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Treatment of VPI with Customized Pharyngeal Flaps: One Size Does Not Fit All

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Background: Failure of complete closure of the velopharyngeal sphincter results in velopharyngeal insufficiency (VPI), which may severely interfere with speech. The pharyngeal flap remains a common procedure for correcting VPI. We aimed to study whether customization of pharyngeal flaps using a dynamic preprocedural assessment can result in successful outcomes in the surgical treatment of VPI, despite variations in surgical technique.
Methods: This is a retrospective review of patients between the ages 4 and 18 years old with VPI who underwent surgical correction by one of four surgeons at our institution. All four surgeons used a superiorly based pharyngeal flap (SBPF) with slight variations in operative technique. All patients also received an evaluation by the speech and language pathologist that included nasometry, multiplanar videofluoroscopy, and flexible videonasopharyngoscopy. Individualized preoperative planning was performed based on the findings.
Results: In total, 158 patients (92%) demonstrated overall successful correction of VPI, defined by a normal post-operative mean nasalance. Thirteen patients (8%) presented with resonance improvement but persistent abnormal mean nasalance. The most common causes of failed VPI correction were inferior migration and/or shrinking of the pharyngeal flap. There was a nonsignificant association between surgical technique and unsuccessful corrections.
Conclusions: The optimal surgical approach for performing pharyngeal flaps to correct VPI is individualized, customizing the procedure based on preoperative imaging. This study demonstrates that despite variations in surgical techniques for performing SBPF, high rates of success can be achieved when adequate surgical planning is based on imaging findings. *(Plast Reconstr Surg Glob Open 2022;10:e4255; doi: 10.1097/GOX.0000000000004255; Published online 14 April 2022.)*

INTRODUCTION

Normal speech is achieved through velopharyngeal competence—described as the complete closure of the velopharyngeal sphincter (VPS)—which acts to separate and seal the nasopharynx from the rest of the vocal tract. Functionally and anatomically, the VPS acts as a muscular valve consisting of the levator veli palatini, musculus uvulae, and superior pharyngeal constrictor muscles of the pharynx. This muscle group acts in coordinated and balanced constriction, allowing for the correct resonance and articulation of phonemes.[1–5] Failure of the VPS to completely close causes velopharyngeal insufficiency (VPI), which manifests as nasal resonance or hypernasality, nasal airway emission, and in some cases, compensatory articulation patterns, which severely interfere with intelligibility and quality of speech.

The etiology of VPI broadly encompasses structural, functional, and dynamic abnormalities. The most common abnormality is seen in patients with cleft palate. Unsuccessful palatal repair can result in inadequate lengthening of the velum, abnormal functioning of the velar musculature, or cicatricial contracture of the velum.[6] Following primary cleft palate repair, as many as 20%–30%

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of cleft palate patients require secondary surgical correction of VPI. After birth, these patients are referred to a cleft/craniofacial clinic and followed by a multidisciplinary service. This is in contrast to patients whose VPI is caused by noncleft etiologies, including submucous cleft palate, velar dysplasia, adenoid atrophy or irregularity, and neurologic conditions (both acquired or congenital) that affect the cranial nerves innervating the VPS.[6] Despite presenting with similar abnormalities in speech, the diagnosis of VPI and recognition of the underlying etiology may be delayed unless the patient is evaluated and treated by a cleft/craniofacial clinic.

The complexity of identifying the etiology of VPI, particularly in patients with noncleft etiologies, is compounded by variation seen in the VPS mechanism.[7] A multidisciplinary approach to testing is needed to identify the anatomic and functional variants that contribute to VPI. This approach includes speech and language evaluation, genetic and immunologic testing, CT or MRA imaging to evaluate for aberrant vasculature, dynamic instrumental assessment using flexible video nasopharyngoscopy (FVNP) and multiplanar video fluoroscopy (MPVF), and evaluation by plastic surgeons and otolaryngologists specializing in VPI correction.[8] FVNP provides in vivo imaging of the vocal tract during articulation. MPVF enhances the three-dimensional analysis of the VPS by measuring the structures and movements of the VPS during speech, visualizing the dynamic motion of both lateral pharyngeal walls and analyzing the depth of pharyngeal closure during speech.[7] The combination of these methods has been shown to be the best approach in both VPI assessment and treatment planning.

The historical perspective of VPI correction is testament to the advancements in diagnostic modalities leading to the evolution in surgical technique over the last century.[8–10] The first attempt at surgical correction of VPI documented in 1865 by Passavant involved the direct adhesion of the soft palate to the posterior pharyngeal wall.[6,7] The concept has undergone countless revisions and permutations. To date, the pharyngeal flap remains as one of the procedures used for correcting VPI and demonstrates some superiority when compared with sphincter pharyngoplasty in a 2012 meta-analysis.[11] Despite continued debate regarding the importance of pharyngeal flap design and orientation, outcomes have demonstrated that VPI correction is not a one-size-fits-all procedure.[7,12–14] Improved outcomes emphasize the surgical customization that suits the individual patient's anatomic and functional pathology.

A recent study by the senior author (PY) demonstrated a high success rate of pharyngeal flap reconstruction using FVNP and MPVF for individualized surgical planning among three different surgeons.[7] In this article, a review of current literature is expanded. The successful correction of VPI by four surgeons with variable technique is studied, evaluating the individualized pharyngeal flap reconstruction based on imaging findings. The purpose of this article is to expand on the concept that the individual customization of superior pedicle pharyngeal flaps based on imaging findings can achieve safe, reliable, and successful outcomes despite varying levels of surgeon experience, approach, and technique.

Takeaways

Question: Does customization of pharyngeal flaps using dynamic preprocedural assessment achieve reliable and successful outcomes in surgical treatment of velopharyngeal insufficiency despite differences in surgeon approach and technique?

Findings: There was overall successful surgical correction of velopharyngeal insufficiency in 92% of patients.

Meaning: High rates of success are achieved in treating velopharyngeal insufficiency when the surgical approach is individualized based on preoperative imaging despite variations in surgical techniques in performing superior pedicle pharyngeal flaps.

MATERIALS AND METHODS

This is a retrospective nonrandomized review of selected cases of VPI treated by customized pharyngeal flaps.

Patients

Children between the ages of four and eighteen years old with VPI undergoing pharyngeal flap surgery between August 2012 and December 2020 were identified. All patients were followed by a multidisciplinary team at the Ian Jackson Craniofacial and Cleft Palate Clinic at Beaumont Hospital in Royal Oak, Michigan. The protocol was approved by the Internal Review Board of Beaumont Hospital Royal Oak.

Preoperative Evaluation

Each patient was evaluated by the multidisciplinary team consisting of plastic surgeon, speech and language pathologist, otolaryngologist, orthodontist, and medical geneticist. Preprocedural MPVF and FVNP were performed. Of the 994 VPI patients identified, the study group included 170 patients with ventral velopharyngeal gaps greater than 20% and a circular shape as determined by MPVF and FVNP.[15] In total, 824 patients with borderline cases (defined as small closure defects in which no gap was discerned but bubbling was observed during the best articulation of the speech sample) and "hourglass" gaps were excluded (Fig. 1). Individualized preoperative planning of pharyngeal flaps was done based on findings on MPVF and FVNP. Polysomnography was indicated when clinical data of sleep disordered breathing were documented. A CT scan of the neck was performed to determine the course of the internal carotid artery.

For the planning of the customized pharyngeal flaps, the following actual size measurements from MPVF were obtained in every case: (a) distance between the tip of the uvula and the hard palate at rest; (b) distance between lateral pharyngeal walls at rest; (c) distance between lateral pharyngeal walls during the articulation of a standardized

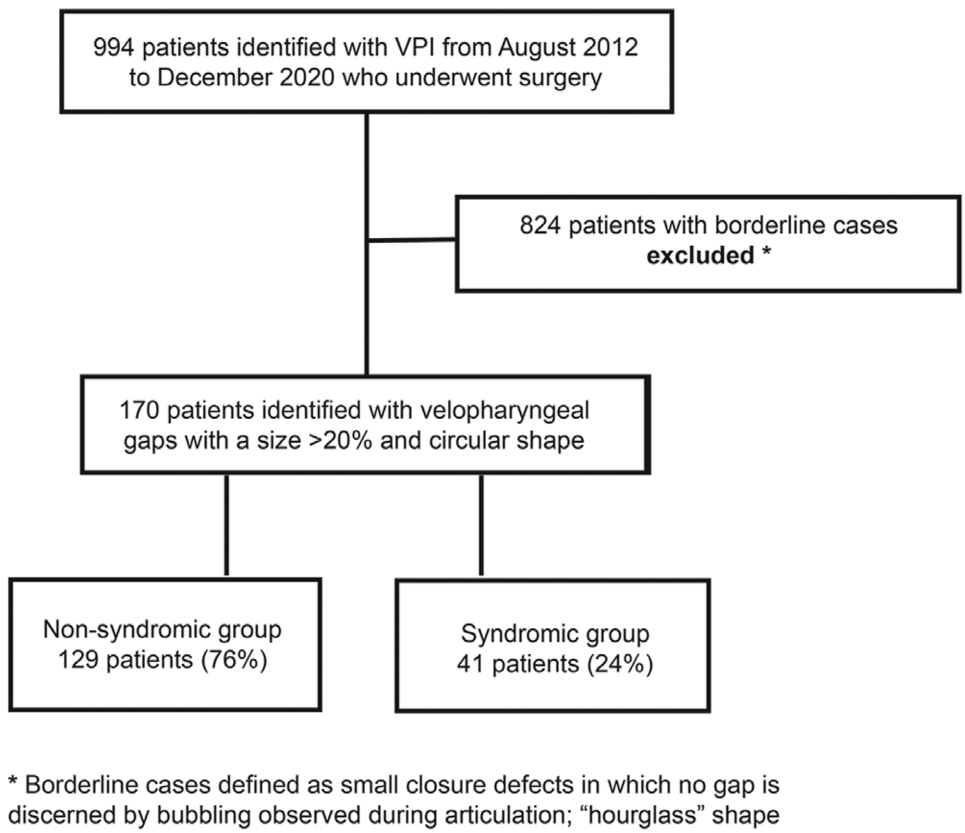

Fig. 1. Inclusion and exclusion criteria. Flow-chart of inclusion and exclusion criteria. Patients with velopharyngeal insufficiency evaluated from August 2012 to December 2020 were evaluated. Borderline cases were excluded from the study. The patients included in the study were further divided between syndromic and nonsyndromic groups.

speech sample, including plosive and fricative phonemes in combination with high and low vowels. Other velopharyngeal closure features during speech were assessed, including adenoid pad, shape of lateral pharyngeal wall motion during speech (balloon-like, shelf-like, longitudinal or irregular), direction of velum motion (posterior or posterior/superior), percentage of velopharyngeal closure gap (or size of the gap), and presence or absence of Passavant's ridge.[7] Besides MPVF, all patients underwent FVNP for assessing velopharyngeal closure during speech. FVNP findings were analyzed as described in previous reports, including velopharyngeal closure pattern, velar movement during articulation expressed as percentage, adenoid size, pharyngeal tonsils size, epiglottis, and vocal cords.[12–15] MPVF and FVNP findings were used for customizing an individualized surgical plan for each case. The length and the width of the flap are customized according to the actual size measurements of the MPVF. The length is corroborated intraoperatively using a needle and a laryngoscopy mirror. The preoperative indication for adenoidectomy and tonsillectomy is based on the imaging assessments.

Outcomes

The primary outcome of interest was successful correction of VPI. Surgical correction was considered successful if the following criteria were fulfilled: mean nasalance less than or equal to 35%, absence of nasal regurgitation, absence of nasal emission, and absence of sleep disordered breathing.

Secondary outcomes included patient demographics, etiology of VPI (syndromic versus nonsyndromic), surgical history, findings on MPVF and FVNP, pre- and postoperative interventions by speech pathology, and postoperative sleep disordered breathing. Preoperative rates of nasal emission, mean nasalance, and nasal regurgitation were also collected. Postoperative variables were collected at routine 2-month appointments where patients underwent nasometry and FVNP. Only those who demonstrated persistent VPI then underwent a postoperative MPVF. All patients with compensatory articulation patterns underwent postoperative speech pathology intervention. Patients were followed postoperatively for a minimum of 2 months and up to 2 years.

Surgical Techniques

A superior-based pharyngeal flap (SBPF) was performed by four surgeons with slight variations in techniques. Three surgeons (Surgeons 1–3) used a surgical technique described by Hogan, with adaption of recent modifications that omit lateral port control with stents.[1,8,16] One surgeon (Surgeon 4) used a technique originally described by Tatum.[17] All techniques utilized general anesthesia, endotracheal intubation, Dingman mouth gags, and local anesthetic. In all cases, the height and width of the flaps were determined by findings on preoperative imaging using MPVF and FVNP.

Three surgeons (surgeons 1–3) began with a midline incision to split the soft palate to the posterior nasal spine. The palatal musculature was then separated from the nasal and oral mucosa. With the posterior pharynx visualized, the SBPF was created. Two longitudinal incisions were made on each side of the posterior pharyngeal wall and connected inferiorly with a transverse incision. The incisions were taken down to prevertebral fascia and the SBPF was raised. All three surgeons then secured the caudal end of the flap to the posterior edge of the soft palate, using horizontal mattress sutures to inset the flap between the nasal mucosa and palatal musculature. Surgeon 3 also emphasized spreading the flap out as widely as possible. Once inset, all surgeons closed the soft palate in layers, using simple interrupted sutures. Surgeons 1 and 2 closed the soft palate in a similar manner: once the nasal layer was closed, the palatal muscle fibers of the posterior third palate were reoriented in a transverse fashion, followed by closure of the oral mucosa. Surgeon 3 closed the soft palate by creating a double opposing Z palatoplasty (one of the nasal layer and one of the oral layer), as described by Furlow to extend the length of the palate, moving the palatal musculature further posteriorly.[18] All three surgeons closed the posterior pharyngeal wall defect vertically, leaving a small area uncovered on the superior end to heal by secondary intention.

Surgeon 4 created a SBPF using a technique described by Tatum.[17] Similar to others, the pharyngeal flap position and width were determined according to findings on preoperative MPVF and FVNP. Instead of splitting the soft palate, surgeon 4 retracted the palate anteriorly and superiorly to provide adequate exposure. Surgeon 4 also emphasized high insertion of the flap on the soft palate, securing the caudal end relatively close to the hard palate-soft palate junction to avoid posterior displacement of the velum. Once the flap was created, a straight, transverse incision was made on the nasal surface of the soft palate, beginning 5–10 mm superior to the free edge of the uvula, wide enough to accommodate the width of the flap. A pocket was created in the soft palate by extending the transverse incision toward the hard palate. The flap was secured within the pocket using horizontal mattress sutures. The posterior pharyngeal wall defect was undermined down to the hypopharyngeal region and closed transversely (as opposed to the vertical repair described by the first three surgeons) to avoid circumferential narrowing of the pharynx. As previously described, a small area on the superior end of the pharyngeal wall defect was left uncovered to heal by secondary intention.

RESULTS

Patient Demographics and Preoperative Variables

Between August 2010 and December 2020, 170 patients were evaluated at the Ian Jackson clinic for VPI, and they underwent a SBPF by one of the four surgeons (Table 1, Fig. 1).

An estimated 76 of the 170 patients (45%) received an SBPF by surgeon 1 and surgeon 2 (59 and 17 patients, respectively) (Table 1). Of the 170 patients, 65 (38%) underwent surgery with Surgeon 3, who closed the soft palate with a modified Furlow's double opposing Z palatoplasty (SBPF + Z). In total, 29 of the 170 patients (17%) were operated on by surgeon 4, who secured the flap to the soft-palate with a transverse pouch (SBPF + P), avoiding division of the midline palate.

The age of the patient population ranged from 4 to 17 years, with a median age of 6 (Table 2). An estimated 110 patients (65%) had a history of tonsillectomy and adenoidectomy (T&A): 40 of 41 syndromic children (98%) and 70 of 129 nonsyndromic children (54%).

In assessing the preoperative speech and language pathology, all patients undergoing surgical correction of their VPI had nasal emission and a mean nasalance greater than 50%. Thirty-four patients (20%) had symptoms of nasal regurgitation. During preoperative imaging, all patients were able to repeat a speech sample without compensatory articulation patterns. However, 43 (25%) patients had compensatory articulation patterns and required speech pathology intervention before imaging procedures. This was done to achieve normal articulation during the speech sample, regardless of abnormal resonance and nasal emission.

Most cases (76%) of VPI were nonsyndromic (Table 2). Of the nonsyndromic etiologies, 85 (50%) were unilateral cleft lip and palate (UCLP), 17 (10%) were bilateral cleft lip and palate (BCLP), 16 (9%) were cleft palate (CP), and 11 (6%) were occult subtotal cleft of the secondary palate (ie, submucosal CP). Only 41 (24%) of VPI cases were syndromic. Of the syndromic cases, 35 (21%) were velocardiofacial syndrome (VCFS), also known as 22q11.2 deletion syndrome, and four (2%) were Stickler syndrome. Only one (1%) patient had 18p deletion, and one (1%) patient had neurofibromatosis 1 (NF1). Of the patients with VCFS, 22 of 35 patients (63%) also had a submucosal CP (Table 2). One hundred percent of patients with VCFS underwent T&A before SBPF.

Postoperative Outcomes

The overall success of surgical correction of VPI was 92%, with a range of 88%–93% (Table 3). VPI was successfully corrected in 115 of 124 patients (93%) by surgeons 1 and 2; 27 of 29 patients (93%) by surgeon 3; and 15 of 17 (88%) patients by surgeon 4. The failure rate of VPI was 13 out of 124 patients, and is broken down by mechanism of injury, VPI etiology, and surgical technique (Tables 4, 5).

The rate of unsuccessful outcomes was higher in patients with syndromic etiology of VPI (Table 4). Of the 13 patients with unsuccessful outcomes, five of the 13 patients (38%) had a syndromic etiology, including VCFS and VCFS with submucosal CP, and eight of 13 patients (62%) had a nonsyndromic etiology. This, however, represents unsuccessful outcomes in five of 41 total patients (12%) with syndromic etiology and eight of 129 total patients (6%) with nonsyndromic etiology. All syndromic

Table 1. Surgeons, Technique, and Patient Distribution

Surgeon	Technique	Soft Palate Incision	Pharyngeal Defect Closure	n	(%)
Surgeon 1	SBPF	Midline	Vertical	59	(35%)
Surgeon 2	SBPF	Midline	Vertical	17	(10%)
Surgeon 3	SBPF + Z	Midline	Vertical	65	(38%)
Surgeon 4	SBPF + P	Transverse	Transverse	29	(17%)
Total				170	

P, Pouch; Z, Z-plasty.

patients with unsuccessful outcomes had VCFS or VCFS with submucosal CP. A chi square test demonstrated a nonsignificant association between surgical techniques, surgeons, and unsuccessful outcomes ($P > 0.05$).

The mechanisms of failure included inferior migration of the flap, complete flap resorption, flap narrower than planned, and unilateral dehiscence (Table 5). All five cases of inferior migration of the flap occurred only in nonsyndromic patients. In the case of complete flap resorption, three of the four cases occurred in syndromic patients. Similarly, in cases with flap narrower than planned, two of the three cases occurred in nonsyndromic patients.

DISCUSSION

The optimal surgery approach to VPI is one that is tailored to the individual and achieves the goal of competent velopharyngeal closure. The purpose of this study was to report the results of a retrospective review of a large number of cases of VPI treated by pharyngeal flap surgery. The procedures were performed by four different surgeons, using different techniques. Preoperative planning and customization of each individual flap is highlighted in this study, although it is not the intention of this study to suggest that every case of VPI be treated with pharyngeal flap surgery. Other techniques include sphincter pharyngoplasty, secondary intravelar veloplasty, secondary Furlow's "Z" plasty, or injection of fat or synthetic materials. All these procedures should be considered as valid options and the ultimate clinical decision be made with consideration of the patient's clinical history, speech evaluation, and imaging findings.

From the results of this study, it seems evident that despite variations in surgical technique, superior pedicle pharyngeal flaps can yield a high success rate when the procedure is planned according to imaging findings. The success rate of 92% is comparable to other studies that place the success rate of pharyngeal flap surgery in the range of 81.5%–97%.[5] The data of this study expand upon previous research through the addition of a surgeon and

Table 2. Patient Demographics and Preoperative Variables

Age, median (range)	6 y	(4–17 y)
Prior T&A, n (%)	110	(65%)
Nonsyndromic, n (%)	70	(41%)
Syndromic, n (%)	40	(24%)
Preoperative speech and language pathology		
Nasal emission, n (%)	170	(100%)
Mean nasalance >50%, n (%)	170	(100%)
Nasal regurgitation, n (%)	34	(20%)
Intervention before imaging, n (%)	43	(25%)
VPI Etiology		
Nonsyndromic, n (%)	129	(76%)
UCLP, n (%)	85	(50%)
BCLP, n (%)	17	(10%)
CP, n (%)	16	(9%)
Submucosal CP, n (%)	11	(6%)
Syndromic, n (%)	41	(24%)
VCFS (22q11.2 deletion), n (%)	35	(21%)
VCFS (22q11.2 deletion) AND submucosal CP, n (%)	22	(63%)
Stickler Syndrome, n (%)	4	(2%)
18p deletion, n (%)	1	(1%)
NF1, n (%)	1	(1%)

UCLP: unilateral cleft lip and palate; BCLP: bilateral cleft lip and palate; CP: cleft palate; VCFS: velocardiofacial syndrome; NF1: neurofibromatosis 1.

Table 3. Postoperative Outcomes

Surgeon: Technique	Patients N	Corrected VPI n (%)	Persistent VPI n (%)
Surgeon 1: SBPF	59	54 (91%)	5 (9%)
Surgeon 2: SBPF	17	15 (88%)	2 (12%)
Surgeon 3: SBPF + Z	65	61 (93%)	4 (7%)
Surgeon 4: SBPF + P	29	27 (93%)	2 (7%)
Total	170	157 (92%)	13 (8%)

Z: Z-plasty; P: Pouch.

Table 4. Unsuccessful Outcomes Based on VPI Etiology

VPI Etiology	n (%)	Mechanism of Failure
Syndromic	5 (38%)	
VCFS	*3 (23%)*	
	1 (8%)	Unilateral dehiscence
	1 (8%)	Flap narrower than planned*
	1 (8%)	Complete flap resorption
VCFS and submucosal CP	2 (15%)	Complete flap resorption
Total	5/41 (12%)	
Nonsyndromic	8 (62%)	
Submucosal CP	*3 (23%)*	
	1 (8%)	Complete flap resorption†
	1 (8%)	Inferior migration of flap
	1 (8%)	Flap narrower than planned
UCLP	***3 (23%)***	
	2 (15%)	Inferior migration of flap
	1 (8%)	Flap narrower than planned
BCLP	***2 (15%)***	Inferior migration of flap
Total	**8/129 (6%)**	

*Normal chromosomal microarray analysis with history of previous unsuccessful flap. Probable Opitz syndrome.
†Ten days of postoperative fever, two courses of antibiotics necessary.

Table 5. Unsuccessful Outcomes Based on Mechanism of Failure

Mechanism of Failure	n (%)	Syndromic, n (%)	Nonsyndromic, n (%)
Inferior migration of flap	5 (38%)	0 (0%)	5 (100%)
Complete flap resorption	4 (31%)	3 (75%)	1 (25%)
Flap narrower than planned	3 (23%)	1 (33%)	2 (67%)
Unilateral dehiscence	1 (8%)	1 (100%)	0 (0%)

a wider variation in technique. A single-surgeon study by Baek et al demonstrated that preoperative planning using FVNP resulted in velopharyngeal competency in 96% of patients.[5] Our study encompassed a larger number of surgeons and surgical techniques but produced comparable outcomes due to preoperative planning and anatomic information using MPVF and FVNP.

All 35 cases of VCFS underwent a neck CT scan with contrast before T&A, followed by pharyngeal flap creation. The CT aimed to assess the anatomical course of the internal carotid arteries, which are frequently medialized in cases of VCFS. In two of the 35 cases, unilateral medialization was detected and both demonstrated internal carotid artery medialization to the left. In both cases, the surgeon was aware of the leftward medialization of the internal carotid arteries and no complications occurred. As a result, it is imperative to implement the routine use of CT imaging before VPI correction for preventing adverse events.

In the 13 patients (8%) who experienced failure of VPI correction, the most common findings were shown to be persistent VPI secondary to inferior displacement, shrinking, or resorption of the pharyngeal flap. Resorption of the flap was more common in patients with a syndromic etiology of VPI while inferior displacement occurred only in nonsyndromic patients. One patient with resorption of the flap experienced postoperative fevers, requiring two courses of antibiotics. While the creation of flaps narrower than planned can be attributed to technical error, other mechanisms of failure beg the question of the underlying physiology leading to unsuccessful outcomes. There may be an inflammatory role causing inferior flap migration or due to embryologic or developmental variance due to VCFS. Further studies with larger sample sizes are needed to elucidate pathology. Multivariate analysis of variables and outcomes was not performed due to the small sample size of patients who were negatively affected. Had this been done, the results would have led to unreliable conclusions.

There are several limitations to this study. First, it is retrospective in nature, and designed to capture the clinical course of patients presenting with VPI. The surgeon and surgical technique could not be randomized. Furthermore, despite the different etiologies and cleft types, the study group was homogenous given the specific inclusion criteria and follow-up occurring at 2 months postoperative. Larger cohorts will be needed to further analyze failure of VPI correction and perform multivariate analysis. It is pertinent that future studies increase the overall sample size of patients undergoing VPI so that a multivariate analysis of variables and outcomes can be conducted.

CONCLUSION

High rates of success are achieved in treating velopharyngeal insufficiency when the surgical approach is individualized based on preoperative imaging despite variations in surgical techniques in performing superior pedicle pharyngeal flaps.

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