Original Investigation

Revision Rates and Speech Outcomes Following Pharyngeal Flap Surgery for Velopharyngeal Insufficiency

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**IMPORTANCE**  Velopharyngeal insufficiency in children with cleft palate (and other causes) contributes to difficulty with communication and quality of life. The pharyngeal flap is a workhorse to address hypernasality and nasal air escape. However, there is a paucity of literature on the characteristics of cases that require revision.

**OBJECTIVE**  To measure the revision rate of pharyngeal flaps, compare the preperceptual and postperceptual speech scores, and identify the characteristics of those patients who required revision.

**DESIGN, SETTING, AND PARTICIPANTS**  A retrospective medical record review was completed for patients who underwent pharyngeal flap surgery from June 1, 2008, through January 31, 2013, at a tertiary academic center.

**MAIN OUTCOMES AND MEASURES**  Perceptual speech analyses and surgical revision rates. Perceptual speech patterns before and after surgery were compared using nasal air emission and resonance scores. The association between requiring revision surgery and covariates was analyzed using multivariable mixed-effects logistic regression.

**RESULTS**  Sixty-one patients were identified, including 24 boys (39%) and 37 girls (61%). The mean (SD) patient age at the time of pharyngeal flap surgery was 8.2 (6.8) years (range, 3-55 years). Velopharyngeal insufficiency was associated with cleft palate in 51 patients (84%), and 17 patients (28%) had a syndrome. The mean (SD) time to surgery after the speech evaluation was 225 (229) days (range, 14-1341 days). The mean (SD) nasal air emission scores decreased by −1.1 (2.0 [1.1] preoperatively to 0.8 [1.1] postoperatively). The mean (SD) resonance score decreased by −1.5 (2.4 [1.1] preoperatively to 0.9 [1.1] postoperatively; P < .001). Flaps were revised in 12 patients (20%), including port revision in 9, complete flap revision in 2, and flap takedown in 1. The only covariate that was significantly associated with revision rates was increased age at surgery, which was associated with a higher probability of revision surgery (odds ratio, 1.31; 95% CI, 1.03-1.66; P = .04).

**CONCLUSIONS AND RELEVANCE**  Pharyngeal flap surgery, when appropriately selected, was effective at improving speech with a revision rate of 20%, which is comparable to previously published studies. Increased age at the time of the pharyngeal flap surgery was associated with an increased need for revision surgery, supporting evidence that cleft centers should encourage early childhood speech evaluations with consistent documentation and prompt treatment.

**LEVEL OF EVIDENCE**  3.
elopharyngeal dysfunction (VPD) necessitating surgery occurs in approximately 15% to 45% of patients with a cleft palate after primary palatoplasty.1 During phonation, the velopharyngeal closure pattern contributes to balanced oronasal resonance.2 Velopharyngeal insufficiency (VPI) describes the inability to close the velopharyngeal sphincter because of an anatomical or structural deficit during the production of oral sounds during speech. Complete closure necessitates coordinated movement of the soft palate and the lateral and posterior pharyngeal walls.3 Patients with VPI present with nasal air escape and hypernasality during phonation. The patient’s quality of life can be negatively affected by decreased overall speech intelligibility from VPI.4

The pharyngeal flap came into common use by surgeons in the 1950s, although Schoenborn originally described it as early as 1876.5 Today it remains one of the most commonly used speech operations for VPI.6 The goal of a successful pharyngeal flap is to create a central subtotal velopharyngeal obstruction. Residual nasal airflow passes through lateral ports between the oral and nasal cavities.5 The size and placement of the pharyngeal flap are chosen to maintain nasal airflow while facilitating velopharyngeal closure for speech. The lateral pharyngeal wall movement controls the airflow through these ports.

In 2008, Cole et al7 reviewed their experience with 222 consecutive patients undergoing pharyngeal flap surgery and concluded that the flap was a safe and reliable option with relatively rare complications. Apart from having a favorable safety profile, the pharyngeal flap was effective in improving speech outcomes, which is equally supported with published improvements in perceptual and objective analysis.8-10

The evaluation of a patient with repaired cleft palate includes perceptual speech assessment by a trained speech-language pathologist with familiarity with patients with cleft.11 Speech-language pathologists typically identify and document resonance, nasal air emission (NAE), consonant production errors, speech intelligibility, and speech acceptability.12 Speech articulation errors may be adequately addressed with the use of intensive speech therapy.

Hypermobility and VPI, on the other hand, often require surgical management. The speech-language pathologist and cleft surgeon complete a standardized VPD evaluation. The velopharyngeal closure pattern is assessed with standardized nasopharyngeal endoscopy and the treatment plan jointly formulated.13 At our institution, the secondary speech surgery treatment is planned based on the VPD evaluation. The velopharyngeal closure pattern and size of the velopharyngeal gap are 2 of the factors that guide the surgical plan to include (1) pharyngeal flap (large central gap with sagittal closure and adequate lateral wall movement), (2) Furlow double-opposing z-plasty palatoplasty (small central gap with notched soft palate, indicating levator veli palatini malpositioning), or (3) sphincter pharyngoplasty (coronal closure pattern).

The pharyngeal flap is designed to fit the defect seen on nasopharyngeal endoscopy as described by Shprintzen et al.14-15 The choice of the flap size must include respect for the potential unintended consequence of nasal obstruction and obstructive sleep apnea (OSA). Thus, if too small of a flap is chosen, a revision procedure may be necessary, whereas a flap that is too large may narrow the nasal airway and require flap take-down. Patients are followed up clinically for OSA symptoms, which would warrant a sleep study.

Some patient characteristics (eg, cleft size or timing of primary palatoplasty) have been investigated in patients with persistent VPI after speech surgery, but specific characteristics that may predispose patients to revision pharyngeal flap are sparsely reported.4,16 Bohm et al17 compared surgical and perceptual speech outcomes in patients treated with pharyngeal flap, sphincter pharyngoplasty, or combined Furlow palatoplasty and sphincter pharyngoplasty. Better speech outcomes were noted after pharyngeal flap or combined sphincter and Furlow procedures compared with the sphincter pharyngoplasty alone; however, the patients who underwent pharyngeal flap required more revisions (29%).

There is a paucity of literature on the characteristics of those patients with VPI who undergo revision of their primary pharyngeal flap surgery. The objectives of this study were to (1) measure the revision rate noted in this sample population, (2) compare the preperceptual and postperceptual speech scores, and (3) identify the characteristics of those patients who required revision.

Methods

After University of California, Davis, institutional review board approval, we performed a retrospective review of all patients with VPI who underwent a superiorly based pharyngeal flap performed by surgeons on the University of California, Davis, cleft team (J.M.S., C.W.S., and T.T.T.) from June 1, 2008, through January 31, 2013. Exclusion criteria included lack of 12 months of postoperative follow-up, previous pharyngeal flap at an outside institution, or other secondary speech surgery procedure (eg, sphincter pharyngoplasty or Furlow double-opposing z-plasty palatoplasty). Data points included age, sex, presence of syndrome, comorbidities, need for revision surgery, age at time of surgery, age at time of revision, time between speech evaluation and flap surgery, and the standardized preoperative and postoperative perceptual speech evaluations.

Operative Technique

All surgeons used the following surgical technique for the superiorly based pharyngeal flap (with only minor variations). Tonsillectomy and adenoidec tomy are completed approximately 3 months before the flap. The velum is split in the midline to the level of the hard palate. The triangular, nasal mucosal flaps are elevated and hinged posteriorly to determine the port size on either side. Red rubber catheters are placed oronasally and exchanged for stents (endotracheal tubes or nasopharyngeal trumpets) to determine the port size. The pentagon-shaped, superiorly based myomucosal pharyngeal flap is elevated in the plane above the prevertebral fascia and elevated as high into the nasopharynx as possible (with care to protect the torus tubarius). The pharyngeal flap is then inset into the oral and nasal flaps and the palatal split reconstructed.
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Table 1. Severity Scale for Nasal Air Emission and Resonance

<table>
<thead>
<tr>
<th>Score</th>
<th>Definition</th>
</tr>
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<tbody>
<tr>
<td>0</td>
<td>Within normal limits</td>
</tr>
<tr>
<td>1</td>
<td>Mild</td>
</tr>
<tr>
<td>2</td>
<td>Mild to moderate</td>
</tr>
<tr>
<td>3</td>
<td>Moderate</td>
</tr>
<tr>
<td>4</td>
<td>Moderate to severe</td>
</tr>
<tr>
<td>5</td>
<td>Severe</td>
</tr>
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</table>

Speech Assessment

Standardized preoperative perceptual speech assessment and VPD assessment with nasopharyngoscopy was documented by the cleft team speech-language pathologist and surgeon. The NAE and resonance severities were graded using the validated Pittsburgh Weighted Values for Speech Symptoms Associated with Velopharyngeal Incompetence Instrument (Table 1). The resonance severity is scored as follows: 0, within acceptable limits; 1, mild hypernasality; 2, mild to moderate hypernasality; 3, moderate hypernasality; 4, moderate to severe hypernasality; and 5, severe hypernasality. Hyponasal resonance was assigned a score of 0 and mixed resonance a score of 2 as per the protocol.19

The secondary speech surgery plan is developed by the consistent interdisciplinary team evaluation, which included at least the following factors: perceptual speech evaluation, nasopharyngeal closure pattern seen on nasopharyngal endoscopic examination, previous surgical treatments, palatal length, comorbidities, and airway obstruction symptoms.

Statistical Analysis

The NAE and resonance scores before and after surgery were compared using paired t tests. The association between requiring revision surgery and patient characteristics was analyzed using multivariable mixed-effects logistic regression, including a random effect for surgeon in the model. Confounding clinical characteristics were selected based on a priori knowledge. Analyses were conducted using the statistical software environment R, version 3.1.1. The mixed-effects logistic regression model was fitted using the R function glmmPQL.

Results

Patient Demographics

After exclusion criteria, 61 patients with surgically managed VPI were identified. Table 2 gives the demographic and surgical characteristics for all patients. The mean (SD) age was 8.2 (6.8) years (range, 3-55 years), including 34 boys (39%) and 27 girls (61%). Fifty-one (84%) had undergone an orofacial cleft and previous palatoplasty, whereas 10 patients (16%) had VPI of a noncleft origin. Complete clefts of the hard palate were the most common, occurring in 16 patients (26%), followed by partial clefts of the hard palate in 15 patients (25%), bilateral complete cleft lip and palate in 8 patients (13%), submucous cleft palate in 6 patients (10%), soft palate clefts in 4 patients (7%), and cleft of the uvula in 2 patients (3%). Pierre Robin sequence was seen in 9 patients. Seventeen patients (28%) had been diagnosed as having a syndrome or developmental or neurologic deficit that could affect velopharyngeal function. The most common condition was velocardiofacial syndrome (n = 7), followed by developmental delay (n = 4), Stickler syndrome (n = 3), cerebral palsy (n = 1), Kabuki syndrome (n = 1), and Diamond-Blackfan syndrome (n = 1). The mean (SD) time to secondary speech surgery (which includes time before tonsillectomy and adenoidectomy and ≥90 days healing time before pharyngeal flap surgery) after the perceptual speech evaluation was 225 (229) days (range, 14-1341 days).

Speech Outcomes

Complete presurgery and postsurgery perceptual speech data sets were available for 37 patients. Table 3 gives the preoperative and postoperative speech outcomes. The preoperative NAE and resonance scores were 3 or higher (moderate to severe) in 16 (43%) of 37 patients and 19 (51%) of 37 patients. All patients who did not require revision surgery had a decrease or no change in the NAE and resonance scores after surgery. The NAE and resonance scores decreased significantly after surgery. The mean (SD) preoperative NAE score of 2.0 (1.1) decreased to 0.8 (1.1) postoperatively. The mean (SD) resonance score of 2.4 (1.1) decreased to 0.9 (1.1) (P < .001). Revision pharyngeal flap surgery was completed in 12 patients (20%) with port revisions in 9, complete flap revisions in 2, and flap takedown in 1 (due to OSA). Eleven of these 12
The association between requiring revision surgery and patient characteristics was analyzed using multivariable mixed-effects logistic regression, including a random effect for surgeon in the model. The model was fitted with the following covariates: age at surgery, cleft type, presence of syndrome, and time to surgery after speech evaluation. Table 4 gives the results of the multivariable mixed-effects logistic regression analysis of revision. Increased age at surgery was associated with a higher probability of revision surgery (odds ratio, 1.31; 95% CI, 1.03-1.66; P = .04). Other covariates were not significantly associated with requiring revision surgery. Presence of a syndrome (eg, velocardiofacial syndrome) was not associated with revision surgery.

Table 4. Multivariable Mixed-Effects Logistic Regression Analysis of Revision

<table>
<thead>
<tr>
<th>Covariate</th>
<th>Odds Ratio (95% CI)</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age at surgery</td>
<td>1.31 (1.03-1.66)</td>
<td>.04</td>
</tr>
<tr>
<td>Cleft type</td>
<td>1.40 (0.81-2.43)</td>
<td>.26</td>
</tr>
<tr>
<td>Syndrome (yes vs no)</td>
<td>0.28 (0.03-2.56)</td>
<td>.29</td>
</tr>
<tr>
<td>Preoperative score</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Nasal airway emission</td>
<td>0.62 (0.26-1.48)</td>
<td>.32</td>
</tr>
<tr>
<td>Resonance</td>
<td>1.34 (0.59-3.06)</td>
<td>.51</td>
</tr>
<tr>
<td>Time to surgery after speech evaluation</td>
<td>0.99 (0.98-1.00)</td>
<td>.10</td>
</tr>
</tbody>
</table>

Discussion

The results of this study are consistent with and extend from previous reports on the effectiveness of speech improvement and revision rates after pharyngeal flap. Although NAE and resonance scores improved after pharyngeal flap procedures, a higher likelihood of needing revision surgery was seen as age of the patient at the time of the pharyngeal flap surgery increased. The mean age of patients at time of pharyngeal flap in our study is significantly older than the recommended age, which is typically 4 to 6 years old. At this age, the child’s ability to cooperate with nasopharyngeal endoscopy and acquisition of consistent speech samples is the rate-limiting step, but earlier interventions seem to be related with better speech outcomes. Fukuhiro and Trindade suggested that patients 6 to 12 years of age had better outcomes than their older counterparts. Our study also has an older age range and mean age for patients at the time of pharyngeal flap than the study by Bohm et al, who found a mean (SD) patient age of pharyngeal flap surgery of 7.3 (3.6) years (range, 3.1-15.6 years).

Older children who had not had adequate velopharyngeal port function with which to develop balanced resonance and the oral air pressure needed for speech sounds are unable to retrain their developed speech patterns after surgery. Several factors may have contributed to the increased age and the prolonged time from speech assessment to surgery in our sample population, which may include medical comorbidities, poor familial adherence and follow-up due to socioeconomic restraints, and overseas adoptions of older children who present with unrepaired primary cleft palate. Late primary palatoplasty increases VPI rates and deserves additional research attention. Surgeons and speech-language pathologists should counsel at-risk patients and families on the increased risks associated with increasing age.

The flap revision rate may be related to increasing age in this study, but a better or worse speech outcome was not identified with increasing age. The revision rate included predominantly port revisions. One could surmise that surgical factors, which were uncommon, influenced the need for a complete flap revision. These factors may include wound healing, surgical technique, infection, or patient dietary adherence, which could be addressed in a large cohort study.

A few potential explanations for the observed effect of increasing age on an increased flap revision rate can be hypothesized but not proved with this study. These explanations may include the following: (1) longer exposure to the inability to create adequate velopharyngeal function, limiting the development of balanced resonance and the oral air pressure needed for speech sounds to train the speech patterns; (2) delayed flap surgery, a secondary effect of children waiting until a later date for their pharyngeal flap surgery because of outstanding circumstances as described above; and (3) potential increased pharyngeal muscular strength capable of affecting the flap’s healing in older patients and possible nonadherence of an adult who can choose his or her own diet, thereby affecting the viability of the flap.

A dreaded secondary effect of pharyngeal flap is OSA. In this series, only one adult developed symptomatic nasal obstruction and sleep apnea symptoms. Children and adults are at risk for OSA after a pharyngeal flap. Liao et al reported no association between the width of the pharyngeal flap and resulting severity of OSA. We prefer to completely take down the flap in the setting of OSA, although others may selectively open the port size as a first step.

This analysis of our cleft center’s experience has limitations that affect the generalizability of the results to other cen-
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Administrative, technical, or material support: Setabutr, Roth, Cervenka, Sykes, Tollefson.

Study supervision: Setabutr, Senders, Tollefson.

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REFERENCES


