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Progressive tightening of the levator veli palatini muscle improves velopharyngeal dysfunction in early outcomes of primary palatoplasty

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Abstract

Background—Velopharyngeal dysfunction is a major morbidity associated with a cleft palate diagnosis. Management of the levator veli palatini with intravelar veloplasty (IVV) has been shown to improve speech resonance. The senior author (ASW) has introduced a more aggressive procedure where the levator is separately dissected, overlapped and tightened. This study compares speech resonance results from 4 separate levator management protocols: Non-IVV, Kriens-IVV, Radical-IVV, and Overlapping-IVV.

Methods—Retrospective chart review was conducted on 252 patients who underwent primary palatoplasty with speech follow-up at 3 years of age. Postoperative velopharyngeal function was evaluated with perceptual speech examinations, and subjects were scored on a 4-point scale (0 = normal resonance; 1 = occasional hypernasality/nasal emission/turbulence/grimacing – no further assessment warranted; 2 = mild hypernasality/intermittent nasal turbulence/grimacing – velopharyngeal imaging suggested; 3 = severe hypernasality – surgical intervention recommended). Fisher's exact test was used to compare outcomes.

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Results—A single surgeon performed all the Non-IVV (n=92), Kriens-IVV (n=103) and Radical-IVV (n=31) procedures while the senior author performed the Overlapping-IVV technique (n=26). Cleft severity proportions were equivalent across the four methods ($p = 0.28$). Patients who underwent Overlapping-IVV demonstrated significantly better velopharyngeal function and none required further velopharyngeal imaging or secondary surgery when compared to the other three procedures ($p < 0.001$ for all comparisons).

Conclusions—Speech resonance outcomes at 3 years of age are improved and the need for secondary VPD management is reduced with more aggressive levator dissection and reconstruction during primary one-stage palatoplasty. Results were best when the muscle was overlapped.

Level of Evidence—III – Retrospective cohort/comparative study.

Introduction

The primary aim of cleft palate repair is to create an intact and functional palate for the purposes of normal speech resonance and articulation. Velopharyngeal competence, the ability to properly and completely close the velopharyngeal sphincter, is required for the normal production of all but the nasal consonants (in English: “m, n, ng”)¹. Velopharyngeal dysfunction (VPD) is defined as the inability to completely close the sphincter, which results in nasal air escape and/or hypernasality. Impaired speech resonance from VPD is one of the major morbidities associated with cleft palate, with 5-30% of patients suffering from abnormal resonance due to structural abnormalities²⁻⁵. While VPD can be diagnosed by both subjective and objective means, perceptual speech evaluation (PSE) by an experienced speech language pathologist remains the gold standard means of evaluation⁶.

Anatomically, the musculature of the soft palate is abnormal in a patient with cleft palate deformity. The levator veli palatini arises from its skull base origin in the petrous portion of the temporal bone, passes inferomedially, and inserts into the cleft margin along the anterior half of the velum¹. This is in contrast to a normal palate, where the levator reaches the midline in the middle 40% of the soft palate⁷. Contraction of the levator makes the velum elevate and extend posteriorly, creating a seal against the posterior pharynx. A patient with an unrepaired cleft palate does not have this normal function as the levator fibers lie in an aberrant orientation.

It has been proposed that reorientation of the levator veli palatini muscles with an intravelar veloplasty (IVV) would facilitate dynamic velar function thereby improving speech resonance by reducing nasal air escape and hypernasality. Two popular surgical techniques for repair of the levator musculature include: 1) submucosal dissection of the undifferentiated palatine musculature (Veau's cleft muscle) on the nasal side, reconstruction of the musculature in the midline and three-layer closure, as advocated by Kriens⁸, and 2) the Radical-IVV^{9, 10} with more aggressive dissection and reapproximation of the levator at the midline, as advocated by Cutting and Sommerlad. Gradually, cleft surgeons began to appreciate the importance of dissection and repositioning of the levator on improving speech outcomes¹¹⁻¹³. The senior author (ASW) has introduced a more aggressive procedure where the levator is separately dissected, significantly overlapped upon itself, and the

muscular sling is tightened, termed Overlapping-IVV¹⁴. The authors believe this creates a tighter sphincter than generated in IVV techniques where the muscles are simply reapproximated in the midline. We aim to compare speech outcomes with progressively increasing aggressiveness in the management of the levator. Based upon our institution's success with this procedure, we postulated that overlapping and tightening of the levator will result in less velopharyngeal dysfunction, when compared to Non-IVV, Kriens-IVV, and Radical-IVV techniques.

Methods

This study was approved by the Institutional Review Board at Washington University School of Medicine. A retrospective chart review was conducted on all patients who underwent primary palatoplasty (Non-IVV, Kriens-IVV, Radical-IVV, Overlapping-IVV) at our institution between 1982 and 2013. Due to a unique prior study at our institution comparing one-stage palatoplasty with either Non-IVV or Kriens-IVV²⁰, an historic comparative population whose palatoplasty was performed or supervised by one senior surgeon (JLM) was available for comparison. In addition, the same senior surgeon was involved with a subsequent set of Radical-IVV palatoplasties. Only patients with postoperative perceptual speech evaluation (PSE) performed at 3 years of age were included. Longer term follow-up data for the Overlapping-IVV technique was not available at the time of this manuscript. Compensatory misarticulations such as glottal stops, pharyngeal stops, pharyngeal fricatives, nasal stops, nasal fricatives and ingressive articulations were not used in the assessment of velopharyngeal function^{15, 16}. Patients were excluded for syndromic diagnosis, submucous cleft palate, and for a history of any previous procedure for correction of VPD. Veau classification was used to categorize cleft severity (Veau 1 - cleft of the soft palate only; Veau 2 - cleft of soft and hard palate; Veau 3 - unilateral cleft of lip and palate; and Veau 4 - bilateral cleft lip and palate)¹⁷. Since 1982, all perceptual speech evaluations were performed by certified speech-language pathologists (SLP). The SLP was blinded to the surgical procedure at the time of the three year old evaluation. Regardless of which SLP conducted the exam, all studies were reviewed in a team setting with a single lead therapist (LMG). Reports discussed key aspects of the exam (i.e. speech resonance, nasal emission, turbulence and grimacing) including the team recommendations (i.e. additional speech imaging, secondary surgery). For the current study, patients were then assigned a score on a 4-point scale based on retrospective assessment of the PSE reports by two speech pathologists (LMG, MBS): (0 = normal resonance, no nasal emission, turbulence or grimacing; 1 = occasional slight hypernasality, nasal emission, turbulence, grimacing – no further assessment warranted; 2 = mild hypernasality, intermittent nasal turbulence, grimacing – velopharyngeal imaging recommended; 3 = severe hypernasality associated with anatomic abnormality – surgical intervention recommended). Patients scoring either “0” or “1” were considered to have a desirable outcome and were functionally normal, whereas “2” or “3” were undesirable outcomes and were functionally abnormal.

ANOVA, performed with SPSS V.22 (Chicago, IL), was used to compare age among all groups. A *p*-value of <0.05 was considered significant. The mid-*p* correction value of Fisher's Exact test was used to compare the proportions of categorical variables. Cleft severity (Veau classification) proportions were compared across all four groups. VPD

outcome of Overlapping-IVV was compared to outcomes of each of the other surgical techniques individually. A total of seven Fisher's Exact comparisons were performed and, applying the Bonferroni Correction to an initial significance level of 0.05, p -values < 0.007 were considered significant. Fisher's Exact testing was performed using an open source script run on MATLAB R2011B (Natick, MA)¹⁸.

Overlapping Intravelar Veloplasty with Oral Z-Plasty

The novel technique developed by the senior author (ASW) has been utilized for both secondary¹⁴ and primary reconstruction of the palate. Overlapping-IVV has been used consistently and exclusively by the senior author since 2010 on all patients requiring palate repair. As a primary procedure, Veau's cleft muscle is identified and dissected out separately from the surrounding mucosa (Figure 1). The levator veli palatini musculature is released and isolated from surrounding muscular attachments, including the palatopharyngeus, palatoglossus and tensor veli palatini (Figure 2). Once separated, the muscle is tubular in structure, frequently taking an appearance similar to that of a lumbrical of the hand. Dissection continues laterally as the levator approaches its origin at the petrous portion of the temporal bone. Of note, the skull base is not directly visualized, but can be safely palpated with the tips of dissecting scissors. Prior to muscle repair, the nasal mucosa is then sutured closed in longitudinal fashion.

A Z-plasty of the oral mucosa is then incised and elevated (Figure 3). With maximal repositioning of the levator, a significant degree of laxity is identified (Figure 4). The levators are then overlapped upon each other – the free end of one levator is extended to the opposing levator body and secured with two interrupted 4-0 Vicryl sutures. Both levators are secured in the same fashion to create a taut reconstruction, but without excessive tension (Figure 5). Based on the surgeon's discretion, additional tightening is frequently performed. More recently, the senior author has begun separate dissection and direct reapproximation of the tensor component. As the tensor muscle moves medially around the hamulus, it becomes tendinous in nature.¹⁹ The soft tissues around this tensor aponeurosis are preserved to optimize suturing – presumably, this bundle includes the remnants of the palatopharyngeus and the aponeurosis itself (Figures 2-6). The oral Z-plasty is then transposed and closed, with the final appearance similar to that of the Furlow technique²⁰ (Figure 7). The main purpose of the Overlapping-IVV technique is to reconstruct the muscular sling involved in velopharyngeal closure.

Results

There were a total of 249 patients who fit our study criteria. A single surgeon (JLM) performed or intra-operatively assisted a plastic surgery resident/fellow surgeon for all of the Non-IVV ($n=92$), Kriens-IVV ($n=103$) and Radical-IVV ($n=31$) procedures, while the senior author (ASW) performed all of the Overlapping-IVV cases ($n=26$). The average age at surgery among the different groups were between 14 to 15 months ($p = 0.429$). The distribution of demographic parameters is shown in Table 1. The cleft types were relatively equivalent across the four procedures (Figure 8A, 8B $p = 0.28$). Patients with unilateral

complete cleft palate and lip (Veau 3) were the most common within each group (40%-55%).

As depicted graphically in Figures 9A & 9B, patients who did not undergo any reconstruction of the levator during primary palatoplasty (Non-IVV) had the worst resonance outcomes and the greatest proportion of individuals requiring either velopharyngeal functional speech imaging (45.7%) or secondary VPD surgery (4.3%) at 3 years of age. A lesser proportion of patients who underwent Kriens-IVV required additional imaging (36.9%) and only 1 patient required secondary surgery to correct VPD by the specified time point. None of the patients who underwent Radical-IVV required additional surgery at 3 years of age. When we compared Non-IVV, Kriens-IVV and Radical-IVV, there appears to be a trend towards improved resonance outcomes as a function of the aggressiveness in levator reconstruction; however, the proportions are not significantly different ($p > 0.243$ for all comparisons).

Postoperatively, patients who underwent Overlapping-IVV demonstrated significantly better velopharyngeal function on the four point scale in head-to-head comparisons compared to Non-IVV, Kriens-IVV, and Radical-IVV ($p < 0.001$ for all comparisons) (Figure 9B). When patient results are stratified into desirable versus undesirable outcomes, patients who underwent Overlapping-IVV again performed better than Non-IVV, Kriens-IVV, and Radical-IVV ($p < 0.001$ for all comparisons) (Figure 10). None of the Overlapping-IVV patients have required further velopharyngeal functional speech imaging or secondary surgery to date. As previously published, there was no significant difference between Non-IVV and Kriens-IVV groups ($p = 0.74$)²¹.

Discussion

Achieving a functional velopharyngeal mechanism, with adequate resonance and articulation development, are well established goals of primary palatoplasty²². In palatal clefts, the muscles of the soft palate are hypoplastic and insert in the cleft margin along the anterior half of the velum and posterior margin of the remaining hard palate, rather than the midline raphe²³⁻²⁵. Many techniques have been described to correct the deformity of the palatine musculature but controversy remains over the most effective technique for cleft palate repair. The ideal procedure should achieve a tension-free multilayer closure of the palatal defect, produce normal velar length, and successful reconstruction of the levator veli palatini resulting in a functional velopharyngeal valve. Pet et al recently reported that procedures that combine both muscle reconstruction and velar lengthening (i.e., Furlow palatoplasty) are most consistently associated with improved speech resonance outcomes²⁶.

The field of cleft palate surgery has been dynamic in the adoption of the intravelar veloplasty to repair the aberrant muscular anatomy. Nevertheless, early studies have suggested that speech resonance improvement was not significant enough to recommend routine performance of the Kriens IVV^{21, 27-29}. Undeterred, surgeons have continued to advocate IVV, increasing their aggressiveness in dissection and/or overlap of the levator muscles in a search for improved results. In line with this concept, we note a trend towards improved speech outcomes as a function of aggressiveness in management of the levator

(Figures 9 & 10). The proportion of patients with normal speech resonance at three years of age in this study gradually increases comparing Non-IVV (28.3%), Kriens-IVV (35.9%), and Radical-IVV (45.2%). The power to determine true differences between these three techniques would be improved with a larger sample size and longer follow-up. Nevertheless, we noted that speech resonance outcomes were best with maximal overlapping and tightening of the levator, when compared to less aggressive approaches. The fistula rates seen in our study cohort at 3 years of age [Overlapping-IVV: 1/26 (3.8%); Non-IVV: 6/92 (6.5%); Kriens-IVV 2/103 (1.9%); Radical-IVV 2/31 (6.5%)] are comparable or less than those reported in the literature.³⁰⁻³² Thus far, no other complications (e.g., dehiscence, infection, sleep apnea, feeding difficulties) have been identified in patients who underwent the Overlapping-IVV technique.

There is vague and inconsistent terminology regarding degree of muscle reconstruction accomplished during palatoplasty in the literature. "Intravelar veloplasty" has been used to describe any degree of muscle dissection³³. It has been clinically observed that poor speech resonance outcomes are associated with incomplete mobilization of the muscle. Andrades et al. proposed a novel classification of muscle reconstruction (Type 0 repair represents techniques where no muscle repair is performed; Type I repair represents an attempt at muscle repair without dissection of the muscle; Type II repair represents partial dissection of the muscle; Type III repair represents complete dissection of the muscle, and extends laterally to the hamulus and pterygoid). The authors also demonstrated in a cohort of 213 patients undergoing two-flap palatoplasty with and without IVV that more muscle reconstruction resulted in improved speech resonance outcomes¹³. We would equate our Non-IVV repair as a Type I, Kriens-IVV as a Type II, and Radical-IVV as a Type III. In an adaptation of their classification, we introduce Overlapping-IVV as a Type IV repair, where the muscles are separately dissected, maximally overlapped and tightened. One potential explanation of why Overlapping-IVV results in the best speech resonance outcomes compared to non-IVV, Kriens-IVV, and Radical-IVV is that the smaller/tighter the oral sphincter complex, the better the closure and the less velopharyngeal dysfunction occurs. Alternatively, it may be thought that overlapping and tightening of the levator in a patient with cleft deformity better addresses the muscle's abnormally increased length and laxity that was clinically observed by Furlow and Randall³⁴ and reported in imaging studies³⁵.

Several factors that determine velopharyngeal function include length and mobility of the velum, medial movement of the lateral pharyngeal walls, anterior movement of the posterior pharyngeal wall, the presence of Passavant's ridge, and the thickness of adenoid tissue^{36, 37}. Mechanistically, the use of the Overlapping-IVV in combination with oral Z-plasty to correct VPD potentially enhances the mobility and length of the soft palate. Occasionally, the oral Z-plasty provides necessary length to aid in retropositioning of muscles in a patient with a short soft palate. Computational 3-D models developed by Inouye et al have shown that retrodisplacement in combination with overlap over the levator were most effective in increasing velopharyngeal closure force³⁸. Although some have cautioned against the use of the Z-plasty in patients with wide clefts (i.e., 10 mm)³⁶, Sie and Gruss have suggested that gap size is not a significant determinant of outcome in patients who receive Z-plasty procedures³⁹. Based upon the results reported here, Overlapping-IVV with single oral Z-plasty appears to successfully treat a wide range of cleft types and severity. It is in our

opinion that one of the benefits of the Overlapping-IVV technique is that the muscle is not limited by its oral and nasal mucosal attachments; this allows for better control of the muscle, in regards to the amount of overlap and its final resting position. Of note, the double-opposing Z-plasty, which leaves the levator attached to the mucosa, also allows for a great degree of mobility and overlap. The extensive dissection of the levator used in our technique potentially increases the risk of scarring, though we have not yet observed any adverse function or complications.

As a retrospective analysis, this study has a number of limitations. Our database of primary palatoplasty does not include patients treated using a double-opposing Z-plasty, a technique commonly used by cleft surgeons. Interestingly, from our own institution, the speech velopharyngeal outcomes of double-opposing Z-plasties were compared retrospectively to Overlapping-IVV, with significant improvement in the second group when performed as a secondary procedure¹⁴. Based on positive clinical results, patients at our institution are continuing to receive the Overlapping-IVV for both primary and secondary management of the cleft palate. Continued implementation of this technique will be essential to the further understanding of the role of levator reconstruction in speech outcomes. Definitive conclusions cannot be made until a larger number of patients with longer followup are studied (e.g. 5-6 years of age). In addition to the Z-plasty, it has been the senior author's preference to reconstruct the tensor veli palatini (i.e., palatal aponeurosis). This description is discussed for the sake of reporting the steps of the operation in its entirety. We do not have data to advocate for its use; however, studies are currently underway to evaluate its effectiveness in regards to hearing and speech.

A notable weakness of our study design is that only one surgeon was involved in the performance of each of the techniques evaluated (ASW for Overlapping-IVV and JLM for the remaining procedures). This leaves open the possibility that the surgeon performing the procedure may be a confounder of the results. Future studies involving additional surgeons utilizing the Overlapping-IVV technique is needed. The authors advocate the Overlapping-IVV technique for its uniqueness in management of the levator and its promising effect on early speech outcomes. The senior author prefers to capitalize on the elasticity of the nasal mucosa, suturing it in a longitudinal fashion, while closing the less elastic oral mucosa with a Z-plasty in order to avoid overlapping scars.¹⁴ It has been suggested that the oral side of the velum shortens as it contracts with velar lift, while the nasal side, which must lengthen for the palatal genu to reach the posterior pharyngeal wall, might theoretically be a better site for a Z-plasty.⁴⁰ Additional studies on the effect of nasal mucosa closure technique on palatal function are needed as there is currently no evidence to either support or refute the use of a linear closure.

Although the perceptual speech examination is considered the gold standard in resonance evaluation, the information it provides is limited. Since the PSE is by nature a subjective examination, it does not provide an objective means of assessing speech production. We know that reconstructing the levator is better than not reconstructing it from anatomical studies and clinical series^{8, 41}. The addition of intravelar veloplasty to classic techniques such as von Langenbeck or Veau-Wardill-Kilner has been shown to significantly improve speech resonance and reduce the need for secondary surgery^{23, 42, 43}. Further investigation

will be needed to show the precise relationship (e.g., step-wise, logarithmic or linear improvement) between extent of muscle reconstruction and degree of improvement in velopharyngeal outcome.

The early age of evaluation remains another weakness of this study. While other papers have used similar criteria^{3, 13, 29}, age 3 is admittedly fairly young and remains the earliest time point at which the majority of children can reasonably participate in structured speech assessment and may also be able to participate in velopharyngeal imaging studies, if needed. The outcomes obtained from speech studies at this age need to be interpreted carefully and may be too early for definitive recommendations for most children. Sullivan et al noted that ideal time for this exam is around 4 years of age because they are unlikely to subsequently develop VPD and children younger than 4 years are often unable to cooperate for appropriate speech assessment⁴⁴. While our experienced speech pathologists have been able to obtain reliable information with our patients at 3 years of age, we plan to corroborate our findings when data at 6 years of age becomes available. Evaluation by speech language pathologist and interpretation of their evaluation is essential to the findings of this study. The conclusions may be confounded by the subjectivity of different therapists. Further studies with standardization of speech exams and inter-rater reliability will help to validate the findings. Despite the retrospective and historic nature of our Non-IVV, Kriens-IVV and Radical-IVV groups, our PSE data is strengthened by the longitudinal care and assessment provided by the same senior speech pathologist (LMG) for every patient in the study.

Conclusion

A novel technique for primary palatoplasty repair (Overlapping-IVV) is found to be a safe and effective procedure for the management of velopharyngeal dysfunction. In this early outcomes study, we noted that maximal overlapping and tightening of the levator has the best speech resonance among palatoplasty techniques evaluated. We also saw an apparent trend towards better outcomes with increasing aggressiveness in management of the levator. Ongoing research is needed to determine the long-term outcomes, durability, and sequelae of our proposed Overlapping-IVV technique.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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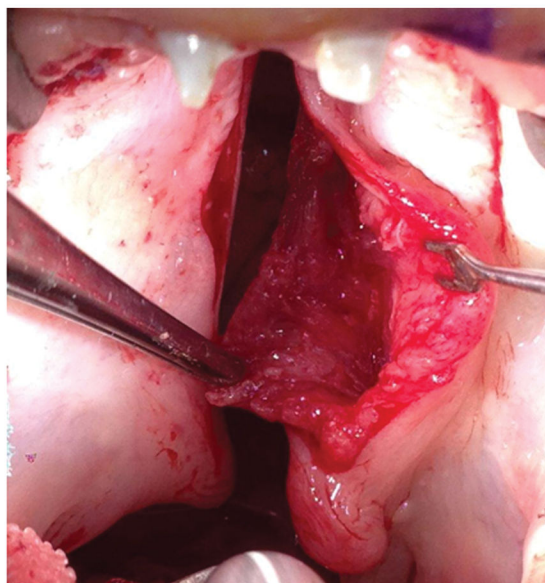
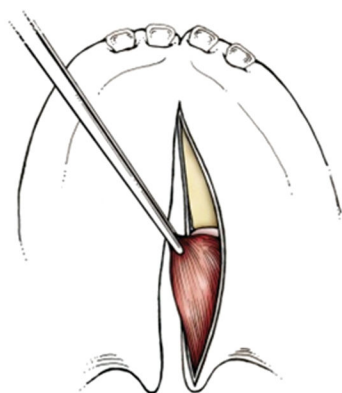


Figure 1. Veau's cleft muscle is identified and separated from surrounding mucosa. In vivo images on the right demonstrate the progressive operative steps and were all photographed during the same procedure.

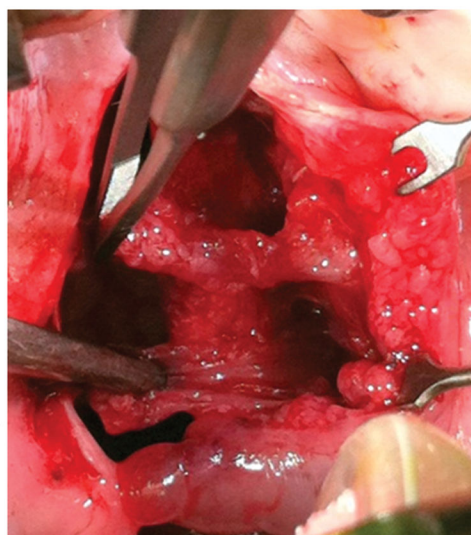
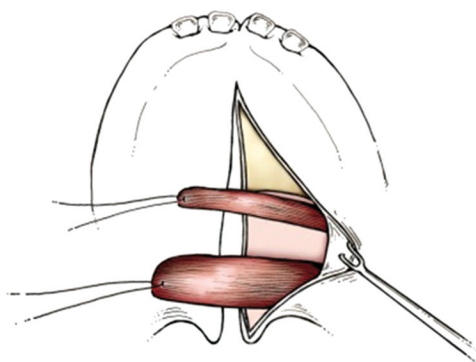


Figure 2. Separation of tensor component (anterior) and levator (posterior) muscle, with repositioning to a transverse orientation.

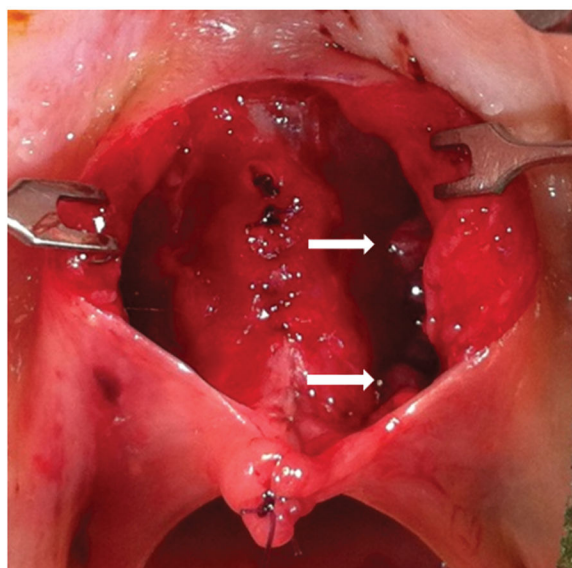
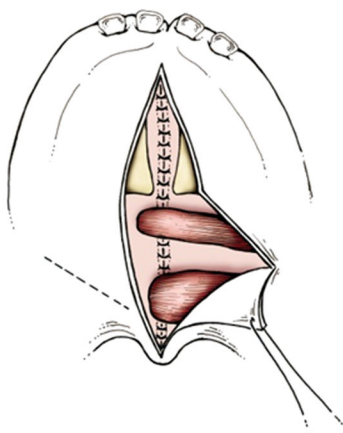


Figure 3. Following dissection of the musculature, the nasal mucosa is closed and oral Z-plasty incisions are made. Arrows indicate tensor aponeurosis and levator veli palatini musculature.

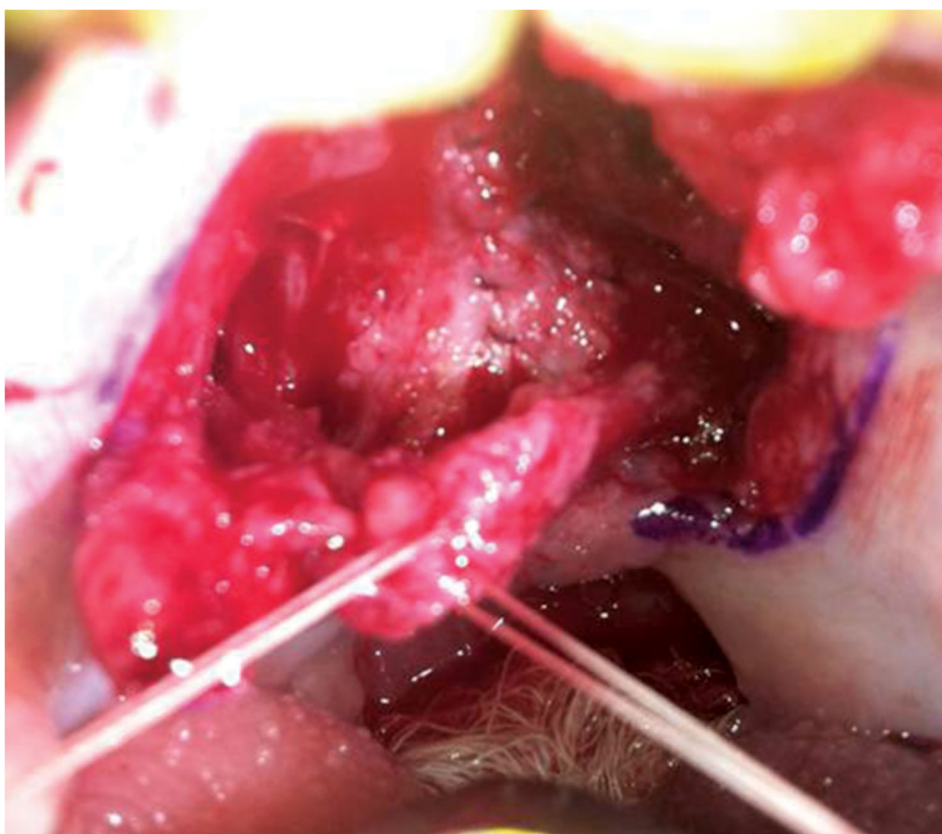


Figure 4.
Significant redundancy can be seen prior to overlapping of the musculature.

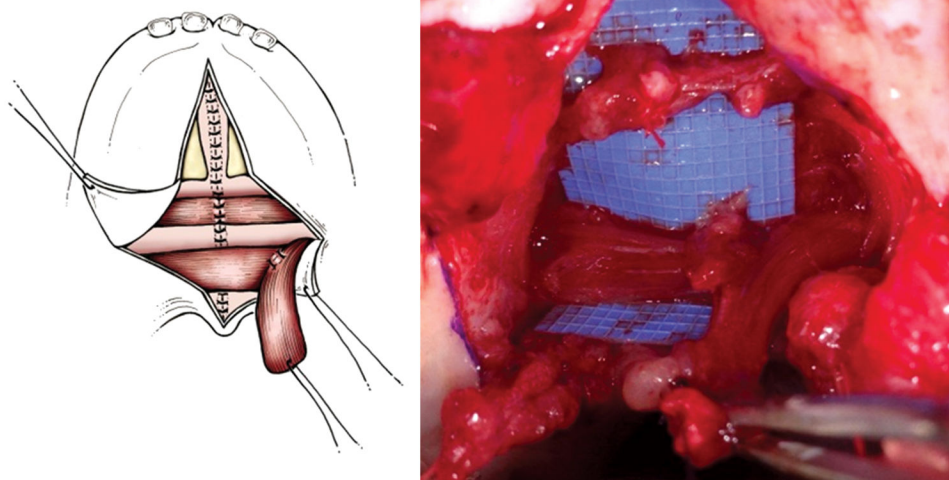


Figure 5.
The tip of right levator has been inset into the body of the opposite muscle. The tensor component has also been reapproximated end-to-end.

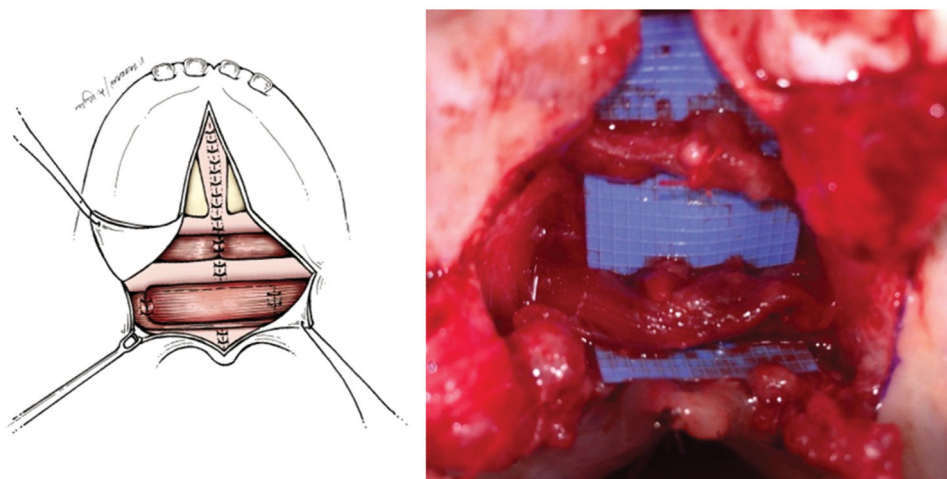


Figure 6.

Both levator muscles have been overlapped upon each other and sutured together using interrupted 4-0 Vicryl. The goal is to achieve a taut muscular sling, without excessive tension. Frequently, additional tightening is performed to achieve appropriate constriction of the levator sling. Again, the tensor component repair is seen anteriorly at the midline.

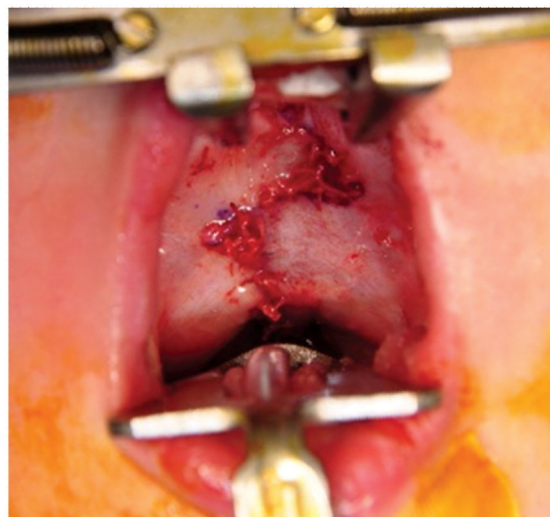
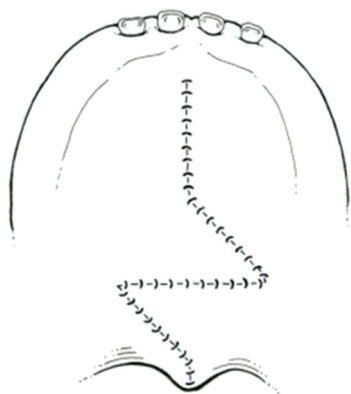


Figure 7.
The appearance of the palate upon completion of the procedure is similar to that of the Furlow palatoplasty repair.

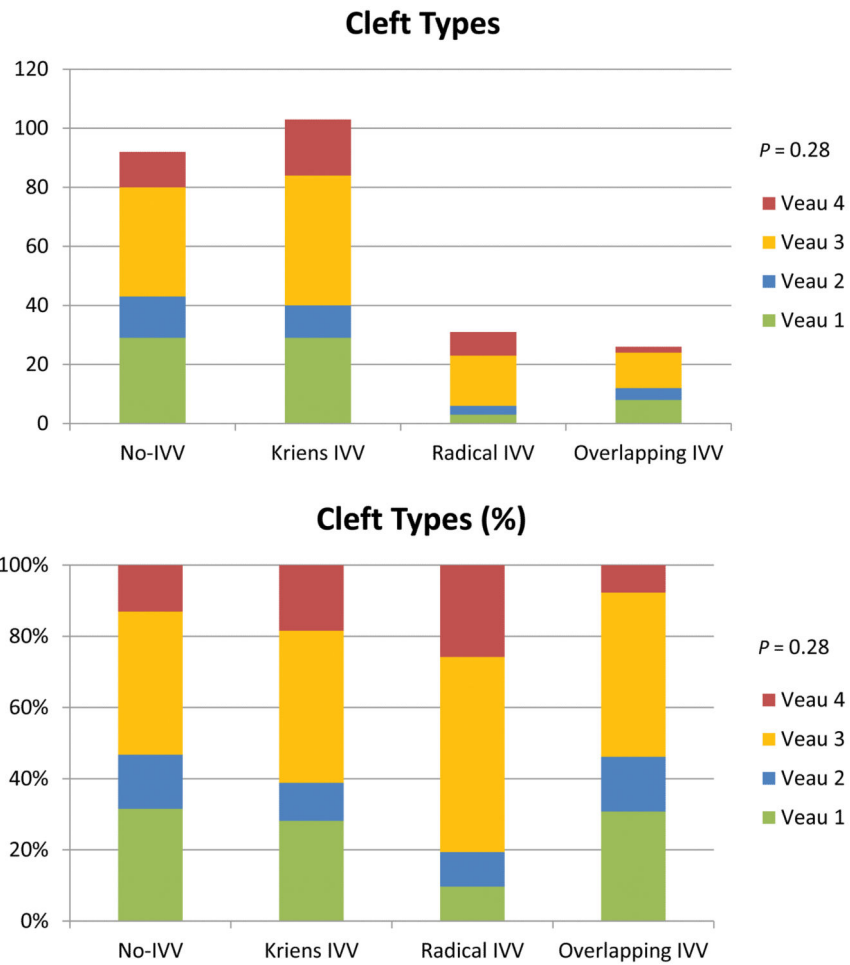


Figure 8.
A. Cleft severity based on Veau Classification among the four techniques.
B. Percentage of cleft types.

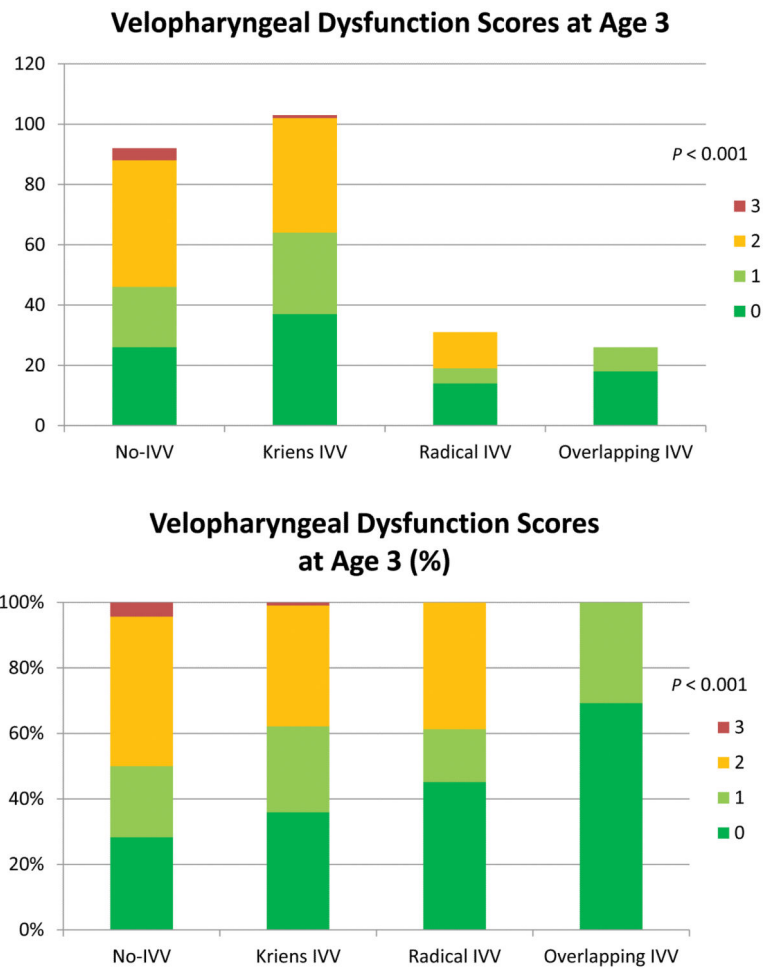


Figure 9.

A. Velopharyngeal dysfunction scores at 3 years of age.

B. Percentage of velopharyngeal dysfunction scores at 3 years of age.

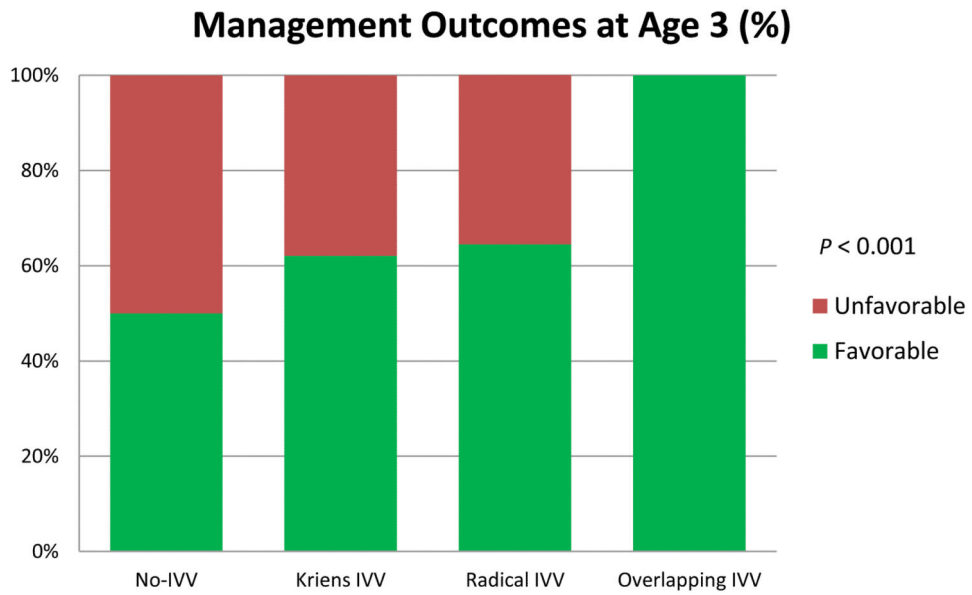


Figure 10. Unfavorable outcomes (e.g., additional velopharyngeal imaging, secondary surgery) are significantly less in patients who underwent Overlapping-IVV.