed satisfaction with speech after intensive speech therapy.

Summary/Conclusion: Present study investigated whether or not 6 hours of short, intensive speech therapy in Ugandan patients with CL/P, known in the CoRSU hospital, had (both short-term and long-term) benefits in terms of articulation, resonance and self-perceived satisfaction with cleft-related features. In summary, speech improvements after speech therapy varied among the three patients. Furthermore, findings suggested that some patients might have benefited from follow-up speech sessions. Further research investigating outcomes of intensive speech therapy in larger patient groups is necessary.
Speech score. A new concept
E Alvarez¹, D Alvarez¹
¹Plastic Surgery, D’artis Plastic Surgery Clinic, Latacunga, Ecuador

Background: Once a defined speech disorder has been identified, it is important to evaluate how it has improved or worsened after establishing a treatment or language therapy. At the moment of this work it is still a qualitative assessment that falls in the subjective to the explorer and for that reason it leads us to not solid conclusions to carry out coherent and definitive analyzes on the severity or not of the speech disorders. The Pittsburgh Scale, has allowed us parameters of quantitative analysis but focused only on the velopharyngeal function, leaving aside the disorders located at another level.

Aims: We propose in this work a simple, simple and complete method that adopts the quantitative modality that allows evaluating the effectiveness of primary, secondary or revision surgery, and the effectiveness of a language therapy.

Methods: this Speech scale was applied in isolation to the same patient by different professionals at different times and their results were compared. Qualification parameters were established for the emission quality of sounds at different levels, for example: NASOLABIAL LEVEL The emission of sounds like "MA-MA" when they were bad at this level with one, acceptable with two and we opt with 3 points PROVOKED NASOLABIAL LEVEL, The emission of sounds like "PA-PA" when they were bad at this level with one, acceptable with two and we opt with 3 points DENTAL PALATINE LEVEL The emission of sounds like "SU-SI" when they were bad at this level with one, acceptable with two and we opt for 3 points VELOPARYNGEAL LEVEL This level of sound modulation like CO CO when they were bad at this level with one, acceptable with two and we opt with 3 points. EFFORT TEST This simple test consists of inflating a balloon when they were bad at this level with one, acceptable with two and with 3 points,. It tests the integrity of all levels so far described working on all of them in full coordination.

Results: the most optimal score it will be 15/15, with this simple quick method, we can put a quantitative numerical score to something that was always qualitative, and had a very subjective appreciation. we finally have an integral assessment instrument of speech at different modulation levels, which has important value in pre-operative diagnosis of the cleft patient, and we also have a useful scale of measurement for postoperative improvement and language therapy evaluation applied to every patient.

Summary/Conclusion: Before this study, there was a quantitative and simple tool in the measurement of the functional result of a cleft patient. "The Alvarez Speech Score" allows us to measure the speech quality of a cleft patient, and at the same time it provides us with information about the effectiveness of surgery and speech therapy. This tool would also allow us to evaluate the technical quality of a surgeon, and a service in general. With this evaluation we would talk about the academic preparation that makes up Plastic Surgery Service and most importantly seek to reinforce this knowledge for the patient benefit. This score can be easily reproduced at all levels without the application of complex technologies. Once the score is tested by others, it could be an instrument that allows measuring and comparing the effectiveness of one technique with another.
Parents’ ratings of intelligibility in 3-year-olds with cleft lip and/or palate using the intelligibility in context scale: findings from the cleft collective cohort studies

M Seifert¹, Y Wren ², A Davies ², S McLeod³, S Baker¹
¹Bristol Speech and Language Therapy Research Unit, ²The Cleft Collective, University of Bristol, Bristol, United Kingdom, ³Charles Sturt University, Bathurst, Australia

Background: The International Consortium for Health Outcomes Measurement (ICHOM) Standard Set of outcome measurements for cleft lip and/or palate (CL/P) includes the Intelligibility in Context Scale (ICS) as a measure of speech outcomes following surgical repair of the palate. The ICS is a free parent-reported measure whereby intelligibility is rated on a five point scale (never, rarely, sometimes, usually, always) in seven different contexts (parents, immediate family, extended family, friends, acquaintances, teachers, strangers). Though the ICS has been validated as a screening tool for use with children with speech sound disorders who speak a range of languages, comparative data for children born with CL/P is not yet available.

Aims: The aim of this study was to provide preliminary data on performance on the ICS for a sample of 3-year-old, English speaking, children born with CL/P. These data will provide a source for comparison with clinical use of the ICS with this population.

Methods: Cross-sectional questionnaire data from the Cleft Collective Birth Cohort study, a national longitudinal clinical cohort study in the UK, was used. Children with orofacial cleft and their families are recruited (providing the mother gave informed consent) at pre/post-natal diagnosis. Mothers are asked to complete questionnaires at various time points, including when the child is aged 3. Recruitment/data collection are ongoing; at the time of this study, data from 239 mothers were available and used in the analyses. Descriptive statistics were used to provide means and standard deviations for each cleft type.

Ethical approval was provided by Central Bristol and London-Central Research Ethics Committees. The Cleft Collective Birth Cohort study is funded by the Scar Free Foundation; the analysis outlined in this paper was funded by the Underwood Trust with support from the University of Bristol.

Results: The mean ICS score for the total sample was 3.72 (SD=0.77) out of a possible total score of 5. For children with cleft lip only the average score was 4.17 (SD=0.62) and for children with cleft palate only it was 3.67 (SD=0.73). For children with cleft lip and palate, the average score was 3.47 (SD=0.77). Overall mothers reported children to be more intelligible to them, mean 4.30 (SD=0.62), than with strangers, mean 3.29 (SD=1.01).

Summary/Conclusion: At age 3, the average score on the ICS for children born with any type of cleft is 3.7. Norms for non-cleft children are not available for this age. These results provide preliminary comparative data for clinical services using the ICHOM outcome measures. They also provide clinical markers for outcomes based on low intelligibility to strangers. Future studies will analyse whether additional syndromes and the presence of hearing difficulties affect intelligibility scores. Analyses with the same cohort of children at 5-years-old will allow comparison with normative data for a non-cleft English speaking population and outcome data which can be compared with the baseline data collected at age 3. These analyses will provide a rich dataset to inform use of the ICS as a standalone outcome measure of surgical repair and a baseline/outcome measure of speech intervention.
Health-related quality of life in patients with cleft palate: validation of the dutch translation of the velo questionnaire

L Bruneel¹, K Bettens¹, C Alighieri¹, M De Bodt², K Van Lierde¹
¹Department of rehabilitation sciences, Ghent University, Ghent, ²Rehabilitation centre of communication disorders, Antwerp University Hospital , Antwerp, Belgium

Background: In health-care, current efforts focus on providing patient-centered care. Specifically for patients with velopharyngeal insufficiency, and by extent patients with cleft palate, the Velopharyngeal Insufficiency (VPI) Effects on Life Outcomes (VELO) questionnaire (Skirko et al., 2012; 2013) allows the clinician to map the impact of speech and swallowing difficulties and speech-related distress on the patient’s functioning and well-being.

Aims: The aim of this project was to translate the VELO questionnaire to Dutch and to evaluate the validity and reliability of this translation.

Methods: The English questionnaire was translated to Dutch following a forward-backward translation procedure. In a first study, the content validity, internal consistency, concurrent validity and discriminant validity were evaluated in 39 parents of patients with cleft palate (parent report) and 14 children with cleft palate (child report). Thirty-five parents completed the VELO questionnaire after one year for the evaluation of the responsiveness. To evaluate the reproducibility, 50 parents and 14 children with cleft palate re-completed the questionnaire after two weeks. Lastly, the relationship between the responses on the VELO questionnaire, and perceptual and instrumental speech assessments in patients with cleft palate was explored. All participants signed an informed consent.

Results: There were no significant differences between the responses of the parent and their child’s. A significant positive correlation was found between the score on the parent report and the patients’ age. Cronbach’s α was 0.955 and 0.817 for the parent report and the child report respectively. Furthermore, a significant negative correlation was found between the parent report and the P-VHI, and analysis showed a significantly worse perception of HRQoL in the patient group compared to an age and gender matched control group. Good reproducibility of the questionnaire was found based on descriptive statistics, results of the Wilcoxon signed rank-test, and the absolute and relative consistency. VELO scores did not significantly differ after one year, neither when performing separate analyses for the intervention (speech therapy) and the non-intervention group. Moderate to strong correlations were found between the total score on the VELO parent report and five speech variables: the VPC-SUM score, speech understandability, passive CSC’s, speech acceptability and the need for cleft-related speech therapy.

Summary/Conclusion: The VELO questionnaire showed good content validity, internal consistency, concurrent validity, discriminant validity, reproducibility and construct validity. Moreover, results confirmed the age effect on VELO scores. In summary, the Dutch VELO questionnaire is a valid, reliable and user-friendly tool. Future research should focus on the identification of factors influencing the patient’s evolution in HRQoL. More specifically, speech features that can be targeted in therapy to achieve the greatest impact on HRQoL should be identified.
Development and validation of a dutch outcome tool for the perceptual evaluation of speech in patients with cleft palate
L Bruneel¹, K Bettens¹, M De Bodt², E D’haeseleer¹, Z Thijss¹, N Roche³, K Van Lierde¹
¹Department of rehabilitation sciences, Ghent University, Ghent, ²Rehabilitation centre for communication disorders, Antwerp University Hospital, Antwerp, ³Department of plastic surgery, Ghent University Hospital, Ghent, Belgium

Background: The past decades, great efforts are being made to define outcomes and related measurements of health conditions. Specifically for patients with cleft palate, speech is a crucial outcome.

Aims: The purpose of the current study was to develop a Dutch outcome tool for the perceptual evaluation of speech in Belgian Dutch-speaking patients with cleft palate with clearly defined speech variables and rating methods, and a standardized speech sample, and to assess the validity and reliability of this outcome tool.

Methods: A listening protocol and Belgian Dutch speech sample were constructed based on a literature review and panel discussions. Following a pilot study, including the evaluation of 20 speech samples from patients with CP±L and ten participants without CP±L, the outcome tool was optimized. In a second phase, a listening experiment with four experienced listeners each evaluating ten speech samples was conducted to assess the final tool’s face and content validity, construct validity, and inter- and intra-rater reliability.

Results: Overall, the listeners acknowledged the face and content validity of the outcome tool. For most parameters good inter- and intra-rater reliability were found. Furthermore, the tool showed good sensitivity, specificity and percentage of agreement when comparing results with nasalance values and the Nasality Severity Index (NSI) 2.0.

Summary/Conclusion: This study showed promising results regarding the validity and reliability of the developed tool. In future studies, the validity and reliability will be optimized, with the eventual aim of standardized speech outcome assessment in Dutch-speaking patients with cleft palate.
Examining predictors and identifying speech and language characteristics of children with cleft palate at 39 months of age
L Beckett¹, K Chapman², M Hardin-Jones³
¹Speech Language Pathology, Riley Hospital for Children, Indianapolis, ²Communication Sciences and Disorder, University of Utah, Salt Lake City, ³Communication Sciences and Disorder, University of Wyoming, Laramie, United States

Background: Children with cleft palate exhibit delays in speech, language, and hearing. However, the extent of the delays are often not well understood because their performance is not compared to non-cleft children of the same age. Further, more information is needed to determine which early speech and language characteristics are most predictive of later performance for early diagnosis of speech and language impairment in this population.

Aims: The purpose of this study was 1) to examine the speech/language skills of children with cleft palate and their noncleft peers at 39 months, and 2) to extend the findings of previous studies that attempted to identify 9 month (presurgery) and postsurgery speech/language skills that predict later speech/language outcomes.

Methods: Participants included 66 children, 43 children with cleft palate and 23 noncleft children. Spontaneous speech/language samples were collected at 9 months (presurgery), postsurgery (approximately 13 months), 21 months, and 39 months of age in the child’s home during an interaction with the caregiver. Samples were phonetically transcribed and entered into Logical International Phonetic Programs (LIPP) (Oller, 1990) for speech analysis. Language samples were analyzed using Systematic Analysis of Language Transcripts (SALT v 6.1a) (Miller & Chapman, 2000). Independent sample t tests and Wilcoxon rank-sum tests were employed to compare the speech and language skills of children with cleft palate and their noncleft peers at 39 months of age. Linear regression analyses using selected factors based on the Least Absolute Shrinkage and Selection Operator (LASSO) approach were used to identify the timepoint(s) and speech/language variables most associated with normal speech/language at 39 months.

Results: A comparison of the speech/language skills of children with cleft palate and their noncleft peers at 39 months revealed the children with cleft palate had fewer consonant sounds, produced less accurate consonants for the majority of the place and manner categories, and had lower mean length of utterances than their noncleft peers. Correlations between 9 month (presurgery) and postsurgery measures and later speech/language outcomes at 39 months revealed negative correlations between 9-month predictors and all outcome measures. All other predictors were positively correlated with the speech outcome measures at 39 months. The strongest predictive values for positive speech outcomes were found at 21 months. Accuracy of stop production was the variable most associated with better speech and language skills at 39 months.

Summary/Conclusion: These results suggest children with cleft palate have poorer speech/language outcomes than noncleft peers at 39 months of age. There is a need for children with cleft palate to receive earlier speech/language intervention to help them catch up with their noncleft peers. Negative correlations were surprising and may be related to presurgery differences or interventions and/or structural deficits occurring in the postsurgical period. Finally, the strongest correlations were found between the 21 month predictors and the 39 month outcome measures.
**Predicting percent consonants correct at three years: evaluation of the long-term predictive value of a screening procedure for toddlers with cleft palate**

L Jørgensen 1,2

1 Department of Nordic Studies and Linguistics, University of Copenhagen, 2 Copenhagen Cleft Palate Center, Copenhagen University Hospital, Copenhagen, Denmark

**Background:** Children with cleft palate (CP) are at risk of speech and language delay. It is important to identify children with delays as early as possible to offer appropriate intervention, but it is also necessary to consider the burden of care for the families and socioeconomic cost in case of unwarranted intervention. Jørgensen and Willadsen (2017) developed and validated a clinically applicable speech-language screening procedure for toddlers with CP based on previously reported predictors of speech-language development (Klintø et al. 2014, Chapman 2004). The screening procedure was designed to evaluate need for intervention based on consonant inventory and productive vocabulary. Consonant inventory was evaluated through naturalistic listening in real time (Ramsdell et al. 2012), and productive vocabulary via the parent questionnaire MacArthur-Bates Communicative Developmental Inventories (Fenson et al. 2007). However, the long-term predictive value of this screening procedure has not yet been investigated.

**Aims:** This study investigates the predictive value of a speech-language screening procedure for toddlers with CP on percent consonants correct (PCC) at 3 years.

**Methods:** 56 toddlers with CP participated in the study. They were recruited from a complete birth cohort of all 103 Danish children with CP born between December 2012 and August 2014. At age 16-24 months, participants were video recorded during 2 x 45 minutes play interaction with a parent. Participants’ need for intervention was evaluated by a screening procedure (Jørgensen and Willadsen 2017) which separated participants into two groups: a “need for intervention” group and a “no need for intervention” group. At 3 years, video recordings of a naming test were obtained. Recordings were phonetically transcribed by a rater unfamiliar with group allocation, and PCC scores were calculated. Inter and intra rater reliability will be reported. Logistic regression analysis will be used to investigate if the outcome of the screening procedure significantly predicts PCC scores at three years.

**Results:** All data have been collected, and analyses are in progress.

**Summary/Conclusion:** If the outcome of the screening procedure can predict PCC scores at 3 years, the screening procedure is a valid instrument for evaluating need for early intervention in toddlers with CP. On the other hand, if the outcome of the screening procedure does not significantly predict low PCC scores at 3 years, the screening procedure may need to be amended, or it may be that age 16-24 months is too early to identify need for intervention in toddlers with CP.
Using borg centiMax scale for perceptual rating of hypernasality
F Favaretto¹, A Fukushiro², R Yamashita¹
¹Laboratory of Physiology, Hospital for Rehabilitation of Craniofacial Anomalies-University of Sao Paulo, ²Department of Speech-Language Pathology, Bauru School of Dentistry-University of Sao Paulo, Bauru, Brazil

Background: Borg centiMax scale (Borg and Borg, 2001) is a verbally level-anchored ratio scale that combines the advantages of ratio scales with those of category scales for level determination. In this method, verbal anchors are placed in congruence with numbers on a ratio scale that covers a biologically natural range of intensities, so that for each category there is a value in a numerical continuum.

Aims: To compare the reliability in auditory-perceptual assessment of hypernasality between the Borg centiMax scale and an ordinal scale and to investigate the influence of speech material on the reliability of the two methods.

Methods: This study was approved by the institutional review board. Audio recording of speech sample was performed in 40 individuals with repaired cleft lip and palate, aged 10 to 45 years. The speech material comprised of 80 audio recordings: 40 stimuli of 12 non-nasal single-word strings and 40 stimuli of 12 sentences repetition both contained high pressure consonants. Four experienced speech-language pathologists analyzed the speech samples and rated hypernasality. Each stimulus was analyzed individually using a 5-point scale and the Borg centiMax scale. Intra- and inter-rater reliability were calculated for both methods and for both types of speech samples. Intra-rater reliability was calculated for each listener within the two methods using the repeated ratings of 16 (20%) speech samples (8 single-word strings and 8 sentences). Inter-rater agreement was calculated comparing the 4 listeners 2-by-2. Intra and inter-rater agreement for ordinal scale was analyzed using weighted Kappa (κ) and for Borg centiMax scale using intraclass correlation coefficient (ICC). The comparison of intra- and inter-rater agreements between the two types of speech samples was calculated using the Z test. The correlation between the two methods was established using Spearman correlation coefficient. For all tests was considered a significance level of 5%.

Results: Statistically significant correlation was found between the methods for both speech samples (single word-string=0.94/p<0.001 and sentences=0.95/p<0.001). Higher intra-rater reliability was found for Borg centiMax scale than ordinal scale in both speech samples. For Borg centiMax ICC ranged from 0.73 to 1.00 for single-word and from 0.42 to 0.99 for sentences. For ordinal scale κ ranged from 0.38 to 1.00 for single-word and from 0.81 to 1.00 for sentences. Additionally, inter-rater reliability was higher for Borg scale than ordinal scale for both speech samples. For Borg centiMax ICC ranged from 0.51 to 0.89 for single-word and from 0.40 to 0.74 for sentences. For ordinal scale, κ ranged from 0.32 to 0.50 for single-word and from 0.10 to 0.53 for sentences. There was a significant difference between the two types of speech samples both for intra- (p<0.011) and inter-rater (p<0.025) reliability using Borg centiMax scale, and for inter-rater reliability (p<0.007) using ordinal scale.

Summary/Conclusion: The Borg centiMax scale was more reliable in the assessment of hypernasality than the ordinal scale showing better intra- and inter-rater agreement coefficients. In addition, the speech material comprising of single-words string influenced the perceptual ratings of hypernasality as compared with sentence repetition showing better reliability in most of the comparisons for both methods.
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Associations between hypernasality, intelligibility, language and reading skills in 10-year-old children with a palatal cleft.

Q Hide¹, T Særvold¹, K Billaud Feragen², R Aukner¹

¹Speech and Language Disorders, Statped, ²Centre for Rare Disorders, Oslo University Hospital, Oslo, Norway

**Background:** This study investigates the associations between hypernasality and intelligibility, and language and reading skills in 10-year old children with a cleft palate +/- lip. Based on previous findings (Havstam et al., 2011; Lohmander et al., 2012; Nyberg et al., 2014), articulation improves with age and few articulation deviations are left at the age of 10. However, our clinical experience often shows reduced intelligibility and nasality deviations, despite speech surgery and / or speech therapy. Therefore, we have chosen to analyse hypernasality as one measure of nasality deviations, and intelligibility as an estimate of the child’s ability to make her/himself understood.

**Aims:** The aim of this study was to explore language and reading skills and their potential associations with the two speech characteristics hypernasality and intelligibility in children with a palatal cleft aged 10 years.

**Methods:** Cross-sectional data collected during routine assessments of speech and language in a centralised treatment setting. The participants were children aged 10, born with cleft palate +/- lip from four birth cohorts (N = 123). The outcome measures were hypernasality and intelligibility, language and reading. The outcome measures were conducted by SVANTE-N, Language 6-16 (Sentence recall, Serial recall, Vocabulary), Word Chain Test and Reading Comprehension Test.

**Results:** A total of 71.3% of the children had no occurrence of hypernasality, and 82.8% had intelligibility scores within the normal range. For all children with hypernasality and intelligibility within the normal range, reading and language scores were also within normal ranges. Children with presence of hypernasality had significantly lower language skills, with mean scores within the lower normal range. Children with reduced intelligibility had lower scores on reading comprehension.

**Summary/Conclusion:** The findings highlight a possible association between hypernasality and language skills, and intelligibility and reading skills. Cleft teams should consider routine assessments of language and reading skills in children with speech impairment, in order to identify potential needs for intervention as early as possible.
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**Speech outcome evaluation of cleft palate patients underwent palatoplasty in plastic surgery division cipto mangunkusumo hospital indonesia.**

P Kreshanti¹, V Sari¹, L Wahyuni², G Wangge³

¹Plastic Surgery, ²Medical Rahabilitation, Universitas Indonesia - Cipto Mangunkusumo Hospital, ³Public Health, Seameo Recfon, DKI Jakarta, Indonesia

**Background:** Speech is the primary goal of palatoplasty, however, there is no current data available about the long term speech evaluation after palatoplasty in our hospital which is the national referral hospital that has the only cleft craniofacial center in Indonesia. The initial data of speech outcome is required for further research which assessment should be standardized and applicable to Indonesian children that mostly speak bahasa.

**Aims:** This study aims to get initial data by evaluating speech outcome of patients that underwent palatoplasty between October 2010 – December 2012 with adapted perceptual assessment words in Indonesian language, and describe factors influencing speech.

**Methods:** Descriptive study with total of 23 samples between October 2010 – December 2012 in Cipto Mangunkusumo Hospital, Indonesia.

**Results:** For articulation rating (74%) patients had normal production of majority of phonemes, while there were 6 (26%) patients had predominantly distortion of phonemes. The hypernasality rating were normal in 12 (52%) patients, mild in 5 (22%) patients and moderate in 6 (26%) patients. The speech intelligibility rating were dominantly normal which all speech is understood in 17 (74%) patients and the rest of 6 (26%) patients were listeners attention needed. The velopharyngeal competence were good in 16 (70%) patients, fair in 1 (4%) patients and poor in 6 (26%) patients.

**Summary/Conclusion:** This study provided initial data that can be used for further research. The good data filing is very important for the sake of education and patients management. By giving the long term follow up to the patients, the optimal outcomes will be achieved. Management of cleft palate patients will be achieved by well integrated services including speech pathologist and orthodontist. This research can be used as reference for speech outcome evaluation in cleft palate patients in Indonesia.
Relationships between early speech skills and later language ability in internationally adopted children with UCLP
A Larsson¹, C Miniscalco², H Mark³, J Schölin³, C Persson ¹
¹Speech and Language Pathology Unit, Department of Health and Rehabilitation, ²Gillberg Neuropsychiatry Centre, Institute of Neuroscience and Physiology, Sahlgrenska Academy, University of Gothenburg, ³Institute of Clinical Sciences, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden

Background: During the years of early 2000, an increase of internationally adopted (IA) children with cleft lip and palate (CLP) was seen by European CLP centres. Many children had unrepaired palates at arrival and many were adopted at an older age than previously seen. Earlier studies have shown a high prevalence of velopharyngeal impairment in this group of children but also other types of speech impairments. The risk for language impairments has also been highlighted, but so far very few studies have investigated the language abilities in IA children with CLP. Longitudinal studies of speech and language development in IA children with CLP are rare and further knowledge is called for.

Aims: The purpose of this project was to longitudinally study the speech production in a group of IA-children with UCLP. Specific research questions were:
1) In what way do speech production in this group of children develop between the ages 3 to 7-8?
2) Are there any correlations between speech production at age 3 and language ability at age 7-8?

Methods: This study investigated the speech skills in a group of IA-children at three time points; age 3 (T1), age 5 (T2) and age 7-8 (T3). Language skills were investigated at age 7-8 (T3). Participants were recruited from a CLP centre at the Sahlgrenska University Hospital in Gothenburg, Sweden. Out of 32 eligible children, 16 participated in this project. All children were adopted from China during 2007-2011 at a mean age of 22 months (min-max 16-33). Data from the routine speech assessments at the hospital was used for T1 and T2, where a standardised test of articulation and nasality was used (SVANTE). At T3, data from a broad test battery of speech and language skills was used. The outcome variable for speech production at all three time points was the PCC (percent consonants correct). For language ability at T3 three outcome variables were set; one for expressive language (the subtest Recalling sentences from the CELF-IV) and two variables for receptive language – vocabulary (PPVT-3) and grammar comprehension (TROG-2). Correlation analyses between PCC at T1 and expressive language and receptive language measures respectively at T3 will be investigated with non-parametric Spearman’s rho correlation.

Results: This is an ongoing project and data has not yet fully been analysed. We will report on the development of speech production/PCC at the three time points (age 3, age 5 and age 7-8). Also we intend to describe any correlations between PCC at T1 and the expressive and receptive language measures respectively at T3.

Summary/Conclusion: We expect our results to contribute to the intervention planning of IA-children with UCLP and to bring more knowledge to the area of speech and language development in all CLP children.
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Early identification of communication and language delays in children between 8-36 months of age with different cleft types

G Musso1, V Di Paolo1, F Aceti1, I fontana1, P Todaro1, L Autelitano1, M Meazzini1, A Rezzonico1
1Smile House, Santi Paolo e Carlo Hospital, Milan, Milano, Italy

Background: Statistically there are significant differences in the receptive and expressive language skills between children with and without cleft lip and palate from 8 to 36 months of age (Scherer et al., 2008; Lamonica et al., 2015). In CLP population, there is a higher prevalence of late talkers than in typically developing (TD) children; consequently, children with CLP have an increased risk for early speech and language delays (Chapman et al., 2003). The purpose of this study is to evaluate CLP patients before 36 months in order to identify children at risk for language delay before their development of a sufficient language level to be analyzed through a standardized perceptual assessment. In our search of the literature, we have not found any studies that compare the communicative and language abilities from 8 to 36 months of age between different cleft types: Unilateral Cleft Lip and Palate (UCLP), Bilateral Cleft Lip and Palate (BCLP) and isolated Cleft Palate (CP).

Aims: This study aims to examine early communicative and language development in Italian-speaking toddlers with Cleft Lip and Palate (CLP) from 8 to 36 months of age, compare it with typically developing (TD) children and then compare communicative and language development in the different cleft types: UCLP, BCLP, CP.

Methods: The sample consisted of 133 children between 8-36 months of age with Unilateral Cleft Lip and Palate (UCLP; n = 34,6%), Bilateral Cleft Lip and Palate (BCLP; n = 9%) and isolated Cleft Palate (CP; n = 56,4%). Exclusionary criteria: presence of syndromes, bilingualism and adoption. Communicative and language development was evaluated using The Italian MacArthur-Bates Communicative Development Inventories (MB-CDI) Complete Form (Caselli et al., 2015). Data on CLP were compared with respect to those of TD children (Caselli et al., 2015). The unpaired Kruskal-Wallis test and Dunn-Bonferroni test were used to compare children with different cleft types.

Results: Word comprehension percent of the population below the 10th percentile (below expectation) was 19,8% at 8-12 months and 22,3% at 14-17 months; A-G production percent of the population below the 10th percentile was 24,5% at 8-12 months and 25,9% at 14-17 months. Word production percent of the population below the 10th percentile was 40,8% at 18-24 months and 42,7% at 27-36 months. Significant difference (p-value=0.048) in word production in different types of cleft was highlighted at 27-36 months of age, where UCLP showed the worse performance. Analysis of MLU highlighted a significant difference (p-value=0.011) and both UCLP and isolated CP showed the worst performances.

Summary/Conclusion: This study showed that 42% of the sample have a language delay compared with the TD peers and it increases in percent with age. Considering that TD children show a prevalence rate of 14-17% language delay (Horwitz et al., 2003), CLP can be considered a risk factor in the development of language disorder. Another result of this study revealed a specific weakness in children with UCLP between 27-36 months of age.
A longitudinal study of speech and lexical development in 4-5 years' children with cleft lip and palate and isolated cleft palate

V Di Paolo¹, G Musso¹, F Aceti¹, I Fontana¹, P Todaro¹, L Autelitano¹, M Meazzini¹, A Rezzonico¹
¹Smile House, Santi Paolo e Carlo Hospital, Milan, Milano, Italy

Background: In CLP population, there is a higher prevalence of late talkers than in typically developing (TD) children; consequently, children with CLP have an increased risk for early speech and language delays.

Aims: The purpose of this study was to verify whether the lexical deficit found in a sample of children of 36 months of age with cleft lip and palate (CLP) was still present at the age of 4 years.

Methods: The sample consisted of 17 children of four to five years of age with unilateral cleft lip and palate (UCLP; n= 58.8 %) and isolated cleft palate (CP; n= 41.2 %) that in a previous study had shown a lexical deficit at 36 months of age found through the Italian MacArthur-Bates Communicative Development Inventories (MB-CDI). All children were recruited from the Smile House in Santi Paolo e Carlo Hospital in Milan, Italy. Exclusion criteria were: presence of syndromes, bilingualism and adoption. Lexical and speech level at 4-5 years of age was evaluated using the BVL 4-12 (Batteria per la valutazione del linguaggio in bambini dai 4 ai 12 anni - Marotta, Fabbro, Bulgheroni, 2015). The Pearson Coefficient was used to compare the results obtained at 3 and 4 years of age.

Results: Probably due to the small sample size, there were no statistically significant correlations, but clinically, it was possible to observe that at 4 years of age none of the children of the sample still showed a lexical deficit. From the analysis of the errors in the speech test, it should be noted that the percent of total errors was 63%, of which 40% were errors secondary to CLP, while 51% were developmental errors, attributable to age. Only 19% of total errors appeared to be typical errors of the specific language disorder.

Summary/Conclusion: It can be concluded that most of the errors are due to CLP or ascribable to age and not typical of a specific language disorder. Furthermore, the lexical deficit evidenced at 3 years of age seemed to have resolved spontaneously at 4 years of age, probably because it was secondary to the pathology and not a predictor of a specific language disorder.
“She sounds very childish – or perhaps she has problems” – 7-years-olds descriptions of speech in peers born with cleft palate

J Nyberg1, E Hagberg2, C Havstam3
1Stockholm Craniofacial Team, Karolinska University Hospital. Division of Speech and Language Pathology, CLINTEC, Karolinska Institute, 2Stockholm Craniofacial Team, Karolinska University Hospital. Division Speech and Language Pathology, CLINTEC, Karolinska Institute, Stockholm, 3Division of Speech and Language Pathology, Sahlgrenska University Hospital and Sahlgrenska Academy at University of Gothenburg, Gothenburg, Sweden

Background: Research on children’s perceptions of various communication disorders indicates that they generally hold negative attitudes towards speakers with speech impairments. Most studies have used dichotomized word pairs created by adults in order to let the children describe how they perceive the specific person behind the speech sample. In a study with 10-year-olds, the children reported on their reflections of speech in peers born with cleft palate. The result showed that peers easily described articulatory difficulties but did not pay attention to minor signs of velopharyngeal insufficiency (VPI).

Aims: The objective of this study was to explore how 7 year-olds describe speech in children born with cleft palate in their own words and to investigate whether they perceive signs of velopharyngeal insufficiency (VPI) and articulation errors.

Methods: Twenty 7-year-olds participated in six focus group interviews where they listened to eight speech samples with different types of cleft speech characteristics and described what they heard. The speech samples were assessed by speech- and language pathologists and comprised normal speech, different degrees of VPI, oral articulation difficulties and glottal articulation. The interviews were transcribed and analyzed with qualitative content analysis.

Results: The analysis resulted in three interlinked categories encompassing different aspects of speech, personality and social implications: Descriptions of speech, Thoughts about causes and consequences and Emotional reactions and associations. Each category contains four to five subcategories with the children’s descriptions and reflections. The comments correspond to the clinicians’ assessments. The result also showed that glottal articulation caused most reflections and comments, but also minor articulatory difficulties were described.

Summary/Conclusion: Descriptions from peers imply that more pronounced signs of VPI were perceived, but referred to in terms relevant for 7-year-olds. The articulatory difficulties, also minor ones, were noted. Peers also reflected on the risk for teasing and bullying and on how children with impaired speech might experience their situation. The findings align with previous related research, but 7-year-olds reflect in a more straightforward and unrestrained way than 10-year-olds. The project is expected to have significant relevance in the clinical work. The result can also be generalized to children with phonological disorders.
Response to articulation therapy by an adult with Nager’s syndrome with speech prosthesis: a single case experimental design.

N Prendeville1, D Sell2
1SPEECH AND LANGUAGE THERAPY, 2ORCHID, GREAT ORMOND STREET HOSPITAL NHS TRUST, LONDON, United Kingdom

Background: Nager syndrome is a rare congenital craniofacial condition of unknown aetiology which is characterised by an absent velum, velopharyngeal insufficiency (VPI), micrognathia, mid-face hypoplasia, early respiratory and feeding problems, ear canal atresia and conductive hearing loss, upper and lower limb malformations, and speech/language delays and disorders. Normal IQ is reported.

Aims: To investigate the response to therapy of entrenched cleft speech characteristics (CSCs) and the amount of treatment in an adult with Nager syndrome following speech prosthetics intervention for velopharyngeal insufficiency (VPI).

Our questions were:
1. Does articulation therapy increase the number of stimulable consonants?
2. Does articulation therapy increase the number of correct consonants on a sentence repetition task?
3. Does articulation therapy reduce the number of posterior and non-oral CSCs?
4. What is the patient’s impression of the psycho-social impact of altered speech production?
5. Does intelligibility in conversational speech improve?

Methods: An adult patient (C.A: 18;03 yrs) with long-term VPI associated with Nager’s syndrome had successful fitting of a speech bulb obturator. She presented with persisting backing-to-uvular (posterior) and pharyngeal (non-oral) CSCs which significantly reduced intelligibility with life-long VPI. A single case experimental design (SCED) was used: ‘A’ = non-treatment phase before and after therapy; ‘B’ = targets /t,s,tʃ,dʒ,k/ treated in therapy and home practice. Sounds /d,z,ʃ,g/ acted as untargeted controls. Individual therapy sessions with a specialist SLT were undertaken. Outcome measures included % correct articulation of targeted and control speech sounds, % posterior and non-oral CSCs and intelligibility. Patient’s speech satisfaction was captured qualitatively in a semi-structured interview and in discussion with family members.

Results: Two trained listeners conducted independent blinded perceptual analyses. Visual analysis and an effect size calculation were used to assess the magnitude of intervention effect. This included percentage of non-overlapping data and percentage of improvement rate difference. Percentage of stimulable sounds and correct sounds in sentence imitation increased, indicating an increase in phonemic contrasts. Visual analysis revealed a decrease in percentage of posterior and non-oral CSCs during the intervention phase. Overall, targeted sounds improved post-therapy resulting in perceptually more acceptable realisations, rendering speech more intelligible. Generalisation to a number of untargeted phonemes was evident. 17 articulation therapy sessions were delivered fortnightly with two 6-week consolidation breaks over 11 months. Qualitative analysis revealed considerable satisfaction with her own speech 6 months post-intervention; while family feedback corroborated this finding.

Summary/Conclusion: Entrenched CSCs resolved with articulation therapy in a motivated adult patient with a speech bulb obturator over an 11-month period, demonstrating a reasonably rapid response to intervention. A single subject experimental design was an appropriate methodology to evidence therapy. The patient reported increased satisfaction with her speech post-intervention and improved communicative interactions with new people while family members reported improved overall speech clarity.
Background: The main goal of team management in cleft palate is to help cleft palate children have an adequate speech development.

Aims: to investigate the prevalence of articulation and resonance disorders following primary palate repair in cleft palate children; and to study the impact of cleft type and age at the time of palatoplasty on speech outcomes.

Methods: Clinical records of 180 preschool children with repaired cleft palate, who were visited in Isfahan cleft palate clinic between the years 2011-2015, were reviewed. Percentage of children demonstrating hypernasality, nasal emission (NE), nasal turbulence (NT), developmental errors, and compensatory misarticulations (CMA) was calculated. The relationship between cleft type and age at the time of palatal surgery, as independent variables, and moderate/severe hypernasality and also CMA were examined.

Results: 67.7 and 64.5 percent of the children demonstrated respectively moderate/severe hypernasality and NE, and 71.1 percent produced CMA. The mean of age at the time of palatal repair was significantly higher in children with moderate/severe hypernasality compared to those without significant hypernasality (17.60±12.94 versus 13.89±8.60, p=0.029). Age at the time of palatal repair was also significantly associated with CMA (p=0.029). The prevalence of CMA and also significant hypernasality was not significantly different in various types of cleft palate.

Summary/Conclusion: we observed the high prevalence of different speech disorders in Iranian preschool children with repaired cleft palate compared to other studies. This can be partly due to late palatal repair in the studied population. Despite many advances in cleft palate management programs in Iran, there are still lots of children who do not access the interdisciplinary team cares in their early childhood. We should, therefore, try to increase accessibility of appropriate and timely management services to all Iranian children with cleft lip/palate.
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**Initial diagnosis of velopharyngeal dysfunction from pediatric otolaryngology consultation: clinical findings**  
S Santos¹, B González Meli², F Lobo Bailón², N González Llorente³, J Cervera Escario¹  
¹Otolaryngology, ²Plastic Surgery, ³Speech Therapy, Hospital Infantil Universitario Niño Jesús, Madrid, Spain

**Background:** Velopharyngeal dysfunction (VPD) without an obvious cleft, is often diagnosed late. The perceptual speech evaluation is considered the gold standard in the evaluation of VPD. When abnormal speech is not treated in the first years of life articulation problems can remain persistent after surgical correction. Otolaryngologists may be the first point of contact for the symptomatic VPD patient.

**Aims:** To assess the role of Pediatric Otolaryngologist in diagnosing VPD as first clinician who may evaluate a resonance disorder.

**Methods:** Retrospective longitudinal charts review of patients from the multidisciplinary Cleft Team of the Niño Jesús University Hospital in Madrid over a 6-year period. One hundred and thirty five charts from patients referred to the Team were reviewed. Clinical findings of patients whose first initial diagnosis of VPD was established at ENT consultation are described.

**Results:** Thirteen patients from a cohort of 135 (9.6%) that received their first VPD diagnosis by Otolaryngology were evaluated. The median age at diagnosis was 3.9 yrs. Reason for referral for ENT consultation were: Speech disorders (n: 7), hearing assessment (n: 5) and stridor (n: 1). Eleven cases of velopharyngeal insufficiency (9 submucous cleft palate and 2 velar dysplasia), 1 case of velopharyngeal incompetence and 1 mixed case of velopharyngeal incompetence (with submucous cleft palate) and mislearning due to severe mixed hearing loss were found. Genetical tests showed 22q deletion in 3 children and chromosomopathies in another 3 ones. Seven children out of 13 were attended previously in another ENT services out of our Cleft Team.

**Summary/Conclusion:** Patients with VPD frequently present to Pediatric Otolaryngologist undiagnosed in the first years of life. Otolaryngologists are uniquely poised to facilitate early diagnosis by perceptual speech evaluation and initiation of therapy, potentially reducing long-term sequelae.
Early speech and language intervention in Brazil: preliminary results from a small randomized study

N. Scherer¹, R. Yamashita², A. Fukushiro³, I. Trindade², K. Lien¹, D. de Oliveira², J. Dilallo¹, R. Scarmagnani²

¹Arizona State University, Tempe, United States, ²University of Sao Paulo, ³University of Sao Paulo, Bauru, Brazil

Background: Background: Young children with palate with or without cleft lip (CP+/−CL) are at risk for early vocabulary and speech sound production delays. Early intervention studies have shown some promising findings to promote early speech and vocabulary development following palate repair; however, we know little about how these interventions can be used in other international contexts. This study adapted an early speech and language intervention developed in the US; Enhanced Milieu Teaching with Phonological Emphasis (EMT+PE) to the Brazilian context at the Hospital for Rehabilitation of Craniofacial Anomalies at the University of Sao Paulo-Bauru.

Aims: Aims: The purpose of this study was to compare the speech and language performance of two groups of toddlers with CP+/−CL randomized into an EMT+PE intervention or a business-as-usual (BAU) comparison group.

Methods: Method: Twenty children with non-syndromic children between the ages of 19-32 months with CP+/−CL were included in the preliminary findings of the study and is part of a larger intervention study which is still ongoing. Inclusion criteria included palate repair prior to 15 months of age, monolingual Brazilian Portuguese spoken at home, absence of a syndrome or sensorineural hearing loss recruited from the Hospital for Rehabilitation of Craniofacial Anomalies. The children in the intervention group received 30-40-minute intervention sessions twice weekly for 24 sessions. Target words for the intervention were selected from the CDI based on sounds omitted or substituted on the Profiles of Early Expressive Phonology: Brazilian Portuguese. The intervention was provided by a speech-language pathologist in one of two sites in San Bernardo or Bauru, Brazil with parents given training in EMT+PE strategies. The Bauru clinicians were trained to criterion on the EMT+PE intervention by the project PI and they trained the clinician in San Bernardo. Assessments of the children in both groups occurred at three time points before, after and 3 months following the end of intervention. Assessments included a standardized language test, single word articulation test, language sample and the CDI parent questionnaire in Brazilian Portuguese. Any interventions received by the BAU group were documented for therapy type, goals and frequency. Inter and intra judge reliability of speech sound production and language sample transcription were obtained for 20% of the samples. Procedural fidelity of the intervention was assessed at 4 time points and showed excellent fidelity.

Results: Results: The pre and post comparison was focused on the changes in single word articulation production. The results indicated a large effect size difference that favored the EMT+PE intervention group for stop consonant production (d = .77) which were the target sounds for all children in the study. Pre-post vocabulary changes favored the EMT+PE group with an effect size of d=1.67. The children in the EMT+PE group had a mean increase in vocabulary of 238 words while the BAU group increased an average of 87 words.

Summary/Conclusion: Conclusions: Preliminary data suggests that EMT+PE adapted for the Brazilian context shows promise as an early speech and language intervention for children with CP+/−CL. However, further data is needed to provide additional insight into changes observed in both groups.
Background: Clinicians agree that children with isolated cleft lip have fewer cleft-associated problems than children with cleft lip and/or palate. Unfortunately, for isolated cleft lip children the risk of cleft-associated problems is unknown and maybe underestimated. Often these children do not get the required follow-up by a multidisciplinary team and thereby not the known benefits in supporting their development.

Aims: This study examines the development of speech and ear problems in children with isolated cleft lip.

Methods: A retrospective study was performed on all children born with an isolated cleft lip and treated at the Wilhelmina Children's Hospital in Utrecht between January 2007 and April 2014. Data were collected for sex, date of birth, genetics, cleft lip type, date of cleft lip repair, type of repair, speech and ear problems.

Results: This study included 75 patients (59% male). Eighteen of the 75 children (24%) developed a speech problem, however only one child (1.3%) had a cleft related speech problem. Fifteen of the 75 patients (20%) reported a history of one or more episodes of acute otitis media (AOM) during the first six years. Six of the 15 children (40%) with a history of AOM received speech and language therapy.

Summary/Conclusion: In this study, children with an isolated cleft lip do not have a higher risk of cleft related speech problems and AOM/OME than other non-cleft children. However, these children do have a higher incidence of speech therapy in general.
Phonological speech intervention study including four children with severe speech sound disorder born with cleft palate using a single case experimental design

H Søgaard¹, M Boers¹, L Dahl Jørgensen¹, E Willadsen²
¹Copenhagen Cleft Palate Center, Copenhagen University Hospital, ²Department of Nordic Studies and Linguistics, University of Copenhagen, Copenhagen, Denmark

Background: Around 50% of children born with cleft palate (CP) have speech problems at preschool age (Lohmander, 2008, Bessel et al., 2013) which is far more than in the population without CP. Still, there is limited knowledge about efficient speech therapy for these children. Multiple Oppositions Intervention (MOI) is described as an effective way of expanding the consonant inventory in children with severe speech sound disorder (SSD) without CP (Williams, 2000).

Aims: The aim of the study is to investigate if MOI is efficient for four selected children with CP and severe SSD.

Methods: Four children with CP and moderate to severe speech sound disorder followed a single case experimental design. The children were between 5;4 and 5;8 years of age and had received years of local speech therapy with limited effect. In the current study, they received between 24 and 29 sessions of MOI with a weekly frequency of one to two sessions including homework. The intervention was carried out by a speech-language pathologist (SLP) from the cleft team. Baselines were established before intervention with three pre-tests. Intervention progress was monitored on trained and untrained consonant sounds every third session. Post-tests were carried out at the end of intervention, and follow-up-tests were done three, six, and twelve months after intervention. Phonetic transcription of the collected data was carried out by two SLPs blinded to intervention time. Percentage consonants correct (PCC) were calculated for consonants in naming tests and spontaneous speech, and inter and intra-rater reliability was calculated. Informed consent was obtained for all participants.

Results: Before intervention, the PCC score for spontaneous speech was 44% in average for the four children (range: 30-57%). At the end of intervention, the average PCC score was 66% (range: 60-68%). PCC scores increased further after intervention and reached 92% in average (range: 84-96%) at the last follow-up test. PCC scores for each child for both trained and untrained sounds in single word naming will be reported.

Summary/Conclusion: All children showed progress during the intervention period, and follow-up tests showed that the progress continued. Baseline recordings and monitoring of consonant production during intervention indicates that the progress cannot be explained by general development. Therefore, we conclude that the intervention was effective although to different degrees across participants.
Spectral tilt measurement of nasality in cleft palate speakers
I. Thammaiah¹, N. Thammaiah¹
¹JSS institute of Speech and Hearing, JSS institute of Speech and Hearing, Mysore, India

Background: Nasality is predominantly an auditory concept (Laver, 1980). Nasalization is normally achieved by lowering the soft palate, resulting in acoustic coupling between the main vocal tract and the nasal cavity. Acoustically describing nasal voice is not a question of identifying one or two parameters which are indicative of nasality.

Aims: The primary objective of the present study was to examine the spectral tilt measures (A1-P0 and A1-P1) between subjects with hypernasality secondary to cleft palate and normal speakers. The second objective was to correlate these spectral measures with the perceptual measures of nasality in cleft palate speakers.

Methods: Two groups of 30 subjects each were selected for the present study. Group 1 consisted of 15 subjects with hypernasality secondary to cleft palate and group 2 included 15 subjects with normal speech and language. Phonation samples and words from Kannada Articulation Test were recorded. The data were analysed both acoustically and perceptually, and the values obtained for the spectral measures, namely A1-P0 and A1-P1 were correlated with the perceptual ratings.

Results: It was inferred from the results of MANOVA that the subjects of group 1 showed significantly higher A1-P0 (F=5.73, p<0.05) and A1-P1 (F=3.83, p<0.05) compared to the subjects of group 2. It was also noted that there was a significant effect of vowel on A1-P0 (F=9.49, p>0.05) and A1-P1 (F=3.83, p<0.05). Further, the results also showed that the spectral parameters correlated well with the perceptual measurements of nasality among subjects of group 1.

Summary/Conclusion: Thus, it was inferred from the results of the present study that nasality directly affects both the spectral parameters considered in the present study. Further, both the spectral parameters correlated well with the perceptual ratings of nasality. Hence the parameters, namely A1-P0 and A1-P1 can be considered as sensitive and reliable measures to identify nasality in cleft palate speakers.
A multidisciplinary approach for children with cleft lip and palate: intelligibility outcomes analysis

G Toniolo¹, F Marcianet¹, F Nardit¹, K Piacentile¹, G Saia², M Brunello¹, U Baciliero¹
¹Regional Centre for Diagnosis and Treatment of Cranio-Maxillo-Facial Malformations, Maxillo-Facial Surgery Unit, AULSS 8 Berica, San Bortolo Hospital, Vicenza, ²Department of Neuroscience DNS, Maxillofacial Surgery Unit, University of Padua, Padua, Italy

Background: Cleft lip and palate is a congenital malformation, which affects many functions such as swallowing, feeding, hearing and language, therefore requiring a multidisciplinary team approach. Different surgical and speech therapy approaches have been described in the literature. The protocol considered in the present study provides for an early surgical treatment supported by an intensive speech therapy counselling focused on the family.

Aims: Investigate the quality of the phono-articulatory production of a sample of children with cleft lip/palate, who underwent the same surgical-speech therapy protocol at our Regional Cleft Centre.

Methods: A retrospective study was carried out at the Regional Centre for Diagnosis and Treatment of Cranio-Maxillo-Facial Malformations of Vicenza (Italy). The children included in the study were 102: 53 males and 49 females, born between 2004 and 2013 with a diagnosis of cleft of the soft palate (Veau type I), soft and hard palate (Veau type II), complete unilateral cleft (Veau type III) and complete bilateral cleft (Veau type IV). All of them were surgically treated by the senior author and adhered to our current surgical-speech therapy protocol. Children with congenital syndromes and/or other associated malformations and children who did not completely adhere to the protocol were excluded. Parental informed consent was obtained for all the children enrolled in the study.

The protocol adopted consists of pre-surgical orthopaedics with palatal plate (Hotz type), early palatal repair at age 3 months, cleft lip repair at 6 months and hard palate repair at 18 months. Surgery is supported by prenatal and neonatal counselling, intensive speech therapy treatment at a set up timing and/or upon request to aid suction and speech development in the early phases and continued at regular follow-up.

After reviewing each patient’s documentation (speech pathology assessments, medical and audio files), children’s intelligibility was rated employing Massari and Mazzocca’s (1985) phonetic classification of the perceptual phono-articulatory assessments carried out at 2-3 and 5-6 years of age.

Results: The analysis of the phono-articulatory production at 2-3 years of age revealed the prevalence of the second intelligibility class rate, corresponding to a globally intelligible speech. Moreover, at 5-6 years the phono-articulatory production analysis suggested a further improvement with the predominance of the first intelligibility class rate, corresponding to a normal speech, comparable to a non-cleft peer. Considering intelligibility outcomes at 5-6 years, 53% of the patients presented a normal speech, 33% a globally intelligible speech, 10% a sufficiently intelligible speech and only 4% a barely intelligible speech.

Three children presented VPI and underwent pharyngoplasty with a subsequent improvement of the phono-articulatory production.

Summary/Conclusion: The present study revealed that most of the children reached the best intelligibility rate at the last evaluation, ensuring a speech comparable to non-cleft peers, highlighting that our surgical-speech therapy protocol characterized by early intervention, monitoring, assessment and regular counselling optimizes speech intelligibility. Particularly relevant was speech therapy counselling, which guaranteed orientation, support and motivation, ensuring a context in which parents felt accompanied, supported and understood.
Nasal speech valve for patients with velopharyngeal insufficiency (vpi): an early evaluation

I Underwood1, L Cafferky2, B Richard3, R Slator4, K Khan5, S Bunn6, H Beswisk6, L Enocson7
1Speech and Language Therapy, Birmingham Women's and Children's NHS Trust, 2Speech and Language therapy, Birmingham Womens and Children's Hospital NHS Trust, 3Cleft Palate/Plastic surgery, Birmingham Womens and childrens Hospital NHS Trust, 4Birmingham Childrens Hospital NHS Trust, 5Birmingham Children's Hospital NHS Trust, Birmingham, 6Shrewsbury and Telford Hospital NHS Trust, Shrewsbury, 7Orthodontist, Birmingham Children's Hospital NHS Trust, Birmingham, United Kingdom

Background: The West Midlands cleft service receives referrals for patients with cleft and non-cleft VPI. Surgery is often the treatment of choice for patients with VPI. Where surgery is not possible; a prosthetic solution can be an alternative.

Aims: To evaluate the effectiveness of a new prosthetic nasal appliance in cleft and non-cleft patients with a structural defect and VPI.

Methods: Patients with VPI who were not amenable for surgery were informed of the prosthetic speech appliance and consent was obtained from those willing to participate in the evaluation. Perceptual speech assessment and recordings were carried out pre-valve fitting. An impression of the patient's nose was taken in order for a custom-made valve to be designed and produced. The valve was then fitted and instructions provided. The following protocol was developed:
- Follow-up telephone interviews to be carried out at one week and at 3 months post valve fitting.
- An informal speech assessment at one month and speech recording at six months post valve fitting.
- Resonance, nasal airflow characteristics and articulation were assessed using the GOS.SP.ASS and conversational speech data.

Results: Eight patient's data was collected: 1 cleft palate; 5 with structural defects due to cancer treatment; 2 non-cleft VPI. Patient feedback has indicated an increase in speech intelligibility leading to greater independence and improved quality of life. Clinical assessment post valve fitting has shown the appliance is effective in reducing hypernasality and nasal air escape in all the patients, whilst also enabling some patients to achieve improved articulatory placements. There were some issues with aesthetics and tolerance of the valve.

Summary/Conclusion: We are encouraged by our initial findings and intend to proceed with a full study and longer term follow-up including patient reported outcomes involving both patient experience and speech related quality of life outcomes. Further investigation into the potential to change speech patterns using the nasal valve in therapy will also be carried out.
Developing a procedure for estimating velopharyngeal closure based on cleft speech characteristics and its correspondence with the velopharyngeal orifice area

R Yamashita1, R Scarmagnani1, A Fukushiro12, I Trindade13

1Laboratory of Physiology, Hospital for Rehabilitation of Craniofacial Anomalies-University of Sao Paulo, 2Department of Speech-Language Pathology, 3Department of Biological Sciences, Bauru School of Dentistry-University of Sao Paulo, Bauru, Brazil

Background: Perceptually rating velopharyngeal competence based on speech symptoms is useful in clinical practice, since it allows making inference regarding velopharyngeal function when there is no access to instrumental evaluation.

Aims: To develop a procedure in order to estimate velopharyngeal closure (VPC) based on the combination of cleft speech symptoms assessed by auditory-perceptual evaluation and its correspondence with the instrumental measurement of velopharyngeal orifice area.

Methods: This study was approved by the institutional review board. Assessments were performed on 62 patients with repaired cleft palate, aged 6 to 45 years. Velopharyngeal orifice area was measured using pressure-flow technique in order to determine VPC (adequate-borderline-inadequate). Audio-visual recording of speech samples were performed in all patients. Speech material comprised 62 stimuli of 12 sentences including 12 pressure consonants produced by Brazilian-Portuguese native speakers. The recordings were edited excluding the examiner speech in order to obtain a set of 12 sentences presented as one stimulus. Hypernasality (H), nasal air emission (NAE), nasal turbulence (NT), weak pressure consonant (WPC), active non-oral errors (AE), nasal grimacing (NG) and overall auditory rating of velopharyngeal competence (VPC-R) were assessed by three experienced speech-language pathologists, first individually. Fifteen days later the raters sat together and performed a final consensus only for the inter-rater disagreements obtained in the individual assessment. H and NG were rated using a 4-point scale, and VPC-R using a 3-point scale. A single score was attributed to the set of 12 sentences. NAE, NT, WPC and AE were rated as absent and present considering 3 consonants in each sentence (total=36 consonants). The final score for each error was defined as the percentage of the affected consonants in each sentence (affected consonant x 100/36). Correlation between the ratings of each variable and the VPC determined by pressure-flow technique was analyzed by Spearman's correlation coefficient. An exploratory model was developed to estimate the VPC. Sensitivity and specificity were calculated in order to verify the clinical applicability of the model.

Results: Significant correlations were found between the VPC and each of the variables: H (r=0.765/p=0.00), VPC-R (r=0.838/p=0.00), NG (r=0.638/p=0.00), NAE (r=0.680/p=0.00), NT (r=-0.305/p=0.00), WPC (r=0.461/p=0.00), and AE (r=0.420/p=0.00). Considering the high correlations with the measurement of VPC, the final scores of H, VPC-R and NG were added resulting into a single variable SOMA. The exploratory model established the following criteria based on the presence and the proportion of each symptom in each category of VPC determined by the instrumental evaluation: Adequate VPC: SOMA=0 ou 1 and, at least 2 variables=0. Borderline VPC: SOMA=2 and NAE<2 (closer to adequate); SOMA=2 and NAE≥2; SOMA=3 and NT>0; SOMA=3 and NT=0 (closer to inadequate). Inadequate VPC: SOMA≥4. Based on these criteria, data analysis showed that the procedure correctly classified the VPC in 88.7% (55/62) of the cases. Sensitivity and specificity of the exploratory model were 96.2% and 94.4%, respectively.

Summary/Conclusion: This procedure may contribute to the diagnosis of velopharyngeal dysfunction in clinical practice. The main idea is to provide the professional with a procedure that allows estimating the velopharyngeal closure based on the speech symptoms.
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**Speech therapy evaluation of oral sensory motor functions in premature children born with cleft lip and palate**

G Zenari¹, F Nardi², T Menegus³, K Piacentile², U Baciliero²

¹Speech Therapist collaborating with the Regional Centre for Diagnosis and Treatment of Cranio-Maxillo-Facial Malformations, Maxillo-Facial Surgery Unit, AULSS 8 Berica, San Bortolo Hospital, ²Regional Centre for Diagnosis and Treatment of Cranio-Maxillo-Facial Malformations, Maxillo-Facial Surgery Unit, AULSS 8 Berica, San Bortolo Hospital, Viale Rodolfi 37, 36100 Vicenza, ³S. Bonifacio Hospital, Rehabilitation Department, Via Circonvallazione 1, 37047 San Bonifacio (VR), Italy

**Background:** Cleft lip and palate is a congenital malformation, responsible of oral, sensory-motor, feeding and language disorders. Only few studies have investigated the isolated forms associated with prematurity.

**Aims:** Evaluate the development of oral functions on premature children born with a cleft, analyze their functional alterations compared to that of non-cleft preterm, and formulate a specific protocol for speech therapy evaluation and therapy.

**Methods:** 35 preterm infants born with different types of cleft were engaged for the study and 20 non-syndromic preterm infants born without a cleft were chosen as sample control. Each child was assessed through a specific protocol at birth and later at five years old. The protocol included a medical history and a specific speech language assessment divided into six areas (breathing, interaction, attention, feeding, morpho-functional assessment of oro-facial structures and communicative-language competences).

**Results:** The data collected showed that prematurity represents a high risk for associated deficits and for alteration of vigilance and attention. In both assessments, the experimental group presented alterations typical of this congenital malformation (regurgitation through the nasal cavity and anomalies of oro-facial morphology). At five years phonetic-phonological and morpho-syntactic levels are compromised in particular in very preterm infants without a cleft.

**Summary/Conclusion:** Nowadays it isn’t possible to determine the variables that characterize the two preterm groups, but the relationship between certain risk factors (birth weight, gestational age and Apgar Index) and certain disorders (worse communicative language skills and neurobehavioural disorders) is conceivable. We propose a multidisciplinary team treatment and a direct speech therapy intervention to improve the development of oral functions and feeding by focusing on posture, improvement of sensory and motility of oro-facial structures and improvement of feeding.
7. Outcome Measurement

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Nasolabial apparence in uclp patients during childhood and at the end of the growth
V Battista¹, M Meazzini², G Rossetti¹, M Ferrari¹, L Autelitano²
¹Maxillofacial Surgery - SmileHouse, ²Ospedale San Paolo, Università degli Studi di Milano, Milan, Italy

Background: The goal in cleft treatment is to provide good results in terms of morphologic apparence, functional outcome and growth, reducing the total burden of care. Results are to be considered definitive at the end of the growth.

Aims: The aim of this study was to evaluate the naso labial appearance in a sample of UCLP patients during early childwood and at completion of growth, in order to evaluate if naso labial complex is stable during growth.

Methods: Nasolabial appearance was qualitatively assessed using the Asher McDade Aesthetic Index at 6 years old and at 20 years of age (or before secondary rhynoplasty). All of the patients were treated and evaluated at San Paolo Hospital, Smile House, Milan. 52 consecutive patients treated by the same surgeon were recalled, 12 patients didn’t come for assessment, and photograph records during childhood were available for 25 patients. The first surgical step (average age of 6 months) was cheilorhinoplasty (Millard modified Delaire technique, without septplasty) and soft palate repair (Pigott). The second step (average age of 35 months) was hard palate and alveolar repair performed simultaneously with an early secondary gengivo alveolo plasty.

Results: In this study labial appearance is quite stable during years, instead nasal appearance especially on the frontal view seems to get worse during growth.

Summary/Conclusion: Labial appearance remains stable, instead nasal symmetry worsens during growth. In order to prevent nasal deformity and to reduce burden of care in these patients we have introduced primary septoplasty, a more invasive primary rhinolasty and the use of nasal retainers. Further studies are needed to demonstrate if this surgical modifications reduce nose asymmetry and need for secondary surgery.
The orthodontic/orthognathic, speech and psychology outcomes in combined orthodontic/orthognathic treatment of cleft lip and palate (CLP) patients.

H Bellardie, V Beale, A Nur Ashikin Abd Rahman, J Davies, P Abeles, Z Foyston

1Orthodontics, University of the Western Cape, Cape Town, South Africa, 2Regional Cleft Lip and Palate Unit, 3Regional Cleft Unit, Royal Manchester Children's Hospital, Manchester, United Kingdom, 4Orthodontics, Universiti Teknologi MARA, Selangor, Malaysia

Background: Abnormal facial growth is common in CLP patients resulting in maxillary retrusion and a class III malocclusion influencing speech resonance and articulation, and masticatory efficiency. Up to 50% of cases require surgical correction by osteotomy when facial growth is complete. The orthognathic team at Royal Manchester Children’s Hospital includes a maxillo-facial surgeon, an orthodontist, a speech and language therapist and a clinical psychologist who assess and treatment plan the surgery as a team.

Aims: To investigate treatment outcomes of surgery, orthodontics, speech and psychology of 27 consecutive CLP patients undergoing orthognathic treatment.

Methods: Radiographs were traced and analysed using Dolphin imaging, the study models were scored by a calibrated observer using the PAR index pre-treatment, pre-surgery and at debond. The psychological outcomes were measured pre-surgery, 6 months and 1 year post-surgery. Patients completed questionnaires assessing anxiety (HADS-A), depression (HADS-D), Satisfaction with Appearance (SWA), orthognathic Quality of Life (QoL), and Self-Image Profile (SIP). Responses were compared across the different assessment time points using descriptive statistics.

High quality speech video/audio samples were taken pre-surgery and 1 year post-surgery. These are to be assessed ‘blind’ by an external, trained specialist speech and language therapist using CAPS-A. A real time assessment was made by the research speech and language therapist for clinical purposes using GOS.SPASS ‘98.

Results: 20 males, 7 females, 16 UCLP, 7 BCLP, 4 ICP. 100% improvement in PAR score, 100% greatly improved. Median age at surgery 20.3 years; orthodontic treatment time 18.9 months; pre- and post-treatment PAR scores 44.78 and 3.59; change in PAR score 39.95; reduction in PAR: 91.48%; Cephalometric outcome measures post surgery: SNA 81.3°, SNB 78.5°, UI/Max plane 114°, LI/mand plane 86°, mean overjet reduction 7.26mm. All 27 patients were more satisfied with their appearance (SWA), and 17 had increased quality of life (QoL) at 1yr post-surgery compared to pre-surgery. 14 remained within the ‘non-clinical’ range at 1yr; one moved from the ‘non-clinical’ to ‘mild’ range; one moved from experiencing ‘moderate’ to ‘mild’ levels of anxiety post-op, one moved from the ‘severe’ range to the ‘moderate’ range and one moved from the ‘mild’ to ‘moderate’ range of anxiety post-op. One remained within the ‘mild’ range. Two reported increased anxiety, with no impact on SWA and QoL scores. All 19 patients’ depression scores remained consistent across the time points: 18 remained within the ‘non-clinical’ range; one remained in the ‘moderate’ range. There was variety in patients’ self-image concerns at 1yr post-surgery, compared to pre-surgery: 6 self-image profile increased; 7 self-image profile score decreased

23 patients were clinically assessed 12 months post-orthognathic surgery using GOS.SPASS’98. Of the 23: 8 had an improvement in their resonance, 11 had no change, 1 became more hypernasal and 3 more hyponasal. 4 had an improvement in nasal airflow, 13 no change, 3 nasal emission was evident, 2 nasal turbulence and 1 had nasal emission and turbulence. 13 had an improvement in occlusal related errors and 10 had no change.

Summary/Conclusion: Combined orthognathic treatment was effective with all orthognathic audit standards being met.
Development and validation of an instrument to outcome measurement effectiveness of presurgical orthopedics

T Castillo

1Fundación de anomalías craneofaciales Fernando Ortiz Monasterio, Ciudad de México, Mexico

Background: Cleft lip and palate (CLP) is characterized by a lack of fusion of the facial processes. Anthropometric measures are used to evaluate the surgery outcomes and nasal asymmetry, considered the main stigma of CLP. Presurgical orthopedic has been claimed to improve this results before of primary repair lip, however, there is not instrument that measures the results of the effectiveness of pre-surgical orthopedics.

Aims: Developed and validity an instrument that outcome measurement of effectiveness presurgical orthopedics.

Methods: Validity study. Ten expert surgeon and orthodontics that have worked minimum 10 years with cleft lip and palate patients were invited to validity an instrument that outcome measurement of effectiveness presurgical orthopedics with delphi panel method.

Results: A total of 110 anthropometric measurements were divided in nasal, lip and alveolar. Only 14 measurements have 75% accord between expert for included that in to instrument: 5 nasal; culmela length, Alar projection length ,Base nasal, culmela angle, nasolabial angle, 1 lip; cleft lip and 8 alveolar; Intersegmental distance, greater distance the fissure, less distance the fissure, protrusion of premaxilla, deviation of incisor point, deviation of premaxilla, extent of posterior fissure, intraoral width.

We found 110 measurements anthropometric but only 14 have 75% accord between experts, this measurements anthropometric will demonstrate if a reliable tool for assessing effectiveness presurgical orthopedics in cleft lip and palate.

Summary/Conclusion: This study showed 75% concordance between expert in 14 measurements anthropometric for evaluate effectiveness of outcomes measurements in treatments with presurgical orthopedics, this study provides benefits that can be taken such as easy and quickly measure.

This instrument use clinical anthropometric and this is considered gold estándar, you benefits because is a method of measure easy , cheap, avoid the need for expensive and sophisticated studies

Validity of an Instrument to Outcome measurement effectiveness of presurgical orthopedics can be used for analysis and multicenter studies.
Using facial symmetry to evaluate surgery in primary cleft lip repair
A Chadha¹, R Bruggink², P Georg³, T Maal², S Berge², P Haers³
¹South Thames Cleft Service, St. Thomas’ Hospital, London, London, United Kingdom, ²UMC Radboud, Nijmegen, Netherlands, ³South Thames Cleft Service, St. Thomas’ Hospital, London, United Kingdom

Background: Several 3D Photographic biomarkers have been developed to appraise cleft lip repair representing technological, methodological and application-based advances. We present a method of symmetry calculation using 3D photographs that demonstrates novelty in terms of methodology and application. The method has the potential to not only evaluate surgical results but also to differentiate between surgical methods of repair.

Aims: This study was granted permission as a Service Evaluation from South Thames Cleft Service. Its aims were to:
1. Apply a novel cropping tool that could be adapted to regions of the face with minimal inherent errors
2. Apply an automated method for standardising facial images to expedite workflow
3. Generate an index, termed the “relative Facial Symmetry Index” capable of reflecting surgical outcomes relative to the preoperative baseline

Methods: This was a retrospective cohort study analysing routinely collected 3D photographs taken before and after unilateral cleft lip repair (with/out alveolar involvement). All procedures were undertaken by a single surgeon at South Thames Cleft Service, London. Each 3D photograph underwent a pre-processing protocol involving automated standardisation before being cropped to the nasolabial region using a standardised tool. Each photo was then analysed using a root mean square methodology to quantify facial symmetry. For each pre/post-operative pair of photographs, the relative Facial Symmetry Index was calculated and distributed graphically. Results were compared to a control age-matched sample without cleft pathology.

Results: This study involved 50 controls and a total of 35+15 non-syndromic UCL patients with a complement of pre- and post-operative 3D photographs of sufficient quality for analysis. All UCL patients had undergone primary cleft lip repair by the same surgeon. The workflow of the described method was found to greatly expedite facial analysis and to have excellent intra- and inter-rater reliability. There was a significant improvement in overall facial symmetry in the UCL cohort relative to controls (p<0.01) but only a moderate correlation between their pre- and post- surgical facial appearance (r=0.46). There was, however, a very strong linear correlation in the relative improvement in facial symmetry, as illustrated by calculation of the rFSI (r = 0.91). Manipulation of this linear relationship enabled accurate prediction of post-operative facial symmetry from pre-operative baseline in a further 15 UCL cases by the same surgeon.

Summary/Conclusion: Availing of the current 3D photographic technology, the novel workflow described represents a methodological advance in facial symmetry calculation based on a cropping tool that reduces the error associated with crops defined by anthropometric landmarks. Furthermore, the automated method of standardisation emphasises the role of the vertical median in the analysis of any facial symmetry in contrast to recent literature. Our findings demonstrate that calculation of a relative Facial Symmetry Index is a far more useful tool than absolute and isolated post-operative facial symmetry calculations and that the tool can actually be used to compare surgical procedures.
Assessment of bilateral buccinator myomucosal flaps in management of velopharyngeal insufficiency in patients with cleft palate

M Elkassaby¹, I Shibli¹, A Ghanem¹, A Sadakah²
¹Oral and Maxillofacial Surgery, ²Faculty of Dentistry, Ainshams University, Cairo, Egypt

Background: Cleft palate is one of the most common causes of velopharyngeal insufficiency (VPI). Different protocols are available for management of VPI. However, posteriorly based buccal myomucosal flaps (BMMF) have been recently in vogue both in literature and in clinical practice.

Aims: This prospective study presents the results of the use of BMMF for the treatment of velopharyngeal insufficiency and assess the quality of speech after palatal lengthening.

Methods: Twelve non-syndromic patients with repaired cleft palate (± lip) suffering from VPI not amenable for speech therapy were included in this study. Age 5-9 years, mean 7.3 years. Patients were submitted to surgical correction of VPI using BMMF under standardized settings. All patients had undergone preoperative, 6 and 12 months postoperative assessment of perceptual evaluation of the speech, nasopharyngoscopy and lateral cephalometric radiographs.

Results: Follow-up after 6 and 12 months postoperatively showed adequate improvement in perceptual evaluation of the speech, nasopharyngoscopy and lateral cephalometric radiographs.

Summary/Conclusion: The results indicate that the BMMF with palatal re-repair are reliable, reproducible, effective with least donor site morbidity and can lead to adequate speech outcomes in children with cleft palate.
Analysis of the results of primary bilateral cleft lip repair in children with congenital bilateral cleft lip and palate with protrusive premaxilla
I Fomenko¹, A Kasatkina², I Timakov²
¹-, - , ² pediatric dentistry department, Volgograd State Medical University, Volgograd, Russian Federation

Background: The number of children with congenital bilateral complete cleft lip and palate is from 12 to 25% in the structure of all congenital malformations of the maxillofacial area. The most complex group of patients is children with protrusive premaxilla. Early orthopedic treatment for patients is not always carried out. Evaluation of the results of primary bilateral lip repair in children remains an urgent problem of cleft surgery.

Aims: The aim of the study was to evaluate the results of primary bilateral cleft lip complete repair in children with congenital bilateral complete cleft lip and palate with protrusive premaxilla without a stage of early orthopedic treatment with intraoral fixation devices.

Methods: 44 children with this pathology were examined and treated. Efficacy of primary bilateral cleft lip complete repair was assessed in children aged 3-4 years before orthodontic treatment. For this, clinical, anthropometric, photometric and biometric methods of investigation were used.

Results: After a primary bilateral cleft lip complete repair in children with congenital bilateral complete cleft lip and palate with protrusive premaxilla, a good and satisfactory aesthetic and functional outcome of the operation (97.6%) was achieved without early orthopedic treatment with intraoral fixation devices. In 100% of the patients after the operation, a significant decrease in the protrusion of the premaxilla was determined. The position of the fragments of the upper jaw was not completely normalized, and the child needed orthodontic treatment in the period of bite of the deciduous teeth.

Summary/Conclusion: If there is no stage of early orthopedic treatment for children with bilateral cleft lip and palate, one-stage cheiloplasty should be performed with the basic elements of the Millard method without osteotomy of the intermaxillary bone
Changes in nostril appearance after le fort i advancement in unilateral complete cleft lip and palate
I Ganske1, R Tan2, O Langa1, C Calabrese1, B Padwa1
1Plastic and Oral Surgery, Boston Children's Hospital, Boston, United States, 2Department of Plastic, Reconstructive, and Hand Surgery, Amsterdam UMC, VU University Medical Center, Amsterdam, Netherlands

Background: Patients with unilateral cleft lip and palate (CL/P) may require Le Fort I advancement to correct class III occlusion after reaching skeletal maturity. Changes in nasal and nostril morphology following Le Fort have been well described in the non-cleft patient population. In unilateral CL/P, the underlying cleft anatomy, previous operations, and scar may affect nostril changes following Le Fort advancement.

Aims: The aim of this study is to compare changes in the nostril dimensions before and after Le Fort osteotomy in patients with unilateral complete CL/P.

Methods: A retrospective study from 2010-2014 compared patients with unilateral complete CL/P who had single-piece Le Fort I advancement to a case-matched control group without CL/P who had similar sagittal advancement. Pre-operative and post-operative 3D photogrammetry was used to measure changes in nostril morphology after orthognathic correction. Anthropometric measurements included nostril width, nostril axis, soft triangle angle, and columellar show, as well as nasal width, columellar length, and columellar width.

Results: Matched cohorts of 19 patients with CL/P and 19 patients without CL/P were identified. The mean age at time of Le Fort advancement was 18.1 +/-2.2 (range 15.0-23.0) for the cleft cohort and 18.7 +/-1.9 (range 15.0-21.0) for controls. The mean sagittal advancement was 7.5mm (range 3-14mm) for patients with cleft lip and palate and 6.28mm (range 2-11mm) for controls. There were no statistically significant differences in the nostril measurements and angles for the cleft side versus the non-cleft side in patients with CL/P. There was a tendency toward less cleft-side nostril widening in patients with CL/P compared to controls, though this did not reach statistical significance.

Summary/Conclusion: In patients with unilateral CL/P, both nostrils respond similarly to the deforming forces of maxillary advancement; thus, baseline nostril asymmetry is not made better or worse by Le Fort advancement. In patients with CL/P, maxillary advancement causes similar changes in nostrils compared to patients who do not have a history of orofacial clefting. Awareness of these changes can aid proper surgical planning for cleft nasal revisions as needed following Le Fort advancement.
Effects of multiple factors on 3d maxillary arch dimension in unilateral cleft lip and palate children: a study of two population
S Haque¹, M Khamis², M Alam³, W Wan Ahmed⁴
¹PhD student, Orthodontic Unit, ²Dean, School of Dental Sciences, Universiti Sains Malaysia, Kota Bharu, Malaysia, ³Head of Department, Orthodontic Department, College of Dentistry, Jouf University, Sakaka, Saudi Arabia, ⁴Department of Biostatistics, Universiti Sains Malaysia, Kota Bharu, Malaysia

Background: Studies have claimed that the maxillary arch dimension of unilateral cleft lip and palate (UCLP) patient are significantly smaller than normal patient. The effects of cheiloplasty and palatoplasty are believed to be the most exacerbated factors of the treatment outcome (maxillary arch retardation) of UCLP patient. However, congenital factors are also thought to have an impact on treatment outcome. A lack of consideration of factors affecting the treatment outcome of UCLP subjects has led to great diversity in protocols and surgical techniques by various cleft groups. Therefore, sound evidence of the effects of factors is required to make sure the success of the treatment outcome. 3D digital model and its analyses have been proven to be an alternative, accurate and reliable method for UCLP research compare to dental casts. However, based on literature, limited researches have been conducted using the 3D digital model in the evaluation of treatment outcome of UCLP subjects. Moreover, most of those treatment outcomes have been assessed based on dental arch relationship and also were on particular center or single based population. Yet, to the best of our knowledge no reported data till date was found for evaluation of treatment outcome by assessing maxillary arch dimension using laser scanned 3D model (LS3DM) of UCLP children between different populations.

Aims: Hence, the aim of the study was to evaluate and compare the effects of multiple factors in treatment outcome based on maxillary arch dimensions using LS3DM in Malaysian and Bangladeshi UCLP children.

Methods: One hundred and seventy (85 for each population) maxillary dental casts were taken before any orthodontic treatment and bone grafting at 5 to 12 years of age. All the dental casts were scanned and converted into LS3DM by Next Engine laser scanner and 510 linear variables [Inter-canine width (ICW), inter-molar width (IMW) and arch depth (AD)] were measured using Mimics software. Multiple linear regression analyses were used to evaluate the association between multiple factors (UCLP types, UCLP side, family history of cleft, family history of class III malocclusion, techniques of cheiloplasty, techniques of palatoplasty) and maxillary arch dimensions (ICW, IMW and AD) between two different populations. The significance level was set at p <0.05.

Results: Significant association was found between two techniques of cheiloplasty (p-value = 0.001) and palatoplasty (p-value = 0.046) with ICW in Malaysian population. Regarding Bangladeshi population, techniques of palatoplasty (p-value = 0.029) and complete type of UCLP (p-value = 0.016) showed significant association with ICW and AD respectively. Among population disparities, no significant association was found.

Summary/Conclusion: The current study suggested that the treatment outcome based on maxillary arch dimension significantly varies in individual population [Malaysian (cheiloplasty and palatoplasty); Bangladesh (type of UCLP and palatoplasty)].
The effectiveness of oral propranolol therapy for infantile hemangioma

P Kreshanti¹, N Putri¹

¹Plastic Reconstructive and Aesthetic Surgery Division, Department of Surgery, Cleft and Craniofacial Center Cipto Mangunkusumo Hospital, Plastic Reconstructive and Aesthetic Surgery Division, Department of Surgery, Medical Faculty, Universitas Indonesia, Jakarta, Indonesia

Background: Hemangioma is the most common tumor in infancy, presenting the majority of proliferating vascular tumors. The tumors that appear on the head and neck region are about 60%. A large size tumor involving the facial region will cause significant esthetic and functional impairment. Propranolol has recently been used as a choice of treatment in infantile Hemangioma.

The RCT and prospective study of the dose and duration of oral propranolol treatment are still limited. Outpatient setting for oral propranolol administration poses a challenge regarding the systemic side effects.

Aims: This evidence based case series were made to evaluate the effectiveness and safety of oral propranolol to treat infantile hemangioma, especially to reduce the tumor size on the head and neck region.

Methods: The literature were collected from Pubmed, EBSCO, and PRS Journal, and then critical appraisal were done to 5 journals which most relevant to the cases.

Results: All of study mentioned that propranolol is a good choice to treat infantile hemangioma. The dose to treat infantile hemangioma are varies; but most of the literatures indicate the initial dose to treat hemangioma on the head and neck region is 1 mg/kg/day, and then it is increased to 2mg/kg/day at one week if well tolerated. It shows significant result in tumor size reduction with minimal side effect.

Summary/Conclusion: Propranolol provides good result to treat infantile hemangioma, however infants who receive propranolol need to be monitored for potential drug toxicity. Frequent blood pressure, heart rate, and respiratory examinations should be performed to the patient who has been administered propranolol in outpatient setting. Serial photographs are also obtained to evaluate the efficacy of propranolol.
Translation and pilot study of the cleft hearing appearance and speech questionnaire (CHASQ)

M Stiernman¹, I Maulina², I Zepa², T Jagomägi³, N Tanaskovic⁴, P Knežević⁵, R Velikova⁶, Y Anastassov⁶, J Radojičić⁷, Z Pesic⁷, M Drevsek⁸, R Spataru⁹, T Boljevic¹⁰, R Dimovska¹¹, S Naumovski¹¹, N Rumsey¹², F Zucchelli¹², N Sharratt¹², M Argyrides¹³, H Svensson¹⁴, M Becker¹⁴, M Persson¹⁵

¹Department och plastic and reconstructive surgery in Malmö, Sweden, Clinical Sciences in Malmö, Malmö, Sweden, ²Riga Stradins Universtity, Riga, Latvia, ³University of Tartu, Tartu, Estonia, ⁴University of Banja Luka, Banja Luka, Bosnia and Herzegovina, ⁵University Hospital Dubrava, Zagreb, Croatia, ⁶Medical University of Plovdiv, Plovdiv, Bulgaria, ⁷University of Niš, Niš, Serbia, ⁸University of Ljubljana, Ljubljana, Slovenia, ⁹University of Medicine and Pharmacy Carol Davila, Bucharest, Romania, ¹⁰University of Podgorica, Podgorica, Montenegro, ¹¹University of Skopje, Skopje, Macedonia, The Former Yugoslav Republic Of, ¹²University of the West of England, Bristol, United Kingdom, ¹³Neapolis University Pafos, Pafos, Cyprus, ¹⁴Clinical Sciences in Malmö, Malmö, ¹⁵Höskolan Kristiandstad, Kristianstad, Sweden

**Background:** Most CL/P-centres have standardized treatment protocols for treatment indication and timing. However, different countries and health care centres have different health care resources. Access to some specialities is therefore limited or non-existing. In particular, access to a psychologist is limited at many centres. In these cases, screening and evaluation of psychological aspects of care by other clinicians with an easily administered test can be helpful so that the limited resources are used in the best way possible. There has however, been a lack of a standardized protocol for detection of and indication for psychological treatment (Klassen et al. 2012).

**Aims:** The aims of this study, which was decided during the EU-project Cost Action Appearance Matters, was to translate Cleft Hearing Appearance and Speech Questionnaire (CHASQ) into nine European languages, to investigate whether patient reported outcomes (PRO) in patients with cleft lip and/or cleft palate (CL/P) were comparable across countries and to investigate clinician experience of the instrument.

**Methods:** The PRO measure CHASQ was translated into Bulgarian, Estonian, Greek, Latvian, Montenegrin, Macedonian, Romanian, Serbian and Swedish and implemented with patients in the respective countries. A focus group discussion was conducted to investigate health care professional experience on participation in the Cost Action Appearance Matters including the strengths and challenges in the use of CHASQ. Data was analysed in accordance with the principles of thematic analysis (Braun and Clarke 2006).

**Results:** Analysis showed statistically significant differences between countries and groups of diagnosis in CHASQ scores. CHASQ helped clinicians gain patient information and informed on treatment decisions, broadened the clinicians’ role as caregivers and was perceived as short and easy to use. Limited time and resources in clinics were limitations in implementing the instrument.

**Summary/Conclusion:** Translation and utilization of CHASQ facilitated international comparison and cooperation. Linguistically valid replicas of CHASQ are now available in many European languages. Results form this study show that CHASQ may be used for screening for patient satisfaction and to spark conversation between clinicians, patients and families.
Outcomes of the Sommerlad palate re-repair for VPI treatment: pre- and postoperative analysis of speech and sleep-related disorders.
I. Trindade¹, C. Bertier², A. Silva², B. Araujo², T. Brosco², I. Trindade-Suedam¹
¹Hospital for Rehabilitation of Craniofacial Anomalies, Bauru School of Dentistry, ²Hospital for Rehabilitation of Craniofacial Anomalies, Bauru, Brazil

Background: Studies have shown that pharyngeal flap surgery, frequently used for the treatment of residual velopharyngeal insufficiency (VPI) in individuals with repaired cleft palate, besides improving speech may lead to breathing disorders during sleep.

Aims: In the present study, the functional outcomes and the associated complications of the Sommerlad palate re-repair - a less obstructive surgery, characterized by radical dissection and retroposition of velar muscles - are prospectively investigated in nonsyndromic individuals with repaired cleft palate±lip, 6 and 12 months after the re-repair.

Methods: Thirty-six individuals, aged 6 to 29 years, with mild to moderate VPI, were included in the study so far, and 11 of them already underwent the first postoperative (6mo) assessment of the surgery complications, the sleep quality, using standardized questionnaires, and the speech-related outcomes, using instrumental methods (nasometry and pressure-flow study). Informed consent was obtained from all participants (institutional ERB #1.905.404).

Results: No surgery complications such as bleeding, airway obstruction, dehiscence and fistula were observed. Regarding sleep quality: snoring was reported by 6 individuals (55%) prior to surgery, with slight worsening after surgery, and excessive daytime sleepiness was reported by 2 individuals (18%) after surgery, and breathing pauses by only one (9%). As for speech: improvement of VP function was seen in 10 individuals (91%), 8 of them (73%) showing adequate closure, and decrease in nasalance scores, suggesting decrease in the degree of hypernasality, was seen in 9 individuals (82%), without compromising pharyngeal patency (subnormal nasopharyngeal area and nasalance suggesting hyponasality were seen in only one case).

Summary/Conclusion: The confirmation of the effectiveness of the Sommerlad palate re-repair on both speech and sleep quality in a statistically significant sample will represent a relevant contribution for the cleft-related VPI treatment.
Digital protocol for analysis and treatment of functional and aesthetic problems in patients with cleft lip and palate

R Velikova¹, V Velikova², D Filchev², Y Anastassov¹, K Gigov¹, I Markova², P Tzarvulanova¹, P Petrov¹
¹Association facial anomalies, ²ALA-Association of facial anomalies, Plovdiv, Bulgaria

Background: The treatment of cleft lip and palate patients is a prolonged process involving a multidisciplinary team of medical specialists. To achieve complete rehabilitation, a planned approach is needed for these patients. Prosthetic treatment should be based on the basic principles of rehabilitation, including physiology, stability and aesthetics, responding to the patient's individual needs and expectations. The prosthetic treatment for patients with cleft lip and palate is intended to provide and support normal speech, swallowing and chewing and to provide lip isolation from the nasal cavity.

Aims: Creating a digital protocol for analysis and treatment of functional and aesthetic problems in patients with cleft lip and palate patients.

Methods: We create a digital list of questions for easier and detailed assessment of soft and hard oral tissues in cleft patients, including medical and dental history, extraoral and intraoral photos, radiology examinations. Upper and lower dental arch were scanned with an intraoral scanner; and the images are transferred into a software program for creating a digital smile design. Then measurements with a facebow was taken. The dental models were put in an articulator and a wax project was done by using the digital smile design in the frontal area and by balancing the occlusal contacts in the distal area. The digital smile design is an instrument that can significantly improve visual communication with the patient and achieve the expected clinical outcomes. There are different techniques for building a digital smile design based on aesthetic principles and rules. However, the end result of the treatment may not meet the patient's expectations due to disharmony between the smile design and the personality of the patient. The wax project can be visualized by making a silicone key and the use of resin material for temporary restorations – mock up.

Results: The Digital Question List is an internet-based platform featuring a menu with several subfields (fields): photo analysis; type of cleft; face analysis, dental, dentolabial and phonetic analysis. The questionnaire provides the opportunity to complete and save a database for each patient, to monitor the outcome of the treatment. It has been also developed a prosthetic classification of the cleft type defects, according to which variants for appropriate treatment were offered.

Summary/Conclusion: The degree of difficulty and the optimal esthetic result in prosthetic treatment are determined by various clinical parameters: face symmetry, degree of disharmony on the middle face, displaced midline. The digital list of questions helps the clinician to decide the type of prosthetic treatment and develop an esthetic, functional and individual treatment plan for patients with cleft lip and palate. According to the severity of the case and the success of the surgical and orthodontic treatments, the typical restoration can be limited to replacing the missing lateral incisor, or the restoration may require undertaking comprehensive treatments involving a much more specialized arsenal of surgical and prosthetic procedures.
8. Pediatric Dentistry/Orthodontics

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Efficacy of anterior maxillary distraction using a tooth borne device in patients with cleft lip and palate

M Cimadevilla¹, S Richardson², B Gonzalez Meli³, J Sastre⁴, A Tejero⁵, F Lobo⁶, M Mejía⁷
¹Orthodontist, Hospital Universitario Niño Jesus, Madrid, Spain, ²Maxillofacial surgeon, Richardson dental&craniofacial hospital, Nagercoil, India, ³Plastic surgeon, Hospital Universitario Niño Jesus, ⁴Maxillofacial surgeon, Hospital Universitario La Princesa, Madrid, ⁵Orthodontist, Dental School University, Valencia, ⁶Plastic surgeon, Hospital Universitario Niño Jesus, Madrid, Spain, ⁷Dentofacial Orthopedic, Nicklaus Children Hospital, Miami, United States

Background: Maxillary hypoplasia is a frequently observed problem in patients born with cleft lip and palate (CLP). Due to multiple surgeries with extensive postoperative palatal scars, they tend to have deformities to the maxillary dental arch, usually presenting class III maloclusion, and a collapsed arch form. The management of maxillary retrusion in CLP patients has been performed using Le Fort I advancement. However, this technique can alters the speech of the patients.

Anterior maxillary distraction was performed to facilitate the forward advancement of only the anterior maxillary segments without affecting the velopharyngeal function. In this study a tooth-borne distractor device was placed preoperatively before performing the osteotomy cuts.

Aims: The study was aimed at investigating the efficacy of tooth-borne distraction of the anterior maxilla and the stability of the advancement achieved over a period of 6 months in 4 patients with cleft maxillary hypoplasia.

Methods: 1 female and 3 male patients presenting CLP were enrolled with this technique; age range between 16-21 years. 2 Bilateral, 2 Unilateral CLP patients. The choice of the use of AMD was decided by the surgeons and the orthodontists, and was performed in patients that presented a mild maxillary retrusion with a potential VPI. A study cast, orthopantomogram and a lateral cephalogram were made before surgery. Lateral cephalograms were analyzed, the initial size of the maxilla and the amount of maxillary deficiency were recorded. The appliance was fabricated on the maxillary dental model by the orthodontist. The premolars and molars on either side were banded. The orientation of the hyrax screw (Dentaurum, Germany) was rotated by 90° such that its activation would result in an anteroposterior movement. A surgical specific technique was used to anterior maxillary distraction. All patients were operated on by a single cleft surgeon.

After a latency period of 5 days, distraction was performed at the rate of 3 turns twice a day. The daily movement was approximately 1.08 mm. The distraction was continued until a positive inter-incisor relation was achieved, after which the appliance was left in situ for a consolidation period of 8 to 10 weeks. After removal of the distractor, a fixed retainer was placed to settle for a period of time before orthodontic treatment.

Results: The amount of maxillary advancement was achieved with the lateral cephalometric radiograph. There was no worsening of speech according to the patients, just because the soft palate was not manipulated. Complications: None of the present patients developed any post-operative fistulas nor bleeding. Occasionaly two of them presented a debonded band of the device during consolidation period but was easily solved. 6 months followed up radiographic evaluation was made. Two patients with mild-severe maxillary retrusion did not get the goal of positive inter-incisor relationship, 2 mm. relapse was got. The other two patients did not develop any relapse and were follow up by the classic orthodontic treatment.

Summary/Conclusion: AMD using tooth borne device is effective correcting midfacial deficiencies in cleft lip and palate maxillary hypoplasia without negative effects on the patient’s speech. Good improvement in facial esthetics was found in all patients. Possible skeletal relapse in mild-severe maxillary hypoplasia using AMD.
Craniofacial and dental characteristics in patients with williams-beuren syndrome
F Danneels¹, K Indencleef², G Hens², D Declerck³, A Verdonck¹
¹Orthodontics, ²Oto-rino-laryngologie, ³pediatric dentistry, KULeuven, Leuven, Belgium

Background: Williams-Beuren syndrome is a rare disease with many medical problems and phenotypical features. Cardio-vascular abnormalities, urogenital and endocrine disorders and neurological problems are common. The average IQ is 55, they often have poor coordination and hyperflexia. Craniofacial characteristics are a wide mouth with full lips and cheeks, small jaw, long philtrum, low positioned nasal root and wide nose wings. The lips remain normally open at rest and patients with this condition often breath through their mouth. The mandibular angle is increased and they have small teeth with spacing. Agenesis and malocclusions are common.

Aims: To quantify the dental, skeletal and facial features of patients with Williams-Beuren syndrome.

Methods: Data-collection of patients with Williams-Beuren syndrome and a control group was conducted. This collection consists of 3 parts: 3D facial surface images, teleradiographics and panoramic images. 3D images were made of 17 study subjects and 33 controls with the VECTRA H1 system. This system has a submillimetric precision and a high repeatability. The pictures are made with the patients standing up, eyes straight ahead and a neutral expression. The analysis is made using a partial least square regression to investigate effects on facial morphology. Cephalometric measurements were made of 6 patients with Williams Beuren syndrome and 8 controls. The measurements consist of a Steiner, Downs and Tweed analysis using Vistadent AT3.1 software (GAC International). Panoramic images were analyzed of 12 patients of the study group.

Results: The 3D image analysis shows us a significant difference between the study group and the control group. The largest effects are seen in the mid-face area, around the nose and lips. This confirms the previous studies and hypothesis. The panoramic images and cephalometric measurements are still being analyzed.

Summary/Conclusion: There are significant craniofacial features in patients with Williams-Beuren syndrome. This can be very important and useful in early diagnosis and treatment of these patients.
The presence of clavicles: a dental examination leading to a diagnosis of cleidocranial dysplasia
C Hardwick1, N Atack1, C Burren1, R John1
1University of Bristol Hospital Trust, Bristol, United Kingdom

Background: Cleidocranial dysplasia is a rare autosomal dominant condition with an incidence of 1:1000000 affecting both skeletal and dental development. Typical signs are the absence of clavicles, delayed fontanelle closure, delayed eruption of the adult dentition and multiple supernumerary teeth. Patients may also suffer from osteoporosis, hearing loss and respiratory infections.

Aims: This case aims to highlight the importance of referring any dental and craniofacial abnormalities to multi-professional teams to allow for a holistic, patient centred approach to their management.

Methods: A 10 year old female was referred by her general dental practitioner regarding multiple un-erupted permanent teeth. She had a history of pneumonia and failed medical referrals to specialist geneticists regarding cleidocranial dysplasia due to her presence of bilateral clavicles. Extra oral findings included joint hyper-mobility, shortened fifth phalanges and frontal bossing. There was also a history of delayed fontanelle closure. Intra oral examination revealed an early mixed dentition, fibrous gingivae, high arched palate and generalised hypo-mineralisation. Radiographic investigations demonstrated multiple supernumerary teeth in all four quadrants of the mouth. These clinical findings prompted a referral to a paediatric endocrinologist.

Results: After genetic profiling and medical examinations a diagnosis of Clediocranial Dysplasia was confirmed. The patient was managed using a multi-professional approach with co-ordination between paediatric dentistry, oral surgery, orthodontists and endocrinologists. After caries management, initial surgical treatment included removal of the anterior supernumeraries, with exposure of the permanent incisors, allowing their eruption. Pre-molar region supernumeraries were subsequently planned for extraction with the provision of fixed orthodontics post eruption.

Summary/Conclusion: This case highlights the importance of holistic dental examinations prompting medical referrals to aid with craniofacial diagnoses.
Applicability of a mathematical equation in tooth size prediction in a sample of Egyptian individuals with cleft lip and palate

M Helmi¹, N Abd El-Rahman², M El-Kassaby³, N Metwalli¹
¹Department of Pediatric Dentistry and Dental Public Health, ²Department of Orthodontics, ³Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Ain Shams University, Cairo, Egypt

Background: Cleft lip and palate (CLP) disorders are regarded as one of the most common birth defects with multiple detrimental medical, social and psychological effects on both the patients and their families. They result from lack of fusion between embryonic processes resulting in discontinuity of the facial structures with esthetic and functional impairment. These abnormalities include –but are not limited to- hindered maxillofacial growth, speech abnormalities, deglutition difficulties, dental abnormalities and malocclusions which require the coordinated interdisciplinary efforts of multiple specialists such as orthodontists, pediatric dentists, oral and maxillofacial surgeons, speech therapists and many more to mitigate the detrimental effects of these disorders.

Mixed dentition space analysis is an orthodontic diagnostic method that allows early detection of signs of arch length discrepancy which is inevitable in individuals with CLP, therefore it is extremely important. However, racial differences of tooth sizes prevent the use of the standard prediction equation developed for Caucasians by Tanaka and Johnston in other populations. This prompted authors to develop similar prediction equations individualized for other populations. However, there haven't been enough studies that investigate whether these equations can be used in tooth size prediction in individuals with CLP.

Aims: The aim of the study was to evaluate the applicability of a mathematical linear regression equation in predicting the combined mesiodistal widths of canines and premolars in a group of Egyptian individuals born with different types of CLP

Methods: Retrospective study was conducted at the Faculty of Dentistry, Ain-Shams University, Egypt. The data was collected from 66 cone beam computed tomography (CBCT) scans (33 males and 33 females) of individuals with CLP categorized into 22 CBCT scans of individuals with unilateral complete cleft lip and palate (UCLP), 22 CBCT scans of individuals with bilateral complete cleft lip and palate (BCLP) with well-developed premaxillae and 22 scans of individuals with bilateral complete cleft lip and palate with rudimentary premaxillae. Mesiodistal widths of maxillary and mandibular permanent canines and premolars were measured using InVivoDental® Ver. 5 software. Later, the combined mesiodistal widths of canines and premolars were calculated using the tested regression equation to be later compared to those measured directly from the scans using Student's paired t-test.

Results: The tested regression equations were found to have variable statistical significance in the studied groups. The differences were statistically significant in the maxillary arches of UCLP males, maxillary cleft sides and mandibular arches of UCLP females. The same was found in the maxillary arches of BCLP males with rudimentary premaxillae and the mandibular arches of BCLP males and females in the same group, as well as both arches of BCLP females with well-developed premaxillae. However, the differences in all groups were found to be clinically insignificant.

Summary/Conclusion: The studied prediction equations developed for the Egyptian population can be used in prediction of mesiodistal widths of canines and premolars in Egyptian individuals with different types of complete CLP as the differences between actual and predicted values didn’t exceed the threshold for clinical significance in all studied groups.
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**Palatal cyst in cleft lip and palate: a case report**  
A Indania¹, P Kreshanti², F Fardizza³, M Mihardjanti⁴, M Saraswati⁵  
¹Medical Faculty, Universitas Indonesia, ²Plastic and Reconstructive Division, Department of Surgery, ³Department of Otolaryngology, Cipto Mangunkusumo Hospital, ⁴Cleft and Craniofacial Center, Cipto Mangunkusumo Hospital, ⁵Department of Anatomical Pathology, Cipto Mangunkusumo Hospital, DKI Jakarta, Indonesia

**Background:** Palatal cyst is a rare condition that affects the hard palate. Only few cases palatal cysts were reported and none were reported in cleft lip and palate patients. The most probable differential diagnoses of palatal cysts include cyst of odontogenic and non-odontogenic origin. Knowledge regarding the workup is essential to provide insight for further follow up and treatment.

**Aims:** We present a case of a 4-month-old male presented to outpatient clinic with left unilateral complete cleft lip and palate and a soft mass in the hard palate.

**Methods:** Soft tissue excision was conducted to enucleate the cyst for histopathological examination. Furthermore, literature searching was conducted through various online databases using keywords such as “Cleft Palate”, “Palatal Cyst”, “Dentigerous Cyst” and “Neurofibroma”.

**Results:** Macroscopically, the extirpated cyst revealed a primary incisive tooth with thick and whitish cream-like consistency. The histopathological examination revealed hypocellular tumor cells with unclear margins and myxoid stroma. The tumor cells have relatively uniformed spindle-shaped core with fine chromatin thus was concluded to have the closest resemblance to neurofibroma. The presence of tooth in the cyst would suggest the diagnosis of dentigerous cyst. Despite dentigerous being the most prevalent type of odontogenic cyst and associated with the crown of an unerupted developing tooth, it is commonly seen in either the mandibular third molar or the maxillary canine and rarely in other teeth. This type of cyst is also rarely found in children younger than 10 years old. In regards to the histopathology examination, neurofibroma is rarely found in intraoral. No other cases were reported for neurofibroma in the palate. However, a study presumed that the origin of the tumor may arise from branches of the fifth and/or seventh cranial nerve. To confirm this diagnosis, we performed immunohistochemistry examination with awaiting results. It may be suggested that the patient has dental growth anomalies that may be associated with cleft palate. Dental growth anomalies can be found in isolated cleft palate patient with tooth agenesis being the most prevalent anomaly. However, the presence of a primary tooth in the palatal cyst may suggest an abnormally located tooth follicle. To confirm this diagnosis, a panoramic radiography is needed to visualize teeth follicles that have yet to erupt. However, this examination may be challenging to be conducted in infants as it requires the patient to maintain a still sitting or standing position for a period of time.

**Summary/Conclusion:** Follow-up examinations are warranted to confirm the diagnosis of palatal cyst which may determine patient’s treatment plan.
Characteristics of cleft lip and palate patients treated in a university orthodontic setting

E Koumpia¹, A Athanasiou ², L Angelis³, M Papadopoulos¹

¹Department of Orthodontics, Dental School, School of Health Sciences, Aristotle University of Thessaloniki, Thessaloniki, Greece, ²Department of Dentistry, School of Medicine, European University Cyprus, Nicosia, Cyprus, ³Department of Informatics, Aristotle University of Thessaloniki, Thessaloniki, Greece

Background: Cleft lip (CL) with or without cleft palate (CL±P) is the most common human birth defect found among living subjects. Such defects are manifested as unilateral or bilateral and they can be non-syndromic or syndromic, with the latter associated with more than 400 syndromes. Epidemiological studies carried out in different regions or countries have shown that the incidence of non-syndromic oral clefts varies greatly with ethnicity, race, gender and type of cleft as well as with the season and year of birth.

Aims: The study aims to explore the characteristics of cleft lip and/or palate patients that received orthodontic treatment at a university clinic, to investigate the prevalence of different cleft types and sex distribution, as well as assess the prevalence of CL±P in relation to gender and to the agricultural occupation of the parents.

Methods: Data were collected from patients’ files of consecutively treated patients at the Clinic of Craniofacial Anomalies, Department of Orthodontics, Xxxxxxxx University of Xxxxxxxx, Xxxxxxxx. Each patient was informed according to the World Medical Association Declaration of Helsinki ethical principles for medical research involving human subjects and provided with a written informed consent. Descriptive statistics were used to explore the distribution of cleft types with the respect to gender and to the parents’ and grandparents’ occupation on agriculture. The statistical significance of the association between cleft types and gender was tested using confidence intervals and the chi-square test.

Results: Cleft lip and palate was the most common defect (92%) followed by cleft lip (6%) and cleft palate (2%). Left-sided CL+P was approximately 1.5 times more common (40%) than right-sided (26%), and more males were affected by CL+P than females, especially left-sided CL+P (chi-square p=0.044, Fisher p=0.065). Independently of cleft type, it was found that 66% of the CL±P patients with information concerning grandparents from father’s side were farmers. On the mother’s side 48% of fathers and 50% of mothers had agricultural occupation.

Summary/Conclusion: The CL±P patients’ characteristics found in this study are in accordance with previous reports, and do not present differences from other populations worldwide.
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**Novel three stage approach in pre-surgical nasal molding on an infant with unilateral cleft lip**

P. Tang, W. Ricky, K. Liu

**Background:** Pre-surgical therapies have been used to mold the maxillary, alveolar and soft tissues of infants with unilateral cleft lip deformity. The conventional naso-alveolar mold (NAM) described by Grayson and Maull in 2004 consisted of an acrylic alveolar plate and stainless steel wire for alveolus and nasal molding respectively. However, the oral plate of the NAM is often poorly tolerated in growing infant, resulting in suboptimal compliance.

**Aims:** Herewith we described our novel pre-surgical nasal molding techniques in an infant with unilateral cleft lip.

**Methods:**

1. **Stage 1:** A tailor made acrylic plate with **two extending arms** was used on an infant within the first week of life. The additional arm was believed to provide a more active pressure to promote the downward movement of the deviated premaxilla and to reduce the lip gap.

2. **Stage 2:** After sufficient downward movement of the pre-maxilla, the next stage of treatment would aim to centralize the deviated columella. Multiple tapes were used in an **asymmetrical** manner to apply active pressure onto the nasal alar of the non-cleft side in order to push the nose back towards the center of the philtrum.

3. **Stage 3:** After the lip gap was reduced significantly and the columella has been pushed almost back to the center of the philtrum, the last stage would involve the use of a novel **extra-oral mold** consisting of stainless steel wire bent according to the curvature of the philtrum. The aim of the extra-oral naso mold was to increase the nostril height and to reduce the nostril width simultaneously at the cleft side.

**Results:** Details of the outcome are shown in figure 1 to figure 5.

**Summary/Conclusion:** The malleability of cartilage was reported to be due to the high maternal osteogen concentrations in the newborn's circulation during the first two weeks of life, implying the time sensitive nature of early treatment planning and the importance of early patient compliance. By adopting our step-wise three stage approach, we are able to produce the desired effects:

1. Reduction of the soft tissue lip gap
2. Centralization of the deviated columella
3. Increase of the cleft sided nostril height

while minimizing discomfort to the baby. We are still in the very preliminary phase in implementing these innovative pre-operative therapies and long term data of its efficacy is lacking. However, the short term outcome is encouraging and we believe these techniques would remain complementary in the future, especially in patients with poor tolerance to the traditional NAM.
Use of the goslon index for the assessment of patients with cleft lip and palate
A Tejero¹, J Gandía¹, M Osca¹, M Cimadevilla², M Mejía³, B González Meli⁴, J Rodríguez de Guzmán¹, F Gómez⁵
¹Orthodontics, University of Valencia, Valencia, ²Orthodontics, Hospital Niño Jesús, Madrid, Spain, ³Orthodontics, Miami Children Hospital, Miami Florida, United States, ⁴Plastic Surgery, Hospital Niño Jesús, Madrid, ⁵Maxillofacial Surgery, Hospital Universitario La Fe, Valencia, Spain

Background: Orofacial clefts are one of the most frequent defects of the newborn. The GOSLON index allows us to categorize the dental relations of these patients in order to evaluate and compare the results of different therapeutic approaches.

Key words: cleft lip, cleft palate, orthodontic index, malocclusion.

Aims: The aim is to determine which type of orofacial cleft is the most predominant, as well as the most frequent type of malocclusion and if there is an association between both sexes. We want to evaluate the severity of the malocclusion by applying the GOSLON index and determining its reliability.

Methods: 100 patients with cleft lip and palate from the Master of Orthodontics of the University of Valencia and the Department of Plastic Surgery of the Hospital Universitario del Niño Jesús in Madrid were selected and divided according to the type of cleft they presented. The 65 plaster models of patients with unilateral cleft lip and palate were analyzed and classified into one of the 5 groups of the GOSLON index.

Results: Unilateral cleft lip and palate was the most predominant. The prevailing malocclusion was Class III and no significant differences were found between both sexes. The GOSLON 3 was the most found. The application of the GOSLON index has turned out to be reliable and trustworthy.

Summary/Conclusion: The GOSLON index is a reliable method that allows us to categorize in an easy way the different malocclusions derived from a unilateral cleft of the lip and palate in order to evaluate the effectiveness of different therapeutic approaches.
9. Other

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Smile house roma reference centre for cleft lip and palate assessment from prenatal diagnosis to the term of the growth: a new concept of the integration with public health system.

P Arangio¹, D Scopelliti¹, R Orsini¹, S Marrocco¹, O Cipriani¹, G Amodeo¹, A Carboni¹, A Agrillo¹

¹Maxillo-Facial Surgery, Ospedale San Filippo Neri - Smile House Roma, Roma, Italy

Background: The Operation Smile foundation is a multinational non-profit organization that deals with the treatment of cleft patients in developing countries. In Italy in collaboration with the NHS and with the Government of the Regions has opened centers of excellence for the treatment of cleft lip and palate from the prenatal period until the end of growth. The patient and his family are in the center of a comprehensive multidisciplinary assessment: maxillofacial surgeon, plastic surgeon, pediatrician, anesthesiologist, resuscitator, pediatric intensivist, nurses, dentist, speech therapist, psychologist

Aims: On the basis of the results obtained from the Milan Smile House, we present the results obtained in the Roma Smile House from December 2017 to today with particular regard to the number of outpatient visits and surgical procedures performed both in primary cases and in secondary surgery.

Methods: Thanks to the agreement between the NHS and the non-profit organization Operation Smile Italia, it has been possible to create a unified multidisciplinary treatment center for cleft lip and palate within the San Filippo Neri Hospital Presidium, Asl Rm1 of Rome.

The multidisciplinary cooperation between the hospital specialists involved is complemented by the clinical experience in the cleft field of the Operation Smile Onlus Foundation. Furthermore, a training program is planned for the components of the maxillo-facial, anesthesiological, intensive care, pediatric, dental and nursing teams for the continuous updating of professionals.

Results: Following the path already implemented by Smile House in Cagliari and Milan, which was established in 2011, has already performed 17,000 visits and over 1000 surgeries, the Smile House Roma was inaugurated in December 2017 and the new Department opened in October 2018. We performed 10 surgery sessions. We treated 19 of primary surgery involving lip and palate and 43 case of lip revision. There were no surgical complications and all patients were discharged the following day. Furthermore in the Smile House Roma a total of 1020 visits were performed in one year divided in multidisciplinary visit (average 15 patients/month), odontoiatric and orthodontic (average 40 patients/month) visits and postop control (average 30 patients/month).

Summary/Conclusion: International guidelines and review of protocols recommend that clinical post-surgical results improve if performed in a single center and by the same team and decrease the number of complications. The concept of Smile House, unique in Italy because it follows the patient affected by prenatal diagnosis until the end of growth, aims to continuously improve the treatment protocol of patients with cleft and unify the treatments of excellence in a single center. It also represents a single point of support and assistance for the patient and the family for centre and south of italy.
The mucosa of the oblique vomer lacks specific signs of nasal mucosa
A Mueller1, B Benitez1, A Brudnicki2, J von Jackowski3, E Bruder4
1Cranio-Maxillofacial Surgery, University and University Hospital Basel, Basel, Switzerland, 2Department of Surgery of Children and Adolescents, Institute of Mother and Child, Warsaw, Poland, 3Department of Biomedical Engineering, University Basel, Allschwil, 4Institute of Pathology, University and University Hospital Basel, Basel, Switzerland

Background: The mucosa overlying the oblique Vomer part (“portion du vomer incurvé”, Veau 1931) in unilateral complete has a similar color as nasal mucosa. Common cleft surgical techniques (e.g. two-flap palatoplasty) therefore use this tissue exclusively to reconstruct the nasal cavity which in turn may lead to tissue shortage for oral layer closure. We search for nasal mucosa specific histologic findings on the vomer mucosa and we study the clinical findings of the vomerine mucosa when partially used for oral layer closure.

Aims: We checked histologically if the vomerine mucosa of the oblique Vomer does show specific signs of nasal mucosa.

Methods: We report a case series of 12 patient, in which the mucosa over the oblique Vomer part was split in two for a balanced use to close the nasal and oral mucoperiosteum. In four patient, a piece of Vomer tissue was studied by histology (stainings of HE, PAS and Alcain) at the time of surgery at 8 month. After 3 to 6 month postoperative, the vomerine mucosa in the oral cavity was documented for visible changes.

Results: All histology results exhibited a stratified squamous epithelium with numerous seromucous glands. Some areas showed palisade arrangement of the basal cells as in metaplasia of respiratory epithelium. However, the search for goblet cells and respiratory cilia was negative. Abundant fibrosis was present. The findings were compatible with oral mucosa as well with squamous metaplasia of nasal mucosa. Postoperative, the vomerine mucosa that healed into to the oral cavity changed its color from dark red to pink, identical to the adjacent masticatory mucosa.

Summary/Conclusion: The histology of the mucosa over the oblique Vomer indicates a strong regenerative and adaptive change of the mucosa, but without clear signs for oral or nasal mucosa. Despite the use of a palatal plate, that separated the Vomerine mucosa from mechanical irritation of the tongue from birth to the time of surgery, the mucosa did not transform into typical nasal mucosa. The Vomerine mucosa should be regarded as a cleft specific wide histologic “transition zone” between typical oral and nasal mucosa. If vomerine mucosa is exclusively used for nasal layer closure, this should be justified other than on its histology.
Treatment outcome of 4-7 years-old patients with cleft lip and cleft palate in Tawanchai Center, Srinagarind Hospital: trend of treatment

W Chantachume¹, C Songsung¹, S Pradubwong¹, B Chowchuen¹
¹Faculty of Medicine Khon Kaen University, Srinagarind Hospital, Khon Kaen, Thailand

Background: The highest incidences of cleft lip and cleft palate are in the Northeast.
Aims: To study the trend and to compare the number of patients with cleft lip and cleft palate receiving treatment at Tawanchai Center, Srinagarind Hospital.
Methods: It was a descriptive study from the medical records of the patients admitted to Tawanchai Center between the ages of 4 – 7 years from 2009-2011. The research instrument was a questionnaire on general information and hometown. Percentage was employed for data analysis.
Results: It was found that there were 125 patients with cleft lip and cleft palate, 81 of them were males (64.8%). According to demographic study, most of the patients or 25 of them (20%) lived in Roi-et, followed by Loei and Sakonakorn with 13 patients each (10.4%), and 11 patients lived in Khon Kaen (8%). Regarding hometown areas, it was found that 106 patients (84.8%) lived outside the city or Muang districts. On the contrary, 19 patients (15.2%) lived in Muang districts. With regard to Health Area, the study revealed that 60 patients (48.8%) lived in Health Area 8, followed by 51 patients (41.5%) in Health Area 7.
Summary/Conclusion: According to the study of the treatment of 125 patients with cleft lip and cleft palate between the ages of 4 – 7 years, the number of the patients tended to decline but likely increase in hometown areas, especially outside of Muang districts. In addition, the number of patients living outside Health Area 7 was likely to increase. Therefore, providing information on the rights of treatment to the patients, families, and general people, will allow more patients to access specialized care centers.
Parents’ experiences of early cleft care – a survey across five European countries

G Davies1, I Akota2, I Bagante3, L Linkeviciene3, M Bigaki4, H Vasilika5, K Bojikova6, N Hashova6, T Nilson7, M Persson7, M Ivanov8, R Spataru8

1Executive Director, European Cleft Organisation, Rijswijk, Netherlands, 2Surgery, Riga Stradins University, Riga, Latvia, 3Orthodontic Department, Vilnius University, Vilnius, Lithuania, 4Project Manager, 5Nurse, Papageorgiou General Hospital, Thessaloniki, Greece, 6Nurse, Asociatcia Licevi Anomali, Plovdiv, Bulgaria, 7Pedagogy, Triskelion, Stavanger, Norway, 8Paediatric surgery, Spitalu Clinic de Urgenta Pentru Copii “M.S. Curie”, Bucharest, Romania

Background: The survey was conducted to inform the development of a course, the Early Cleft Care Programme (ECTP), for nurses and front line health professionals working with babies with clefts. In 2015 the European Cleft Organisation finalised a set of Early Care Cleft Guidelines that were endorsed by the European Committee for Standardisation (CEN). Subsequent to this, in 2017, ECO was awarded a grant by the EU Erasmus+ programme to lead a consortium of six partners to develop the ECTP course.

Aims: In preparation for developing the course material a survey of parents experiences was conducted across five of the project’s partner countries: Bulgaria, Greece, Latvia, Lithuania and Romania. The families were asked about their, and their child’s needs, following birth and diagnosis. The results of the survey would inform curriculum development.

Methods: The survey was developed by all partners. Family background information was followed by questions relating to: quality and sensitivity of communication, clarity of information, information needs, feeding support, overall satisfaction with care and affordability of treatment. Additionally a focus group meeting took place in Plovdiv, Bulgaria attended by parents, members of the cleft team an external facilitator. The participants came up with a set of common themes that needed addressing.

Results: There were in total 94 respondents from the five countries, broken down as follows: Bulgaria 17; Greece 16; Latvia 23; Lithuania 20; Romania 18. Six families participated in the focus group meeting in Bulgaria. The results were summarised under six main headings: communication, understanding of the diagnosis, feeding, meeting with team, overall treatment satisfaction and, finally, cost and affordability. In terms of communication, around a quarter of families felt that the health professionals giving information after diagnosis were poor communicators and that facts were not presented in a clear and positive manner. On feeding, around a fifth of respondents were not given any information, nearly a half were not shown how to feed their baby and three quarters were not given any special bottles or equipment to feed their child. It was clear that most distress faced by parents is during the period between diagnosis (before or after birth) and the eventual meeting with the cleft team.

Summary/Conclusion: The provision of accurate information communicated in a clear, and positive manner at point of diagnosis is crucial. Handled badly, the effects can be devastating and in some cases lead to abandonment. Front line health professionals must have a basic knowledge of clefts and treatment pathways. There must be rigorous referral mechanisms ensuring referrals are made to the right place and specialist at the right time. The provision of feeding advice in the early hours and days is essential. Written information needs to be available, with staff rapidly at hand, to give practical feeding support in the maternity units. There needs to be better communication with, and training for, gynaecologists, obstetricians, neonatologists, nurses, midwives, and social workers.
Treating a person with a cleft, not the patient's cleft
R. Dei Castelli, R. Dei Castelli
I.O.E., Formosa, Argentina

Background: Cleft lip and palate is a very often pathology in our region. Sometimes they get a diagnosis before the child is born but most of the time due to social reasons they get the diagnosis during the delivery. We need to treat not a child but the whole family.

Aims: Show how better are our results when we work on a team

Methods: Compare different techniques with a long time follow up

Results: Working on a team have better results than treating just the cleft by a surgeon

Summary/Conclusion: The treatment of a cleft should begin intra uterine. With an early diagnosis we can teach the parents how to front face the disease. As a team we treat more than 200 patients a year. Every patient is special and have his own treatment.
Nasal speech valve for patients with velopharyngeal insufficiency (vpi): manufacturing of the appliance
S Bunn¹, H Beswick¹, L Cafferky², I Underwood², L Enocon³
¹Maxillo Facial Prosthetics, Shrewsbury and Telford Hospital NHS Trust, Shrewsbury, ²Speech and Language Therapy, Birmingham Womens and Childrens NHS Foundation Trust, ³Orthodontist, Birmingham Childrens Hospital NHS Trust, Birmingham, United Kingdom

Background: The West Midlands cleft service receives referrals for patients with cleft and non-cleft VPI. When surgery is not possible; a prosthetic solution can be an alternative.

Aims: To describe a new prosthetic appliance to eliminate VPI in patients with a structural defect in cleft and non-cleft patients. This presentation is to demonstrate impression and manufacturing technique for this appliance.

Methods: Patients with VPI that were not amenable for surgery were invited to be part of an evaluation of a new prosthetic speech appliance to help reduce VPI. An impression of the patient’s nose and nostrils was conducted with light body silicone. The Maxillofacial Prosthetists cast the impression in Crystacal Stone and modify the internal lateral walls. A wax replica of the one piece valve is created from which the final Odontosil 50 shore silicone nasal speech valve is produced and then activated.

The custom fitted valve was fitted with instructions. Follow-up interviews at one week and 3 months. Follow-up speech assessment at one and six months.

Results: Initial results are encouraging. Most patients accept wearing the silicone appliance.

Summary/Conclusion: A simple but effective nasal speech valve in design and fabrication, requiring one appointment prior to fit.

We aim to follow up the long term wear and tear of the appliance and report on patient quality of life outcomes to determine what patient’s best benefit from this appliance
Geospatial epidemiology of cleft lip with/without palate as an alternative population approach, case colombia
S Guarnizo-Peralta¹, H Rengifo-Reina¹, E Suarez-zuñiga¹
¹Nacional University of Colombia, BOGOTA, Colombia

Background: The current clinical attention approach in congenital anomalies focus on the creation of excellent inter and multidisciplinary reference centers. However, the reality of people with cleft lip with/without palate is overflowing with great diversity of access barriers including limitations in health access and discontinuity in health services. For that reason, it is important to understand which is the real dimension of the event (magnitude and frequency) and to design specific and integral strategies according to people's needs. Nevertheless, traditional public health approach starts from inconclusive and individual diagnoses, which it do not satisfy the population diagnosis needs, as require all complex phenomena, such as congenital anomalies and the cleft lip and palate.

Aims: To describe the event of cleft lip with/without palate using geospatial epidemiology strategies in Colombia.

Methods: It is a mixed ecological exploratory study. It includes a comparative analysis of several sources of information in the country (the National Oral Health Study, the Colombian health information system of the Ministry of Health and Social Protection, the Public Health Surveillance of the country, the Registry of Service Providers, and public environmental information of the country). The data included national registries and reports between 2010 and 2015 from the Public Health Surveillance System (SIVIGILA), the Individual Healthcare Provision Records (RIPS) and the official environmental behavior reports. The statistical analysis was performed by the software Stata version 13, Microsoft Excel 2016 and geospatial with Epi Info ™ 7 and ArcGis 10.5.

Results: In this study, the national prevalence of oral cleft in Colombia is 2.78x10,000 inhabitants with an incidence of 5.47x10,000 births. Most of the cases are in the mountain range and the main centers of attention are in capital cities. Additionally, there are spatial-temporal coincidences of environmental risk factors such as illegal mines and coca crops and people with cleft lip with/without palate.

Summary/Conclusion: The geospatial approach in people with cleft lip with/without palate gives advantages over the traditional approach of the event. The traditional epidemiology provides limited information, which it does not satisfy population needs in a complex approach. Geospatial epidemiology allows better approximation of the event from a population perspective. It helps with the formulation of public policies, localization of reference centers, monitoring routes of people attention and control of areas with the greatest incidence. Whereas, the use of traditional methodologies reveals a lack of national information about the event. Then, a positive guidance makes an impact on HealthCare Access and the types of services provided, making the difference between a correct rehabilitation and social integration or a simple surgical correction without sequelae management.
Overview of patients born with a submucous cleft palate
C Havstam\textsuperscript{1,2}, L Nyman\textsuperscript{2}, C Persson\textsuperscript{2}
\textsuperscript{1}Speech-Language Pathology, Sahlgrenska University Hospital, \textsuperscript{2}Speech-Language Pathology, Sahlgrenska Academy at University of Gothenburg, Gothenburg, Sweden

Background: The key anatomical symptoms of a submucous cleft palate are a bifid uvula, a thinness in the midline of the velum and an indentation in the posterior midline of the hard palate. In some individuals, this results in velopharyngeal incompetence (VPI). For those who do suffer from VPI, different surgical methods are used and previous research has shown little evidence to support any specific surgical intervention.

Aims: To present an overview of number of patients, age at diagnosis, any additional syndrome, number of operations and outcomes of primary palatal surgery in patients born with submucous cleft palate at Sahlgrenska University Hospital in Gothenburg, Sweden.

Methods: A chart review encompassing the years 2004-2017 was performed with a search for the ICD10-diagnosis Q353A. A re-analysis of available pre- and postoperative recordings of the patients who had undergone primary palatal surgery was performed blindly and independently by three speech-language pathologists specialized in cleft care. They assessed velopharyngeal function on a three-point scale (competent, marginally incompetent or incompetent). Statistical comparison of pre- and postoperative speech results was performed with the Wilcoxon signed rank sum test.

Results: 68 patients (39 males, 29 females) born with a submucous cleft palate were identified. Their mean age at diagnosis was 7.6 years, ranging from birth to 56 years. Eight individuals were diagnosed with a syndrome; five had 22q11-deletion syndrome, one had van der Woude syndrome, and one had partial trisomy. 37 out of 68 patients (54\%) had undergone primary palatal surgery and 18 of them had additional palatal surgery due to unsatisfactory results of the first operation; 13 patients had two operations, four patients had three and one patient had five operations all in all. The additional surgical methods used were palatal re-repair (n=7), pharyngeal flap (n=12), fistula closure (n=4), cartilage transplant (n=1) and fat injection (n=1). Re-analyses of the 20 patients with pre- and postoperative speech recordings from their primary operation revealed no statistically significant differences. Five individuals improved their velopharyngeal function, nine were unchanged and six deteriorated.

Summary/Conclusion: Patients born with a submucous cleft palate form a heterogeneous group. Surgical outcomes indicate a need for revision of surgical methods but available data was incomplete. Suggested modifications will be discussed.
Association between maternal smoking and laterality of orofacial clefts
T Kruse¹, E Mangold², B Braumann¹
¹Department of Orthodontics, University of Cologne, Cologne, ²Institute of Human Genetics, University of Bonn, Bonn, Germany

Background: Epidemiological evidence suggestive of the origins of the laterality of orofacial clefts is sparse. Whereas gender-specific incidences hint at the existence of endogeneous factors (1), little evidence exists concerning exogeneous factors determining the laterality of orofacial clefts. The association between maternal tobacco smoking and cleft lip and palate has been shown repeatedly (2). Following this association and arguments related to asymmetric facial artery development (3), it seems possible that maternal smoking leading to an additional vasoconstriction may affect the laterality of orofacial clefts.

Aims: Aim of this study was to test if maternal smoking is associated with the laterality of cleft lip (CL) and cleft lip and palate (CLP). In a second step, we wanted to know if this association is the same for males and females.

Methods: 605 mothers of 233 girls and 372 boys with nonsyndromic unilateral CL/CLP were interviewed after they had given informed consent to participate in the study. 191 mothers were classified as smokers during the first trimester of pregnancy. The data entail 605 infants with an unilateral CL/CLP, 407 among them being left-sided (67.3 %) and 198 being right-sided (32.7 %). To assess differences in laterality between the different groups two-sample t-tests were used.

Results: The share of left-sided clefts was higher in boys than in girls (70.7 % vs. 61.8 %, p=0.02549), aligning with previous findings (4,5). The share of left-sided clefts was smaller among children of smoking mothers than of non-smoking mothers (58.3 % vs. 69.1 %, p=0.04057). In other words, smoking was positively associated with unilateral CL/CLP being right-sided. Gender-specific analyses revealed that this association only held among girls (41.5 % vs. 67 %, p=0.0039) but not among boys (68.7 % vs. 70.4 %, p=0.7772).

Summary/Conclusion: Our results suggest that maternal smoking, as an exogenous factor, affects the laterality of CL/CLP differently for boys and girls. A better understanding of the laterality of orofacial clefting may help us to reveal the mechanisms underlying teratogenesis.

**Epidemiology of cleft lip/palate and cleft palate in Switzerland: data from the Swiss Cleft Registry**

G La Scala¹, E Dorie¹, C Staudt², W Gnoinski³, I Schnyder⁴, J Hohlfeld⁵, M Tönz⁴, A de Buys Roessingh⁵, O El Ezzi⁵, J Kuttenberger⁶, A Müller⁷

¹Pediatrics (Pediatric Surgery), University of Geneva Hospitals, Genève, ²Orthodontics, Center of Dental Medicine, University of Zürich, ³Limmat Center for Cleft Lip and Palate, Zürich, ⁴Pediatric Surgery, InselSpital, University of Bern, Bern, ⁵Pediatric Surgery, CHUV, University Center of Pediatric Surgery of Western Switzerland, Lausanne, ⁶Clinic for Oral and Maxillofacial Surgery, Cantonal Hospital, Luzern, ⁷Oral and Maxillofacial Surgery, University of Basel, Basel, Switzerland

**Background:** National epidemiologic data on facial clefts was lacking in Switzerland. A Swiss National Cleft Registry was created in 2011, enrolling patients born with a cleft lip/palate.

**Aims:** Collect accurate Swiss national epidemiological data on cleft lip and palate (types of cleft, incidence, associated anomalies, potential risk factors and possible clustering of cases).

**Methods:** Inclusion criteria:
- Children born since 01.01.2011 in Switzerland with a cleft lip ± palate or a cleft palate.

Exclusion criteria:
- Complex facial clefts
- Children followed in Switzerland but living abroad and for whom the mother spent the first trimester of pregnancy outside Switzerland.

The study is conducted with the support of the Swiss Association for Cleft Lip and Palate and Related Craniofacial Anomalies, with the participation of all the member centres.

In each centre, the responsible physician obtains parental consent usually at the time of the preoperative consultation and records on a specific form administrative data for the child and the parents, the details of the cleft, any associated anomalies / syndromes, pregnancy and family history.

**Results:** 633 cases were enrolled from 1.1.2011 to 15.10.2018. 57.2% of patients have a cleft lip ± palate, 42.8% a cleft palate. Data is still collected, therefore results suffer of an under-reporting bias.

In Switzerland, overall oral cleft incidence from 2011 to 2017 (592'813 live births) is at least 10.0/10'000. We noted a wide variation (0 to 19/10'000) in regional incidence, studying the regions where the patients’ mother spent the first trimester of pregnancy. Cleft lip ± palate incidence is 5.7/10'000 with a 1.83 M/F sex ratio. Cleft laterality is 48% left, 29% right, 23% bilateral. Cleft palate incidence is 4.3/10'000; 63% soft and hard palate, 37% involving only the soft palate, with a 0.8 M/F sex ratio. Pierre Robin sequence is present in 18% of patients with cleft palate. Overall, congenital anomalies are reported in 19.9% of patients. Anomalies are mostly craniofacial (26%) or cardiovascular (26%) and are more frequent in cleft palate patients (27% vs. 15% for cleft lip ± palate). Syndromes are noted in 9.6% of patients (21% van der Woude, 13% 22q11⁻) and are present in 14.4% of cleft palate and 6.1% of cleft lip ± palate patients. Prenatal ultrasound identified 66% of cleft lip ± palate but only 6% of cleft palate patients. 3.3% of mothers and 4% of fathers have a cleft, with a positive family history for a cleft in 11.4% of mothers and 10.4% of fathers. Congenital anomalies (clefts and others) are present in the family history in 29% of patients. The study and data analysis are underway; updated and more detailed results will be presented.

**Summary/Conclusion:** This study provides the first national data on the epidemiology of cleft lip and palate in Switzerland. While it suffers from an under-reporting bias as case registration depends on the goodwill of centres’ professionals and on parental consent, it provides important data on cleft epidemiology and identifies areas with higher incidence of this congenital anomaly.
The cleft collective: a unique set of cleft cohort studies recruited from centres across the UK
S University Of Bristol1, K Humphries2, A Davies2, Y Wren2, J Sandy2, E Stergiakouli2, G Sharp2
1Population Health Sciences, 2Bristol Dental School, University of Bristol, Bristol, United Kingdom

Background: Funded by The Scar Free Foundation, The Cleft Collective is one of the largest multidisciplinary cleft lip and palate research programme to date. Despite being one of the most common birth anomalies, little is known about the causes of cleft lip and/or cleft palate (CL/P). Treatment involves a considerable burden of care from birth onwards, together with a variety of social and psychological challenges.

Aims: The study addresses three key questions commonly posed by parents of a baby born with a cleft:
- What has caused my child's cleft?
- What are the best treatments for my child?
- Will my child be OK?

Methods: Capitalising on the centralisation of cleft care in the UK, this research is carried out in close collaboration with each of the NHS cleft centres and over 100 maternity units. Children and their families are being recruited to the Cohort Studies by centralised cleft teams; Biological samples and questionnaire data are collected from all family members. In setting up the study, the research team has worked in partnership with patients, representative organisations, Clinical Research Networks (CRNs) and experts in the field, including nurses, psychologists, speech and language therapists, surgeons, orthodontists, geneticists and midwives. This has ensured that research decisions are informed by clinical experience, existing literature and patient perspectives.

Results: Recruitment commenced in December 2013 and is ongoing. We are currently recruiting from all 16 cleft teams and to date 7482 individuals (mothers, fathers, affected children, siblings) from 2667 families have been recruited into the study, with around 80% of these individuals donating blood, tissue or saliva samples from which DNA has been extracted. We have recently extended our recruitment to include babies whose clefts are detected by antenatal screening and we are collecting cord blood from these babies. Extensive phenotype data is available from questionnaires, surgical forms, patient notes, and detailed speech phenotypes are available from a subset of the cohort. In addition we plan to link to external sources such as the National Pupil Database, NHS Digital and the CRANE database.

Summary/Conclusion: By working closely with cleft teams and maternity units across the UK we have created a unique cohort of children born with a cleft and their families, with questionnaire data and biological samples which can be used by academics, clinicians and trainees to address the questions pertinent to those affected by CL/P.
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**Visiting homes and renovation of houses of children with cleft lip and palate by multidisciplinary team in Thailand**

S Maneeganondh¹, A Theeyoung¹, S Pradubwong¹, N Patjanasootorn¹

¹Faculty of Medicine Khon Kaen University, Srinagarind Hospital, Khon Kaen, Thailand

**Background:** Children with cleft lip and palate common born in poor families, cause of lack of opportunities in psychosocial development and living in poor conditions.

**Aims:** To visit and renovate their houses.

**Methods:** The descriptive and qualitative studies of children with cleft lip and palate living in KhonKaean, Thailand and being patients of Tawanchai center of treatment of cleft lip and palate and craniofacial anomalies of Srinagarind hospital, faculty of Medicine, KhonKaen University, Thailand. After visiting home and evaluation the conditions by multidisciplinary team, and selecting the houses with seriously poor conditions need to renovate immediately. There were 4 houses in this survey need to reconstruction. After permitting to report from Institutional review broad (HE591110) the data were collected in 1. visiting homes evaluation 2. Interviewing datas, 3. planning and processing in house renovations 4. content analysis for learning in many problems in process of renovations.

**Results:** 4 patients and their houses were choosing by team for immediately to renovate, age between 2-12 years old. 3 of 4 were male, number of family members living in a house was 6-12 persons, living expense were not enough and had debt for all 4 families. Total number of traveling to visit was 8 times, in 10 homes. Number of meeting with community leadership, social security departments was 6 times, number of telephone calling and e-mail for processing renovation houses was 15 times. 3 house were renovated, 1 house need to make a new one. With a good corporation in many government and non government organizations such as Tawanchai foundation, Local administration organization, provincial administration organization, Vocational collage of KhonKaen for supporting funds and house builders to make satisfaction, succession and well being in in the mission for these children.

**Summary/Conclusion:** Visiting homes and renovation houses is not simply process, need many afford to accommodation to complete the missions. Teams need to include multidisciplinary team who good in communication, leadership, organization, and strenuous. The Families and community trust need to be created and sharing opinion.
Children's and parent's perception and experiences of hospital care at the plastic surgery department, Sahlgrenska university hospital

A Paganini, M Millback, J Sahlsten Schölin, C Nielsen

Plastic Surgery, Sahlgrenska University Hospital, Göteborg, Sweden

Background: When a child is cared for in a hospital designed for adults, the natural access to an environment adapted and designed for children's needs is not present. Hospital admission can be frightening for children, so therefore healthcare professionals need to be aware of this in order to create a child-friendly setting. The demands on doctors, nurses and assistant nurses are high, in order to make the child's meeting with the health care environment should be as calm and adapted as possible. The children and their families should feel safe and well cared for by professionals in order to facilitate the hospital stay. At the pediatric plastic surgery department at Sahlgrenska University Hospital, approximately 550-600 children/year are treated between 0-18 years of age. The major patient groups that's treated at the department is CLP and craniofacial malformations including craniosynostosis.

Aims: To explore children's and parents' experience of being cared for at a children's ward in an adult hospital

Methods: A structured work have been performed during several years, using a multitude of methods ranging from the Sahlgrenska adaptation to value based care as well as interviews of families. At the end of 2017, a questionnaire (modified National Patient Survey, SKL) was also sent out to 80 children and parents between 0-16 years of age who had been cared for at pediatric plastic surgery ward. The study was conducted retrospectively between year 2016 and 2017. Data from children and parents interviews was used and followed up by a modified National Patient Survey.

Results: The response rate on the survey was measured at 60% and in summary, the results are better than the national average. The questionnaire consists of eight different domains, seven of which were used in this study; Information, Availability, Participation, Treatment, Overall Impression, Recommend and Trust. Eg in the domain Information a slight difference was noted in the correlation between preoperative information given and the postoperative outcome in regards to surgical results, pain and nutrition. The results have been used to develop a local health care quality register, aiming to follow up participation, treatment and information.

Summary/Conclusion: The local health care registry is implemented and a valuable tool for feedback from children and parents. It gives an immediate response as how the care is provided and the possibility to improve for instance information channels, such as Facebook and websites. Furthermore, a web application based on the registry is under development.
Non-syndromic cleft lip and/or cleft palate: epidemiology and risk factors in Lubumbashi (DR Congo), a case-control study.

M Sébastien¹, H Reychler², K Devriendt³, P Kalenga⁴, P Lukusa⁵, F Tshilombo¹, M Kashal¹, T Kayembe⁶

¹Pediatric and maxillofacial surgery, University of Lubumbashi, Lubumbashi, Congo, The Democratic Republic of the, ²Maxillofacial surgery, Université catholique de Louvain, Brussels, ³Centre of human genetics, KU Leuven, Leuven, Belgium, ⁴Gynecology, University of Lubumbashi, Lubumbashi, Congo, The Democratic Republic of the, ⁵Genetics, KU Leuven, Leuven, Belgium, ⁶Toxicology, University of Lubumbashi, Lubumbashi, Congo, The Democratic Republic of the

Background: Orofacial clefts are the most common congenital malformation of the head and neck region. Their prevalence is approximately one in 700 live births. A multifactorial cause is most common, with an interaction of multiple genetic and environmental factors.

Aims: To determine the incidence and risk factors of occurrence of non-syndromic cleft lip and/or cleft palate (NSCLP) in Lubumbashi.

Methods: A case-control study was conducted in the health district of Lubumbashi from February 2012 to December 2015. An exhaustive sampling, collecting all newborns with cleft lip and/or cleft palate (CL ± P) in maternity wards was conducted. From a total of 172 cases, 162 non-syndromic cases were recruited. For each case, one clinically normal newborn control was selected.

Results: Non-syndromic cleft lip and/or cleft palate P had an incidence of 1/1258 live births (0.8/1000). We found significant associations with a family history of cleft lip and palate (CLP) (χ² family history = 11.5, p = 0.0007), maternal alcohol intake (OR = 19.3, 95% CI: 1.9-197.1), paternal alcohol during the periconceptional period and the first trimester of pregnancy (OR = 18.7, 95% CI: 3.9-89.2), maternal educational level lower than high school (OR = 9.5, 95% CI: 2.0-44.7), clay (Pemba) consumption during pregnancy (OR = 38.3, 95% CI: 9.3-157.0), the use of insecticides in the evening (OR = 130.3, 95% CI: 13.2-1286.9), indoor cooking with charcoal (Makala) (OR = 6.5, 95% CI: 1.22-34.5), and regular consumption of Kapolowe fish, supposedly contaminated with heavy metals (OR = 29.5, 95% CI: 7.4-116.7).

Summary/Conclusion: Several environmental risk factors highly prevalent in Central Africa for facial clefting were found.
Appearance-related gender differences in patients born with uclp
A Paganini¹, H Mark¹, M Persson²
¹Plastic Surgery, Sahlgrenska University Hospital, Göteborg, ²Faculty of Health Science, University of Kristianstad, Kristianstad, Sweden

Background: Gender based differences in medical treatment have been recognized, due to conscious or unconscious perceptions i.e. gender-bias. There has been shown differences in surgical cleft treatment between genders, as well as indications that women in general are more dissatisfied with their appearance than men. However, the results regarding gender differences in patients with CLP the literature are scarce and contradictory.

Aims: The aim of this study was to investigate demographic gender differences in a consecutive population of patients born with UCLP and investigate self-reported appearance-related gender differences in the same population using the DAS24.

Methods: The study sample consisted of 180 consecutive patients, 58 women and 122 men, with unilateral cleft lip and palate and no associated syndromes or malformations, born 1966 to 1986 and treated at Sahlgrenska University Hospital. All patients were asked to participate in the study and to answer a set of questionnaires, including demographic data and the DAS24.

Results: 81 patients participated and the results show that statistically significant gender differences occurred in the indicators income, living arrangements, alcohol consumption and number of esthetic surgeries. Women also reported a significantly higher appearance related social anxiety and social avoidance than men. The aspects of appearance that was most disturbing was cleft-related among both genders, where dissatisfaction with the nose was most common.

Summary/Conclusion: This study indicates that gender influences appearance related dissatisfaction in patients with CLP, as well as in regards to demography. Further studies are needed to see the impact of gender in relation to CLP and appearance.
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**Positive impact on parents’ anxiety of a specialized nurse in a multidisciplinary team in the context of a child with cleft lip or palate**

M Perrot¹, G La Scala¹

¹Pediatric Surgery, HUG University Center of Pediatric Surgery of Western Switzerland, Genève, Switzerland

**Background:** The medical history of a child with a cleft lip or palate is often long and stressful since his birth it may be the cause of high anxiety for parents.

In order to improve our quality of care, a specialized nurse joined the multidisciplinary team in 2013. Initially the nurse was mostly in charge of follow-up coordination, but rapidly a specific consultation was implemented to allow a personalized follow-up of the child and his family with the reference nurse.

This consultation is based on a global family approach, considering the patient and his family as partners with resources, and uses concepts of the therapeutic education; it allows the families to identify their difficulties, mobilize their resources and strengthen the link and the trust towards the multidisciplinary team.

The follow-up begins in the prenatal period if applicable or postnatally once the cleft has been diagnosed. The following appointments are flexible and depend on the family needs. Surgery is a critical step for the parents, we help them identifying their needs before the procedure, they visit the surgical ward, the operating room area and post-anesthesia care unit, and we begin the therapeutic teaching for the postoperative care. The day of surgery the specialized nurse is in charge of the child on the ward and is a resource for the family during the whole admission. The nurse plans and organizes with the family the discharge and the follow-up appointments. Once the child returns home the nurse is available by e-mail or telephone in case of questions.

**Aims:** Evaluate the impact of a specialized nurse’s care on parental stress and anxiety at the time of surgery admission for cleft surgery.

**Methods:** Data regarding parental stress/anxiety about the hospital admission for cleft surgery was prospectively collected at 3 to 6 weeks after the operation in a single center. We compared the period before and after the implementation of care with the specialized nurse. Parents evaluated their stress and anxiety on a 0-5 Likert scale.

**Results:** Data was available for 54 consecutive families from January 2012 to April 2015, 27 before and 27 after the implementation of specialized nurse care. The median level of stress/anxiety reported before implementation was 5 (IQR 4-5) and afterward was 2 (IQR 2-4) \((p<0.0001)\).

**Summary/Conclusion:** This study shows a significant decrease in the parental anxiety with a nurse-coordinated global family approach within the multidisciplinary team. This result encourages us to pursue the development of our project and underlines the importance of having a specialized nurse involved in the inpatient and outpatient cleft children care.
Gender differences in the prevalence of clefts by using an embryologically based classification for clefts: a retrospective cross-sectional study from 2006 to 2016
S Pool1, L van der Lek2, K de Jong3, C Vermeij-Keers4, C Mouës5
1Plastic Surgery, University Medical Center Groningen, Groningen, 2Medical Center Leeuwarden, Leeuwarden, Netherlands, 3Epidemiology, Medical Center Leeuwarden, Leeuwarden, 4Dutch Association for Cleft Palate and Craniofacial Anomalies, Netherlands, 5Plastic Surgery, Medical Center Leeuwarden, Leeuwarden, Netherlands

Background: Traditionally, facial clefts are divided into two categories: cleft lip with or without cleft palate (CL ± P), and cleft palate only (CP) or into three categories: CL with or without alveolus (CL ± A), CLAP, and CP. Using these classifications may hamper further knowledge on the causes and treatment of clefts, since important subphenotypes like palatal hypoplasia, submucous/subcutaneous and incomplete clefts are often not distinguished. A validated new classification system, however, divides all subphenotypes into categories based on underlying developmental mechanisms, i.e. fusion and differentiation, and their timing, i.e. early and late periods, in embryogenesis of the primary and secondary palates. During the early embryonic period, only the fusion defects of the primary palate (e.g. complete CL) develop. In the late embryonic period both the differentiation defects of the primary palate (e.g. incomplete CL), and the fusion and differentiation defects (e.g. (in)complete and submucous CP, respectively) of the secondary palate occur1.

Aims: The aim of our study was to define the gender differences for the subphenotypes in newborn cleft children in the Netherlands.

Methods: This was a retrospective cross-sectional study on children with facial clefts born from 2006 to 2016. Children with a syndrome, sequence or association, children with non-Caucasian or consanguineous parents, and adoptive or foster children were excluded. Data were provided by the Dutch Association for Cleft Palate and Craniofacial Anomalies that records anonymously cleft subphenotypes and other craniofacial abnormalities through a validated system. Clefts were classified in early (E-), late (L-) and early/late (EL-) embryonic periods, in primary (P-), secondary (S-) and primary/secondary (PS-) palates, and further divided into fusion (F-), differentiation (D-) and fusion/differentiation (FD-) defects, respectively.

Results: In total 2089 children were analysed: 1311 males and 778 females. Cleft subphenotypes in females occurred significantly more often in the L-period compared to males (66% vs. 55%, p=0.000), whereas clefts in males significantly more frequent occur in the EL-periods compared to clefts in females (40% vs. 27%, p=0.000). In addition, females had significantly more often S-clefts compared to males (42% vs. 23%, p=0.000), while males significantly more often had PS-clefts compared to females (44% vs. 30%, p=0.000). Furthermore, the clefts in females were significantly more often the result of a F-defect compared to males (60% vs. 52%, p=0.000).

Summary/Conclusion: Clefts in females mainly occur in the L-period, are most often S-clefts only, and are usually the result of a F-defect. Compared to females, clefts in males more common occur in the EL-periods, are therefore more often PS-clefts, and are less frequently the result of only F-defects.

Evaluation of hub and spoke model of cleft care in Liverpool, UK
M Seshu¹, G Hitchens¹, M Cooney¹, S Dominguez-Gonzalez¹, J Russell¹, C Penfold², L Crowder¹
¹orthodontics, Alder Hey children’s hospital, UK, Liverpool, ²maxillofacial surgery, Glan Clwyd Hospital, Rhyl, United Kingdom

Background: Liverpool (Hub unit) is a part of the North West and Isle of Man Cleft network and provides outreach service to North Wales (3 Spoke units). Primary surgery is centralized to the Hub but orthodontics is carried out locally. The three spoke units in North Wales - Rhyl, Wrexham and Bangor covers a population of around 690,000 spread across 2500 square miles.

Aims: This service evaluation was carried out to compare the services in the Hub and spoke units as well as outcomes. The comparison parameters were divided into 4 domains – location and travel to clinics, patient assessment at time points as per the cleft protocol, alveolar bone grafting and orthodontic outcomes.

Methods: Patients who had alveolar bone grafting in 2006-2014 carried out by a single surgeon were identified in the database. The Hub has digital records and the Spokes have paper records. Data was collected so that each patient journey could be mapped from birth till present for this cohort. This data was collated with outcomes to complete the assessment. Patient journey and timelines were mapped for around 80 patients treated by the team.

Results: It was found that on average patients travelled 9-10 miles to attend clinics in the Hub but 23-25 miles for the Spoke units. Clinicians travelled an average of 60 miles (one way) to attend the outreach clinics. There was good compliance with the cleft protocol at the different time points during treatment.

The ABG outcome, Kindelan score 1, was achieved for majority of patients irrespective of where the surgery was carried out. Canine eruption took an average of 44 months after bone grafting for patients treated in Hub but the Spokes showed a wide variation. Percentage of cases of who had pre surgical orthodontics was similar but there were significantly more number of orthodontic treatments carried out in 1 Spoke.

Most of the patients have comprehensive orthodontic treatment and PAR scores were similar for Hub and Spoke units. Orthodontic treatment was carried out locally for Welsh patients to reduce the burden of care with liaison with the orthodontist from the hub unit.

Summary/Conclusion: There were differences in service provision between the Hub and Spoke units but the outcomes were similar. In spite of clinicians travelling long distances for the Spoke clinics in 3 different locations across North Wales, patients have to travel longer to attend clinics, which reflects on the geographical area covered.
Human umbilical blood lineage-negative stem and progenitor cells as potential adjuvant therapy for cleft lip and palate surgical treatment protocol

A Zawiślak¹, M Kawa¹, S Wnęk¹, D Rogińska¹, E Paczkowska¹, B Machaliński¹
¹General Pathology Department, Pomeranian Medical University, Szczecin, Poland

**Background:** Orofacial cleft is one of the most common facial birth defects and represents a significant public health problem. Its treatment requires compound and comprehensive surgical, orthodontic, dental, phoniatic, and psychological management strategies. While surgical intervention is crucial for whole treatment process, functional closure of the cleft palate and esthetic reconstruction of nasolabial components still represent enormous challenge for pediatric surgeon. Cleft lip and palate reconstructive surgery protocol demands several steps. It begins at 6-month old with rhinocheiloplasty and soft palate closure and later involves the hard palate surgery, alveoloplasty, and secondary rhinoplasty. New possibilities of using selected stem and progenitor cells from the umbilical cord blood are discussed to improve final surgical results. Human umbilical cord blood is an easily obtainable and rich source of stem cells. We hypothesized that population of umbilical blood-derived lineage-negative stem and progenitor cells due to their potential to release numerous trophic factors related to connective tissue growth and development could play a role in partial recovery of both, fibrous and osseous compartments, in therapeutically reconstructed cleft lip and palate.

**Aims:** The aim of this study was to determine whether human umbilical cord blood lineage-negative cells are able to produce noticeable concentrations of trophic factors related to connective tissue, including osseous and chondral tissue compartments, that could have practical implication for stem cell-based adjuvant therapy to repair orofacial clefts.

**Methods:** We investigated the expression of the several trophic factors, such as FGFs, IGFs, HGF, OPTGRN, TGFs, MMPs and angiopoetins or interleukins in lineage-negative stem and progenitor cell population isolated from human umbilical cord blood under steady state conditions. The expression of investigated trophic factors was detected by QRT-PCR, western blotting or immunofluorescence. Gene expression profiles of lineage-negative stem cells were determined using genome-wide RNA microarray technology.

**Results:** Expression of trophic factors related to connective tissue at both the mRNA and protein level was observed. Global gene expression analysis revealed considerably higher expression of genes associated with the production and secretion of proteins, migration, proliferation, and differentiation in lineage-negative cells than in other cell populations from human umbilical cord blood.

**Summary/Conclusion:** Our study shows that umbilical blood cord lineage-negative stem and progenitor cells could express trophic factors related to connective tissue metabolism, growth and development under steady-state conditions. Therefore, there is a noticeable need for further dedicated research in this field. These findings may indicate the possible role of human stem and progenitor cells in the new therapeutic procedures for regeneration of connective tissue components in reconstructed orofacial clefts in addition to performed surgical procedures.
Language and neurodevelopmental measures in 7-9 week old infants with isolated oral clefts
A Conrad¹, K Wermke², E Kuhlmann¹, M Eisenmann²
¹Pediatrics, University of Iowa, Iowa City, United States, ²Center of Pre-Speech Development & Deve. Disorders, Bavarian Julius-Maximilians-University, Wurzburg, Germany

Background: Previous neuroimaging research has demonstrated significant structural and activation differences between children and adults with isolated oral cleft of the lip and/or palate (iCL/P) and age-matched unaffected controls. However, there remain questions as to how much these differences may be due to developmental differences, increased exposure to anesthesia, or other medical experiences.

Aims: The purpose of this research study was to evaluate the earliest markers of language functioning and neurological development in infants with isolated oral cleft of the lip and/or palate (iCL/P).

Methods: Participants were recruited through advertisements and clinic visits at a local mid-western university for this case/control study. A total of 8 participants (4 unaffected and 4 with iCL/P), ranging from 1.68 to 2.66 months age, were enrolled and completed at demographic and pre-speech language measures. A subset of 6 (4 unaffected and 2 with iCL/P) successfully completed structural MRI scans. Main outcome measures included: Family functioning ratings, social-cognitive development ratings, vocal recordings, and structural MRI.

Results: Given the highly novel method of obtaining unseated MRI scans on infants (7-9 weeks old), the sample size is limited. There were no significant differences in family or cognitive functioning between groups. Infants with cleft demonstrated a different rhythmicity (had a higher rate of oscillatory breaks) in their vocalizations. Descriptive data by participant group are provided for family functioning, cognitive functioning, language development, and structural brain measures (gray and white matter volume and cortical thickness).

Summary/Conclusion: The findings of this study provide a foundation from which to build further research on the neural-structural development of infants with oral clefts: the need to evaluate measures of cortical development, inclusion of information on anesthesia exposure and oxygenation, and suggestions for avoiding identified pitfalls BLOCKS to obtaining data.
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