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Review Article

**CLEFT LIP AND PALATE SURGICAL MANAGEMENT IN
PEDIATRIC: SYSTEMATIC LITERATURE REVIEW****Wejdan Ali Mobasher^{1*}, Bashair Mohammed Asiri¹, Lujain Ahmed Asiri¹, Layla
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Mohammed Muteb Alharbi⁵**¹ King Khaled University, Abha, Saudi Arabia,² King Faisal University, Alhasa, Saudi Arabia,³Ibn Sina National College for Medical Studies, Jeddah, Saudi Arabia,⁴ King Abdulaziz Univeristy, Jeddah, Saudi Arabia,⁵ King Saud bin Abdulaziz University for Health Sciences, Riyadh, Saudi Arabia.**Abstract**

This review is aiming to discuss the cleft lip and palate surgical management in pediatric, the presented review was conducted by searching in Medline, Embase, Web of Science, Science Direct, BMJ journal and Google Scholar for, researches, review articles and reports, published over the past years. were searched up to November 2018 for published and unpublished studies and without language restrictions, if several studies had similar findings, we randomly selected one or two to avoid repetitive results. On the basis of findings and results this review found importance of surgical technique, age at repair, and cleft type for velopharyngeal function, three-dimensional (3D) facial morphology in patients surgically corrected for unilateral cleft lip and palate (UCLP) following pre-surgical nasoalveolar molding (NAM), features of dental occlusion in patients born with a unilateral cleft lip and palate (UCLP), and the quality of outcomes in patients with unilateral cleft lip and palate (UCLP)

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INTRODUCTION:

Incomplete understanding of factors affecting outcome of treatment in children with unilateral cleft lip and palate (UCLP) has resulted in large variety of protocols and surgical techniques employed by various cleft teams worldwide. A survey of European cleft centers [1], demonstrated that 201 cleft teams practiced 194 different protocols. Although approximately 43% of them were two-stage, in which lip closure was followed by simultaneous repair of hard and soft palate, the number of primary surgeries ranged from 1 (when all cleft structures are repaired simultaneously) to 4 (when cleft structures are closed at different timings). Comparison of treatment outcome of several European cleft centers—the Eurocleft studies—showed that one of the best treatment outcomes was achieved by a center practicing a three-stage treatment protocol with hard palate closure delayed until 8–11 years of age. [2]

Nollet et al. [3] who examined dental arch relationship in a sample of 9-year-olds treated at the Radboud University Nijmegen Medical Centre according to three-stage protocol with delayed hard palate closure (DHPC), also found a very good treatment outcome compared to the Eurocleft study. Also, studies by Lilja et al. [4] and Sinko et al. [5] revealed favorable dental arch relationships following protocols including DHPC. Lilja et al. reviewed treatment results in a sample of 104 patients treated consecutively by the Gothenburg cleft team, Sweden, and found that 85% of them were rated as having good or very good outcome. Sinko et al. examined dental arch relationship in 123 9-year-olds treated according to the Vienna concept—four-stage protocol including DHPC at 6 years—and found that 71.5% of the patients were assessed as having good or very good outcome. Few studies have so far examined the long-term results following one-stage repair of UCLP. In a cephalometric study, Corbo et al. [6] compared two small samples of preadolescent children with complete UCLP that were operated according to the Malek procedure. In 11 children, the complete cleft was closed in one operation at 3 months of age, and in ten children, a two-stage repair was used where the soft palate was closed at 3 months and lip and hard palate closed at 6 months of age. No difference between the two protocols was observed. Savaci et al. [7]

Successful management of cleft lip and palate requires special attention to the soft tissues of the lip and nose, as well as the hard tissues of the maxilla, including dental abnormalities. [8] It is thought that good techniques can provide a foundation upon which the results of lip and primary nasal surgery can be built. For example, when using nasoalveolar molding (NAM) it is

suggested that primary surgical repair of the nose and lip heals under minimal tension, reducing scar formation and improving the esthetic results. [9,10] Thus, NAM is used to reshape or remodel the nasal cartilages and mold or remodel the maxillary arch before cleft lip repair and primary rhinoplasty. It has also been suggested that NAM provides esthetic benefits in terms of nasal tip and alar symmetry, and functional benefits in terms of improved dental arch form. Grayson and Cutting [11].

There is at present a surge of interest in the development of strategies to improve the outcome of treatment for patients born with a cleft lip and palate.¹² It has become clear that poorly performed primary surgery is likely to compromise facial growth and dental development. [13] It is also evident that a wide range of surgical techniques exist to correct this anomaly but with no clear-cut guidelines for optimal timing or method. As a result, when outcome with one technique appears disappointing, surgeons are likely to make modifications to, or radical departures from, their current regimes. These changes are often made with little data or rationale. Until recently it has been perceived that the effects of primary surgery on the maxilla and facial growth cannot be determined until facial development is complete, usually in late adolescence. However, a large European study involving six centres demonstrated that it is possible to detect differences in outcome as early as 10 years of age. [14] The ability to predict the outcome even earlier would provide surgeons with a sound basis on which to judge their results and a rational indication that modification of a technique was justified. Recently, some evidence from a study of radiographs suggested that soft-tissue outline may be a useful indicator to evaluate quality of treatment [15]

Timing of palatal surgery in patients with cleft lip and palate has long been a controversial issue. Improved facial growth has been the aim of a persistent push for primary veloplasty with late closure of the hard palate [16,17,18]. However, severe speech deficiencies and functional difficulties have been reported in patients treated by delayed closure of the hard palate the debate about early compared with late palatal surgery during the last two decades has been based mainly on information from studies that have shown extremely bad speech after delayed hard palate closure. [19,20]

The outcome of primary repair surgery for cleft lip and palate is often expressed in the subsequent quality of speech, hearing, facial appearance, and the dentoskeletal relationships of the mandible and maxilla. It is recognized that poor growth of the

maxillary region is related to the effects of primary repair surgery [21], and this is of particular concern for the orthodontist who must correct any residual dentofacial discrepancies during early adolescence. Although those patients who have displayed favorable facia growth may require only relatively routine orthodontic treatment, patients with unfavorable facial growth often also need orthognathic surgery for complete correction of

METHODS:

The present review was conducted November 2018 in accordance with the preferred reporting items for systematic reviews and meta-analyses (PRISMA) declaration standards for systematic reviews. We reviewed all the topics on cleft lip and palate surgical management, such as importance of surgical technique, age at repair, and cleft type for velopharyngeal function, three-dimensional (3D) facial morphology in patients surgically corrected for unilateral cleft lip and palate (UCLP) following pre-surgical nasopalveolar molding (NAM), features of dental occlusion in patients born with a unilateral cleft lip and palate (UCLP), and the quality of outcomes in patients with unilateral cleft lip and palate (UCLP). To achieve this goal, we searched Medline, Embase, Web of Science, Science Direct, and Google Scholar for, researches, review articles and reports, published over the past 15 years.

Our search was completed without language restrictions. Then we extracted data on study year, study design, and key outcome on cleft lip and palate surgical management. The selected studies were summarized, and unreproducible studies were excluded. Selected data is shown in the Table 1.

Studies has been rated as being high quality by an established evaluation process based on the DyunaMed criteria and it's based on the level of evidence as following:

Level 1 (likely reliable) evidence: representing research results addressing clinical outcomes and meeting an extensive set of quality criteria which minimize bias. example: Randomized controlled trial/meta-analysis.

Level 2 (mid-level) evidence: representing results addressing clinical outcomes and using some methods of scientific investigation but not meeting the quality criteria to achieve level 1 evidence labeling. Example: well-designed non-randomized clinical trials.

Level 3 (lacking direct) evidence: representing reports that are not based on scientific analysis of clinical outcomes. Examples include case series, case reports, expert opinion and conclusions extrapolated indirectly from scientific studies.

dentofacial discrepancies. The quality of skeletal and dental relationships can be assessed in a standardized way using cephalometric analysis of lateral skull radiographs and rating the dental arch relationships using the Goslon scoring system. [22,23,24] These measurement techniques are now widely accepted and have been used in a recent U.K. study of clinical outcomes of cleft lip and palate care. [25]

Inclusion criteria

Inclusion criteria were cleft palate: unilateral, surgical management, children.

Exclusion criteria

Irrelevant articles [not related to the aim of this review and articles that did not meet the inclusion criteria in this review.

Data extraction and analysis

Information relating to each of the systematic review question elements was extracted from the studies and collated in qualitative tables. Direct analysis of the studies of cleft palate.

RESULTS:

Pharyngeal flap was necessary in 14% of von Langenbeck and 15% of VY repaired patients. There was a significant linear association ($p = 0.025$) between age at repair and velopharyngeal insufficiency (VPI). Patients with an attached vomer, soft cleft palate (SCP), and unilateral cleft lip/palate (UCLP) had a 10% flap rate, whereas those with an unattached vomer, hard/soft cleft palate (HSCP), and bilateral cleft lip/palate (BCLP) had a 23% flap rate ($p = 0.03$). Age at repair was critical for the unattached-vomer group ($p = 0.03$) but was not statistically significant for the attached-vomer group ($p = 0.52$).²⁶

Using the first two principal components, which accounted for 63% of the total shape-change, UCLP and control groups showed similar distributions in the modal space ($p > 0.05$). For the UCLP group, the mean 3D facial form was smaller and less protrusive when superimposed on the non-cleft mean. Using FESA, reductions in facial volume were found in the UCLP group, involving the columella (29%), labial tubercle (51%), lower lip (29%) and lateral aspects of the face (19%). The UCLP group also showed increases in size above the tip of the nose (25%) and laterally to the columella directly below the nares (29%) [27].

The mean age of the 5-year-old records was 5.3 SO 0.4 years (range 5-6.4 years) and for the 10 year olds, 9.9 SD 0.8 years (range 8.3-12 years). Intra-examiner agreement for the two separate Goslon rankings of 5 and 10-year-old records is shown in Table 1. It is evident that for both the 5 and 10-year old records, agreement was good and at a similar

level. When the inter-examiner correlation for both 5 and 10-year-old records was examined (Table 2), agreement was at worst moderate and at best very good (Altman, 1991). The relationships between the 5 and 10-year-old Goslon scores were then compared. Since the Goslon Yardstick was developed for 10-year-old, it is not entirely appropriate to examine agreement statistics between the Yardstick and the 5-year-old records. A more representative evaluation of this relationship is to compare the number of records which either retained their Goslon score or become worse from 5 to 10 years of age (Table 3). It was found firstly that in two scoring exercises on both 5 and 10-year-old models, the observers were all reasonably consistent. Secondly, for two examiners (B and D), 93 per cent of the scores they made on 5-year-old models stayed the same or became worse in the 10-year-old models. The weakest prediction

for this was 70 per cent (examiner C). Finally, from the 27, 5-year-old records, those cases where agreement was high between the examiners were selected. Two representative cases for each of the five groups were then chosen to represent a range for each category from excellent (Group 1) to very poor (Group 5). [28]

Twenty-nine of the 34 subjects had good or satisfactory arch relationships. Comparison between the surgeons revealed that the Wardill-Kilner group had a greater proportion of Goslon grades of greater than 3, indicating poor arch relationships. However, this difference failed to reach statistical significance. There were no significant cephalometric differences between patients treated by the two surgeons. [29]

Table (1) Results from Sequencing Studies:

Author and year	Sample	Surgical management	Key point	Level of evidence
EILEEN M, et al. 1997. ²⁶	228 patients	importance of surgical technique, age at repair, and cleft type for velopharyngeal function	Surgical technique was not a significant variable either in aggregate or for the Veau types	Level 2
Singh GD, et al 2007. ²⁷	15 patients with left UCLP	three-dimensional (3D) facial morphology in patients surgically corrected for unilateral cleft lip and palate (UCLP) following pre-surgical nasoalveolar molding (NAM).	Following surgical repair of UCLP in patients previously treated with NAM, 3D facial morphology was virtually indistinguishable from the non-cleft mean	Level 1
Nikki A, et al. 2016. ²⁸	27 subjects (17 males and 10 females)	features of dental occlusion in patients born with a unilateral cleft lip and palate (UCLP).	70 per cent of cases at 5 years of age remained in the same category or deteriorated by 10 years of age	Level 1
CHRIS D, et al. 2003. ²⁹	Thirty-four children	The quality of outcomes in patients with unilateral cleft lip and palate (UCLP)	Although the difference was not statistically significant, the highest proportion of patients likely to require orthognathic surgery was found in those treated using the Wardill-Kilner technique.	Level 2

DISCUSSION:

Does statistical analysis of our experience bring us any closer to the goal of optimal management of cleft palate? We found that surgical technique (von Langenbeck versus VY repair) was not a determinant. We substantiated the trend toward earlier palatal repair in attaining velopharyngeal closure. Lack of correlation between velopharyngeal function and extent of skeletal clefting was an unexpected finding [26]

The systematic errors of the imaging method employed in this study have been validated with a mean distance error of 0.04 mm and a RMS of 0.36 mm (16), and submillimeter accuracy (17). Random errors were addressed by duplicate digitization on two occasions, which yielded similar results ($p > 0.05$). Thus, the systematic and random digitization errors were considered in this study but found to be non-significant. The NAM is undertaken after correction of the alveolar segments, and considerable success is claimed when deploying NAM. However, the use of NAM is limited to a few centers and the numbers of patients for a study of this nature are limited. In a previous study (4), improvements in nasal morphology following NAM before surgical correction were noted. Therefore, the present study was performed to study 3D facial morphology following surgical repair of UCLP in patients after NAM compared with a non-cleft, control group. Prasad et al. (18), using 3D dental casts, concluded that different regimens in the management of UCLP can significantly affect maxillary growth. Thus, the management of the patients in this study followed that of Cutting et al. (19), who also used a combined protocol of pre-surgical NAM with a one-stage lip, nose, and alveolus repair for bilateral clefts [27]

The aim of this study was to develop a robust and reliable index for dental relationships of 5-year-old patients born with a UCLP. It is emphasized that the sample was longitudinal and that none of the records from the 5 or 10-year-old groups had received any orthodontic treatment or secondary alveolar bone grafting. The position of the teeth was therefore not influenced by these variables. At the time, the standard procedures for children born with UCLP and referred to Frenchay Hospital was to repair the lip at 3 months of age and the palate at 6 months. Palatopharyngoplasty was performed in cases with speech problems and in all there were five such patients in the sample. There is no definitive proof that this latter procedure can influence facial growth. [28]

There is still no agreement on the best surgical approach to the primary repair of cleft lip and palate (Molsted, 1999). Decisions about clinical treatment in health care should ideally be guided by

evidence from randomized controlled trials (Cochrane, 1972; Richards and Lawrence, 1995), and the randomized, prospective clinical trial has been recommended as the most appropriate way of assessing the efficacy of cleft palate treatment regimens (Semb and Shaw, 1998). Retrospective multicenter studies such as the Eurocleft study (AsherMcDade et al., 1992; Mars et al., 1992; Molsted et al., 1992; Shaw et al., 1992a, 1992b) have enabled the collection of large samples. However, analysis is often hindered by the large number of potentially confounding variables including differences in surgeons and in the timing and techniques of surgical repair between centers and also within individual centers. Furthermore, other treatment interventions following the primary repair surgery may also influence outcome, and these interventions can vary depending on individual surgeon and center protocols. It can therefore be difficult to identify any factors that may be responsible for successful or unsuccessful outcomes when using multicenter data. [29]

CONCLUSION:

Finally, the results of this studies show the cleft lip and palate surgical management in pediatric. On the basis of findings and results this review found importance of surgical technique, age at repair, and cleft type for velopharyngeal function, three-dimensional (3D) facial morphology in patients surgically corrected for unilateral cleft lip and palate (UCLP) following pre-surgical nasoalveolar molding (NAM), features of dental occlusion in patients born with a unilateral cleft lip and palate (UCLP), and the quality of outcomes in patients with unilateral cleft lip and palate (UCLP) are most common surgical management in pediatric .

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