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A Summary of the Existing Data on Cleft Surgical Outcomes: What Do We Not Know?

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Summary: Globally, cleft surgical protocols have nuanced differences between centers, yet the goals of cleft lip and palate (CLP) repair are similar. These protocols all aim to achieve optimal aesthetic appearance and speech outcomes while limiting significant complications such as velopharyngeal insufficiency and midface hypoplasia. Variability in complication rates following primary CLP repair has been well documented within the literature. A series of systematic reviews investigate the impact of factors on surgical outcomes, all demonstrating an inability to comment on the ideal technique and timing to optimize outcomes. This article presents a summary of the current state of the literature on surgical outcomes following CLP repair. Studies were limited by small samples sizes; standardized measurement tools; study design; and lack of diversity in cleft phenotype, severity, and patient population. These limitations highlight the need for further research with more representative populations globally, standardized measurement tools, and a global consortium of cleft surgeons to make recommendations based on improved data. As the need for training in cleft surgery expands across the globe, evidence-based algorithms are essential to optimize outcomes and limit costly complications.
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INTRODUCTION

The goals of cleft lip and palate (CLP) repair have focused on optimizing aesthetic appearance and speech outcomes while limiting velopharyngeal insufficiency (VPI) and midface hypoplasia (MFH). Many surgeons developed their own protocols and modifications to optimize these outcomes, inciting great diversity in technique and timing and moving us further away from a gold-standard approach.[1–3] As a result of these modifications over time, there are numerous cleft care protocols practiced worldwide, with variable consensus and data fraught with limitations that hinder our ability to evaluate outcomes. CLP phenotypes, severity, and presentation differ vastly; while aspiring to 1 standard that fits every setting is unreasonable, there is a need for evidence-based recommendations on consistent techniques and specific timings that will allow us to optimize surgical outcomes in any setting.[4] To develop these recommendations we need high-quality data.[5–8]

A series of systematic reviews and meta-analyses have been conducted by our research group to evaluate the evolution of CLP repair and existing data on surgical outcomes after primary CLP repair.[1,2,9–13] This article summarizes those findings and the current state of the literature on the role of technique and timing on surgical outcomes such as VPI and MFH.

THE CURRENT STATE OF THE LITERATURE

Cleft Lip Repair Outcomes

The subjective nature of evaluating lip outcomes has continuously impaired direct comparison between outcomes and protocols. The Asher-McDade scale is arguably the most well-known of available metrics and has been both modified and used in novel methods, such as crowd-sourcing in attempts to improve reliability, but intrinsic biases of raters cannot be fully eliminated.[14–16] Current evaluation methods have been shown to poorly correlate with patients' satisfaction of their results.[17] This lack of consensus in evaluation and decision for secondary lip surgery is reflected by a systematic review

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from Sitzman et al[18] reporting lip revision surgery rates from 0% to 100%. Novel options such as 3-dimensional image analysis or artificial intelligence may standardize classification of lip outcomes in the future, but are not yet widely available, even in high-resource settings.[19,20] Improving understanding of lip outcomes will require simple, validated tools that can be easily implemented across multiple centers, allowing for collection of data in a standardized format.

Velopharyngeal Insufficiency

Speech is critical to childhood development and is prioritized in many centers around the world.[21–23] Our group investigated VPI rates between surgical protocols globally, and discovered that there is great variability ranging from 8% to 20%.[9] The systematic review included 35 articles, totaling 10,795 patients with a mean age of 15.7 months at primary palate repair and an overall mean VPI rate of 20%.[9] Primary repair between 6 and 12 months demonstrated the lowest rate of VPI.[9] The review was unable to analyze rates of VPI by surgical technique due to insufficient detail in selected articles.[9] Less than half the articles gave a written or graphical description of their method (43%), whereas the rest simply named a technique (26%), or cited a technique article (20%).[9] Although there was great variability in the results, these studies all shared the same limitations: lack of standardized measurement tools to evaluate speech outcomes, limited long-term follow-up, lack of technique description details, small sample sizes with lack of diversity in population, and inability to account for important factors such as severity.[24]

Midface Hypoplasia

The Natural History of Midface Hypoplasia in Unoperated Cleft Palates

The impact of the natural history of maxillary growth is not fully understood. With the aberrancies in embryological development, tissue deficits, and often-missing lateral incisor, we can hypothesize that there may be an element of inherent MFH in patients with cleft palate before any surgical intervention; however, we lack sufficient data to validate this. A systematic review by Wlodarczyk et al[10] evaluated impact of cleft surgery on maxillary growth comparing sella-nasion-A point (SNA) angles of unoperated unilateral CLP (UCLP), bilateral CLP (BCLP), and isolated cleft palate (ICP) to controls. Control patients were without craniofacial pathology and of similar age to those with clefts.[10] Before surgery, unoperated UCLP and BCLP demonstrated significantly larger angles than controls, whereas unoperated ICP had significantly smaller angles than controls.[10]

The findings suggest that ICP may inherently be associated with an element of MFH as the intact lip could restrict growth anteroposteriorly.[10] All phenotypes of repaired cleft palates had significantly smaller angles than controls, reinforcing our understanding that surgery is associated with scarring and development of MFH.[10] The review was limited by the lack of large-scale, high-powered studies to differentiate impact of repair techniques or protocols on the development of MFH.[10]

Takeaways

Question: What gaps in knowledge exist regarding cleft surgical outcomes within the current literature?

Findings: Conclusions regarding the ideal technique and timing to prevent velopharyngeal insufficiency, midface hypoplasia, and fistula, were limited by small sample sizes, lack of standardized measurement tools, and lack of diversity in phenotype, severity, and patient population. The cleft lip and palate surgical community needs higher-quality data to inform standards of care that can be applied in any setting.

Meaning: Optimization of cleft surgical outcomes is only achievable through a collaborative global consortium of surgeons setting forward evidence-based algorithms that are based in robust data.

Impact of Cleft Palate Repair Timing on Midface Hypoplasia

Variation exists between cleft centers, as each team attempts to balance palatal length and speech while minimizing scarring and periosteal dissection to limit MFH.[11] Some centers believe in prioritizing speech and aim to repair the palate before 18 months,[21,22] whereas others believe speech outcomes are not worse after 18 months and prioritize limiting MFH and the need for orthognathic surgery.[25,26] This variability in timing compounds in cases of severe cleft, with greater tissue deficiencies requiring greater dissection to achieve anatomical closure and palatal lengthening at the risk of greater MFH.

There are minimal data directly comparing early versus delayed palatoplasty and their effects on surgical outcomes. One systematic review aimed to investigate ideal palatal repair timing with respect to limiting MFH. SNA and A-point-nasion-B point (ANB) angles were analyzed across 34 articles (1271 patients total).[11] Cleft groups included patients with UCLP and BCLP, and controls, which were patients without any craniofacial pathology that had been matched.[11] As expected, SNA for all patients with a repaired palate, regardless of timing, was significantly lower compared with controls.[11] Hard palate repair at 13–18 months had a greater SNA angle than all other repair timings.[11] These results suggest that palatal repair timing contributes to the development of MFH and that repair at 13–18 months may mitigate some risk. Unfortunately, MFH was studied in isolation, and we are unable to evaluate the impact of this timing on speech.[11] This is true for most studies, where either MFH or VPI are studied in isolation; however, these outcomes are often at odds, and balancing them is critical to optimal outcomes. Again, the review lacked power due to low sample size (1271 patients) and was limited in its ability to evaluate severity and impact of technique in addition to timing of repair.[11]

Sagittal Growth Restriction Following Isolated Lip Repair

MFH is often evaluated in the context of palate repair; however, the impact of lip repair on MFH is poorly

documented.[12,27,28] A systematic review identified that isolated lip repair in patients with cleft palate was associated with MFH.[12] The presence of the cleft palate, even if unrepaired, contributed to greater MFH in the setting of lip repair than if there was no cleft of the palate.[12] Interestingly, in unoperated cleft lips, there was a protrusive maxilla, suggesting unrestricted anteroposterior growth.[12] Although the studies included were, again, limited by sample size and quality, this review begets further research questions: Is the presence of a cleft palate inherently associated with maxillary growth restriction even if left unrepaired? Does lip repair act as a physiological sling similar to nasoalveolar molding or lip taping that could restrict growth of the premaxilla and reduce preoperative severity? This logic is the foundation of the neonatal lip repair practiced by some institutions, but long-term effects of these interventions on MFH is not well documented.[29]

THE IMPACT OF SEVERITY ON OUTCOMES

Although technique and timing are important variables in evaluating surgical outcomes, cleft severity is crucial to account for, yet often neglected in studies reporting on outcomes. Wide clefts and more severe phenotypes are associated with worse outcomes more frequently requiring secondary operations to improve outcomes or address complications. Impact of severity is demonstrated by the Smile Index, an assessment scale which classifies clefts as "severe" or "nonsevere" based on the cleft width ratio.[30–32] The index shows that patients with severe clefts have a higher likelihood of unacceptable surgical outcomes and lip revision.[30–32] A similar scale, the palatal index, also demonstrates that wider clefts are associated with fistula development and higher rates of orthognathic surgery, as demonstrated in a systematic review.[13,33] In patients with more severe tissues deficits, as in BCLP and UCLP, orthognathic surgery rates were 38.1% and 30.2%, respectively, versus 4.4% for ICP and 1.8% for isolated cleft lip.[13] However, due to low sample size, definitive conclusions regarding mitigation of risk were unable to be determined.[13]

WHY THE NEED FOR HIGHER QUALITY DATA?

These systematic reviews demonstrated that the ability to reach consensus on optimal technique and timing protocols for CLP is limited by (1) small sample sizes, (2) minimal diversity in study populations, (3) lack of standardized measurements and evaluation tools, and (4) lack of accounting for severity or confounders like surgeon skill. A summary of the included reviews can be found in Table 1.

Although validated evaluation tools exist in cleft care for surgical outcomes, there is minimal standardization, making comparison of outcomes challenging.[34–39] Current tools also focus on evaluation of singular outcomes such as lip appearance, VPI, fistulas or MFH. However, the impact of timing and surgical technique on outcomes is intricately related. We cannot talk about lengthening the palate to facilitate speech without wondering about the impact of the extensive dissection on MFH. Updated tools should allow for complete evaluation of patient outcomes, to determine the full impact of surgical protocols.

Additionally, future research should aim to include global patient populations, particularly low- and middle-income country (LMIC) populations where the burden of disease is high. Long-term follow-up and resource availability can be challenging for any center but is more difficult in LMICs where barriers to care are higher and need to be accounted for.[40–42] We need standardized evaluation tools and higher quality data with larger sample sizes, greater diversity in population, and representation of varying cleft severity and phenotype. To achieve global consensus, we need improved and higher quality data to inform algorithms for cleft care. Our recommendations for improving data collection can be found in Table 2.

Table 1. Summary of Included Reviews

Study Title	Study Author and Year	Rationale for Inclusion
The Burden of Care for Children with Unilateral Cleft Lip: A Systematic Review of Revision Surgery	Sitzman et al. (2016)[18]	Reports secondary surgery rate in patients with a unilateral cleft lip
Systematic Review of Postoperative Velopharyngeal Insufficiency: Incidence and Association with Palatoplasty Timing and Technique	Xepoleas et al. (2023)[9]	Reports rate of postoperative VPI while trying to stratify for technique and timing
Midface Growth Potential in Unoperated Clefts: A Systematic Review and Meta-Analysis	Wlodarczyk et al. (2022)[10]	Reviews mean SNA and ANB between different phenotypes of unoperated clefts, while comparing rate to control patients without craniofacial pathology
A Meta-Analysis of Palatal Repair Timing.	Wlodarczyk et al. (2021)[11]	Reviews mean SNA and ANB between different phenotypes of clefts while stratifying for the timing of palatal repair. The study aimed to highlight the ideal repair timing to limit MFH
Sagittal Growth Restriction of the Midface Following Isolated Cleft Lip Repair: A Systematic Review and Meta-Analysis	Celie et al. (2022)[12]	Investigated the impact of lip repair on SNA angles across different phenotypes to determine the impact of lip repair on MFH
The Smile Index Part 2.	Yao et al. (2019)[31]	A simple set of measurements demonstrating worse clinical outcomes in severe clefts following unilateral lip repair
The Likelihood of Orthognathic Surgery After Orofacial Cleft Repair	Choi et al. (2021)[13]	A review that demonstrates different rate of orthognathic surgery between cleft phenotypes, highlighting the need to consider phenotype in treatment protocols

Table 2. Summary of Recommendations to Improve Data Collection

Secondary Deformity	Unsolved Issues	Recommendations
Secondary deformities of the lip	1. Safety of repairing at earlier ages 2. Timing and technique for various phenotypes	Future outcomes studies should focus on cleft lip repair timing with: 1. Inclusion of all cleft phenotypes 2. Stratification of cleft severity 3. Detailed reporting of techniques and technique modifications 4. Objective criteria/tools dictating when revision surgery is required, less reliance on subjective surgeon opinions
Palatal fistula	1. Timing of primary repair 2. Impact of technique on occurrence of fistula (optimal technique to minimize fistula)	Future outcomes studies should focus on cleft palate repair timing with: 1. Multicenter studies evaluating location, size, and functional impact of palatal fistula, 2. Inclusion of all cleft phenotypes 3. Stratification of cleft severity 4. Detailed reporting of techniques and technique modifications 5. Objective evaluation tools dictating when revision surgery is required, less reliance on subjective surgeon opinions
Velopharyngeal insufficiency	1. Optimal timing and technique of repair to optimize length while limiting MFH	Future outcomes studies should focus on cleft palate repair timing with: 1. Stratification of cleft severity 2. Multicenter studies using a single,standardized speech evaluation protocol 3. Inclusion of all cleft phenotypes 4. Larger and more diverse sample sizes 5. Ensuring that an adequate technique description is provided 6. Studies correlating both speech and midface outcomes within the same cohort
Midface hypoplasia	1. Natural history of midface hypoplasia in cleft disease 2. Optimal timing and technique of repair to optimize speech while limiting MFH	Future outcomes studies should focus on cleft lip and palate repair timing with: 1) Examining populations of older patients with unrepaired clefts 2) Stratification of cleft severity 3) Multicenter studies using objective evaluation tools dictating when revision surgery is required, less reliance on subjective surgeon opinions 4) Inclusion of all cleft phenotypes 5) Larger and more diverse sample sizes

CONCLUSIONS

Despite centuries of innovation in CLP repair with the aim of optimizing outcomes, there is still variable consensus on primary CLP protocols that can optimize surgical outcomes. This variability is fueled by a lack of high-quality data. The current state of the literature is fraught with limitations. With a high burden of unoperated clefts and an increased need for training in cleft surgery in LMICs, we need data-driven algorithms that can be modified for various severities, phenotypes, and resource settings. This article provides recommendations for future research.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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