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"Flip-over flap" in two-stage cleft palate repair☆

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ABSTRACT

Purpose: This study served to evaluate a two-stage concept in cleft palate repair, including key use of a triangular hinge ("flip-over") flap, in order to prevent palatal fistulae. It uses data from a prospective registry established in 1991.
Materials and methods: The concept entails Furlow soft palate repair (at 1 year of age) and hard palate closure (at 4 years) by a three-pronged approach [paring of the edges with or without postero-lateral relaxing incisions, peninsula (Veau) flap(s)], plus a triangular hinge flap. The latter is elevated from the oral layer of the already-repaired soft palate, stays based anteriorly, and is flipped over to close the posterior nasal layer defect. The case series is compared with data from the literature.
Results: The palatal fistula rate for Veau II to IV types (two-stage surgeries) was 4.3%. The overall fistula rate in the cleft population (Veau I–IV) was 2.9%. Meta-analyses describe 4.9 and 8.6% on average. There was no difference between sample A in which the flip-over flaps were used only when modified Veau flaps were indicated (until 2006) and sample B in which it was used regardless of the technique of hard palate closure applied (2006–2018). The fistula rate decreased to zero after 2010, which may reflect also an influence of other factors such as the interpositioning of a collagen membrane and also of improved surgical judgment.
Conclusions: Using a flip-over flap in two-stage cleft palate repair may contribute to prevent fistula formation at the hard/soft palate junction.
Level of evidence: III.

1. Introduction

Since its inception in 1826 by Johann Friedrich Dieffenbach, surgical closure of both soft and hard palate has become fundamental in cleft palate care (Dieffenbach, 1826). The focus has shifted from prevention of fistulae over improved speech results to favoring maxillo-facial growth, and the achievement of normal dental occlusion, as well as smile aesthetics. As such, over time, attention has gravitated from hermetic closure to levator muscle reconstruction (e.g. intravelar veloplasty, Kriens, 1969), less aggressive and less hasty flap dissection (e.g., two-stage Zurich approach, Hotz and Gnoiski, 1976; Perko, 1979), and bony repair of alveolar defects (e.g., Loma Linda early secondary bone transplantation, Boyne and Sands, 1972). The debate over timing and technique continues, marked by extremes of position (speech vs. growth) that seemingly defy resolution. Consequently, the senior author adopted a two-stage approach (Schweckendiek, 1955) in 1991, incorporating the Furlow (1986) technique for soft palate repair in 1995. Hard palate repair has typically involved a paring of the edges (Brusati and Manucci, 1994), with or without postero-lateral relaxing incisions, occasionally raising a unilateral peninsula flap from the greater segment (Veau, 1931) in patients with unilateral cleft lip, alveolus, and palate (CLAP; Veau III clefts) or bilateral Veau flaps in those with bilateral CLAP (Veau IV clefts). In 1995, a triangular hinge flap originating from the oral layer of the already repaired soft palate and based at the anterior aspect of the already repaired soft palate was introduced to facilitate closure of the nasal layer at the hard/soft palate junction (Mommaerts et al., 2006). As of 2006, the author periodically added a collagen membrane (TissuDura; Baxter

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International, Deerfield, IL, USA) between nasal and oral layer flaps to help oppose the pressure fluctuations of Bernoulli effect.[1] This paper was aimed at presenting this hinge flap (so-called "flip-over flap") concept and reporting its use in a single surgeon's consecutive case series, focusing on avoiding palatal fistula formation.

2. Materials and methods

2.1. Timing and technique

A Furlow double-opposing Z-plasty (Furlow, 1986) is performed at the age of 1 year, sometimes earlier (not less than 9 months) if canonical babbling has started (/məˈmɑ:/for "mama,"/ká:kɑ:/for "papa"), as sometimes happens in girls. At the age of 4 years (not less than 3 years for non-syndromic isolated complete palatal clefts, Veau II), the hard palate is closed surgically. By that time, the palate has sizably grown, and the residual cleft in the hard palate often has narrowed by appositional bone growth (albeit unlikely in Veau II, for reasons not known to the authors). Palatal mucosal surface has also increased, allowing for broader flaps, supraperiosteal alveolar (gingival) dissection, and subperiosteal (fibromucosal) dissection just lateral to the cleft (underlying nasal cavity and covering the remainder of the hard palate up to the alveolus; Delaire et al., 1989). This is needed for careful transpositional flap elevation. However, this was not always a necessary maneuver. Paring of the edges, perhaps with postero-lateral release, most often sufficed in patients with Veau II and III clefts.

In any event, the residual hard palate defect is widest at its posterior aspect, where dissection of layers along the horizontal portion of palatine bone is hindered by fibrous fixation. Formation of a nasal layer, in part by cranially based vomer flaps but also using nasal floor mucosa, may be difficult in that posterior pocket, because the postero-inferior edge of the vomer and each side of the nasal crest of the palatine bones are hypoplastic. The difficulty encountered in approximating deficient nasal and fibrous oral layers creates a risk of a Pittsburgh type III fistula (Smith et al., 2007).

By the time the hard palate is to be repaired, the soft palate mucosa has become pliable. Edges of the hard palate defect are delineated (Fig. 1; video) by markings that run posteriorly into the mid–soft palate and taper, defining a triangular flap based posteriorly to the defect. This triangular flap is raised first, using a #15 blade on a #7 shaft and Metzenbaum scissors. Then the lateral edges of the cleft are incised, using a lamellar blade (Beaver-Visitec International Inc, Waltham, MA, USA) bevel-up at 60° on the lateral side; a #15 blade on a # 7 shaft at the vomer side; and a #12 blade on a Barron shaft at the anterior alveolus. The oral and nasal layers are elevated subperiosteally, using a Mitchell trimmer (osteocarver #4). In highly arched palates, the nasal layer flaps are rotated upward, combining medial and lateral plus the triangular (flip-over) flaps. The latter nicely fills the posterior nasal layer defect (Fig. 2a and b). Approximation of epithelial edges is achieved using resorbable interrupted sutures (polyglactin 5-0) anchored in postero-anterior direction. The oral layer flaps, based on palatal gingiva, are rotated downward to meet in the middle, at least in instances of high palatal vaults (Fig. 3a and b; Fig. 5a). If tension is evident at the junction of the soft palate, postero-lateral release incisions and submucosal dissection posterior to the greater palatal artery are required to shift that bridge flap medially (Figs. 4a and 5b). Given the inherent stretch of mucosal layers, approximating sutures remarkably produce a downsizing of postero-lateral defects (Fig. 4b). Rarely, a Veau transpositional (peninsula) oral mucosa flap dissected from the greater segment, then elevated supraperiosteally over the alveolus and subperiosteally over the palatal vault, is necessary in patients with Veau III clefts (Fig. 5c). Postero-lateral release and submucosal dissection behind the palatal pedicle are similarly performed. Elevation of a peninsula flap reveals the palatal artery. Bleeding of its branches is common laterally at the transition between supraperiosteal and subperiosteal planes. Careful bipolar cautery is performed for hemostasis, and the bony spikes of palatal grooves are removed via rongeur. This modified Veau flap may now be advanced as needed, being a palatal peninsula flap based on a pedicle and soft palate mucosa. Both structures give way when pulling the flap forward. This feature is convenient in Veau IV defects, where the palate posterior to the premaxilla is usually devoid of useful mucosa for coverage. Of course, such modified Veau flaps must be generated bilaterally to repair Veau IV clefts (Fig. 5d). The oral layer is subsequently closed using resorbable interrupted sutures using polyglactin 4-0 and 5-0, backed by 5-0 horizontal mattress sutures. Dead space is avoided by applying fibrin glue between layers and atop the oral suture line. Once applied between layers, they are digitally compressed. This technique was thought to be an alternative to those mattress sutures adjoining oral and nasal layers like Veau, Schuchardt and others did it. Use of a compression bandage was instituted only recently, and quickly abandoned because of being impractible (2018). After 2006, the flip-over flap was used in all three types of oral layer closure, not only in modified Veau flap techniques (group A).

Supplementary video related to this article can be found at https://doi.org/10.1016/j.jcms.2018.10.019.

The necessity to shift from paring of the edges to adding postero-lateral incisions or raising a flap in Veau III clefts may become indicated during the operation. There are no detriments in shifting to the next method. This concept therefore embodies "cut as you go" versatility.

Beginning in 2006, the author occasionally added a collagen membrane (TissuDura; Baxter) between layers, spanning the length of defects. Although this biomatrix is beneficial in resisting Bernouilli forces and offers further prevention of dehiscence, it is also foreign material that will delay healing and adjoining of layers. Unfortunately, the merits or drawbacks of doing so could not be fully assessed through practice-based analysis. The same is true of paring the edges versus raising a flap for cleft repair. The question remains: will the palatal vault be as high after paring as after flap surgery?

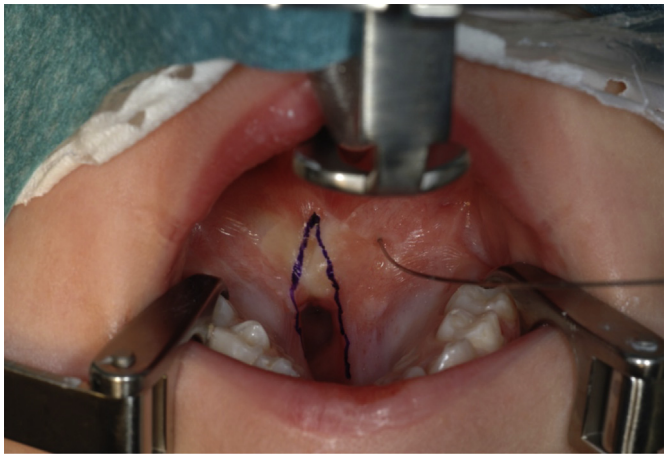

Fig. 1. Markings of anteriorly based, triangular flap behind residual hard palate cleft (arrows), and incisions required to separate oral and nasal layers lateral to cleft. Note: the flip-over flap is tailored to occupy the posterior half of a nasal layer defect. Infiltration anesthesia is achieved here using a long Whitacre spinal needle.

[1] Bernoulli effect is the occurrence of what is stated by Bernoulli's principle, that in a flowing fluid, an increase in velocity happens simultaneously with a decrease in pressure (en.wictionary.org, July 3, 2018).

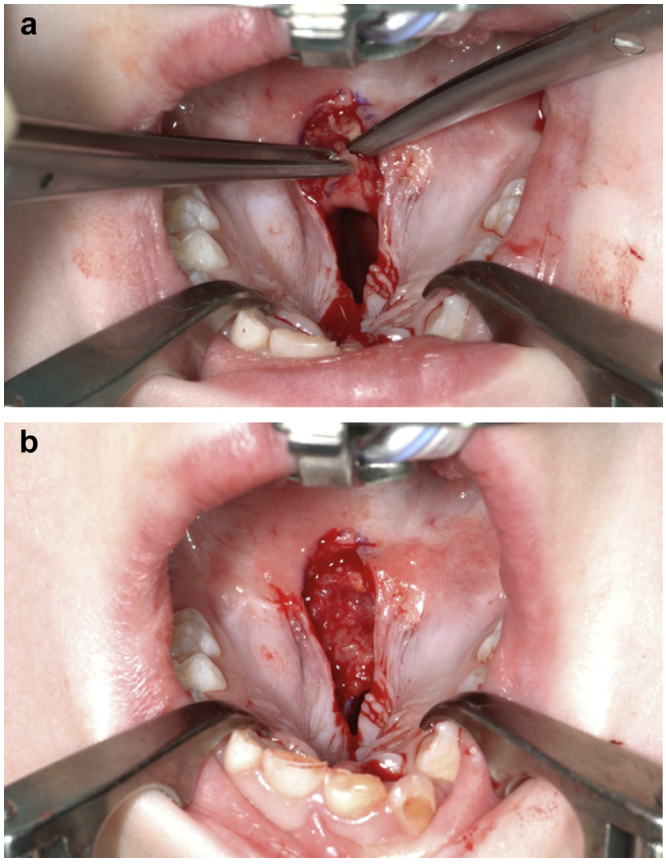

Fig. 2. (a) View of palate as flip-over flap is raised in Veau III cleft. (b) View of palate in Veau III cleft after suturing of flip-over flap over posterior half of nasal defect.

2.2. Patient population

Between 1991 and 2018, a total of 139 two-staged palate repairs (Veau II–IV) were undertaken by the author, a trainee or another staff surgeon under his guidance. The Widmaier–Perko palatoplasty was used until 1995, each hard palate closure protected by a compression gauze pack fixed to deciduous teeth. From 1995 to 2017, a total of 116 palate defects of Veau II (49), III (46) and IV (21) types were repaired using the technique described here (groups A and B). A compression pack was not used. Follow-up monitoring for palatal fistula formation in locations Pittsburg I–V in this group ranged from 6 months to 16.8 years after the second stage surgery (performed at the age of approximately 4 years). Until 2005, the flip-over flap was used only if oral flaps were required (group A). Later, it was also applied if paring of edges sufficed, with or without postero-lateral release incisions (group B). In this second sampling, there were 30 syndromic patients.

2.3. Methodology

From 1993 on, demographic, recall, referral and operative data were registered in the Microsoft Access database management system with a self-defined front-end. Complications needing surgical intervention were as such registered prospectively. In 2011, the senior author (MM) relocated his database to the Brussels Cleft & Craniofacial Centre. Many patients from the Bruges database continued to seek treatment in Brussels. Fistula not needing surgical intervention were retrieved from the electronical medical records. Missing data were retrieved by contacting the parents or the patients. The diagnosis of a naso-palatal fistula was made by clinical inspection only.

Fig. 3. (a) View on the palate of a Veau III cleft 3 years after soft palate repair. (b) View of palate in Veau III cleft after turn-down of oral layer flaps, without lateral incisions (ie, paring of edges).

3. Results

Only five out of the 115 patients with a cleft palate developed a fistula. Thus the overall rate of palatal fistula formation (types I through type IV fistulae according to the Pittsburgh Classification System; Smith et al., 2007) in cleft palate only, unilateral and bilateral CLAP (Veau II–IV palatal cleft type) cases (1995–2018) was 4.3%. Outcomes of flip-over flaps in the intermittent group A (modified Veau flaps, until 2006) and in group B (routine usage, (2006–2018) did not differ. Rates for Veau IV, Veau III and Veau II clefts were 4.7% (1/21), 0% (0/46) and 8.2% (4/49), respectively. However, the overall palatal fistula rate in Veau I to IV cases was 2.9% only. The palatal fistula rate was 10% in syndromic and 2.3% in non-syndromic cleft patients for Veau II to IV; 1.4% in male and 8.5% in female patients also for Veau II to Veau IV.

One patient had a fistula located in the hard palate (Pittsburgh type IV) and the other four patients at the junction between hard and soft palate (Pittsburgh type III fistula). One of these patients developed two fistulae (one Pittsburgh type III fistula, one Pittsburgh type V). We excluded three patients (3/116) with type V fistulae according to the Pittsburgh Classification from the

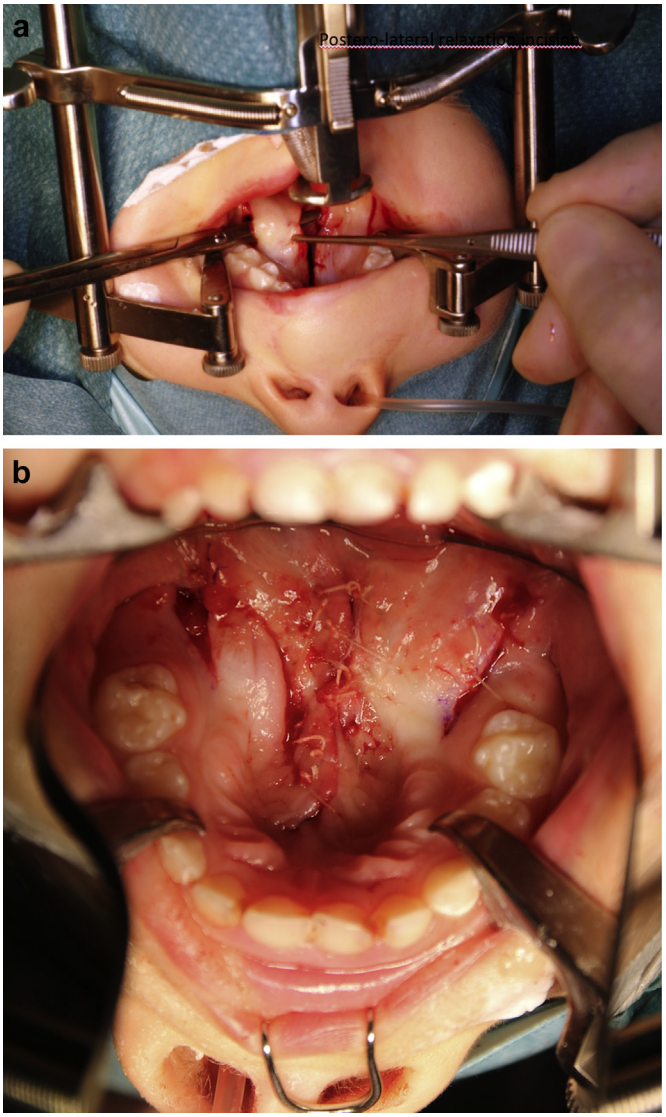

Fig. 4. (a) View of palate in Veau III cleft as submucosa is undermined by Metzenbaum scissors (posterior to greater palatal neurovascular bundle). Note: undermining is done here from the posterolateral incision but is equally permissible from the incised defect. (b) View of palate in Veau III cleft after final closure and application of fibrin glue, demonstrating postero-lateral release incisions of which the epithelial edges could partially be approximated after closure of the midline defect.

statistical analysis. Those fistulae were closed during the alveolar bone grafting procedure and did not require a specific secondary surgery.

One other patient with a palatal fistula was also excluded from the study the soft palate was repaired according to Furlow, but the hard palate was closed using the Perko palatal mucosal flap technique (Perko, 1974) with secondary elongation of the soft palate. That patient developed two type IV Pittsburgh fistulae.

Pittsburgh type VI and VII fistulae were not considered here, as they were deliberately left open to facilitate orthodontic/orthopedic alignment of the alveolar cleft margins up to the age of osteoplasty of the alveolar cleft.

The shapes (sagittal/transversal) and sizes of the six fistulae in these five patients were 1 mm/1 mm, 4 mm/0 mm, 3 mm/1 mm, 3 mm/1 mm, 5 mm/1 mm and 4 mm/2 mm. They were asymptomatic in two. Within the fistula group there was no type I or II.

It should be noted that there were no fistulae encountered after second-stage surgery performed after 2010.

4. Discussion

Because the flip-over flap is confined to a two-stage repair, the rationale for the latter must first be explained. The main goal of soft palate repair is to allow for normal velopharyngeal function and speech achieved by construction of a levator muscle sling before an infant develops compensatory glottal stops, glottal fricatives, and "backing." Children without craniofacial malformations or mental retardation begin level-I babbling (vocalizations containing a vowel, glottal stop, or glide) between ages 6 and 10 months (Morgan and Wren, 2018); and level-II babbling (canonical production of well-formed syllables containing at least one consonant and one vowel, including reduplicated babbling) commences between ages 7 and 12 months (Stark, 1980; Van Beinum & 't Hullenaar-Doppem, 2010; Morgan and Wren, 2018). Those with clefts start a few months later (Chapman et al., 2001). The ideal time for levator muscle sling construction depends on two factors: the onset of babbling on the one hand side (which must be noted and considered for a timely repair to prevent compensatory vocalization), and the ability to surgically identify and control orientation of the velopharyngeal musculature on the other. Allowing mucous membranes to strengthen, muscles to increase in mass, and maxilla to grow up to 9 months or 1 year of age eliminates reliance on magnification (provided the surgeon has normal sight). Careful dissection, when done at a younger age using an operating microscope and a single-stage approach, heightens the number of ensuing palatal fistulae (14.3% by Sommerlad's technique vs 4.9% general average - Bykowski et al., 2015). Robotic surgery has been attempted by some, although clearly for commercial reasons. This option has not been proved more effective in any surgical domain.

Likewise, impaired sagittal growth of the maxilla is related to lip rather than (hard) palate repair (Oxford Centre of Evidence Based Medicine Level of Evidence [OCEBM LOE] I; Bichara et al., 2015), and not only timing but also techniques of lip and palatal closure may impact physical development. There are intrinsic growth disturbances as well, accounting for the variability seen within cohorts under identical treatment protocols. For example, sagittal growth impairment (measured cephalometrically by SNA) is known to correlate with the absence of a lateral incisor—a sign of inherent tissue hypoplasia and possibly lack of migration by neural crest cells (OCEBM LOE III, Meazzini et al., 2011). On the other hand, Liao and Mars (2006, OCEBM LOE II) have concluded from a systematic review of retrospective studies that there is "inconclusive evidence about the effect of timing of hard palate repair on facial growth," adding that "in some circumstances, the surgeon's skill may have a greater influence than the timing."

Facial bone growth occurs circumferentially and along sutures. If all or part of the midpalatal suture is missing, as manifested at alveolar and palatal cleft sites, transverse maxillary development of anterior (more than posterior) regions is disturbed. McCance et al., (1990); (OCEBM LOE III) found the non-operated unilateral cleft lip and palate (UCLP) group to be narrower in the second molar region by a mean of 1.6 mm and by a 5.0 mm mean in the canine region when compared with a control group of normal Sri-Lankan adults. These findings were corroborated in a study of non-operated Indonesian adult patients with UCLP and BCLP (OCEBM LOE III, Diah et al., 2007). Early and aggressive surgery may also lead to collapse. Early (<18 months of age) one-stage palatal repair applying Wardill's or von Langenbeck's techniques often produces lateral crossbite during adult years in patients with CLAP (bilateral, 70%; unilateral, 20%; OCEBM LOE III, Farzaneh et al., 2008, 2009). In surgical closure of the hard palate, the void is partly replaced by fibrous tissue but may include small bony bridges. Delayed hard palate closures (at 8–10 years of age) have instead resulted in

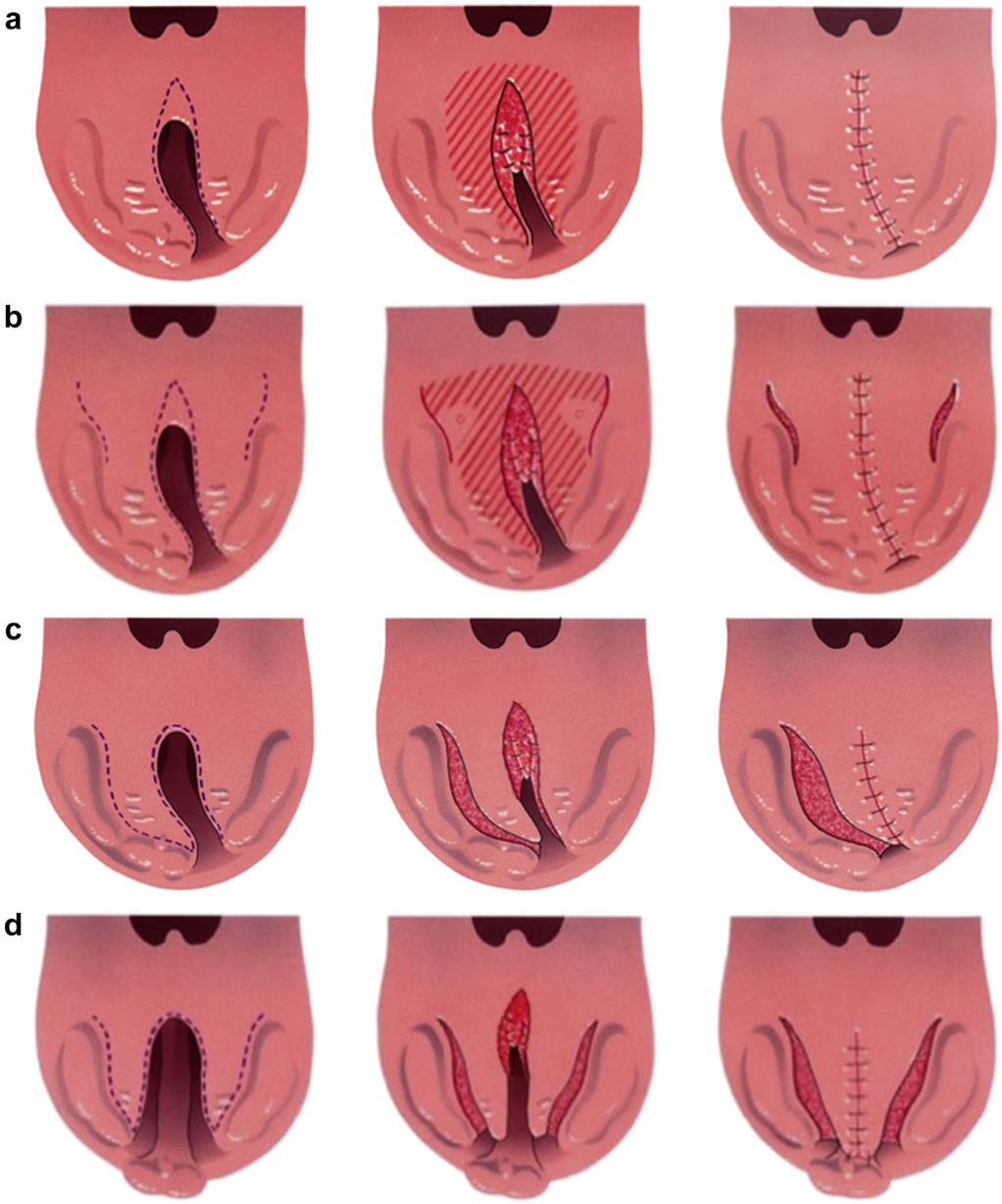

Fig. 5. Demonstration of three-pronged approach: (a) pairing of edges in Veau III cleft, without lateral release incisions; (b) pairing of edges and postero-lateral release incisions in Veau III cleft; and modified Veau flaps in Veau type III (c) and IV clefts (d).

significantly deeper palatal vaults and better speech when compared with early closures (at 16 months) using a Wardill–Killner push-back technique (OCEBM LOE III, Bakri et al., 2012). Hence, delayed hard palate closure and optimal speech therapy (if possible to be provided) represent by far the preferred treatment strategy (OCEBM LOE III, Gundlach et al., 2013).

The question then becomes: What is the optimal age to close the hard palate as a second-stage procedure? Certainly, it must be when most of transverse growth has occurred. Maxillae of children without clefts attain 80% growth by the age of 4 years, after which the momentum slows (OCEBM LOE III, Edwards et al., 2007). To date, however, there are no reported studies addressing growth in patients with isolated and unrepaired palatal clefts. Four years should be the optimal age if normal sutural growth is the basis. Further transverse growth disturbances demonstrated in the present patient sampling have been orthodontically (not surgically) corrected. The age of hard palate repair can be reduced to 3 years only if the hard palate has only been partially affected by the cleft; and it has not been proved that leaving the hard palate open until age 4 or even longer has a negative effect on speech development, although many authors are claiming this. However, one must provide speech therapy in those cases. Incidentally, Hardin-Jones et al. (2002) found no benefit of applicating an obturator.

The overall incidence of fistulae varies in the literature between 4.9% (OCEBM LOE III, meta-analysis by Bykowski et al., 2015) and 8.6% (OCEBM LOE III, systematic review by Hardwicke et al., 2014). Rates of fistulae after primary cleft palate surgery for Veau I, II, II and IV are: 2.6%, 8%, 6.9%, and 9.1% respectively (meta-analysis by Bykowski et al., 2015). The location of fistulae, based upon the Pittsburgh Fistula Classification System (Smith et al., 2007) was described as: uvula (type I), 0%; soft palate (type II), 15.7%; soft palate–hard palate junction (type III), 50%; and hard palate (type IV), 32.6% (meta-analysis by Bykowski et al., 2015). In our patients, fistula formation happened also preferably at the transition

between hard and soft palate (44%), but the overall number remained small.

There has been a lot of debate in the literature about two-stage versus one-stage palatoplasty. A systematic review published in 2017 by (Reddy et al., 2017) showed that there was no conclusive evidence for the effects of one-stage versus two-stage palate repair on the frequency of fistulae.

Using the described protocol, our overall palatal fistula rate of 2.9% is comparable with the rates published by other authors. The overall fistula rate presented in this paper for Veau III and IV defects is remarkably less, perhaps validating the premise of this study: the flip-over flap technique reduces the occurrence of palatal fistulae. It seems especially indicated in wider clefts, when Veau flaps are indicated.

Unfortunately, many other factors could not be weighed; the reduction in cleft width and increase in palatal tissue due to growth could have contributed to the outcome, as well as the use of fibrin glue and the increasing surgical expertise. The need for an interposed collagen membrane in preventing Pittsburg type IV fistulae should be investigated. Although this biomatrix is beneficial in resisting Bernouilli forces and offers further prevention of dehiscence, it is also foreign material that will delay healing and adjoining of layers. Unfortunately, the merits or drawbacks of doing so could not be fully assessed through practice-based analysis. The same is true of paring the edges versus raising a flap for cleft repair. It is currently not known if the palatal vault will be as high after paring as after flap surgery. Only analysis of a large patient series, not to mention randomized clinical trials might bring enlightenment. Considering the currently declining rates of congenital clefts, it is questionable whether such research is feasible at all or even necessary in the end.

5. Conclusion

Use of a flip-over (triangular hinge) flap in two-stage repairs of cleft palate seems to contribute to a reduction of palatal fistula formation.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jcms.2018.10.019.

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