

Reconstructive

SPECIAL TOPIC

Cleft Palate Repair: A History of Techniques and Variations

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Summary: Orofacial clefting is a common reconstructive surgical condition that often involves the palate. Cleft palate repair has evolved over three centuries from merely achieving anatomical closure to prioritizing speech development and avoiding midface hypoplasia. Despite centuries of advancements, there is still substantial controversy and variable consensus on technique, timing, and sequence of cleft palate repair procedures. Furthermore, evaluating the success of various techniques is hindered by a lack of universal outcome metrics and difficulty maintaining long-term follow-up. This article presents the current controversies of cleft palate repair and details how the history of cleft palate repair has influenced current techniques commonly used worldwide. Our review highlights the need for a global consortium on cleft care to gather expert opinions on current practices and outcomes and to standardize technique classifications. An understanding of global protocols is crucial in an attempt to standardize technique and timing to achieve anatomical closure with optimal velopharyngeal competence, while also minimizing the occurrence of maxillary hypoplasia and palatal fistulae. (Plast Reconstr Surg Glob Open 2022;10:e4019; doi: 10.1097/GOX.000000000004019; Published online 28 March 2022.)

INTRODUCTION

Orofacial clefting is one of the most common congenital anomalies worldwide and accounts for a considerable portion of the global burden of reconstructive surgical disease.^{1,2} Involvement of the palate is common and can have a deleterious effect on health and childhood development, including malnutrition, speech abnormalities, psychological consequences, and social isolation.^{1,3,4} Surgical correction is the standard treatment when attempting to restore normal palatal form and function. However, cleft palate repair is technically challenging. Complications following repair are significant when they occur and include speech disturbance and maxillary growth deficiency.⁴ Strengths and weaknesses of various palatoplasty techniques as they relate to outcomes are continually debated. This article presents the current controversies of cleft palate repair and provides an understanding of how commonly used

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Copyright © 2022 The Authors. Published by Wolters Kluwer Health, Inc. on behalf of The American Society of Plastic Surgeons. This is an open-access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal. DOI: 10.1097/GOX.00000000004019 techniques have been influenced by the history behind cleft palate repair.

GOALS OF CLEFT PALATE REPAIR

Over the years, goals of cleft palate repair have remained constant and focus on three areas: anatomical closure of the palatal defect, producing normal speech, and minimizing growth disturbance.^{5–7}

Separation of the oral and nasal cavities and reconstruction of the velopharyngeal valve assist with mastication, feeding, and preventing malnutrition.⁸ Adequate speech development and early restoration of articulation are crucial in childhood development and social integration. However, prioritizing speech through early cleft palate repair can lead to maxillary growth restriction, often warranting surgical correction.^{6,9} Conversely, prioritizing midface growth by delaying hard palate repair could potentially result in speech errors that may or may not be corrected by further surgery or speech therapy. The relative importance of growth and speech are constantly debated among proponents of the various approaches.

Experts debate the advantages of different techniques but generally agree that the following principles are dictated by the goals of repair^{5,8,10,11}:

- 1. Anatomical closure of the defect
- 2. Tension-free suturing

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- 3. Reorientation of the abnormally positioned soft palate musculature to reconstruct of the levator veli palatini
- 4. Lengthening and retro-positioning of the soft palate
- 5. Minimizing denuded areas of bone and nasal or oral mucosa
- 6. Layered closure of the hard and soft palate

Surgeons differ in their approach regarding the age of closure, the sequence and timing of hard versus soft palate repair, the number of anatomical layers required to close the defect, and the amount of acceptable retro-positioning of tissues. There is debate on the consequences of denuded areas post repair and the most appropriate dissection technique to reorient the palatal muscles. Cleft surgeons postulate the relative contributions of these surgical decisions to fistulae, speech, and maxillary growth. Understanding modern-day techniques begins with examining the history of cleft palate repairs.

HISTORY OF PALATE REPAIRS

Cleft palate surgery has been defined over the last three centuries by numerous surgeons (Fig. 1). Before the 18th century, the mainstay of treatment was an obturator.¹² Soft and hard palate repairs were considered separate entities initially.¹⁰ Soft palate repair techniques were described as early as the 1700s and centered on normal speech production and velopharyngeal competence.⁵ Hard palate repair came over 70 years later and prioritized tension-free anatomical closure and normal facial growth and dentition.^{10,13,14}

Soft Palate Repair Techniques

Velopharyngeal function and speech production have been the focus of soft palate cleft repair, and aim to

Takeaways

Question: Cleft palate repair techniques have developed over three centuries to ensure anatomical closure, while attempting to normalize speech and limit midface hypoplasia. These variations are often regional.

Findings: Despite advancements, there is variable consensus on technique, timing, and sequence of cleft palate repair procedures.

Meaning: We lack high-quality long-term data to evaluate outcomes of technique variations.

lengthen the palate while closing the defect and reconstructing the levator sling.⁸ The first description of soft palate closure in 1764 was provided by a French dentist, Le Monnier, who cauterized the cleft edges and sutured the defect closed.¹⁵ In the 1820s in Berlin, Von Graefe described the soft palate as the most important "voiceforming and voice-influencing structure" and urged the scientific community to consider the importance of soft palate repair to correct speech anomalies in cleft patients.¹⁶ Von Graefe de-epithelialized the cleft margins before bringing the edges together. Following his success in 1816, many other surgeons reported successful palatal repairs using a similar technique.^{16,17}

These early repairs employed very basic techniques of approximating cleft edges with heavy sutures and focused solely on anatomical closure, neglecting principles of palatal function. This resulted in a short, immobile palate that impaired speech production. Subsequent techniques prioritized principles that would lengthen the soft palate.

In the 1900s, palatal lengthening became even more important, especially following hard palate repair. Wardill and Kilner modified Veau's hard palate repair technique



Fig. 1. Timeline of cleft palate repair technique variations.

by retro-positioning the soft palate through an incision in the nasal mucosa and relaxing incisions anteriorly and laterally. The resultant two triangular flaps could be advanced posteriorly in a V-Y pushback to increase palatal length.^{18,19} However, the defect in the nasal mucosa reportedly caused shortening of the palate through scarring and contracture, ultimately causing speech defects.²⁰ To correct this, Veau proposed a two-layer closure of the nasal lining.²¹

Since then, many methods have been suggested to close the often-deficient nasal mucosa, including pharyngeal and vomer flaps.⁸ Vomer flaps are advantageous because they are simple, well-vascularized, and provide an effective nasal lining.²² Despite conflicting evidence as to whether these flaps result in maxillary growth disturbance,^{8,23,24} the Oslo Cleft Palate Team has used vomer flaps in a single-layer closure of cleft palates with great success and minimal facial growth disturbance.²⁵

In 1931, Veau described the abnormal arrangement of soft palate musculature in the cleft palate patient, which runs longitudinally and parallel to the cleft as opposed to transversely in the normal patient.^{10,26} In one of the most important contributions to improving speech, Veau advocated separating soft palate musculature from its insertion on the posterior hard palate to lengthen the soft palate

and reduce tension on the mucosal closure.⁵ However, the orientation of muscles was still oblique and abnormal.

In 1969, Kriens described a technique to reconstruct the velopharyngeal muscular sling by re-orienting the soft palate musculature from oblique to transverse without causing significant disruption to the muscles related to the Eustachian tube.²⁷ His technique, known as intravelar veloplasty, was a milestone for soft palate repair as it restored function of the muscular sling to reduce middle ear dysfunction and improve motion of the palate. The intravelar veloplasty allowed three-layer closure of the soft palate: nasal mucosa, soft palate musculature, and oral mucosa. Since then, Leonard Furlow and Brian Sommerlad have made important contributions to muscle repair.

Furlow's double-opposing Z-plasty, described in 1978, involves the creation and transposition of two mirrored z-flaps—an anterior mucosal flap and posterior myomucosal flap—to create an overlapping muscular sling without the need for relaxing incisions (Fig. 2).^{10,28} The Z-plasty allows for closure of the hard palate in one procedure while (1) lengthening the soft palate without the need for pushback, (2) re-aligning the musculature and reconstructing the velopharyngeal sling, (3) reducing palatal scarring and increasing palatal mobility, and (4) decreasing negative effects on maxillary growth.²⁸



Fig. 2. Furlow's double-opposing Z-plasty technique (drawn by the first author, modified from Hill MA et al).

Furlow's double-opposing Z-plasty is commonly used today and achieves good speech outcomes.^{6,28} However, this technique may struggle to close wider clefts and require greater dissection and multiple hard palate flaps to achieve anatomical closure.^{29,30} Sommerlad³¹ positioned velar muscles as anatomically as possible through radical retro-positioning of the muscle and tensor tenotomy. He limited dissection of the hard palate to prevent scarring and midface hypoplasia. In a 10-year follow-up study, 80% of Sommerlad repairs did not need lateral relaxing incisions or mucoperiosteal flap elevations. Using this technique, rates of secondary velopharyngeal surgery for speech decreased from 10.2% to 4.6% over a 15-year period.³¹ These speech outcomes are superior in comparison with Langenbeck's palatoplasty, where velopharyngeal insufficiency has been reported in up to 30% of cases,³² and comparable to Furlow's z-plasty that boasts up to 98% velopharyngeal competence.^{33,34} Although a 2014 systematic review reported no difference in fistula rates between techniques, fistula rates using Sommerlad's technique occur in up to 15% of cases, which may be a result of limited hard palate dissection.³⁵ Furlow's z-plasty has been shown to have relatively low fistula rates, up to 9.7%, whereas fistula rates using von Langenbeck's palatoplasty range vastly between 7% and 42%.³⁶⁻³⁸ In comparison with these other techniques, fistula rates using Sommerlad's repair have been considered acceptable in light of the benefit of decreased maxillary growth disturbance.^{35,38,39} Furthermore, both Sommerlad's and Furlow's techniques were found to be associated with the lowest rates of middle ear dysfunction and need for tympanostomy tubes.⁴⁰

Hard Palate Repair Techniques

Dieffenbach in Berlin pioneered hard palate mucosal elevation as a technique for closure of the hard palate in 1826.⁴¹ This technique was further advanced by the introduction of relaxing incisions and lateral osteotomies to ease hard palate closure. Closure of the cleft with the least amount of tension has always been an important principle. In 1889, Billroth suggested fracturing of the hamulus to aid in achieving this goal and increasing palate mobility.^{42,43} Although still in use, this technique, along with postoperative scarring and contracture, resulted in poor facial growth and increased complications of hearing and middle ear function.²³

von Langenbeck revolutionized hard palate repair by introducing the bipedicle mucoperiosteal flap. The technique involved incision along the oral side of the cleft edges and a lateral relaxing incision along the posterior alveolar ridge to create two mucoperiosteal flaps elevated from hard palate bones.⁴⁴ These flaps could then be mobilized medially to close the hard palate while ensuring improved vascular supply and tension-free closure (Fig. 3).^{9,45} Principles of closure were combined from Dieffenbach and von Langenbeck in a technique commonly known today as Langenbeck's palatoplasty.⁵ However, this technique has been prone to contracture, impeding speech production.⁴⁵ To counter these complications, Langenbeck's palatoplasty is often combined with intravelar veloplasty.⁴⁴ In addition to describing techniques of soft palate repair, Veau also modified von Langenbeck's bipedicle flap technique to a unipedicle mucoperiosteal flap based on the posterior greater palatine artery which connected the lateral relaxing incisions to the anterior cleft margins.⁵ This technique prioritized tension-free closure of the anterior cleft defect extending through the primary palate, but impaired maxillary growth through scarring of the denuded bone areas.^{11,46} In contrast, von Langebeck's bipedicle mucoperiosteal flap technique requires decreased dissection of the anterior palate and therefore less disturbance of dentition and facial bone growth.^{9,11}

In 1967, Bardach, a Polish surgeon, modified Langenbeck's two-flap technique in an attempt to decrease scarring and maxillary growth deficiency by minimizing hard palate bony exposure.¹¹ Mucoperiosteal flaps are based on the greater palatine artery posteriorly. Once the cleft is closed, the two flaps are sutured back to the alveolar margins to reduce the amount of bone exposure (Fig. 4).^{7,47} This technique successfully reduced palatal scarring and minimized maxillary hypoplasia but did not correct abnormalities of speech.

Technique Modifications

Principles of soft and hard palate repair interact intricately. Furthermore, as techniques have been passed down from mentor to trainee, they have been modified and combined over the years to achieve optimal results.

Soft palatoplasty variations can be considered in two broad categories: (1) Furlow and its many modifications, including the Children's Hospital of Philadelphia modification,⁴⁸ the Mann technique, and others; and (2) intravelar veloplasty techniques that range from simple to more aggressive attempts at gaining length. This includes radical intravelar veloplasty, and Cutting's modification thereof, which has evolved from a one-stage repair to a two-repair utilizing vomer flaps.⁴⁹ Radical intravelar veloplasty has been associated with improved speech outcomes, particularly when combined with the two-flap palatoplasty.⁵⁰ Sommerlad, in particular, deserves credit for trying to gain length while keeping the nasal layer intact.⁵¹ Modifications of hard palate techniques are also evident in recent years, including the hybrid palatoplasty and minimal incision technique, which have reported improved preservation of maxillary growth, with lower fistula rates compared with the use of relaxing incisions.⁵²⁻⁵⁵

The Veau-Wardill-Kilner technique was one of the first combined variations in the early 1900s, which combined Veau's unipedicle mucoperiosteal flap for hard palate closure with Wardill-Kilner's V-Y pushback approach to lengthen the soft palate (Fig. 5). Similarly, Bardach's two-flap hard palate repair was also combined with intravelar veloplasty.^{5,13}

In more recent years, Robert Mann has combined a modified Furlow technique with interposing buccal flaps for hard palate closure. This technique aimed to achieve tension-free closure and palatal lengthening whilst limiting fistula complication rates.^{56,57} Buccal flaps have been used with success in closure of wider clefts.⁵⁸



Fig. 3. Von Langenbeck's palatoplasty technique (drawn by the first author, modified from Sato FRL et al¹⁹).

Timing and Sequence of Operations

Timing of cleft palate repair is complex and debated, as it affects speech and midface growth.^{14,59} While early palatoplasty prioritizes speech production, delayed palatoplasty minimizes midface growth disturbance.⁶⁰

The one-stage palatoplasty (also known as the "holein-one" repair) has become an increasingly popular technique in recent years, particularly in resource-limited settings where repeated procedures are often not feasible.⁶¹ This technique advocates for both hard and soft palate closure around 10 months of age with minimal disturbances in facial bone growth.14,62 In comparison, other centers such as the Great Ormond Street Hospital (GOSH) in London and Oslo Cleft Palate Team advocate for a two-stage repair: early lip repair and singlelayer hard palate closure using a vomer flap at the age of 3 months and posterior palate closure at 18 months, using Langenbeck's technique.^{24,63,64} A systematic review of the literature showed that the need for orthognathic surgery following a two-stage palatoplasty is comparable to a one-stage palatoplasty, with orthognathic surgery rates of 21% and 20.8%, respectively. However, velopharyngeal insufficiency rates and the need for corrective speech surgery using the two-stage palatoplasty were significantly higher than the one-stage palatoplasty (23.9% compared with 15.1%).⁶⁵ Results within two-stage palatoplasty protocols also differ. The Milan Cleft Protocol also advocates for a two-stage repair; however, lip and soft palate repair are scheduled between the age of 4 and 6 months and hard palate repair between the age of 18 and 36 months. This protocol has resulted in increased maxillary hypoplasia when compared with the Oslo Protocol (difference in SNA greater than 2.6 degrees and ANB greater than 2.9 degrees).⁶⁶

Schweckendiek introduced two-stage palatoplasty between 1944 and 1951.^{9,67,68} His approach demonstrated good results by repairing the soft palate at 3–6 months and the hard palate at 11–12 years.¹³ Early palatoplasty is often performed between 6 and 9 months; however, in some centers, it is performed as early as 3–6 months of age to facilitate good speech outcomes.^{10,28,47} Late palatoplasty, between the age of 18 months to 15 years, prioritizes maxillary growth, and is center- and surgeon-dependent,⁶⁹ but may be associated with increased speech delays.⁷⁰ Other centers choose to repair the hard palate earlier, within the first year or 18 months of life; they report minimal maxillary growth disturbance and cite that facial growth benefits of repair after 5 years of age do not outweigh the



Fig. 4. Bardach's two-flap palatoplasty technique (drawn by the first author, modified from Leow AM et al⁹).



Fig. 5. Veau-Wardill-Kilner palatoplasty technique (drawn by the first author, modified from. Sato FRL et al¹⁹).

detrimental effects on speech development.^{62,71,72} While some studies have reported poor speech outcomes following Scheckendiek's technique, many European centers (such as Goteburg) and surgeons (such as Talmant) have championed delayed closure,^{73,74} achieving acceptable speech outcomes while minimizing maxillary hypoplasia.⁷⁵

DISCUSSION

As techniques evolved, principles became more refined with greater attention to restoring precise anatomy to achieve the goals of repair. Technique and timing of cleft palate repair are important concepts to ensure good outcomes, but there is no consensus on either.⁷⁶ Cleft palate repair variations differ between surgeons and cleft centers worldwide. Original techniques are often modified and combined to create variations, such as the combination of Von Langenbeck's procedure with other techniques to reconstruct the velopharyngeal muscle sling or lengthen the palate.^{9,77} As a result, comparison of techniques between centers is particularly difficult and often incomparable.⁹

Techniques also differ between regions. Bardach's twoflap palatoplasty and Furlow's double-opposing z-plasty are most commonly used in the United States.¹⁰ In Brazil and the United Kingdom, the most commonly performed techniques are Veau-Wardill-Kilner and Langenbeck's palatoplasty.⁷⁸ Renowned European centers such as GOSH and Oslo utilize the single-layer closure vomer flap technique.²⁴ Furlow's double-opposing z-plasty is a common procedure worldwide due to good speech outcomes⁷; however, studies have reported obstructive sleep apnea and difficulty utilizing the technique in wider clefts.^{79,80}

Most institutions agree that early palatoplasty should occur before the age of 18 months,^{24,59,81-83} but best timing is still debated and lacks high-quality evidence.¹⁰ Variations in timing and sequence are also regional, with North America tending toward early palatoplasty and Europe tending toward delayed palatoplasty or the Oslo Protocol. Different institutions have different protocols for timing and sequence of operations, often centered around improving speech outcomes while limiting maxillary hypoplasia. However, the lack of standardized speech measurements and variable techniques have hampered the ability to objectively determine optimal timing.^{61,69} Some suggest that timing should be based on the patient's condition and associated syndromes, the type of cleft, and the capabilities of the cleft team.⁴ Surgery is often delayed or simpler techniques are used when associated syndromes will not allow prolonged anesthesia.⁵ The one-stage palatoplasty is favored in both low- and high-resource settings.^{61,63,69} However, recent studies have shown that the two-stage palatoplasty confers improved maxillary growth.73,74

There is no single technique or recommendation for timing that prioritizes speech development, while limiting midface hypoplasia and preventing palatal fistulae. This has resulted in controversy compounded by a lack of standardized techniques and outcome metrics, and difficulties in long-term follow-up and data collection. Many studies are underpowered and lack generalizability. Outcomes of cleft palate repair are not easily measured and require sufficient numbers, long-term follow-up, and vigilant collection of data to evaluate outcomes of a technique. Evaluation of speech is often subjective and lacks standardized reporting mechanisms, while effects on facial growth require 15–20 years of follow-up, which is often not achieved.⁵

We lack universal metrics to assess appropriate speech development, define thresholds of the acceptable limits of midface hypoplasia, and evaluate the impact of technique on different cleft phenotypes. Furthermore, cleft severity, extent of hypoplasia, and cleft width are highly variable. Techniques are often not analyzed in the context of cleft palate severity or width. Limiting hard palate dissection mitigates midface hypoplasia; however, this is often not possible in wider clefts.^{84,85} Many traditional techniques are inadequate to address wider clefts, resulting in higher rates of oronasal fistulae,³⁰ and necessitating more aggressive dissection that impairs maxillary growth.⁸⁵

CONCLUSIONS

Since the 18th century, there have been phenomenal improvements in cleft palate repair with a more refined understanding of the anatomy. Cleft palate repair has evolved from merely striving for anatomical closure to balancing speech improvement while minimizing midface hypoplasia and preventing oronasal fistulae. Despite this, we still lack a universally accepted technique and protocol for timing to optimally achieve the goals of cleft palate repair while limiting complications. The variation and abundance of techniques is a result of different goals of cleft palate repair being prioritized at different times and might suggest that no single technique is best for every case. Furthermore, these techniques have traditionally been passed on in a master-apprentice fashion, with very few surgeons being exposed to the long-term sequelae of more than one technique. As a result, there have been numerous modifications of techniques. A few regional groups (such as Eurocleft, Scandcleft, and Americleft) have acknowledged the need for greater collaboration in standardizing best practice; however, these groups are still limited to high-income regions, with surgeons in these regions sharing similar opinions. In addition to standardizing protocols, a reclassification and standardization of technique types and names would be beneficial in comparing technique outcomes and training in various techniques. Orofacial clefting is a global problem requiring worldwide collaboration to address these knowledge deficiencies. This highlights the need for a global consortium on cleft care to gather expert opinions on current techniques and outcomes measurements in an attempt to concede on a gold standard.

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