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The Effects Of Early And Delayed Palatal Repair On Maxillary Growth And Speech Outcome In Unilateral Cleft Lip And Palate Patients: A Systematic Review

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Abstract. *Some cleft clinics use a two-stage method to correct the cleft palate, which helps avoid reduced maximum growth after early hard palate surgery and impaired speech development after late soft palate repair.. This study sought to evaluate the effects of early vs late palatoplasty on speech and maximum growth results. The following databases were searched: Cochrane Library, Science Direct, Pubmed, and NCBI. The primary objective of this study was to determine whether unilateral, non-syndromic cleft lip, alveolus, and palate patients who underwent early or late hard palate surgery before or after 18 months of age had an impact on speech and maxillary development. Things like case studies, reviews, editorials, textbooks, and studies conducted on animals weren't included. The following systematic observations consist of eight studies. Most studies had methodological quality that ranged from fair to good. This review discusses the experience of cleft centers using a variety of one and two stage palatoplasty procedures and timing, and shows inconsistent results regarding maximal progression. The seemingly inconsistent results of the studies included in this observation of speech outcomes suggest that there is insufficient evidence to conclude whether one- or two-stage palatoplasty has a significant effect on maximal development for a variety of reasons. The review provides inconclusive evidence on the impact of hard palate repair timing on facial growth and speech outcomes in unilateral cleft lip and palate patients, highlighting the need for further controlled studies.*
Keywords: *UCLP, palatoplasty, maxillary growth, speech*

INTRODUCTION

The debate in cleft palate repair revolves around achieving optimal speech development while preventing abnormal midfacial growth, which often occurs out of sync. Different cleft centers worldwide employ varied protocols, disagreeing on the ideal timing and techniques for repair to achieve the best outcomes in speech and midfacial growth (Alonso & Raposo-Amaral, 2018; Smith & Losee, 2014).

To overcome this disparity, several cleft hospitals use a two-stage palate surgery strategy that divides hard palate repair from soft palate restoration. The plan's objective is to mitigate the negative effects of delayed soft palate repair (impaired speech development at an earlier age) and premature hard palate repair (limited maxillary growth due to scarring on the bony palate) (Alonso & Raposo-Amaral, 2018). After primary veloplasty, the primary benefit of this

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two-stage treatment is the shrinking of the hard palate gap. By employing a smaller flap (minimizing mucoperiosteal elevation) and lowering the hard palate gap following the first repair, this technique seeks to minimize any potential adverse effects on subsequent development and allow for a tension-free repair (Alonso & Raposo-Amaral, 2018; Smith & Losee, 2014).

Varied midfacial development outcomes have been observed as a result of cleft centers using varied procedures and timing for early soft palate surgery (between 3 and 18 months) and late hard palate repair (between 3 and 12 years) (Al-Ghatam et al., 2015; Long et al., 2011; Bakri et al., 2014). Interestingly, while some centers report improved speech outcomes with the two-stage approach, others show worse results (Sell et al., 2015; Dissaux et al., 2015; Lohmander et al., 2012). Contrarily, one-stage repairs, where both palates are closed in a single surgery, tend to yield better speech outcomes overall (Randag et al., 2014; Klintö et al., 2014; Funayama et al., 2014). While there is discussion in support of two-stage surgery, avoiding the opportunity to improve the soft palate's morphology and facilitate proper speech development in cases of early cleft palate is not warranted due to insufficient evidence of stunted midface growth caused by early repair. In particular, eighty-eight percent of the cleft palate surgeons in the United States who participated in the survey preferred a one-stage procedure (Katzel et al., 2009).

In order to better understand when palatoplasty is most beneficial for maxillofacial development and speech quality in people with non-syndromic full unilateral cleft lip and palate, a comprehensive study was carried out.

RESEARCH METHOD(S)

Eligibility Criteria

In this meta-analysis and systematic review, we adhered to the PRISMA guidelines (Page et al., 2021). All patients were eligible for inclusion if they had a non-syndromic complete unilateral cleft lip, alveolus, and palate and had hard palate surgery either before or after the 18th month of life. The research used dental casts or cephalometric analysis to assess maxillary development.

Case reports, case series, reviews, textbooks, editorials, and animal research were all excluded from the study. Furthermore, manual research and citation were done to make sure that every study that was accessible was included.

Search Strategy and Study Selection

On March 5, 2023, the literature search was started and finished. Using search phrases such as ((palatoplasty) OR (palatal repair) OR (palatal surgery) AND ((unilateral) OR (cleft palate) OR (cleft lip)) AND ((growth) OR (speech)), we methodically retrieved publications from Pubmed, NCBI, Science Direct, and Cochrane Library. The specific Medical Subject Heading (MeSH) phrases that are utilized are shown in Table 1. The Rayyan program was utilized to enter all information in order to detect duplicates and enable the writers to pick pertinent research collaboratively. The first search was carried out by the writers, who also imported papers into Rayyan software from a number of academic databases. Each author individually screened the studies, and a second author confirmed the results of all the initial searches. Conflicts that arose throughout the screening process were settled by group discussion until an agreement was achieved. The respective authors were contacted once by email if more information was needed.

Data extraction and Quality assessment

Each writer independently extracted data, and a third author verified the results. Authors, publication year, location, design, duration, participant age range, intervention scheduling and protocols, surgical methods, age at evaluation, and statistical methodologies were all well documented. The standard error of the mean (SEM) data was transformed into mean and standard deviation using the Cochrane procedure (Cochrane, 2024).

Each author assessed the included studies' quality independently using the New Castle Ottawa Scale. Three or four stars for selection, one or two stars for comparability, and two or three stars for outcome/exposure were used to determine high-quality research publications. Two stars in the selection domain, one or two stars in the comparability domain, and two or three stars in the outcome/exposure domain were rated fair quality studies by researchers. In the categories of selection, outcome/exposure, and comparability, research with scores of 0 or 1 was deemed to be of low quality (Ottawa Hospital Research Institute, 2024). Internal disputes were settled until an agreement was achieved.

FINDINGS AND DUSCUSSION

Study selection and characteristics

After eliminating duplicates, 2523 records were left out of the initial 2957 that were obtained. 2190 studies were eliminated from the group based on screening of the abstract and title. 333 reports were left over from this process for additional eligibility review. Studies using

animals, improper research designs, unsuitable results, studies without complete publications, and studies involving bilateral cleft lip were among the exclusions. Using a manual search, two more studies by Liao et al. and Funuyama et al. were located and added to the review. The systematic review contained eight studies (Figure 1). Von Langenbeck, Wardill-Killner, modified Furlow, Veau procedure, closing the residual hard palate in a case of mixed dentition, performing intravelar veloplasty at the same time as closing the mucoperiosteal and alveolus, and soft palate closure utilizing a vomer flap were all methods used for palate repair. A number of other procedures were also considered..

Quality of study assessment

Predetermined criteria were used to evaluate the papers that are discussed here. The remaining studies were categorized as "good quality," while four of the included studies were rated as "fair quality" (Table 2). In summary, every study satisfies the requirements to be included in the review.

Maxillary growth

The results of maxillary development at age twenty were only published in one research (Liao et al., 2010). Two studies, Liao et al. (2010) and Yamanishi et al. (2011), aimed to determine the effects of one- or two-stage palatal surgery on the maxilla's relationship to the cranial base. Liao et al. (2010) suggested that one-stage repair led to a longer adult cranial base length compared to two-stage repair. However, Yamanishi et al. (2011) did not find any difference in outcomes between the two approaches. Thus, there's contradictory evidence regarding the effect of these surgeries on the cranial base.

The study found that the group that had delayed hard palate repair at 18 months had longer anterior-posterior maxillar length (ANS-ptm'), a longer distance from the anterior nasal spine to projection point N in the palatine (N'-ANS), and a greater anterior facial height (N-N') (Yamanishi et al., 2011). Furthermore, a two-stage palatal repair improved the sagittal jaw connection, according to one study (Liao et al., 2010). Bakri et al. (2014) evaluated vertical maxillary increase in another trial. As opposed to the 9-month treatment group, the delayed two-stage palatoplasty group showed improved outcomes in terms of anterior facial height (n-sp'), maxillary anterior height (baseline of the maxilla (NL)-is), overbite, and maxilla tilt (Bakri et al., 2014).

The study conducted by Holland et al. (2007) discovered statistically significant variations in maxillary growth between patients who had delayed palatoplasty and those who had one-stage palatoplasty at 12 months. The early palatoplasty group in particular improved their SNA and ANB angles (Holland et al., 2007). In contrast, patients treated later were more

likely to present with Class III malocclusion and required Le Fort I surgical advancement more frequently (Holland et al., 2007). This suggests that early one-stage palatoplasty may have advantages in terms of maxillary growth compared to delayed palatoplasty.

An analysis of the impact of a single-stage palate repair on mandibular length was conducted using cephalometric measures, namely Articulare-Menton (Art-Me) and Articulare-Gnathion (Ar-Gn). One-stage palate repair significantly affected mandibular length in a research done at age 20 (Ar-Gn, $p = 0.05$) (Liao et al., 2010). However, in another study, which evaluated Art-Me and ramus length at 4 years of age, no significant effect was observed (Yamanishi et al., 2011). On the contrary, Daskalogiannakis et al. (2006) and Zemann et al. (2011) found no statistically significant changes between the treatments when comparing the sagittal development of two distinct centers. Daskalogiannakis et al. found that the early-one-stage palatoplasty group had greater SNB, mandibular length, and SNA point. These variations were limited to mandibular points (Daskalogiannakis et al., 2006).

Bakri et al. (2012) discussed the height of the palatal vault. The study by Bakri et al. revealed that the palatal vault height was noticeably larger in the group that underwent early one-stage closure rather than the delayed two-stage approach. In this study, we found no statistically significant difference in the height of the palatal vault between the right and left sides (Bakri et al., 2012).

Speech outcome

A retrospective research comparing one-stage and two-stage palatal surgery revealed that at the age of 15, the two-stage repair group had higher cases of velopharyngeal insufficiency (VPI), nasal emissions, hypernasality, and less desirable phonation values (Holland et al., 2007). Additionally, a different research found that the two-stage group performed worse on speech outcomes related to velopharyngeal function or subsequent operations for velopharyngeal insufficiency than the one-stage group ($p < 0.001$ for both comparisons) (Liao et al., 2010). Yamanishi et al., on the other hand, found no statistically significant difference between the one-stage and two-stage repair groups at 4 years of age in the frequency of articulation mistakes and velopharyngeal insufficiency (VPI) (Yamanishi et al., 2011). Similarly, at 4 and 8 years of age, Funayama et al. assessed velopharyngeal function and found no significant changes between the one-stage and two-stage palatoplasty groups (Funayama et al., 2014).

DISCUSSION

A number of variables influence maxillofacial growth in children with clefts, but the main ones are intrinsic growth problems and treatments, including non-surgical and surgical procedures. Surgical operations are frequently linked to maxillary hypoplasia because they disrupt growth centers and cause the production of scar tissue. While cleft lip surgery prevents sagittal maxillary length in patients with cleft lip and palate, early cleft palate repair has been shown to impede maxillary development in all dimensions (Shi & Losee, 2015; Berkowitz, 2015). The methods that reduced growth the most were Veau's pushback and Langenbeck's relaxing incision cleft palate repairs. Furlow's cleft repair, on the other hand, showed little negative consequences on maxillary development. Rather than the order of hard or soft palate surgery, the time of hard palate closure was the main factor influencing postoperative growth (Shi & Losee, 2015).

The initial veloplasty narrows the hard palate cleft, which is a major advantage of two-stage surgeries. Due to the reduced defect size, tension-free healing with smaller flaps and less mucoperiosteal elevation is made possible later in the growing period. It is assumed that this strategy will have a less negative effect on future growth (Alonso & Raposo-Amaral, 2018). The results of one- or two-stage palatoplasty on maxillary development are unclear, according to this evaluation of studies. The age at which the last evaluation was completed may be a significant factor contributing to the inconsistent outcomes, in addition to differing surgical techniques. Of all the studies, only one evaluated the result after growth had stopped at the age of twenty (Liao et al., 2010). There is still insufficient scientific evidence to support the claim that two-stage palatal closure improves the sagittal-jaw relationship.

In addition, the two techniques that are most frequently employed to assess maxillofacial development are dental cast analysis and cephalometry. Despite being frequently utilized, cephalometric studies have intrinsic methodological flaws that cause variance based on the kind of landmark, the observer, and the radiographic projection. Furthermore, it may be difficult to determine anatomical landmarks in individuals with unilateral cleft lip and palate (UCLP) due to the use of different X-ray equipment (Alonso & Raposo-Amaral, 2018; Bongaarts et al., 2008).

Ensuring that cleft patients achieve normal speech, including proper resonance, reduced nasality, and improved intelligibility, is a crucial objective in cleft care. The unintelligible speech of cleft patients can influence how typically developing peers perceive them socially and personally (Lee et al., 2017). Additionally, cleft children experiencing fewer speech difficulties tend to report higher scores in the Pediatric Quality of Life Inventory, encompassing

both physical and psychosocial health domains (Damiano et al., 2007). Consequently, cleft surgeons have a responsibility to regularly and critically evaluate the outcomes of cleft repair procedures, particularly focusing on speech quality, such as velopharyngeal insufficiency rates and the need for secondary speech surgeries, as well as palatal integrity, including fistula rates (Alonso & Raposo-Amaral, 2018; Smith & Losee, 2014).

discovered statistically significant variations in maxillary growth between patients who had delayed palatoplasty and those who had one-stage palatoplasty at 12 months. The early palatoplasty group in particular improved their SNA and ANB angles (Timbang et al., 2014). The study finds conflicting scientific data about the effectiveness of early one-stage palatal repair vs delayed two-stage palatal repair in treating nasal resonance, nasal emission, hypernasality, phonation, and speech intelligibility.

The studies involved in this review present contradictory results on speech outcome (Funayama et al., 2014; Liao et al., 2010; Yamanishi et al., 2011; Holland et al., 2007)**.** In this regard, it is essential to draw attention to variables such as the surgical operation's kind and timing, the patient's age at the time of the treatment, and the surgeon's expertise determining speech results. It should be mentioned that while several of the examined studies included nasal endoscopy as an extra diagnostic technique, none of them carried out reliability assessments (Funayama et al., 2014; Liao et al., 2010).

There are many limitations that should be taken into account when evaluating the results of this systematic study. The examined literature did not offer conclusive data about the impact of hard palate repair timing on speech and facial development outcomes in cleft lip and palate patients. A number of variables, including the age range of patients, the timing of evaluations, and the variations in hard palate repair timing between trials, contributed to this lack of clarity. To address these limitations and improve reliability in future research, there should be controlled studies that include age cut-offs for the timing of delayed and early palatoplasty. This will lead to more reliable conclusions and potentially improving the management and treatment of cleft lip and palate patients.

Given that studies done in English are more likely to be published with significant results, biases in research selection and publishing may have had an impact on the small number of evaluated studies. All of the studies reviewed were non-randomized which may carry a high risk of bias. This is due to ethical considerations as it could be deemed unethical to delay surgery when speech is adversely affected after a certain age. It is important to take these limitations into account when evaluating the results of the systematic review.

Furthermore, no meta-analysis was performed due to the diversity of the samples, variables, and quality of the studies reviewed. Combining such diverse results may be inappropriate from a clinical standpoint. Despite the absence of a meta- analysis, the review offered insights into the sources of diversity among the studies. According to this study, maxillary development may be more significantly impacted by hard palate repair during the pubertal growth period. Furthermore, in certain circumstances, the surgeon's experience may have a greater impact on facial development results than the time of hard palate repair (Alonso & Raposo-Amaral, 2018; Smith & Losee, 2014).

CONCLUSION AND RECOMMENDATION

In conclusion, the review's findings remain equivocal on how the time of hard palate repair influences speech and facial development outcomes for people with unilateral cleft lip and palate. The theory that waiting to have hard palate surgery will help maxillary development is not supported by the available data. The passage underscores the need for further well-designed, controlled studies with reliable methods, particularly focusing on long-term results, to address these limitations and provide more definitive conclusions.

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