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Long-term outcomes of sphincter pharyngoplasty in patients with cleft palate☆

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Summary

Objective: The purpose of this study was to evaluate long-term outcomes of sphincter pharyngoplasties, including speech outcomes, revision surgeries, and postoperative incidence of obstructive sleep apnea (OSA).

Design: Retrospective matched-cohort study

Setting: Two craniofacial centers in Los Angeles, CA

Patients: Patients (n = 166) with cleft lip and palate (CLP) or isolated cleft palate (iCP) who underwent sphincter pharyngoplasty from 1992 to 2022 were identified. An age- and diagnosis-matched control group of 67 patients with CLP/iCP without velopharyngeal insufficiency (VPI) was also identified.

Interventions: The pharyngoplasty group underwent sphincter pharyngoplasty, whereas the non-VPI group had no history of VPI surgery or sphincter pharyngoplasty.

Main outcome measures: Postoperative speech outcomes, revision surgeries, and incidence of OSA were evaluated. Multivariable regression was used to evaluate independent predictors of OSA.

Results: Among the patients in the pharyngoplasty cohort, 63.9% demonstrated improved and sustained speech outcomes after a single pharyngoplasty, with a median postoperative follow-up of

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Statement of ethical approval
This multi-institutional retrospective study was approved by the University of California, Los Angeles, Institutional Review Board (no. 11-000925).

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8.8 years (interquartile range [IQR], 3.6-12.0 years). One-third of the patients who underwent pharyngoplasty required a revision surgery, with a median time to primary revision of 3.9 years (IQR, 1.9-7.0 years). OSA rates increased significantly among the pharyngoplasty cohort, from 3% before surgery to 14.5% after surgery ($p < 0.001$). The average time from sphincter pharyngoplasty to OSA diagnosis was 4.4 ± 2.4 years. Multivariable analysis results indicated that sphincter pharyngoplasty surgery was independently associated with a fourfold increase in OSA ($p = 0.03$).

Conclusions: Although sphincter pharyngoplasty remains successful in improving long-term speech outcomes, persistent OSA is a sequela that should be monitored beyond the immediate postoperative period.

Keywords

Sphincter pharyngoplasty; Velopharyngeal insufficiency; Cleft palate; Obstructive sleep apnea; Revision surgery

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Sphincter pharyngoplasty is an established surgical technique for the treatment of velopharyngeal insufficiency (VPI) in children with cleft palate (CP) owing of its historically high success rate, ease of revision, and relatively low incidence of postoperative airway complications.[1] However, few studies have evaluated these outcomes within the same cohort or beyond the early postoperative period. Larger retrospective studies have generally been limited to speech outcomes and revision rates.[2-8] Studies have also been limited by follow-up period; in a recent systematic review, the largest study comprising 250 patients had a mean follow-up period of 2.4 years.[1,2] However, as noted by Orticochea,[3] velopharyngeal incompetence can reoccur upon completion of puberty due to an increase in the size of the pharynx, resulting in separation of the posterior pillars down the midline of the posterior pharyngeal wall. Thus, long-term studies assessing VPI reoccurrence and indications for revision surgeries are needed.

Furthermore, a less studied outcome of sphincter pharyngoplasties is the postoperative incidence of persistent obstructive sleep apnea (OSA). The dynamic port created in a sphincter pharyngoplasty procedure, through elevation and transposition of bilateral myomucosal flaps, decreases the cross-sectional area of the velopharyngeal port.[9,10] This clinically evident reduction in velopharyngeal diameter, by definition, increases airway obstruction to a certain degree, especially in the immediate postoperative period. However, it is unclear whether postoperative OSA is transient due to postoperative edema or whether it can become persistent. Studies that have evaluated OSA rates after sphincter pharyngoplasty have been limited by a lack of comparison groups, small sample sizes, and/or short-term follow-up.[11-14] Others have reported obstructive sleep symptoms, such as snoring, as a reason for revision, although no OSA diagnosis was made using polysomnography.[15,16] The largest study evaluating OSA rates after sphincter pharyngoplasty by Ettinger et al.[17] included 146 patients, with a mean follow-up time of 4.5 years, and demonstrated a significant postoperative increase in OSA incidence (1.4% before surgery to 22% after surgery). However, the timing of the postoperative OSA diagnosis was not reported; therefore, it is unclear whether the cases were acute versus persistent.

Given the limitations of follow-up that exist within the literature, the purpose of this study was to evaluate the long-term outcomes and complications of sphincter pharyngoplasty, including speech outcomes, revision surgeries, and postoperative incidence of OSA.

Methods

Patient selection

This retrospective matched-cohort study adhered to Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) guidelines. Medical records from 1992 to 2022 were retrieved from the Craniofacial/Cleft Palate Program at the Orthopaedic Institute for Children and the Craniofacial Clinic, University of California, Los Angeles, to identify sphincter pharyngoplasty cases. Patients with a history of cleft lip and palate (CLP) or isolated cleft palate (iCP), VPI diagnosis, and sphincter pharyngoplasty (n = 187) met the inclusion criteria. We excluded patients who underwent sphincter pharyngoplasty surgery at the age of > 21 years (n = 9) and patients with less than 6 months of postoperative follow-up (n = 12). As a control group, we identified an age- and diagnosis-matched cohort of patients with a history of CLP or iCP without VPI. The control group was age-matched based on the age at the last follow-up.

Patients were retrospectively reviewed for demographics, speech evaluations, VPI diagnosis and surgical treatment, pharyngoplasty revision procedures, and sleep study evaluations using multidisciplinary team notes, operative reports, and consultation reports. This multi institutional retrospective study was approved by the Institutional Review Board of the University of California, Los Angeles (no. 11-000925).

Diagnosis and surgical correction of velopharyngeal insufficiency

Diagnosis of VPI was determined via nasoendoscopy (i.e., an observed velopharyngeal gap during an oral loaded speech task) and/or a perceptual rating of hypernasality on speech pathology evaluation. Preoperative nasoendoscopy assessment was routinely performed, except during the coronavirus disease 2019 pandemic. Indication for VPI surgery was determined by both the surgeon and the speech pathologist.

Secondary Furlow palatoplasty is typically used as first-line treatment in patients with a short palate and excellent wall motion or those treated with primary intravelar velo-plasty. Sphincter pharyngoplasty is typically performed when patients undergo primary or secondary Furlow palatoplasty or when wall motion is deficient. Pharyngeal flaps and buccal myomucosal interposition flaps are not part of our surgical algorithm.

The Jackson modification of the Hynes sphincter pharyngoplasty (Figure 1) is performed as follows[18]: A transverse incision is made across the posterior pharyngeal wall at the level of the C1 vertebral body. Parallel incisions are made just behind the posterior tonsillar pillars. The lateral pharyngeal walls are elevated as superiorly based myomucosal flaps containing the palatopharyngeus muscles. The flaps are interpolated into the defect in the posterior pharyngeal wall and inset at the C1 level, at or above Passavant's ridge, in a double-breasted fashion, narrowing the pharyngeal port. The donor sites are then closed in layers.

Primary outcomes

This study assessed speech outcomes, revision surgeries, and incidence of postoperative OSA after sphincter pharyngoplasty.

At the annual multidisciplinary team clinic, patients underwent preoperative and postoperative speech evaluations, which included assessment of resonance balance and voice quality, by a licensed speech pathologist. Resonance balance was categorized as normal (oral), hypernasal, hyponasal, culde-sac, or mixed and could include an audible nasal air emission, if detected. Voice quality was categorized as normal, hoarse, harsh, strained, or aphonic. Preoperative hypernasal resonance was noted for all included patients. Postoperative speech improvement was defined as normal resonance and voice quality, without nasal air emission, in the most recent speech evaluation after pharyngoplasty.

Revision surgeries after sphincter pharyngoplasty were reviewed using patient operative records. Revision surgery indications were categorized as either persistent VPI (requiring tightening or repositioning of the port) or obstructive symptoms (requiring enlarging or releasing of the port).

Patients were referred for polysomnography studies based on pertinent history and clinical suspicion for OSA. Sleep study reports and pulmonology consultation notes were reviewed for diagnosis of OSA based on polysomnogram, apnea-hypopnea index (AHI), and prescription of continuous positive airway pressure (CPAP) or bilevel positive airway pressure (BiPAP).

Statistical analysis

Descriptive statistics were performed to evaluate all patient characteristics. The pharyngoplasty and control groups were compared; differences in the distribution of demographic and diagnostic variables were assessed using chi-square tests and independent t tests. When there were fewer than five patients in a subgroup and/or the data were not normally distributed, Fisher's exact tests and Mann-Whitney U tests were used in place of chi-square and t tests, respectively.

Among patients in the sphincter pharyngoplasty cohort, incidences of OSA and OSA treatment with CPAP/BiPAP were compared before and after surgery using McNemar's tests. The polysomnogram-based OSA data variables were compared between the pharyngoplasty and control groups using Fisher's exact tests, independent t tests, and Mann-Whitney U tests, where appropriate.

We conducted univariable analyses to evaluate predictor variables of OSA. The association between all potential explanatory variables and the presence of OSA was determined using chi-square tests, Fisher's exact tests, t tests, or Mann-Whitney U tests. All explanatory variables with p values < 0.07 were incorporated in multivariable logistic regression to model independent predictors of OSA. All statistical analyses were performed using SPSS Version 28 (IBM Corp., Armonk, NY) with an alpha level of $p < 0.05$.

Results

Patient characteristics

Two hundred thirty-three patients (54.9% men) with CLP or iCP (mean age at the last follow-up, 19.0 ± 2.6 years) were reviewed. One hundred sixty-six patients who underwent sphincter pharyngoplasty were compared to an age- and diagnosis-matched control group of 67 patients without a history of VPI. Baseline patient characteristics were comparable between the two groups (Table 1). Most patients in both groups had unilateral cleft lip and palate (UCLP) (50.0% pharyngoplasty group vs 47.8% control group) and were non-syndromic (77.7% vs 80.6%). The only significant difference between the groups was primary CP repair type given that a larger percentage of the pharyngoplasty cohort had unknown repair types (such as repairs performed at other institutions or countries). However, all basic demographic characteristics (such as sex and age) and variables related to OSA risk (such as cleft diagnosis, Pierre Robin sequence, and history of tonsillectomy or adenoidectomy) were comparable.

Sphincter pharyngoplasty speech outcomes and revision rates

One hundred sixty-six patients underwent sphincter pharyngoplasty at a median age of 10.3 years (interquartile range [IQR], 7.4-14.2 years), with a median postoperative follow-up time of 8.8 years (IQR, 3.6-12.0 years; Table 2).

After sphincter pharyngoplasty, 63.9% of the patients demonstrated improved speech on clinical examination. Fifty-five patients (33.1%) underwent revision surgery at a median postoperative time of 3.9 years (IQR, 1.9-7.0 years). The pharyngoplasty revision indications included persistent VPI requiring tightening or repositioning of the port (n =43) and airway obstruction requiring enlarging or releasing of the port (n = 11). Of the 55 primary revision cases, 39 patients (70.9%) had sustained improved speech outcomes at the most recent follow-up. After primary revisions, improved speech outcomes for the total cohort increased from 63.9% to 87.3%.

Twelve patients (7.2%) required additional surgical intervention. On an average, secondary revisions were required 2.2 ± 1.6 years after primary revision. Half of these patients demonstrated sustained improved speech outcomes after secondary revision. Improved speech outcomes for the entire cohort increased to 91.0% after secondary revisions.

Tertiary pharyngoplasty was required for three patients at an average of 5 years after the secondary revision. One of the three patients (33.3%) demonstrated improved speech after tertiary revision. Improved speech outcomes for the entire cohort increased to 91.6% after tertiary revisions.

Obstructive sleep apnea

Next, we reviewed the available polysomnogram data to evaluate OSA incidence between the pharyngoplasty and control groups (Table 3). Among the patients in the sphincter pharyngoplasty cohort, OSA incidence increased after surgery, that is, OSA prevalence increased from five patients before surgery to 24 patients after surgery (3.5% vs 14.5%, respectively; $p < 0.001$). In comparison, the control group had a baseline OSA incidence of 4.5%. The median AHI was 3.4 for the pharyngoplasty cohort and 1.8 for the control group. Given the pediatric OSA diagnostic criteria (a AHI score > 1 is abnormal), most of these cases were classified as mild OSA (AHI, 1-4.9 events/hour).[19,20] The average time from sphincter pharyngoplasty to OSA diagnosis was 4.4 ± 2.4 years. In patients with at least 3 years of follow-up after pharyngoplasty (n = 135), the rate of postoperative OSA increased to 17.0%.

Total OSA treatment requirements (CPAP or BiPAP) within the sphincter pharyngoplasty cohort increased from two patients before surgery to 19 patients after surgery (1.2% vs 11.4%, respectively; $p < 0.001$). In comparison, the OSA treatment requirement rate in the control group was 1.5%.

After surgery, we compared the pharyngoplasty and control groups in terms of OSA prevalence, age at OSA diagnosis, AHI at diagnosis, and rate of OSA treatment with CPAP/BiPAP. The postoperative pharyngoplasty cohort had significantly greater prevalence of OSA diagnoses (14.5% vs 4.5%; $p = 0.021$), older age at OSA diagnosis (14.6 vs 9.4 years; $p = 0.021$), and greater rate of OSA treatment with CPAP/BiPAP (13.6% vs 1.5%; $p = 0.004$).

Given our clinical suspicion that a history of VPI surgery or combined surgery methods could be associated with greater OSA risk, we separated the sphincter pharyngoplasty cohort into two subgroups: 1) pharyngoplasty only and 2) pharyngoplasty and secondary Furlow, which included those who underwent combined sphincter pharyngoplasty and Furlow palatoplasty as well as those who had a history of secondary Furlow palatoplasty for VPI before sphincter pharyngoplasty.

When comparing OSA data between the pharyngoplasty only and pharyngoplasty and secondary Furlow subgroups, there were no significant differences in the incidence of postoperative OSA, postoperative time to OSA diagnosis, median AHI, or postoperative OSA treatment.

Identifying predictors of obstructive sleep apnea

To evaluate whether sphincter pharyngoplasty was independently associated with OSA, we first performed univariable analyses to identify all potential predictors of OSA. As shown in Table 4, on univariable analysis, OSA was significantly more likely among patients with a diagnosis of iCP and Pierre Robin sequence. Meanwhile, there was a nonsignificant trend toward greater likelihood of OSA among patients with a history of sphincter pharyngoplasty, primary palate repair with Furlow palatoplasty, and Stickler syndrome.

To determine which variables were independently predictive of OSA, all explanatory variables with a p value < 0.07 in univariable analysis were included in multivariable logistic regression (Table 5). The predictor variables included were cleft diagnoses, primary CP repair type, Pierre Robin sequence, and pharyngoplasty surgery.

In multivariable analysis, sphincter pharyngoplasty surgery was significantly associated with higher OSA incidence (model $p < 0.01$; area under the receiver operating characteristic curve=0.76). Patients with a history of pharyngoplasty were four times more likely to exhibit OSA compared to the controls (odds ratio [OR]=4.24; $p = 0.03$). Patients with a history of pharyngoplasty and secondary Furlow for VPI were eight times more likely to exhibit OSA compared to the controls (OR=8.17; $p = 0.01$).

Discussion

In this study, we evaluated the long-term outcomes of sphincter pharyngoplasties, including speech and revision surgeries, and focused on the potential of persistent OSA after surgery. Overall, the mean age at the last follow-up was 19 years, likely at skeletal maturity, and the median postoperative follow-up period was 8.8 years. Sixty-four percent of patients demonstrated improved and sustained speech outcomes after a single pharyngoplasty. One-third of patients who underwent pharyngoplasty required revision surgery, with a median time to primary revision of 3.9 years. Finally, although the overall postoperative OSA rate remained relatively low, multivariable analysis demonstrated that sphincter pharyngoplasty was independently associated with a fourfold increase in persistent OSA. The effect of surgery was compounded after completion of both sphincter pharyngoplasty and secondary Furlow surgery for VPI, which demonstrated a greater than eightfold likelihood of OSA.

Speech outcomes and revision surgery

Sustained correction of VPI was noted in approximately two-thirds of sphincter pharyngoplasty cases. Although these data are consistent with those in the literature supporting the high success rate of sphincter pharyngoplasties,[1,21,23] many existing studies may have overestimated the postoperative outcomes due to short follow-up periods. A recent meta-analysis found an overall speech improvement rate of 78.4%; however, no average follow-up time was reported.[1] The largest study included in the meta-analysis by Losken et al.[2] evaluated 250 patients who underwent sphincter pharyngoplasty and reported a success rate of 87.2% after a mean follow-up of 2.4 years. Our median follow-up period was 8.8 years. Given the existing knowledge on VPI recurrence risk after completion of growth, the lower speech improvement rate of 64% in our study after a greater than threefold increase in follow-up time compared to those in existing studies underscores the need for long-term follow-up after secondary speech surgery.[3,24]

In our analysis, one-third of pharyngoplasty patients required a revision surgery. Our findings are consistent with studies on VPI surgery with longer follow-up times. Makar et al.[24] conducted a national claims database analysis that evaluated multiple types of VPI surgery (including sphincter pharyngoplasty, pharyngeal flap, and palatal lengthening surgeries) and demonstrated that an overall VPI surgery revision rate of 12.7% increased to 21.7% among patients with 3 years of postoperative enrollment. This work and our findings over a median postoperative follow-up period of 8.8 years highlight the risk of recurrent VPI over longer periods.

In our cohorts, primary revisions occurred four years after initial pharyngoplasty. A subset of patients remained competent for multiple years and then required repositioning of their pharyngoplasty ports after a growth spurt or completion of growth. Given that velopharyngeal structures reach peak growth at different rates, including the velum and pharyngeal depth not reaching peak growth until 16-17 years of age,[25,26] we hypothesize that growth in pharyngeal structures contributes to an increased need for revision when following patients until completion of growth and adulthood. Our group has previously highlighted the relationship between higher rates of revision surgeries required and longer-term follow-up among the population with CP, specifically those with alveolar bone regrafting averaging 4.4 years after initial grafting.[27] Our study paralleled those findings, with a median time to primary pharyngoplasty revision of 3.9 years. Thus, studies with follow-up periods under four years may underestimate revision rates.

Persistent obstructive sleep apnea

Although sphincter pharyngoplasty has previously been associated with OSA in retrospective studies,[11,15,17] there is a paucity of literature evaluating the persistence of postoperative OSA. It has been questioned whether OSA after VPI surgery may be transient, hypothesized as being primarily caused by acute postoperative edema of the upper airway that may resolve within days to months.[12,13,28] Therefore, we sought to understand whether there was a relationship between sphincter pharyngoplasty and long-term incidence of OSA. In our total pharyngoplasty cohort, OSA rates increased significantly, from 3% before surgery to 14.5% after surgery. Notably, most cases of OSA fell into the mild category (median AHI=3.4; mild OSA was defined as an AHI of 1-4.9).[19,29,30] These findings can help providers counsel patients on the risk of OSA before surgery.

Our work builds on the limited literature on predictors of postoperative OSA for patients undergoing sphincter pharyngoplasty. Ettinger et al.[17] previously examined OSA after sphincter pharyngoplasty; however, their mean follow-up period was 4.5 years, whereas this study had a median follow-up period of 8.8 years. While previous studies have not distinguished between acute (immediate postoperative) and persistent OSA, this study documents the diagnosis of postoperative OSA occurring within a mean time of 4.4 years after initial pharyngoplasty.

Furthermore, this study includes prior or concurrent secondary Furlow palatoplasty surgery for VPI as a risk factor for OSA (OR=8.17; $p = 0.01$), indicating potentially additive effects of both surgeries on OSA compared to patients who underwent sphincter pharyngoplasty alone. There are limited studies evaluating the outcomes of combined sphincter pharyngoplasty and Furlow palatoplasty.[15,31,32] Our findings contrast with the work by Bohm et al.[32] that compared 38 patients who underwent combined Furlow and sphincter pharyngoplasty, 20 patients who underwent sphincter pharyngoplasty, and 38 patients with pharyngeal flap and found no statistically significant differences in overall complication rates, which included transient and persistent OSA. Notably, their study did not report follow-up time, raising the possibility that differences in persistent OSA may have not been detected with limited follow-up.

Our results may underestimate the incidence of postoperative OSA given some of the limitations of our methodology. Our study identified OSA cases based on polysomnograms, which were ordered following clinical suspicion. Although universal polysomnography may be beneficial in capturing all instances of OSA, limitations in healthcare resources of our economically disadvantaged patient population (> 80% on Medi-Cal) and the burden of care for the patient and family members preclude such a possibility.

Additional limitations of this retrospective study include lack of standardized speech scoring tools and limited availability of data on the type of primary CP repair given that the repairs were performed at outside institutions or in other countries. Furthermore, although this dataset included multiple diagnoses with varying pathologies (i.e., iCP vs unilateral CLP), we controlled for cleft diagnosis in our multivariable regression and did not observe significant differences in postoperative OSA between the diagnoses included.

A strength of our study is in the use of overnight polysomnography as it is the gold standard for diagnosing airway obstruction.[33,34] However, universal delivery of a time-consuming and resource limited diagnostic tool is impractical.[35,36] Beyond routine screening questions, one potential avenue to more accurately diagnose patients who definitively need polysomnography would be to incorporate quantitative, validated patient-reported outcome measures or objective measures using wearable technology that may record similar metrics as polysomnography.[37,38]

Conclusions

In this two-institution retrospective cohort analysis, almost two-thirds of patients demonstrated who underwent pharyngoplasty improved and sustained speech outcomes after a single pharyngoplasty, whereas the remaining one-third required a revision surgery over a median follow-up period of 8.8 years. History of sphincter pharyngoplasty and history of pharyngoplasty/secondary Furlow palatoplasty were significantly associated with OSA. Notably, these cases of OSA were generally long-term in nature, with an average diagnosis in 4.4 years after initial pharyngoplasty. This work highlights the importance of following patients with VPI longitudinally as patients may develop recurrent VPI and/or obstructive sleep symptoms that require intervention.

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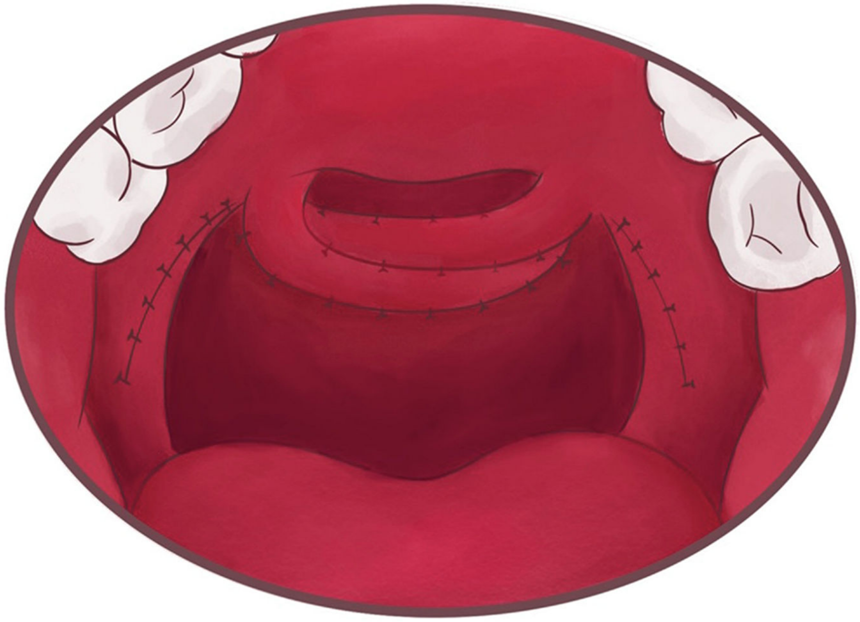

Figure 1.

Sphincter pharyngoplasty. The posterior pillar flaps (including the palatopharyngeus muscle) are rotated 90 degrees and inset into the posterior pharyngeal wall mucosa defect and to each other in a Z pattern.

Table 1

Patient characteristics in pharyngoplasty and control groups.

Characteristic	All Patients	Pharyngoplasty	Controls–No VPI	*p*
No.	233	166	67	
Sex, n (%)				
Female	105 (45.1)	71 (42.8)	34 (50.7)	NS
Male	128 (54.9)	95 (57.2)	33 (49.3)	
Cleft diagnosis, n (%)				
Isolated cleft palate	70 (30.0)	50 (30.1)	20 (29.9)	NS
Unilateral cleft lip and palate	115 (49.4)	83 (50.0)	32 (47.8)	
Bilateral cleft lip and palate	48 (20.6)	33 (19.9)	15 (22.4)	
Genetics, n (%)				
Nonsyndromic	183 (78.5)	129 (77.7)	54 (80.6)	NS
Syndromic	50 (21.5)	37 (22.3)	13 (19.4)	
Stickler syndrome	21 (9.0)	14 (8.4)	7 (10.4)	NS
Velocardiofacial syndrome	10 (4.3)	10 (6.0)	0 (0.0)	NS
Other syndromes	19 (8.2)	13 (7.8)	6 (9.0)	NS
Pierre Robin sequence, n (%)	26 (11.2)	17 (10.2)	9 (13.4)	NS
Insurance status, n (%)				
Public	198 (85.0)	143 (86.1)	55 (82.1)	NS
Private	35 (15.0)	23 (13.9)	12 (17.9)	
Primary palate repair age, median mo (IQR)	12.0 (10.9-18.3)	12.0 (11.0-19.8)	12.0 (10.0-15.3)	NS
Primary palate repair type, n (%)				0.01
Furlow palatoplasty	96 (41.6)	66 (39.8)	31 (46.3)	
Intravelar veloplasty	55 (23.6)	33 (19.9)	22 (32.8)	
Other/Unknown	81 (34.8)	67 (40.4)	14 (20.9)	
History of tonsillectomy/adenoidectomy, n (%)	5 (2.1)	3 (1.8)	2 (3.0)	NS
Mean age at last follow-up ± SD, yr	19.0 ± 2.6	19.2 ± 2.6	18.4 ± 2.8	NS

VPI, velopharyngeal insufficiency; NS, not significant; SD, standard deviation; IQR, interquartile range.

Table 2

Sphincter pharyngoplasty speech and revision surgery outcomes among patients with velopharyngeal insufficiency (n = 166).

Characteristic	n (%)
History of prior secondary Furlow for VPI	9 (5.4)
Sphincter pharyngoplasty method	
Pharyngoplasty only	150 (90.4)
Combined pharyngoplasty and Furlow	16 (9.6)
Median age at initial pharyngoplasty, yr (IQR)	10.3 (7.4-14.2)
Median postoperative follow-up, yr (IQR)	8.8 (3.6-12.0)
Revision surgeries	
None	111 (66.9)
Primary revision	55 (33.1)
Median time to primary revision, yr (IQR)	3.9 (1.9-7.0)
Indication (n = 55)	
Persistent VPI	43 (78.2)
Obstructive symptoms	11 (20.0)
Unknown	1 (1.8)
Secondary revision	12 (7.2)
Mean time to secondary revision ± SD, yr	2.2 ± 1.6
Indication (n = 12)	
Persistent VPI	9 (75.0)
Obstructive symptoms	3 (25.0)
Tertiary revision	3 (1.8)
Mean time to tertiary revision ± SD, yr	5.0 ± 4.4
Indication (n = 3)	
Persistent VPI	3 (100.0)
Obstructive symptoms	0 (0.0)
Speech outcomes *	
Improvement following single pharyngoplasty	106 (63.9)
Improvement following primary revisions	145 (87.3)
Improvement following secondary revisions	151 (91.0)
Improvement following tertiary revisions	152 (91.6)

VPI, velopharyngeal insufficiency; IQR, interquartile range; SD, standard deviation.

*Improvement in speech was defined as normal resonance and voice quality on the most recent speech evaluation after pharyngoplasty.

Table 3

Obstructive sleep apnea and polysomnogram characteristics.

Characteristic	Pharyngoplasty all, n = 166	Pharyngoplasty only, n = 141	Pharyngoplasty and secondary Furlow, n = 25	Controls-No VPI, n = 67
Sleep study ordered, n (%)				6 (9.0)
Preoperative sleep study	3 (1.8)	2 (1.4)	1 (4.0)	-
Postoperative sleep study	37 (22.3)	29 (20.6)	8 (32.0)	-
OSA diagnosis, n (%)				3 (4.5)
Preoperative OSA	5 (3.0)	5 (3.5)	0 (0.0)	-
Postoperative OSA	24 (14.5)	19 (13.5)	5 (20.0)	-
Mean age at OSA diagnosis ± SD, yr	14.6 ± 3.3	14.8 ± 3.6	13.8 ± 1.6	9.4 ± 4.7
Mean postoperative time to OSA diagnosis ± SD, yr	4.4 ± 2.4	4.6 ± 2.5	3.5 ± 2.1	-
Median AHI of OSA cases (IQR)	3.4 (1.8-9.6)	3.1 (1.7-10.9)	5.0 (2.7-32.5)	1.8*
OSA Treatment, n (%)				
CPAP				0 (0.0)
Preoperative CPAP	2 (1.2)	2 (1.4)	0 (0.0)	-
Postoperative CPAP	16 (9.6)	12 (8.5)	4 (16.0)	-
BiPAP				1 (1.5)
Preoperative BiPAP	0 (0.0)	0 (0.0)	0 (0.0)	-
Postoperative BiPAP	3 (1.8)	2 (1.4)	1 (4.0)	-

VPI, velopharyngeal insufficiency; OSA, obstructive sleep apnea; AHI, apnea-hypoxia index; CPAP, continuous positive airway pressure; BiPAP, bilevel positive airway pressure; SD, standard deviation; IQR, interquartile range

*Given that n = 3, there is no IQR for this value

Table 4

Univariable analysis of factors associated with obstructive sleep apnea.

Characteristic	OSA No	OSA Yes	*p*
No.	206	27	
Mean age at last follow-up ± SD, yr	18.8 ± 2.7	19.8 ± 1.9	0.07
Sex, n (%)			0.63
Female	94 (45.6)	11 (40.7)	
Male	112 (54.4)	16 (59.3)	
Insurance, n (%)			0.59
Medicaid	176 (85.4)	22 (81.5)	
Private	30 (14.6)	5 (18.5)	
Diagnosis, n (%)			*0.01*
Isolated cleft palate	55 (26.7)	15 (55.6)	
Unilateral cleft lip and palate	108 (52.4)	7 (25.9)	
Bilateral cleft lip and palate	43 (20.9)	5 (18.5)	
Pierre Robin sequence, n (%)	19 (9.2)	7 (25.9)	*0.01*
Syndromic, n (%)	41 (19.9)	9 (33.3)	0.11
Stickler syndrome	16 (7.8)	5 (18.5)	0.07
Velocardiofacial syndrome	7 (3.4)	3 (11.1)	0.10
Median primary palate repair age (IQR), mo	12.0 (8.1)	12.0 (8.0)	0.96
Primary palate repair type, n (%)			*0.06*
Furlow palatoplasty	80 (38.8)	17 (63.0)	
Intravelar veloplasty	51 (24.8)	4 (14.8)	
Other/Unknown	75 (36.4)	6 (22.2)	
History of tonsillectomy/adenoidectomy, n (%)	3 (1.5)	2 (7.4)	0.10
Pharyngoplasty Surgery Type, n (%)			*0.06*
None	64 (31.1)	3 (11.1)	
Pharyngoplasty only	122 (59.2)	19 (70.4)	
Pharyngoplasty and secondary Furlow	20 (9.7)	5 (18.5)	

OSA, obstructive sleep apnea; SD, standard deviation; IQR, interquartile range.

Table 5

Multivariable predictors of obstructive sleep apnea for all patients.

Variables	OR	SE	95% CI	*p*
Cleft diagnosis				
Isolated cleft palate	1.53	0.64	0.43-5.40	0.51
Unilateral cleft lip and palate	0.60	0.63	0.17-2.06	0.42
Bilateral cleft lip and palate	Ref	Ref	Ref	-
Primary palate repair type				
Furlow palatoplasty	3.10	0.58	1.01-9.56	0.05
Intravelar veloplasty	1.63	0.72	0.40-6.65	0.50
Other/Unknown	Ref	Ref	Ref	-
Pierre Robin sequence	1.92	0.62	0.57-6.41	0.29
Pharyngoplasty Surgery				
None	Ref	Ref	Ref	-
Pharyngoplasty only	4.24	0.67	1.15-15.59	0.03
Pharyngoplasty and secondary Furlow	8.17	0.85	1.54-43.5	0.01

Area under the receiver operating characteristic curve = 0.76 OR, odds ratio; SE, standard error; CI, confidence interval, Ref, Reference value