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Current Awareness Bulletin – Cleft lip and palate
April 2015

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Guidelines

Depression in children and young people: Identification and management in primary, community and secondary care
NICE guidelines [CG28] Published date: March 2015

Cochrane Systematic Reviews

New Reviews – March 2015

Non-speech oral motor treatment for children with developmental speech sound disorders

Cleft Palate-Craniofacial Journal – Latest Issue

Cleft Palate-Craniofacial Journal
ISSN: 1055-6656 Latest issue available from Allen Press in Journals@Ovid (Athens Authorization)

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Titles highlighted in green may be of particular interest to Speech and Language Therapists
Titles highlighted in orange may be of particular interest to Clinical Psychologists

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1. Title: A retrospective study on a hospital-based cleft care center in Shanghai.


Author(s): Wang, Daohe, Xu, Haisong, Qi, Fazhi, Chen, Yang, Gu, Jianying

Abstract: The objective of this investigation was to describe the characteristics of the current cleft treatment situation in a hospital-based cleft center in Shanghai and provide references to clinical diagnosis, treatment, and nursing. A total of 1584 patients from the Center for Cleft Lip and Palate, Department of Oral and Cranio-Maxillofacial Science, Ninth People’s Hospital, Shanghai Jiao Tong University School of Medicine during June 2006 to February 2008 were analyzed retrospectively. Data regarding sex, native place, type of cleft, cleft side, accompanied malformations, family history, and age at surgery were analyzed in detail. Length of stay after surgery, the primary operation fee, and some other hospitalized information were also investigated. From 1584 patients (1590 operations; 6 patients had 6 operations), there were 939 male and 645 female patients (M: F = 1.46: 1). The number of Shanghai local patients is 249 (15.72%), whereas the other 1335 patients were from out of Shanghai. Approximately 15% of the patients had certain family history. The age at operating varied from 2 months to 36 years; the mean value was 6.95 years. The postoperation hospital stay varied from 1 day to 15 days; the mean value was 5.54 days. The primary operation fee was 235 to 673 USD depending on the different surgical procedures. The number of cleft types or other malformation, which had not been treated in the statistics varied from zero to 3; the mean value was 0.4375. The cleft morphology was classified as follows: cleft lip, 591 cases (37.31%); cleft palate, 651 cases (41.10%); alveolar cleft, 144 cases (9.10%); facial traverse cleft, 27 cases (1.70%); velopharyngeal insufficiency, 105 cases (6.63%); velocardiofacial syndrome, 57 cases (3.60%); and Pierre Robin sequence, 15 cases (0.95%). In all the classifications, left was more than right (L: R = 2.10 : 1). As a busy hospital-based cleft care center, most of the patients are from out of Shanghai. The current multidisciplinary protocol for cleft care in such specialist cleft center is cost-effective. There may be a tendency that the patients with cleft palate are more than the patients with cleft lips in recent years, which may due to the popularization of prenatal examination in China.

Source: Medline

2. Title: A survey of assessment and management of velopharyngeal incompetence (VPI) in the UK and Ireland.

Citation: Journal of Plastic, Reconstructive & Aesthetic Surgery: JPRAS, Apr 2015, vol. 68, no. 4, p. 485-491 (April 2015)

Author(s): Hodgins, N, Hoo, C, McGee, P, Hill, C

Abstract: A questionnaire designed to survey methods of assessment and management of velopharyngeal incompetence (VPI) was circulated to all surgical members of the Craniofacial Society of Great Britain and Ireland. 45 questionnaires were distributed yielding 30 respondents (66.6% response rate). 27 respondents performed surgery for both cleft and non-cleft forms of VPI. Multi-planar videofluoroscopy and nasendoscopy were the most frequently used methods of assessing and diagnosing VPI. The most frequently utilised corrective surgical procedure was palatal re-repair, followed by the Hynes pharyngoplasty and the Furlow double opposing z-plasty technique. Orticcohea and Jackson pharyngoplasties were less commonly performed. Splitting the palate during pharyngoplasty procedures was an inconsistent practice. 20 palatal surgeons referred patients to an otolaryngology specialist when tonsillectomy and/or adenoidectomy was indicated prior to their corrective palatal procedure. This was most frequently performed 3 months prior to palatal surgery. This survey identified normal patient speech as the criterion of success after corrective surgery for VPI. Assessment and management of VPI in Great Britain and Ireland is a highly varied practice. When a palatal procedure is indicated, surgical approaches are tailored to address each individual’s pattern of velopharyngeal closure deficiency. The surgeons surveyed reported this as being most accurately demonstrated by direct nasendoscopic visualisation and dynamic multi-planar videofluoroscopic studies. Copyright © 2014 British Association of Plastic, Reconstructive and Aesthetic Surgeons. Published by Elsevier Ltd. All rights reserved.

Source: Medline

3. Title: Absent maxillary lateral incisor as evidence of poor midfacial growth in unilateral cleft lip and palate.

Citation: Oral Surgery, Oral Medicine, Oral Pathology & Oral Radiology, 01 April 2015, vol./is. 119/4(392-395), 22124403

Author(s): Hardwicke, Joseph, Chhabra, Purnima, Richard, Bruce

Abstract: OBJECTIVE: The absence of a maxillary permanent lateral incisor in patients with unilateral cleft lip and palate (UCLP) may affect the dental arch relationship. An analysis is performed to investigate the relationship between the maxillary-mandibular dental relationship and the status of the maxillary permanent lateral incisor. STUDY DESIGN: Patients with non-syndromic UCLP were analysed using the GOSLON Yardstick to assess maxillary-mandibular dental relationship on pre-expansion orthodontic study models. Absence of the permanent upper lateral incisor on the cleft side was assessed from medical records and dental radiographs. RESULTS: A total of 83 subjects
were identified: 54 males and 29 females, with 54 having a missing lateral incisor on the cleft side. There was a significant relationship between the GOSLON Yardstick score and the absence of the maxillary permanent lateral incisor on the cleft side (p<0.05). Of those in GOSLON categories 4 and 5, 78% had a missing lateral incisor compared to 42.3% in GOSLON 1 and 2. CONCLUSIONS: Missing lateral incisor may be an indicator of severity of dental arch discrepancy.

**Publication type:** journal article  
**Source:** CINAHL

4. **Title:** Analysis of alar balance in secondary cleft lip with z-plasty technique of nasal cartilage.  
**Citation:** The Journal of craniofacial surgery, Mar 2015, vol. 26, no. 2, p. 354-357 (March 2015)  
**Author(s):** Yang, Junyi, Chen, Lingfeng, Wu, Yingzhi, Mu, Xiongzheng  
**Abstract:** The cleft nasal deformity is a prevailing problem of complex challenge for plastic surgery, especially in the secondary correction. We mainly chose 40 patients with unilateral secondary cleft lip nasal deformity with alar collapse. Based on biomechanics and anatomy of nasal cartilage, we adapt a Z-plasty with cartilage mucosa using the deformed lateral crus of the upper lateral cartilage to support the collapse of lower lateral cartilage. All of our patients were satisfied with the aesthetic morphology after surgery, so we are confident that this method should be considered as an auxiliary treatment to rhinoplasty.  
**Source:** Medline

5. **Title:** Asymmetric Maxillary Protraction for Unilateral Cleft lip and Palate Patients Using Finite Element Analysis.  
**Citation:** The Journal of craniofacial surgery, Mar 2015, vol. 26, no. 2, p. 388-392 (March 2015)  
**Author(s):** Chen, Zhengxi, Pan, Xiaogang, Zhao, Ning, Chen, Zhenqi, Shen, Gang  
**Abstract:** Unilateral cleft lip and palate (UCLP) patients frequently present with an asymmetry in the nasomaxillary complex and a maxillary hypoplasia. The aim of this study was to investigate biomechanical effects of asymmetric maxillary protraction in UCLP patients using finite element method. A finite element model of a UCLP patient's skull was generated using data from spiral computed tomographic scans. On the basis of this finite element model, three groups of orthopedic forces were loaded. All forces were applied in a direction that was 30 degrees downward and forward to the occlusal plane on the region of the alveolar of the maxillary canine. The value of orthopedic force was 5 N in cleft side and 5 N in noncleft side (group A), 6 N in cleft side and 5 N in noncleft side (group B), and 7 N in cleft side and 5 N in noncleft side (group C), respectively. All 3 groups were effective in promoting maxilla forward. In group B, the displacement difference between the cleft side and the noncleft side was the smallest. The largest value difference between the cleft side and the noncleft side was found in group C. Maxillary protraction with a loading of 6 N in the cleft side and 5 N in the noncleft side produced the most favorable outcome. It can be suggested that it might be advantageous to perform asymmetric maxillary protraction on UCLP patients.  
**Source:** Medline

6. **Title:** Automatic evaluation of hypernasality based on a cleft palate speech database.  
**Citation:** Journal of medical systems, May 2015, vol. 39, no. 5, p. 242., 0148-5598 (May 2015)  
**Author(s):** He, Ling, Zhang, Jing, Liu, Qi, Yin, Heng, Lech, Margaret, Huang, Yunzhi  
**Abstract:** The hypernasality is one of the most typical characteristics of cleft palate (CP) speech. The evaluation outcome of hypernasality grading decides the necessity of follow-up surgery. Currently, the evaluation of CP speech is carried out by experienced speech therapists. However, the result strongly depends on their clinical experience and subjective judgment. This work aims to propose an automatic evaluation system for hypernasality grading in CP speech. The database tested in this work is collected by the Hospital of Stomatology, Sichuan University, which has the largest number of CP patients in China. Based on the production process of hypernasality, source sound pulse and vocal tract filter features are presented. These features include pitch, the first and second energy amplified frequency bands, cepstrum based features, MFCC, short-time energy in the sub-bands features. These features combined with KNN classier are applied to automatically classify four grades of hypernasality: normal, mild, moderate and severe. The experiment results show that the proposed system achieves a good performance. The classification rates for four hypernasality grades reach up to 80.4 %. The sensitivity of proposed features to the gender is also discussed.  
**Source:** Medline

7. **Title:** Cleft lip/palate, short stature, and developmental delay in a boy with a 5.6-Mb interstitial deletion involving 10p15.3p14  
**Citation:** Molecular Syndromology, March 2015, vol./is. 6/1(39-43), 1661-8769;1661-8777 (06 Mar 2015)  
**Author(s):** Gamba B.F., Rosenberg C., Costa S., Richieri-Costa A., Ribeiro-Bicudo L.A.
8. Title: Congenital intra-oral adhesions: A surgical approach to cleft palate lateral synechia syndrome.


Author(s): Mascarella, Marco A, Schwartz, Joseph, Manoukian, John J

Abstract: An array of genetic syndromes has been associated with intra-oral adhesions in neonates. The primary medical issues arise from airway obstruction, feeding difficulties and poor oral development, specifically with cleft palate lateral synechia syndrome (CPLSS). Despite this, a paucity of data exists for the clinical management of intra-oral adhesions in this population. We report the cases of a father and daughter diagnosed with CPLSS who presented with respiratory and feeding difficulties at birth undergoing surgical correction. Early surgical ligation of intra-oral bands allows for a stabilization of the airway, improved feeding and oral development with a good long-term outcome. Copyright © 2015 Elsevier Ireland Ltd. All rights reserved.

Source: Medline

9. Title: Contribution of malocclusion and female facial attractiveness to smile esthetics evaluated by eye tracking.

Citation: American journal of orthodontics and dentofacial orthopedics : official publication of the American Association of Orthodontists, its constituent societies, and the American Board of Orthodontics, Apr 2015, vol. 147, no. 4, p. 472-482 (April 2015)


Abstract: There is disagreement in the literature concerning the importance of the mouth in overall facial attractiveness. Eye tracking provides an objective method to evaluate what people see. The objective of this study was to determine whether dental and facial attractiveness alters viewers' visual attention in terms of which area of the face (eyes, nose, mouth, chin, ears, or other) is viewed first, viewed the greatest number of times, and viewed for the greatest total time (duration) using eye tracking. Seventy-six viewers underwent 1 eye tracking session. Of these, 53 were white (49% female, 51% male). Their ages ranged from 18 to 29 years, with a mean of 19.8 years, and none were dental professionals. After being positioned and calibrated, they were shown 24 unique female composite images, each image shown twice for reliability. These images reflected a repaired unilateral cleft lip or 3 grades of dental attractiveness similar to those of grades 1 (near ideal), 7 (borderline treatment need), and 10 (definite treatment need) as assessed in the aesthetic component of the Index of Orthodontic Treatment Need (AC-IOTN). The images were then embedded in faces of 3 levels of attractiveness: attractive, average, and unattractive. During viewing, data were collected for the first location, frequency, and duration of each viewer's gaze. Observer reliability ranged from 0.58 to 0.92 (intraclass correlation coefficients) but was less than 0.07 (intrarater) for the chin, which was eliminated from the study. Likewise, reliability for the area of first fixation was kappa less than 0.10 for both intrarater and interrater reliabilities; the area of first fixation was also removed from the data analysis. Repeated-measures analysis of variance showed a significant effect (P

Source: Medline

10. Title: Design of a haptic model for the training of cleft treatment procedures.


Author(s): Rau, Andrea, Nobis, Christopher-Philipp, Behr, Alexandra V, Kesting, Marco R

Abstract: Cleft lip and palate (CLP) defects are considered to be the most common facial birth defect. Despite the relevance of this global health issue, the knowledge level on CLP is alarmingly low. Therefore, it was our aim to construct a realistic haptic anatomic CLP model that can be used for 3-dimensional visualizing CLP and practicing the first steps of CLP treatment. Models of newborns with CLP are not commercially available so far. Therefore, construction was based on a purchased baby doll of a healthy newborn. After fabrication of the model, we used it in a hands-on course for medical students. A total of 138 students were asked to perform practical tasks such as like taking intraoral impressions and fixing drinking and nasoalveolar plates on the CLP model. To evaluate the didactic
benefit, preteaching and postteaching multiple-choice tests were performed. A suitable patient's plaster model from our archive served as a template for shaping a unilateral CLP in the face of a baby doll by means of a scalpel and a handheld rotating milling machine. Hard and soft palate were milled out and replaced by a hard stone cast of a patient with cleft palate. When analyzing the preteaching and postteaching scores of the students' multiple-choice tests, an improvement was achieved in 69.6%, which proved to be statistically significant (P

Source: Medline

11. Title: Distraction osteogenesis versus orthognathic surgery for the treatment of maxillary hypoplasia in cleft lip and palate patients: a systematic review.

Citation: Orthodontics & craniofacial research, May 2015, vol. 18, no. 2, p. 96-108 (May 2015)

Author(s): Austin, S L, Mattick, C R, Waterhouse, P J

Abstract: To compare the effectiveness of distraction osteogenesis to orthognathic surgery for the treatment of maxillary hypoplasia in individuals with cleft lip and palate. A systematic review of prospective randomized, quasi-randomized or controlled clinical trials. MEDLINE, EMBASE, Scopus, Web of Science, CINAHL, CENTRAL, trial registers and grey literature were searched. Hand searching of five relevant journals was completed. Two reviewers independently completed inclusion assessment. Data extraction and risk of bias assessment were completed by a single reviewer and checked by a second reviewer. Five publications all reporting different outcomes of a single randomized controlled trial are included within the review. The quality of the evidence was low with a high risk of bias. Both surgical interventions produce significant soft tissue improvement. Horizontal relapse of the maxilla was statistically significantly greater following orthognathic surgery. There was no statistically significant difference in speech and velo-pharyngeal function between the interventions. Maxillary distraction initially lowered social self-esteem, but this improved with time resulting in higher satisfaction with life in the long term. The low quality of evidence included within the review means there is insufficient evidence to conclude whether there is a difference in effectiveness between maxillary distraction and osteotomy for the treatment of cleft-related maxillary hypoplasia. There is a need for further high-quality randomized controlled trials to allow conclusive recommendations to be made. © 2014 John Wiley & Sons A/S. Published by John Wiley & Sons Ltd.

Source: Medline

12. Title: Effectiveness of a parent-implemented intervention program for young children with cleft palate.


Author(s): Ha, Seunghhee

Abstract: This study investigated the effectiveness of a parent-implemented intervention on children's speech-language development and parents' interaction styles. Seventeen children with cleft palate (CP) and their mothers participated in all sessions of a parent-implemented intervention program. Nine children with CP and their mothers who did not receive the intervention were included to examine the full effectiveness of the program. The intervention program consisted of four phases, pre-intervention test, parent training, parent-implemented intervention at children's home for 3 months, and post-intervention test. Children's language and speech measures and maternal measures from pre- and post-intervention tests were compared between groups (intervention vs. no intervention). Children who received a parent-implemented intervention exhibited significant improvement in language measures based on standardized tests and quantitative language and speech measures from spontaneous utterances. The children in the intervention group showed a significantly greater extent of change in expressive vocabulary size, number of total words, and mean length of utterance than did those who did not receive the intervention. Mothers who received the training showed a significantly decreased number of different words, increased responsiveness, and decreased non-contingent utterances for children's communication acts compared to those who did not receive the training. The results of the study support the effectiveness of parent-implemented early intervention on positive changes in children's speech-language development and mothers' use of communication strategies. Copyright © 2015 Elsevier Ireland Ltd. All rights reserved.

Source: Medline

13. Title: Establishment of a rat model for alveolar cleft with bone wax

Citation: Journal of Oral and Maxillofacial Surgery, April 2015, vol./is. 73/4(733.e1-733.e10), 0278-2391;1531-5053 (01 Apr 2015)

Author(s): Xu Y., Sun J., Chen Z.

Language: English

Abstract: Purpose Although treatment of cleft lip and cleft palate is becoming standardized, treatment of alveolar cleft remains controversial. Thus, preclinical animal work remains necessary to improve clinical outcome. This study established an alveolar cleft model in the rat. Materials and Methods Maxillary first molars were extracted to create
a 4- x 4- x 3-mm complete alveolar cleft on the right and left sides in 25 8-week-old Sprague-Dawley rats. The left cleft was filled with bone wax and the right side remained untreated. Animals were sacrificed at 0, 4 and 8 weeks after surgery. Harvested alveolar cleft samples were evaluated by micro-computed tomographic and histologic analyses. Results The healing rate and osteoblast activity of the left cleft were less than those of the right cleft. Conclusion This model of a critical-size alveolar cleft can be used efficiently for the therapeutic evaluation of novel techniques for the treatment of alveolar cleft.

Publication type: Journal: Article
Source: EMBASE

Citation: Journal of plastic, reconstructive & aesthetic surgery : JPRAS, Apr 2015, vol. 68, no. 4, p. 479-484 (April 2015)
Author(s): Sakamoto, Yoshiaki, Soga, Shigeyoshi, Jinzaki, Masahiro, Yamada, Yoshitake, Ogata, Hisao, Kishi, Kazuo
Abstract: Current imaging techniques for velopharyngeal closure (VPC) evaluation are two-dimensional, static, or distressing, thus necessitating multiple procedures to understand this three-dimensional and dynamic area. We validated the use of a novel four-dimensional (4D) computed tomography (CT) technique for the morphological and kinematic evaluation of VPC in cleft palate patients based on dynamic volume scanning with 320-detector-row CT. Five patients aged 4-10 years (40% males) with persistent velopharyngeal insufficiency post palatoplasty underwent conventional tests (cephalometry and video-nasal endoscopy) and 4D-CT. For each patient, complete multiplanar reconstruction, 4D airway CT, and 4D-CT endoscopy data for all scanning phases were compared with cephalometric and video-nasal endoscopy data. The movements of the velum and posterior pharyngeal walls were graded by each modality. 4D airway CT revealed higher anatomical detail than cephalometry, additionally providing dynamic images. 4D-CT endoscopy and video-nasal endoscopy were in agreement for all patients regarding the patterns of VPC, with complete visualization of VPC in five versus one patient, respectively. 4D airway CT and cephalometry showed a discrepancy in one case, wherein grading by cephalometry was overestimated. 4D-CT was also useful in determining the width and length of a proposed pharyngeal flap. The examination time (mean ± standard deviation (SD), seconds), including patient preparation time, was 224 ± 73, 492 ± 145, and 718 ± 123 for cephalometric radiographs, CT, and video-nasal endoscopy, respectively. The mean estimated radiation dose during 4D-CT was 4.44 ± 1.64 mSv. 4D-CT provides detailed morphological and kinematic analysis of VPC and may offer advantages over conventional procedures. Copyright © 2014 British Association of Plastic, Reconstructive and Aesthetic Surgeons. Published by Elsevier Ltd. All rights reserved.
Source: Medline

15. Title: Facial tissue depths in children with cleft lip and palate.
Citation: Journal of forensic sciences, Mar 2015, vol. 60, no. 2, p. 274-284 (March 2015)
Author(s): Starbuck, John M, Ghoneima, Ahmed, Kula, Katherine
Abstract: Cleft lip and palate (CLP) is a craniofacial malformation affecting more than seven million people worldwide that results in defects of the hard palate, teeth, maxilla, nasal spine and floor, and maxillodental asymmetry. CLP facial soft-tissue depth (FSTD) values have never been published. The purpose of this research is to report CLP FSTD values and compare them to previously published FSTD values for normal children. Thirty-eight FSTDs were measured on cone beam computed tomography images of CLP children (n = 86; 7-17 years). MANOVA and ANOVA tests determined whether cleft type, age, sex, and bone graft surgical status affect tissue depths. Both cleft type (unilateral/bilateral) and age influence FSTDs. CLP FSTDs exhibit patterns of variation that differ from normal children, particularly around the oronasal regions of the face. These differences should be taken into account when facial reconstructions of children with CLP are created. © 2014 American Academy of Forensic Sciences.
Source: Medline

16. Title: Factors associated with high hospital resource use in a population-based study of children with orofacial clefts
Citation: Birth Defects Research Part A - Clinical and Molecular Teratology, February 2015, vol./is. 103/2(127-143), 1542-0752;1542-0760 (01 Feb 2015)
Language: English
Abstract: Background: Little is known about population-based maternal, child, and system characteristics associated with high hospital resource use for children with orofacial clefts (OFC) in the US. Methods: This was a statewide, population-based, retrospective observational study of children with OFC born between 1998 and 2006, identified by the Florida Birth Defects Registry whose records were linked with longitudinal hospital discharge records. We
stratified the descriptive results by cleft type [cleft lip with cleft palate, cleft lip, and cleft palate] and by isolated versus nonisolated OFC (accompanied by other coded major birth defects). We used Poisson regression to analyze associations between selected characteristics and high hospital resource use (>90th percentile of estimated hospitalized days and inpatient costs) for birth, postbirth, and total hospitalizations initiated before age 2 years. RESULTS: Our analysis included 2,129 children with OFC. Infants who were born low birth weight (<2500 grams) were significantly more likely to have high birth hospitalization costs for CLP (adjusted prevalence ratio: 1.6 [95% confidence interval: 1.0-2.7]), CL (adjusted prevalence ratio: 3.0 [95% confidence interval: 1.1-8.1]), and CP (adjusted prevalence ratio: 2.3 [95% confidence interval: 1.3-4.0]). Presence of multiple birth defects was significantly associated with a three- to ten-fold and a three- to nine-fold increase in the prevalence of high costs and number of hospitalized days, respectively; at birth, postbirth before age 2 years and overall hospitalizations. Conclusion: Children with cleft palate had the greatest hospital resources use. Additionally, the presence of multiple birth defects contributed to greater inpatient days and costs for children with OFC.

Publication type: Journal: Article
Source: EMBASE

17. Title: Fryns anophthalmia-plus syndrome: Two rare cases
Citation: Genetic Counseling, 2015, vol./is. 96/3(397-398), 1015-8146 (2015)
Author(s): Bozkurt O., Bidev D., Sarj F.N., Dizardh E.A., Ulu O., Uras N., Oguz S.S., Canpolat F.E., Dilmen U.
Language: English
Abstract: Fryns anophthalmia-plus syndrome: two rare cases: Fryns anophthalmia-plus syndrome is a rare syndrome with clinical diversity primarily including anophthalmia/microphthalmia, facial clefts, cleft lip/palate, ear and nasal deformities. Here we present two different cases of APS with anophthalmia/microphthalmia, cleft palate, low set ears, ventriculomegaly and one of which had intestinal non-fixation anomaly not described in the literature before.

Publication type: Journal: Article
Source: EMBASE
Full text: Available Genetic counseling (Geneva, Switzerland) at Genetic Counseling

18. Title: Genetic and non-genetic factors that increase the risk of non-syndromic cleft lip and/or palate development
Citation: Oral Diseases, April 2015, vol./is. 21/3(393-399), 1354-523X;1601-0825 (01 Apr 2015)
Language: English
Abstract: Objectives: We investigated the relationship between non-syndromic cleft lip/palate (NSCLP) and polymorphisms in methylenetetrahydrofolate reductase (MTHFR), methionine synthase (MTR), methionine synthase reductase (MTRR), and RFC1, as well as the corresponding interactions with environmental factors. Subjects and Methods: One hundred and forty NSCLP patients and their mothers, as well as 175 control individuals and their mothers, were recruited. Information regarding smoking and alcohol consumption was recorded. Blood samples were obtained in order to measure serum folate and cobalamin, as well as, plasma total homocysteine concentrations and to extract DNA. Polymorphisms in MTHFR(677C>T and 1298A>C), MTR(2756A>G), MTR(66A>G), and RFC1(80A>G) were analyzed by PCR-restriction fragment length polymorphism. Results: Among the patients, 59.5% had cleft lip and palate, 22.0% had cleft palate, and 18.5% had cleft lip only. Maternal alcohol consumption was recorded. Blood samples were obtained in order to measure serum folate and cobalamin, as well as, plasma total homocysteine concentrations and to extract DNA. Polymorphisms in MTHFR(677C>T and 1298A>C), MTR(2756A>G), MTR(66A>G), and RFC1(80A>G) were analyzed by PCR-restriction fragment length polymorphism. Results: Among the patients, 59.5% had cleft lip and palate, 22.0% had cleft palate, and 18.5% had cleft lip only. Maternal alcohol consumption and reduced folic acid concentrations in both children and mothers (P < 0.001 and P = 0.003, respectively) were risk factors for NSCLP. Patients and their mothers carrying the MTHFR 667T allele showed lower serum folate than CC (P = 0.011 and P = 0.030, respectively). Mothers who carried the MTHFR 1298C allele exhibited increased risk of having a child with NSCLP, after adjusting for alcohol consumption (OR: 1.75, 95% CI: 1.03-2.99, P = 0.038). Conclusions: Reduced folic acid levels, alcohol consumption, and the MTHFR 667T and 1298C alleles may have contributed to NSCLP development in this sample population from Rio Grande do Norte.

Publication type: Journal: Article
Source: EMBASE

19. Title: Identification of functional variants for cleft lip with or without cleft palate in or near PAX7, FGFR2, and NOG by targeted sequencing of GWAS loci
Citation: American Journal of Human Genetics, March 2015, vol./is. 96/3(397-411), 0002-9297;1537-6605 (05 Mar 2015)
Boys with midline malformations, even without proven cobalamin disorder, profound neurocognitive impairment, this familial observation suggests that HCFC1 gene should be considered in future studies. Both presented with dysmorphic features (flat profile, cleft lip for one), increased nuchal translucency, prenatal findings, and respiratory distress. We further identified significant clusters of non-coding rare variants near NTN1 and NOG and found several rare coding variants likely to affect protein function, including four nonsense variants in ARHGAP29. We confirmed 48 de novo mutations and, based on best biological evidence available, chose two of these for functional assays. One mutation in PAX7 disrupted the DNA binding of the encoded transcription factor in an in vitro assay. The second, a non-coding mutation, disrupted the activity of a neural crest enhancer downstream of FGFR2 both in vitro and in vivo. This targeted sequencing study provides strong functional evidence implicating several specific variants as primary contributory risk alleles for nonsyndromic clefting in humans.

**Abstract**

Although genome-wide association studies (GWASs) for nonsyndromic orofacial clefts have identified multiple strongly associated regions, the causal variants are unknown. To address this, we selected 13 regions from GWASs and other studies, performed targeted sequencing in 1,409 Asian and European trios, and carried out a series of statistical and functional analyses. Within a cluster of strongly associated common variants near NOG, we found that one, rs2277277, disrupts enhancer activity. We furthermore identified significant clusters of non-coding rare variants near NTN1 and NOG and found several rare coding variants likely to affect protein function, including four nonsense variants in ARHGAP29. We confirmed 48 de novo mutations and, based on best biological evidence available, chose two of these for functional assays. One mutation in PAX7 disrupted the DNA binding of the encoded transcription factor in an in vitro assay. The second, a non-coding mutation, disrupted the activity of a neural crest enhancer downstream of FGFR2 both in vitro and in vivo. This targeted sequencing study provides strong functional evidence implicating several specific variants as primary contributory risk alleles for nonsyndromic clefting in humans.

**Publication type:** Journal: Article

**Source:** EMBASE

**Full text:** Available [American journal of human genetics](http://www.salisburyhospital.org) at Salisbury District Hospital Healthcare Library

**Full text:** Available [American journal of human genetics](http://www.salisburyhospital.org) at American Journal of Human Genetics, The

**20. Title:** Midfacial volumetric and upper lip soft tissue changes after Le Fort I advancement of the cleft maxilla

**Citation:** Journal of Oral and Maxillofacial Surgery, April 2015, vol./is. 73/4(708-718), 0278-2391;1531-5053 (01 Apr 2015)

**Author(s):** Susarla S.M., Berli J.U., Kumar A.

**Language:** English

**Abstract:** Purpose To analyze, using 3-dimensional photogrammetric data, midfacial soft tissue and volumetric changes in the cleft maxilla after 1- or 2-piece Le Fort I (LF1) advancement. Materials and Methods This was a retrospective study of patients with cleft and maxillary hypoplasia who underwent LF1 advancement. The primary predictor variable was the type of advancement (1 piece vs 2 pieces). Outcome measurements were changes in soft tissue linear measurements (subnasale [Sn], labium superius [LS], and stomion [SO]) and midfacial volume after maxillary advancement. Results Eleven patients (7 male, 4 female) underwent LF1 advancements (4 underwent 2-piece advancement). The mean maxillary advancement was 6.2 +/- 1.7 mm. Soft tissue changes at the Sn, LS, and SO were 5.2 +/- 2.0, 5.8 +/- 2.5, and 5.2 +/- 1.8 mm, respectively. The average volume change was 12.2 +/- 5.7 cm<sup>3</sup>.

**Publication type:** Journal: Conference Paper

**Source:** EMBASE

**21. Title:** Multiple congenital anomalies in two boys with mutation in HCFC1 and cobalamin disorder

**Citation:** European Journal of Medical Genetics, March 2015, vol./is. 58/3(148-153), 1769-7212;1878-0849 (01 Mar 2015)

**Author(s):** Gerard M., Morin G., Bourillon A., Colson C., Mathieu S., Rabier D., Billette de Villemeur T., Ogier de Baunly H., Benoist J.F.

**Language:** English

**Abstract:** The cobalamin type C deficiency is a rare condition that results from impaired biosynthesis of both methylcobalamin (MeCbl) and adenosylcobalamin (AdoCbl). Hemizygous mutations of the HCFC1 gene explain the majority of clinically and biologically compatible cblC patients without MMACHC mutations (OMIM 309541). We report a family with two maternal half-brothers with multiple congenital anomalies and HCFC1 gene mutation in the second Kelch domain. Both presented with dysmorphic features (flat profile, cleft lip for one), increased nuchal translucency, prenatal onset microcephaly and hypospadias. Additionally to early onset intractable epilepsy and profound neurocognitive impairment, this familial observation suggests that HCFC1 gene should be considered in boys with midline malformations, even without proven cobalamin C deficiency.
22. Title: Muscle tension line concept in nasolabial muscle complex-based on 3-dimensional reconstruction of nasolabial muscle fibers.

Author(s): Yin, Ningbei, Wu, Jiajun, Chen, Bo, Song, Tao, Ma, Hengyuan, Zhao, Zhenmin, Wang, Yongqian, Li, Haidong, Wu, Di

Abstract: Plastic surgeons have attempted various ways to rebuild the aesthetic subunits of the upper lip in patients with cleft lip with less than perfect results in most cases. We propose that repairing the 3 muscle tension line groups in the nasolabial complex will have improved aesthetic results. Micro-computed tomographic scans were performed on the nasolabial tissues of 5 normal aborted fetuses and used to construct a 3-dimensional model to study the nasolabial muscle complex structure. The micro-computed tomographic (CT) scans showed the close relationship and interaction between the muscle fibers of nasalis, pars peripheralis, levator labii superioris, and pars marginalis. Based on the 2-dimensional images obtained from the micro-computed tomographic scans, we suggest the concept of nasolabial muscle complex and muscle tension line group theory: there is a close relationship among the alar part of the nasalis, depressor septi muscle, orbicularis oris muscle, and levator labii superioris alaeque nasi. The tension line groups are 3 tension line structures in the nasolabial muscle complex that interlock with each other at the intersections and maintain the specific shape and aesthetics of the lip and nose.

Source: Medline

23. Title: Nasoalveolar molding in cleft care - Experience in 40 patients from a single centre in Germany

Citation: PLoS ONE, March 2015, vol./is. 10/3, 1932-6203 (03 Mar 2015)
Author(s): Rau A., Ritschl L.M., Mucke T., Wolff K.-D., Loeffelbein D.J.
Language: English

Abstract: Nasoalveolar molding (NAM) has gained wide acceptance and evidence in cleft therapy. However, standardized treatment protocols and experiences recorded from European centres are lacking. The results of 40 infants with cleft lip and palate treated with presurgical NAM according to the Grayson technique were analyzed. Standardized parameters of cleft width and nasal symmetry were measured in pre- and posttreatment plaster casts and in digitalized 3-dimensional STL models. Statistical analyses were performed by using Student’s t-test in a per-protocol manner. 27 out of 40 infants completed NAM and were analyzed. In 13 patients NAM was either temporarily interrupted or terminated prematurely due to skin irritations or lack of parental support. These cases were excluded from statistical analysis, resulting in a drop-out rate of 32.5%. Intersegmental alveolar distance (ISAD), intersegmental lip distance (ISLD), nostril height (NH), nostril width (NW) and columella deviation angle (CDA) were significantly changed in unilateral cleft lip and palate (UCLP) (n = 8). In unilateral cleft lip (UCL) (n = 9), only ISLD, NH and CDA were significantly changed. ISAD of the right and left side, ISLD of the right and left side, premaxilla deviation angle, nostril height and columella length were changed significantly in bilateral cleft lip and palate (BCLP) cases (n = 10). NAM is a suitable presurgical treatment modality. A positive effect has been seen in UCLP and BCLP infants, as compared with their birth status.

Publication type: Journal: Article
Source: EMBASE
Full text: Available ProQuest at PLoS ONE
Full text: Available ProQuest at PLoS One

24. Title: Non-cleft causes of velopharyngeal dysfunction: Implications for treatment

Citation: International Journal of Pediatric Otorhinolaryngology, March 2015, vol./is. 79/3(286-295), 0165-5876;1872-8464 (01 Mar 2015)
Author(s): Kummer A.W., Marshall J.L., Wilson M.M.
Language: English

Abstract: Although a history of cleft palate is the most common cause of velopharyngeal dysfunction (VPD), there are other disorders that can also cause hypernasality and/or nasal emission. These include other structural anomalies of the velopharyngeal valve (velopharyngeal insufficiency), neurophysiological disorders that result in inadequate velopharyngeal movement (velopharyngeal incompetence), and even faulty articulation placement in the pharynx (velopharyngeal mislearning). Unfortunately, individuals with non-cleft causes of hypernasality and/or nasal emission do not typically present at a cleft palate/craniofacial center where there are professionals who specialize in the evaluation and treatment of these disorders. As a result, they are often misdiagnosed and do not receive appropriate treatment. In this review, we present various conditions that can cause hypernasality and/or
nasal emission during speech. We discuss appropriate treatment based on the underlying cause of the condition. It is important that pediatric otolaryngologists are able to recognize these disorders so that affected patients are referred to specialists in velopharyngeal dysfunction for treatment.

**Publication type:** Journal: Article

**Source:** EMBASE

25. **Title:** Oblique lip-alveolar banding in patients with cleft lip and palate.

**Citation:** The British journal of oral & maxillofacial surgery, Apr 2015, vol. 53, no. 4, p. 390-392 (April 2015)

**Author(s):** Naidoo, S, Bülow, K-W

**Abstract:** We report an oblique lip-alveolar band, a rare banding of soft tissue that involves the lip and alveolus, which we have found in five patients with cleft lip and palate (0.2%), compared with an incidence of the Simonartz lip-lip band of 5.7%). To our knowledge this has not been reported previously. In two patients the bands affected the cleft lip and alveolus bilaterally, with or without the palatal cleft, and in three the bands were unilateral cleft lip and alveolus with or without the palatal cleft. Copyright © 2015 The British Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

**Source:** Medline

26. **Title:** Passive smoking in the etiology of non-syndromic orofacial clefts: A systematic review and meta-analysis

**Citation:** PLoS ONE, March 2015, vol./is. 10/3, 1932-6203 (11 Mar 2015)

**Author(s):** Sabbagh H.J., Hassan M.H.A., Innes N.P.T., Elkodary H.M., Little J., Mossey P.A.

**Language:** English

**Abstract:** Background Studies have found a consistent positive association between maternal smoking and nonsyndromic orofacial clefts (NSOFC). However, no comprehensive assessment of the association between NSOFC and passive smoking has been undertaken. This systematic review and meta-analysis explores the relationship between maternal passive smoking and NSOFC, and compares the associations between passive and active smoking.

Methods and Findings Search strategy, inclusion / exclusion criteria, and data extraction from studies reporting maternal passive smoking and NSOFC was implemented without language restrictions. Risks of bias in the identified studies were assessed and this information was used in sensitivity analyses to explain heterogeneity. Meta-analysis and meta-regression of the extracted data were performed. Egger's test was used to test for small study effects.

Fourteen eligible articles were identified. Maternal passive smoking exposure was associated with a twofold increase in risk of NSOFC (odds ratio: 2.11, 95% confidence interval: 1.54-2.89); this was apparent for both cleft lip with and without palate (OR: 2.05, 95% CI: 1.27-3.3) and cleft palate (OR: 2.11, 95% CI: 1.23-3.62). There was substantial heterogeneity between studies. In the studies that provided data enabling crude and adjusted odd ratios to be compared, adjustment for potential confounders attenuated the magnitude of association to about a 1.5-fold increase in risk. Conclusion Overall, maternal passive smoking exposure results in a 1.5 fold increase in risk of NSOFC, similar to the magnitude of risk reported for active smoking, but there is marked heterogeneity between studies.

This heterogeneity is not explained by differences in the distribution of cleft types, adjustment for covariates, broad geographic region, or study bias/quality. This thorough meta-analysis provides further evidence to minimize exposure to environmental tobacco smoke in policy making fora and in health promotion initiatives.

**Publication type:** Journal: Review

**Source:** EMBASE

**Full text:** Available ProQuest at PLoS ONE

27. **Title:** Periodontal status among patients with cleft lip (CL), cleft palate (CP) and cleft lip, alveolus and palate (CLAP) in Chennai, India. A comparative study

**Citation:** Journal of Clinical and Diagnostic Research, March 2015, vol./is. 9/3(ZC53-ZC55), 2249-782X;0973-709X (01 Mar 2015)

**Author(s):** Nagappan N., John J.

**Language:** English

**Abstract:** Background: Long term health of the stomatognathic system as well as esthetic aspects is the therapeutic goals in patients with oro facial clefts. Aim: The aim of this study was to assess and compare the periodontal status of patients with cleft lip (CL), cleft palate (CP) and cleft lip, alveolus and palate (CLAP) reporting to a hospital in Chennai, India. Materials and Methods: The study group consisted of 80 cleft patients. Subjects were divided into three groups. Group 1: patients with cleft lip (CL), Group 2: subjects with cleft palate (CP) and Group 3: subjects with cleft lip alveolus and palate (CLAP). Community Periodontal Index for Treatment needs CPITN Index was recorded.

Results: Among the 80 study subjects, 51 (63.8%) were males and 29 (36.2%) were females. Among the 26 study
subjects with cleft lip, 10 (38.5%) had healthy periodontium, 4 (15.4%) had bleeding on probing and 12 (46.1%) had calculus. Mean number of sextants coded for healthy and bleeding was maximum among the subjects with cleft palate. Mean number of sextants coded for calculus was maximum among the subjects with cleft lip alveolus and palate. Prevalence of periodontal disease is high among patients with cleft lip, alveolus and palate (35%) than in Cleft lip (32.5%) and Cleft Palate (32.5%). Conclusion: Gingivitis and Calculus is predominantly high in patients with Cleft Palate and Cleft Lip respectively.

**Publication type:** Journal: Article  
**Source:** EMBASE

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**28. Title:** Post septorhinoplasty custom-made unilateral nasal stent for nasal cleft deformity  
**Citation:** North American Journal of Medical Sciences, 2015, vol./is. 7/2(73-76), 2250-1541;1947-2714 (2015)  
**Author(s):** Rathee M., Bhoria M., Boora P.  
**Language:** English  
**Abstract:** Context: Nasal cleft deformity is a complicated problem. Utilization of nasal stent in postseptorhinoplasty aims at establishing and maintaining airway patency, tissue position, and reduces tissue contracture after surgery. Case Report: A 16-year-old female patient presented with history of surgical reconstruction of congenital cleft lip and cleft palate with secondary septorhinoplasty of nasal cleft deformity. Patient was referred for nasal stent 1 week after septorhinoplasty. This case report provides a novel technique for fabrication of esthetic nasal stent after postseptorhinoplasty for secondary cleft nose deformity correction. Conclusion: This case report presents a simple, convenient technique for nasal stent fabrication for prevention of restenosis for cleft nose deformity post secondary septorhinoplasty. Provision of nasal stent allows breathing, maintains esthetics, comfort, nasal patency, and contour with minimal discomfort.  
**Publication type:** Journal: Article  
**Source:** EMBASE

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**29. Title:** Presumed Larsen syndrome in a child: a case with a 12-year follow-up.  
**Author(s):** Mei, Haibo, He, Rongguo, Liu, Kun, Wu, Jiangyan, Tang, Jin, Yan, An  
**Abstract:** Larsen syndrome (OMIM 150250) was first described in 1950 as an entity characterized by distinct facial features and dislocations of the multiple large joint, and cleft palate, hearing loss, and spinal abnormalities were occasionally observed. The prevalence of Larsen syndrome is estimated to be one in 100 000 live births. Management of multiple large-joint dislocations often proves difficult with a tendency toward recurrence, particularly if a patient has complete dislocation of the knee. We treated a boy with the clinical phenotype of Larsen syndrome using 10 orthopedic procedures, but failed to achieve a satisfactory outcome. The aim of this report is to review the surgical course and report results of surgical treatments for this patient with 12 years of follow-up.  
**Source:** Medline

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**30. Title:** Prevalence of depressive symptoms in patients with cleft lip and palate  
**Citation:** Brazilian Journal of Otorhinolaryngology, March 2015, vol./is. 81/2(177-183), 1808-8694;1808-8686 (01 Mar 2015)  
**Author(s):** Lima L.S., Ribeiro G.S., de Aquino S.N., Volpe F.M., Martelli D.R.B., Swerts M.S.O., Paranaiba L.M.R., Martelli Junior H.  
**Language:** English  
**Abstract:** Introduction: Cleft lip and/or palate (CL/P) represent the most common congenital anomalies of the face. Objective: To evaluate the prevalence of depressive symptoms in children and adolescents with nonsyndromic cleft lip and/or palate (nsCL/P). Methods: We conducted an observational, case-control study, with a case study group composed of 61 patients with nsCL/P, aged 7-17 years, and a control group of 61 clinically normal patients. Both groups were selected at the same institution. Results: Depressive symptoms were observed in the case group (nsCL/P), but there were no statistically significant differences compared to the control group. No association was found between the two groups (case and control) in relation to sociodemographic variables: gender, age, and education. Conclusions: This study identified the prevalence of depressive symptoms in children and adolescents with nsCL/P from a localized geographic population, although the results were not statistically significant when compared to the control group, not justifying the use of CDI (Child Depression Inventory) as a screening instrument for depressive symptoms in the examined population.  
**Publication type:** Journal: Article  
**Source:** EMBASE
31. Title: Retinoic acid inhibits histone methyltransferase Whsc1 during palatogenesis
Citation: Biochemical and Biophysical Research Communications, March 2015, vol./is. 458/3(525-530), 0006-291X;1090-2104 (13 Mar 2015)
Author(s): Liu S., Higashihiro N., Yahiro K., Moriyama K.
Language: English
Abstract: Cleft lip with or without palate (CL/P) is a common congenital anomaly in humans and is thought to be caused by genetic and environmental factors. However, the epigenetic mechanisms underlying orofacial clefts are not fully understood. Here, we investigated how the overdose of retinoic acid (RA), which can induce cleft palate in mice and humans, regulates histone methyltransferase, Wolf-Hirschhorn syndrome candidate 1 (WHSC1) during palatal development in mice. We treated mouse embryonic fibroblasts (MEFs) with 1 μM all-trans RA and discovered that the global level of H3K36me3 was downregulated and that expression of the H3K36 methyltransferase gene, Whsc1, was reduced. The expression level of WHSC1 in embryonic palatal shelves was reduced during palatogenesis, following maternal administration of 100 mg/kg body weight of RA by gastric intubation. Furthermore, the expression of WHSC1 in palatal shelves was observed in epithelial and mesenchymal cells at all stages, suggesting an important role for palatal development. Our results suggest that the pathogenesis of cleft palate observed after excessive RA exposure is likely to be associated with a reduction in the histone methyltransferase, WHSC1.
Publication type: Journal: Article
Source: EMBASE
Full text: Available Elsevier at Biochemical and Biophysical Research Communications

32. Title: Retinoic acid inhibits histone methyltransferase Whsc1 during palatogenesis
Author(s): Kopcsányi, Gábor, Vincze, Olga, Bagdán, Viktor, Pytel, József
Abstract: Contradictory experience has been published on the outcomes of ear surgery in patients with cleft palate. The authors of this study investigated whether there were differences in the short- and long-term outcomes of tympanoplasty performed due to cholesteatoma in children with or without cleft palate. Tertiary care medical centre. The authors retrospectively analyzed the first author’s 24-year experience of paediatric tympanoplasty using the software programme developed by the fourth author. The outcomes of 268 tympanoplasties on 172 ears with cholesteatoma in 151 'NoCleft' patients were compared to the outcomes of 35 tympanoplasties on 20 ears of 19 'Cleft' patients. The average age of the patients was 10.7±3.6 years and 9.5±2.7 years respectively. The average follow-up time was 4 and 4.1 years. Preoperative PTA-ABGs (31.22/34.88dB; p=0.058), best postoperative PTA-ABGs (17.04/16.4dB; p=0.499), last postoperative PTA-ABGs (19.93/20.98dB; p=0.298), the final hearing improvement (11.29/13.9dB; p=0.193) and postoperative PTA-ABG deterioration with time (2.89/4.58dB; p=0.117) were statistically compared between the 'NoCleft' and 'Cleft' groups. The same parameters were analyzed separately in the case of tympanoplasty performed with intact ossicular chain and the different type of columella ossiculoplasty. No significant differences were found in the necessity for grommet insertion (8-fold difference, p=0), and conversion to open techniques (p=0). The authors conclude that the achievable audiological outcomes of tympanoplasty in children with cleft palate and cholesteatoma do not differ significantly from those of the general child population. However, this more frequently requires ventilation tube insertion and more frequent follow-up visits. The latter is ensured by patient care within the frameworks of the 'Cleft Palate Team'. We have to accept that in some cases Eustachian tube dysfunction caused by the underlying disease (cleft palate) ‘takes over’ and we have to resort to open techniques.
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Source: Medline

33. Title: Rhinopharyngeal autologous fat injection for treatment of velopharyngeal insufficiency in patients with cleft palate.
Author(s): Piotet, Elsa, Beguin, Céline, Broome, Martin, Iglesias, Katia, Olivier, Frédéric, Leuchter, Igor, Zbinden, Chantal, Hohlfeld, Judith, de Buys Roessingh, Anthony, Schweizer, Valérie, Pasche, Philippe
Abstract: Patients with cleft palate are prone to velopharyngeal insufficiency. In minor cases or when hypernasal speech does not resolve after velopharyngoplasty, an augmentation pharyngoplasty with autologous fat can be proposed. The aim of the present study is to evaluate the short-term (within 2 months) and long-term efficiency
(during the 24 months following the procedure) of our procedure in the setting of velopharyngeal insufficiency related to a cleft palate. Twenty-two patients with cleft palate related velopharyngeal insufficiency were included in this retrospective study. All patients were operated following the same technique, in the same institution. The pre- and postoperative evaluations included a nasometry, a subjective evaluation using the Borel-Maisonny score, and a nasofibroscopy to assess the degree of velopharyngeal closure. Scores of Borel-Maisonny and nasometry were compared before, shortly after the procedure (within 2 months) and long term after the procedure (within 24 months). Forty-one procedures in 22 patients with a cleft palate performed in our institution between October 2004 and January 2012 were included in the study. Nine patients had a previous velopharyngoplasty with persistent rhinolalia despite intensive speech therapy. In 14 patients the procedure was repeated because of recurrent hypernasal speech after the first injection. The average number of procedures per patient was 1.8. Postoperative nasometry and Borel-Maisonny scores were statistically significantly improved and remained stable until the end of the follow-up (median 42 months postoperative) in most patients. Complications were rare and minor. Autologous fat injection is a simple procedure for treatment of minor velopharyngeal insufficiencies in patients with cleft palate, with good long-term results and few complications.

Source: Medline

34. Title: Risk of persistent palatal fistula in patients with cleft palate.
Citation: JAMA facial plastic surgery, Mar 2015, vol. 17, no. 2, p. 126-130 (March 1, 2015)
Author(s): Ahmed, Mairaj K, Maganzini, Anthony L, Marantz, Paul R, Rousso, Joseph J
Abstract: Many individuals with a cleft palate also have an associated craniofacial syndrome or anomaly. To investigate the predictive associations of persistent palatal fistulas in patients with previously repaired cleft palate. We performed a case-control study of patients with cleft palate repairs from January 1, 1986, through December 31, 2000, at a major tertiary care hospital center in the Bronx, New York. The study population consisted of patients who had their primary surgery before the age of 3 years and had all their cleft-related treatment completed at the same hospital center. Palatal fistula was defined as a breakdown of the primary surgical repair of the palate, resulting in persistent patency between the oral and nasal cavities. Data collection was conducted by using the hospital centers' electronic medical records and patient tracking systems and confirmed by review of hard copies of patient records. The Veau classification system was used to classify the preoperative cleft severity. A total of 130 patients were identified-23 patients with palatal fistula and 107 controls. A total of 12 girls and 11 boys were identified in the palatal fistula group and 56 girls and 51 boys in the control group. The mean patient age at the time of palatoplasty was 12.6 and 14.5 months in the palatal fistula and control groups, respectively. A statistically significant association was found between the outcome of fistula and severity of cleft, as defined by the Veau classification system (P = .01). Furthermore, for each Veau class increase, the odds of a palatal fistula increased by 2.64 (95% CI, 1.35-5.13; P = .004). No statistically significant associations were found between the outcome of fistula and the following independent variables: patient sex (P = .98), patient age at palatoplasty (P = .82), type of palatoplasty (P = .57), surgeon (P = .15), orthodontic treatment (P = .59), ear infection (P = .30), or clefts associated with syndromes (P = .96). Palatal fistulas are reliably associated with severity of cleft, as defined by the Veau classification system. This knowledge gives the health care professional a more reliable method of preoperatively assessing the risk of postoperative palatal fistula in the cleft palate population.

Source: Medline

35. Title: Skeletal and soft tissue changes and stability in cleft lip and palate patients after distraction osteogenesis using a new intraoral maxillary device
Citation: Journal of Cranio-Maxillofacial Surgery, April 2015, vol./iss. 43/3(323-328), 1010-5182;1878-4119 (01 Apr 2015)
Author(s): Ansari E., Tomat C., Kadlub N., Diner P.A., Bellocq T., Vazquez M.-P., Picard A.
Language: English
Abstract: Background The authors have recently reported on the use of an internal maxillary distraction device. In this study, we report on the hard and soft tissue movements achieved with this intraoral distraction device, and the stability changes after distraction osteogenesis for maxillary hypoplasia in patients with cleft lip and palate. Methods Ten male patients with severe hypoplasia of the maxilla, with complete uni- or bilateral cleft lip and palate were included. The mean age of the patients at the time of operation was 11.91 years (+/-3.41). To evaluate the distraction process and stability, superimpositions on the preoperative lateral cephalograms were performed. The mean follow-up (FU) was 15.42 months (+/-3.94). Results Cephalometric measurements at all of the maxillary hard and soft tissue points improved significantly. Maxillary point A was advanced by 8.25 mm (+/-3.17; P < 0.001). After distraction soft tissue point A' had advanced 7.10 mm (+/-2.69; P < 0.001). The soft tissue to hard tissue ratio at point A was 0.86:1 after distraction. Maxillary horizontal relapse at point A was 14.1% at FU. Vertical relapse was not
significant. Conclusion This rigid intraoral distraction device can be successfully used in the correction of severe maxillary hypoplasia. The marked aesthetic improvement and low psychological encumbrance make this device viable for the treatment of cleft-related hypoplasia of the maxilla.

**Publication type:** Journal: Article

**Source:** EMBASE

### 36. Title: Smell deficits as an endophenotype in patients with non-syndromic cleft lip and/or palate and their non-affected first-degree relatives: A pilot study

**Citation:** Chemical Senses, March 2015, vol./is. 40/3(288), 0379-864X (March 2015)

**Author(s):** Roosenboom J., Claes P., Hens G.

**Language:** English

**Abstract:** Cleft lip and/or palate (CL/P) is one of the most frequent congenital birth defects with an incidence of 1/700 live births and a multifactorial etiology. Although recent studies such as linkage and association studies, have given insight in the genetic etiology of CL/P, most of the causal genes remain unidentified. A candidate gene approach, by the study of endophenotypes, is a unique and promising manner to reveal more of the genetic etiology of CL/P. Endophenotypes are characteristics that are associated with a condition and are considered to be an expression of the underlying susceptibility genes of this condition. One of the possible endophenotypes of CL/P is a higher frequency of olfactory dysfunction in patients and their non-affected first-degree relatives. Although olfactory function has not extensively been investigated in patients with non-syndromic CL/P, there is some evidence for a decreased capacity to smell in patients with CL/P and their non-affected relatives. Furthermore, olfactory dysfunction within syndromic CL/P could be an indication for reduced smell capacity within non-syndromic patients. When smell dysfunction is determined in non-affected relatives, it could be an indication for an underlying genetic cause. In this pilot study with 48 patients, 41 non-affected relatives and 23 controls, smell capacity was tested using the Sniffin' Sticks (Burghardt), testing for smell threshold, discrimination and identification. Furthermore, a questionnaire for olfactory dysfunction was used to compare objective and subjective perception of the smell capacity. To confirm the central etiology of the smell deficits, an MRI was taken for volumetry of the olfactory bulb, expecting smaller olfactory bulb volumes in subjects with a decreased smell capacity. Structural defects were examined using acoustic rhinometry and rhinomanometry in subjects showing an olfactory deficit. The pilot study revealed a significant olfactory dysfunction in patients with CL/P (p=0.018) and their non-affected relatives (p=0.023), compared to the control group. More olfactory dysfunction was seen in patients and relatives with a familial history of CL/P, indicating a genetic origin of this feature. This study is the first to show decreased smell capacity in patients with CL/P and their non-affected first-degree relatives, indicating that olfactory dysfunction could be considered to be an endophenotype of non-syndromic CL/P.

**Publication type:** Journal: Conference Abstract

**Source:** EMBASE

### 37. Title: Temporal characteristics of nasalization in Persian speaker children with and without cleft palate

**Citation:** International Journal of Pediatric Otorhinolaryngology, April 2015, vol./is. 79/4(546-552), 0165-5876;1872-8464 (01 Apr 2015)

**Author(s):** Baghban K., Torabinezhad F., Moradi N., Asadollahpour F., Ahmadi N., Mardani N.

**Language:** English

**Abstract:** Objectives: The purpose of this study was to measure and compare temporal patterns of nasalization in Persian children with and without cleft palate in three vowel contexts. Methods: A Sample of 14 children with repaired cleft palates with or without cleft lip with moderate to severe hyper nasality and 14 children without cleft palate was chosen as subjects. The subjects were chosen from the ages of 4 to 12 years. The nasal onset interval, nasal offset interval and total nasalization duration were obtained from acoustic waveforms and spectrograms in three vowel contexts using Praat Software. For eliminating the effect of different speed of speech in the cleft palate group and control group, the ratio of nasalization duration was calculated. Results: Total nasalization duration are demonstrated by acoustic signals which shows the total significant different temporal patterns in children with cleft palate and without cleft palate and across the vowel contexts (. P<.0000). Conclusions: Longer nasalization durations in children with cleft palate in comparison to children without cleft palate show the delayed or deviant temporal patterns in children with cleft palate. The duration of nasalization reflecting temporal patterns of the oral-nasal acoustic impedance in children with cleft palate may have an influence on the perception of hyper nasality.

**Publication type:** Journal: Article

**Source:** EMBASE

### 38. Title: Ten-year experience of more than 35,000 orofacial clefts in Africa

**Citation:** International Journal of Pediatric Otorhinolaryngology, October 2015, vol./is. 79/5(587-592), 0165-5876;1872-8464 (01 Oct 2015)

**Author(s):** Chemical Senses, March 2015, vol./is. 40/3(288), 0379-864X (March 2015)

**Language:** English

**Abstract:** The purpose of this study was to measure and compare temporal patterns of nasalization in Persian children with and without cleft palate in three vowel contexts. Methods: A Sample of 14 children with repaired cleft palates with or without cleft lip with moderate to severe hyper nasality and 14 children without cleft palate was chosen as subjects. The subjects were chosen from the ages of 4 to 12 years. The nasal onset interval, nasal offset interval and total nasalization duration were obtained from acoustic waveforms and spectrograms in three vowel contexts using Praat Software. For eliminating the effect of different speed of speech in the cleft palate group and control group, the ratio of nasalization duration was calculated. Results: Total nasalization duration are demonstrated by acoustic signals which shows the total significant different temporal patterns in children with cleft palate and without cleft palate and across the vowel contexts (. P<.0000). Conclusions: Longer nasalization durations in children with cleft palate in comparison to children without cleft palate show the delayed or deviant temporal patterns in children with cleft palate. The duration of nasalization reflecting temporal patterns of the oral-nasal acoustic impedance in children with cleft palate may have an influence on the perception of hyper nasality.

**Publication type:** Journal: Conference Abstract

**Source:** EMBASE
Abstract: Background: Surgical correction of orofacial clefts greatly mitigates negative outcomes. However, access to reconstructive surgery is limited in developing countries. The present study reviews epidemiological data from a single charitable organization, Smile Train, with a database of surgical cases from 33 African countries from 2001-2011. Methods: Demographic and clinical patient data were collected from questionnaires completed by the participating surgeons. These data were recorded in Excel, analyzed using SPSS and compared with previously reported data. Results: Questionnaires were completed for 36,384 patients by 389 African surgeons. The distribution of clefts was: 34.44% clefts of the lip (CL), 58.87% clefts of the lip and palate (CLP), and 6.69% clefts of the palate only (CP). The male to female ratio was 1.46:1, and the unilateral: bilateral ratio 2.93:1, with left-sided predominance 1.69:1. Associated anomalies were found in 4.18% of patients. The most frequent surgeries included primary lip/nose repairs, unilateral (68.36%) and bilateral (11.84%). There was seasonal variation in the frequency of oral cleft births with the highest in January and lowest by December. The average age at surgery was 9.34 years and increased in countries with lower gross domestic products. The average hospital stay was 4.5 days. The reported complication rate was 1.92%. Conclusions: With the exception of cleft palates, results follow trends of worldwide epidemiologic reports of 25% CL, 50% CLP, and 25% CP, 2:1 unilateral:bilateral and left:right ratios, and male predominance. Fewer than expected patients, especially females, presented with isolated cleft palates, suggesting that limitations in economic resources and cultural aesthetics of the obvious lip deformity may outweigh functional concerns and access to treatment for females. A fewer than expected associated anomalies suggests either true ethnic variation, or that more severely-affected patients are not presenting for treatment. The epidemiology of orofacial clefting in Africa has been difficult to assess due to the diversity of the continent and the considerable variation among study designs. The large sample size of the data collected provides a basis for further study of the epidemiology of orofacial clefting in Africa.

Publication type: Journal: Article
Source: EMBASE
Full text: Available BioMedCentral at BMC Psychiatry

39. Title: TGFbeta3 Regulates Periderm Removal Through DELTANp63 in the Developing Palate
Citation: Journal of Cellular Physiology, June 2015, vol./is. 230/6(1212-1225), 0021-9541;1097-4652 (01 Jun 2015)
Author(s): Hu L., Liu J., Li Z., Ozturk F., Gurumurthy C., Romano R.-A., Sinha S., Nawshad A.
Language: English
Abstract: The periderm is a flat layer of epithelium created during embryonic development. During palatogenesis, the periderm forms a protective layer against premature adhesion of the oral epithelia, including the palate. However, the periderm must be removed in order for the medial edge epithelium (MEE) to properly adhere and form a palatal seam. Improper periderm removal results in a cleft palate. Although the timing of transforming growth factor beta3 (TGFbeta3) expression in the MEE coincides with periderm degeneration, its role in periderm desquamation is not known. Interestingly, murine models of knockout (-/-) TGFbeta3, interferon regulatory factor 6 (IRF6) (-/-), and truncated p63 (DELTANp63) (-/-) are born with palatal clefts because of failure of the palatal shelves to adhere, suggesting that these genes regulate palatal epithelial differentiation. However, despite having similar phenotypes in null mouse models, no studies have analyzed the possible association between the TGFbeta3 signaling cascade and the IRF6/DELTANp63 genes during palate development. Recent studies indicate that regulation of DELTANp63, which depends on IRF6, facilitates epithelial differentiation. We performed biochemical analysis, gene activity and protein expression assays with palatal sections of TGFbeta3 (-/-), DELTANp63 (-/-), and wild-type (WT) embryos, and primary MEE cells from WT palates to analyze the association between TGFbeta3 and IRF6/DELTANp63. Our results suggest that periderm degeneration depends on functional TGFbeta3 signaling to repress DELTANp63, thereby coordinating periderm desquamation. Cleft palate occurs in TGFbeta3 (-/-) because of inadequate periderm removal that impedes palatal seam formation, while cleft palate occurs in DELTANp63 (-/-) palates because of premature fusion. J. Cell. Physiol. 230: 1212-1225, 2015.
Publication type: Journal: Article
Source: EMBASE

40. Title: The detection of areas in Poland with an increased prevalence of isolated cleft lip with or without cleft palate
Citation: Annals of Agricultural and Environmental Medicine, 2015, vol./is. 22/1(110-117), 1232-1966;1898-2263 (2015)
Author(s): Wieckowska B., Materna-Kiryluk A., Wisniewska K., Kossowski T., Latos-Bielenska A.
41. Title: Uncommon oral cleft in wolf-hirschhorn syndrome.

Citation: Brazilian dental journal, Mar 2015, vol. 26, no. 2, p. 203-206 (2015 Mar-Apr)

Author(s): Aquino, Sibele Nascimento de, Machado, Renato A, Paranaiba, Lívia Maris R, Coletta, Ricardo D, Aguiar, Marcos J Burle de, Fernandes, Cassandro, Martelli Júnior, Hércilio

Abstract: Wolf-Hirschhorn syndrome (WHS) is a syndrome with craniofacial and systemic abnormalities, which is related to 4p deletion. A 3-month old girl with an undiagnosed syndrome was referred for evaluation of the cleft lip and palate. Hypotonia, short stature, cardiac malformation, hypertrophied clitoris, and atypical thumb of both hands was observed. Microcephaly, low-set ear, prominent glabella, downsloping palpebral fissures, a characteristic "Greek warrior helmet" appearance, micrognathia, ears with pits/tags and bilateral incomplete cleft lip apart from incomplete cleft palate were observed as craniofacial findings. With clinical diagnosis of WHS, blood was subjected to karyotyping, which showed a 4p15.2 deletion, consistent with the condition. Here is reported the case of this WHS patient with an uncommon oral cleft extending the phenotypic spectrum of the disorder. The child was referred to a multidisciplinary team to reparative surgery of the cleft lip and palate. The patient is on regular medical follow-up and will be further assisted by dentists, physical therapists, occupational therapists and psychologists. The genotype-phenotype correlation of the affected patient with previous WSH syndrome reports is described.

Source: EMBASE

42. Title: Unilateral microform cleft lip repair: application of muscle tension line group theory.

Citation: The Journal of craniofacial surgery, Mar 2015, vol. 26, no. 2, p. 343-346 (March 2015)

Author(s): Yin, Ningbei, Song, Tao, Wu, Jiajun, Chen, Bo, Ma, Hengyuan, Zhao, Zhenmin, Wang, Yongqian, Li, Haidong, Wu, Di

Abstract: In microform cleft lip repair, reconstructing the elaborate structures is difficult. We describe a new technique of unilateral microform cleft lip repair that is based on the muscle tension line group theory. According to the shape of Cupid bow, a different small incision is used without creating an obvious cutaneous scar. First, the nasolabial muscle around the nasal floor (the first auxiliary tension line group) is reconstructed, and then the orbicularis oris muscle around the philtrum (the second auxiliary tension line group) is reconstructed based on the muscle tension line group theory. From June 2006 to June 2012, the technique was used in 263 unilateral microform cleft lip repairs. For 18 months, 212 patients were followed up. The appearance of the nasal alar, nasal sill, philtrum, and Cupid bow peak improved. Most patients had a satisfactory appearance. Based on the muscle tension line group theory, using this technique offers the ability to adduct the nasal alar effectively to form a good nasal sill and philtrum.

Source: Medline

43. Title: Use of buccal myomucosal flap for palatal lengthening in cleft palate patient: Experience of 20 cases.

Citation: Contemporary clinical dentistry, Mar 2015, vol. 6, p. S36, 0976-237X (March 2015)

Author(s): Varghese, Don, Datta, Shubharanjan, Varghese, Annie

Abstract: The purpose of this review was to assess the effectiveness of the buccal myomucosal flap in secondary
repairs of cleft palate in 20 patients. Totally, 20 patients, who underwent secondary palatoplasty between 5 years and 8 years in which a buccal myomucosal flap was used, were reviewed retrospectively. All patients had undergone at least one previous attempted repair at other institutions. Indications for the secondary repair included velopharyngeal incompetence and/or oronasal fistula. Patients were evaluated preoperatively for oronasal fistula status, velopharyngeal competence, nasal resonance, speech quality, and nasal escape. The buccal myomucosal flap was used in all 20 patients, and there was marked increase in the quality of speech as well as nasal regurgitation decreased. In patients with levator dysfunction due to poor primary surgery and glottal speech the results were inconclusive. Palate re-repair combined with a buccal myomucosal flap, occasionally in conjunction with other techniques, is an effective method for correcting failed cleft palate repairs. Minimum donor site morbidity and complication makes the buccal flap a useful armamentarium of a cleft surgeon.

Source: Medline

**Full text:** Available ProQuest at Contemporary Clinical Dentistry

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**44.** Title: Use of hyperdry amniotic membrane in operations for cleft palate: a study in rats.

**Citation:** The British journal of oral & maxillofacial surgery, Apr 2015, vol. 53, no. 4, p. 358-363 (April 2015)

**Author(s):** Tsuno, Hiroaki, Noguchi, Makoto, Okabe, Motonori, Tomihara, Kei, Yoshida, Toshiko, Nikaido, Toshio

**Abstract:** The growth of maxillary bone and the development of dentition are often impaired in patients who have had pushback operations for repair of a cleft palate. There has been considerable discussion about the most suitable technique or material used in such repairs to resolve the problem. Hyperdry amniotic membrane, a new preservable material derived from human amnion, has recently been introduced in several procedures. We have evaluated its use during pushback surgery in animal studies to try to correct the inhibition of growth and development of the maxilla. Mucosal defects were created in 3-week-old rats, and then covered with hyperdry amniotic membrane or not. Healing was assessed by histological and morphological examination at 1 week and 7 weeks postoperatively. In the group treated with hyperdry amniotic membrane, submucosal tissue was reconstructed successfully during the early postoperative period. Lateral palatal growth was not inhibited as much, and medial inclination of the teeth was less, after a period of growth using this material. The results suggest that hyperdry amniotic membrane is a suitable new dressing material for use in the treatment of cleft palate. Copyright © 2015 The British Association of Oral and Maxillofacial Surgeons. Published by Elsevier Ltd. All rights reserved.

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