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Multilevel Sleep Surgery Including the Palate in Nonsyndromic, Neurologically Intact Children with Obstructive Sleep Apnea

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Abstract

Objective. To examine the effects of multilevel sleep surgery, including palate procedures, on obstructive sleep apnea parameters in the pediatric population.

Study Design. A case series with chart review was conducted to identify nonsyndromic, neurologically intact pediatric patients who underwent either uvulectomy or uvulopalatopharyngoplasty as part of multilevel sleep surgery from 2011 through 2017.

Setting. A tertiary care, university children's hospital.

Subjects and Methods. Unpaired Student t test was used to compare average pre- and postsurgical apnea-hypopnea index (AHI) and oxygen saturation nadir (OSN). Paired Student t test was used to compare the mean pre- and postsurgical AHI and OSN within the same patient for the effects of adenotonsillectomy (T&A) vs multilevel sleep surgery.

Results. In patients who underwent T&A previously, multilevel sleep surgery, including palate procedures, resulted in improved OSA severity in 6 (86%) patients and worsened OSA in 1 (14%) patient. Multilevel sleep surgery, including palate procedures, significantly decreased mean AHI from 37.98 events/h preoperatively to 8.91 events/h postoperatively (P = .005). However, it did not significantly decrease OSN.

Conclusion. This study includes one of the largest populations of children in whom palate procedures as a part of multilevel sleep surgery have been performed safely with no major complications and a low rate of velopharyngeal insufficiency. Therefore, palatal surgery as a part of multilevel sleep surgery is not necessarily the pariah that we have traditional thought it is in pediatric otolaryngology.

Obstructive sleep apnea (OSA) is a form of sleep-disordered breathing characterized by intermittent nocturnal upper airway obstruction. In 1988, the successful use of uvulopalatopharyngoplasty (UPPP) in a child was first described.[1] Thereafter, the use of UPPP and uvulectomy has been reported only in either syndromic or neurologically impaired children with OSA.[2-9] We present a small cohort of nonsyndromic and nonneurologically impaired pediatric patients with OSA who underwent either uvulectomy or UPPP. Our primary objective was to demonstrate the effects of multilevel sleep surgery, including palate procedures, on apnea-hypopnea index (AHI) and oxygen saturation nadir (OSN). Our secondary objectives were to highlight the type of multilevel sleep surgeries our

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cohort received, as well as any available drug-induced sleep endoscopy (DISE) data, and compare OSA parameters of multilevel sleep surgery to previously performed adenotonsillectomy (T&A) in children.

Materials and Methods

A case series with chart review was conducted at a single institution, our tertiary care university children's hospital from 2011 through 2017. Thirty-eight nonsyndromic, neurologically intact children with OSA who underwent uvulectomy or UPPP as a part of multilevel sleep surgery were identified at our institution using *Current Procedural Terminology* (*CPT*) codes 42140 and 42145, respectively. The decision to perform a multilevel sleep surgery was determined by either DISE results and/or in-office physical examination performed by the treating physician. Some patients in the cohort did not undergo DISE because it was not commonplace at our institution prior to 2013. Basic demographic information was collected, including patient age, sex, race, body mass index (BMI), medical history (especially of genetic disorders and neurologic conditions), prior surgeries, age at the time of surgery, and relevant information regarding patient outcomes (ie, surgical complications, airway status, continuous positive airway pressure [CPAP] use). Additional testing information was collected, including sleep endoscopy results and pre- and postprocedural polysomnography (PSG) data. PSG was scored in accordance with the American Academy of Sleep Medicine guidelines.[10] Institutional review board approval was obtained at Drexel University College of Medicine.

Patients with a genetic syndrome, with a neurologic impairment, or without PSG results were excluded. However, the only syndrome we encountered was Down syndrome. Unpaired Student *t* test was used to compare average pre- and postsurgical AHI and OSN. Paired Student *t* test was used to compare the mean pre- and postsurgical AHI and OSN within the same patient for the effects of T&A vs multilevel sleep surgery, including palate procedures. Two-factor to multifactor linear repeated mixed models assessed changes in pre- and postoperative outcomes over time after controlling for sex, BMI, and age. An unstructured variance-covariance structure was imposed in the statistical models. The 95% confidence intervals (CIs) for the difference around the means were calculated. Statistical significance was set at a *P* value less than or equal to .05 with 1- and 2-tailed hypothesis testing. Statistical analysis software (IBM *SPSS*, Cary, NC) was used for analysis.

Results

Out of the total population (n = 38), 26 patients were excluded, resulting in a final cohort of 12 patients. Twenty-four children were excluded for missing PSG data, of whom 2 had hydrocephalus with neurologic impairment. An additional 2 were excluded for having Down syndrome (**Figure 1**). Demographic information about our cohort is illustrated in **Table 1**.
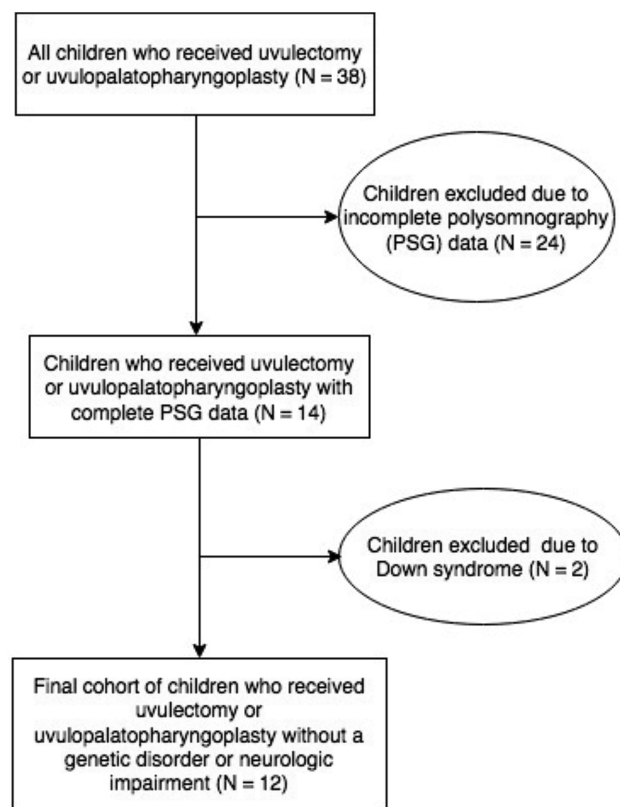
Figure 1. Study patient selection.

Table 1. Patient Demographic and Characteristics (n = 12).

Characteristic	Value[a]
Age, mean (SD), y	6.92 (4.32)
Sex	
Male	9 (75)
Female	3 (25)
Race	
African American	8 (67)
Hispanic/Latino	4 (33)
Average BMI, kg/m^2	22.75
Obese (>95th percentile for age)	6 (50)
Palatal surgery type	
UPPP/combination surgery	7 (58.3)
Uvulectomy/combination surgery	4 (33.3)
UPPP alone	1 (8.3)

Abbreviations: BMI, body mass index; UPPP, uvulopalatopharyngoplasty.
[a]Results are reported as number (%) unless otherwise specified.

Palate surgery was combined with a variety of other procedures, including adenoidectomy (primary or revision), tonsillectomy (primary or revision after intracapsular tonsillectomy), and inferior turbinate reduction (ITR). Overall, 6 (50%) patients had improved OSA severity while only 1 (8%) patient worsened after multilevel sleep surgery, including palate procedures (**Table 2**). Sleep endoscopy was

Table 2. Obstructive Sleep Apnea Parameters before and after Combination Palatal Surgery (n = 12).

Patient No.	Age, y/Sex	Procedure	Preoperative			Postoperative		
			AHI	OSN	Severity	AHI	OSN	Severity
1	4/F	Uvulectomy + revision T&A	78.3	71	Severe	5.8	87	Moderate
2	8/M	Uvulectomy + revision A	51.5	80	Severe	17.5	85	Severe
3	5/M	Uvulectomy + revision A + ITR	15.8	68	Severe	6.6	72	Severe
4	5/M	Uvulectomy + revision A	8.9	90	Moderate	10.9	69	Severe
5	3/M	UPPP + revision A	2.9	92	Mild	0.9	91	Normal
6	13/M	UPPP + T + revision A	26.4	86	Severe	0.7	94	Normal
7	6/M	UPPP + A	4.3	87	Mild	2.6	90	Mild
8	15/F	UPPP + T&A + ITR	16.7	75	Severe	2.5	88	Mild
9	4/M	UPPP + revision T&A	15.1	77	Severe	1.2	80	Moderate
10	3/M	UPPP	36.1	61	Severe	5.7	85	Moderate
11	13/M	UPPP + T&A	101.8	60	Severe	28.9	85	Severe
12	4/F	UPPP + T&A	98	65	Severe	23.6	72	Severe

Abbreviations: A, adenoidectomy; AHI, apnea-hypopnea index; F, female; ITR, inferior turbinate reduction; M, male; OSN, oxygen saturation nadir; T, tonsillectomy; T&A, adenotonsillectomy; UPPP, uvulopalatopharyngoplasty.

Table 3. Sleep Endoscopy Results (n = 5).

Patient No.	Age, y/Sex	Sleep Endoscopy Findings	Procedure
3	5/M	Oropharyngeal obstruction from uvular hypertrophy, nasopharyngeal obstruction from adenoid and inferior turbinate hypertrophy	Uvulectomy + revision A + ITR
4	5/M	Uvular and adenoid hypertrophy causing oropharyngeal and nasopharyngeal obstruction, respectively	Uvulectomy + revision A
5	3/M	Uvular and soft palate redundancy causing oropharyngeal obstruction, adenoid hypertrophy causing nasopharyngeal obstruction	UPPP + revision A
9	4/M	Uvular and soft palate redundancy causing oropharyngeal obstruction	UPPP + revision T&A
10	3/M	Uvular and soft palate redundancy causing oropharyngeal obstruction	UPPP

Abbreviations: A, adenoidectomy; F, female; ITR, inferior turbinate reduction; M, male; T&A, adenotonsillectomy; UPPP, uvulopalatopharyngoplasty.

performed on 5 patients immediately prior to palate surgery. Results included nasopharyngeal obstruction from the posterior inferior turbinate and adenoid hypertrophy as well as oropharyngeal obstruction from uvular and soft palate redundancy (**Table 3**).

To further evaluate the effects of palate surgery, OSA severity was compared in each patient between his or her prior T&A and subsequent multilevel sleep surgery, including palate procedures (**Table 4**). Five patients who underwent multilevel sleep surgery did not undergo previous surgery; therefore, they were not included in this comparison. There were 7 patients who underwent previous T&A and subsequent multilevel sleep surgery. Prior to T&A, 2 (28.5%) children had mild OSA, 2 (28.5%) had moderate OSA, and 3 (43%) had severe OSA. After T&A, 1 (14.3%) child had mild OSA, 2 (28.6%) had moderate OSA, and 4 (57.1%) had severe OSA. Prior to multilevel sleep surgery, 2 (28.6%) children had mild OSA, 1 (14.3%) had moderate OSA, and 4 (57.1%) had severe OSA. After multilevel sleep surgery, 1 (14.3%) child was cured of OSA, 1 (14.3%) had mild OSA, 3 (42.9%) had moderate OSA, and 2 (28.5%)

had severe OSA. Overall, T&A resulted in worsening OSA in 3 (43%) patients and improved OSA in 4 (57%) patients. Multilevel sleep surgery, including palate procedures, resulted in worsening OSA in 1 (14%) patient and improved OSA in 6 (86%) patients.

After adjusting for sex, age, and BMI, multilevel sleep surgery, including palate procedures, significantly decreased mean AHI from 37.98 events/h preoperatively to 8.91 events/h postoperatively ($t = 3.48$, $P = .005$). However, it did not significantly improve mean OSN postoperatively. In children who underwent prior T&A, multilevel sleep surgery did not significantly improve mean AHI or OSN compared to T&A alone after adjusting for age, sex, and BMI percentile (**Table 5**).

Zero patients from the final 12-patient cohort experienced acute, postsurgical complications. Of the original 38 patients, only 1 patient (2.6%) sustained a complication, which was velopharyngeal insufficiency. This patient was evaluated and treated by our speech and language pathology department and did not require surgical intervention. Six patients (50%) indicated better postoperative CPAP

Table 4. Effect of Adenotonsillectomy vs Combination Palatal Surgery on Obstructive Sleep Apnea (n = 7).[a]

		Adenotonsillectomy					Combination Palatal Surgery			
Patient No.	Age, y	Preseverity	ΔAHI	ΔOSN	Postseverity	Age, y	Preseverity	ΔAHI	ΔOSN	Postseverity
1	1.8	Mild	−74	−13	Severe	4	Severe	72.5	16	Moderate
3	5	Severe	17.8	8	Severe	5	Severe	9.2	4	Severe
4	4	Mild	−6.3	−5	Moderate	5	Moderate	−2	−21	Severe
5	2	Moderate	1.9	7	Mild	3	Mild	2	−1	Normal
7	1.9	Severe	10.7	7	Moderate	6	Mild	1.7	3	Mild
9	3	Moderate	−5.6	0	Severe	4	Severe	13.9	2	Moderate
10	2	Severe	28.3	−9	Severe	3	Severe	30.4	24	Moderate

Abbreviations: AHI, apnea-hypopnea index; OSN, oxygen saturation nadir.
[a]Negative values represent lack of improvement in a parameter: an increase in AHI and a decrease in OSN.

Table 5. Overall Surgical Improvements in Obstructive Sleep Apnea Parameters.[a]

Comparison (A vs B)	Sample Size	Mean A	Mean B	DM	t Value	95% CI	P Value
AHI pre- vs post-multilevel U/UPPP	12	37.98	8.91	29.07	3.48	10.71 to 47.44	.005[b]
OSN pre- vs post-multilevel U/UPPP	12	76.00	83.17	−7.17	−2.04	−14.89 to 0.55	.066
ΔAHI post-T&A vs post-multilevel U/UPPP	7	−3.89	18.24	−22.13	−1.05	−73.61 to 29.35	.333
ΔOSN post-T&A vs post-multilevel U/UPPP	7	−0.71	3.86	−4.57	−0.64	−22.04 to 12.89	.546

Abbreviations: AHI, apnea-hypopnea index; CI, confidence interval; DM, difference of means; OSN, oxygen saturation nadir; T, tonsillectomy; T&A, adenotonsillectomy; U, uvulectomy; UPPP, uvulopalatopharyngoplasty.
[a]Negative values represent lack of improvement in a parameter: an increase in AHI and a decrease in OSN.
[b]Denotes statistical significance ($P \leq .05$) from unpaired and paired t testing.

compliance while 1 patient was initiated on CPAP for residual OSA. Therefore, we feel that multilevel sleep surgery that includes palate procedures can be performed safely in the pediatric OSA population.

Discussion

There are limited data for the use of palate surgery in pediatric OSA. In 1 study by Wiet et al,[2] UPPP with tonsillectomy or T&A was performed on 3 and 13 patients, respectively. However, all of these patients had comorbidities, including obesity, Down syndrome, asthma, and cerebral palsy (CP). Although there were promising results, T&A alone resulted in a better improved AHI. Kosko and Derkay[3] studied 15 patients with neurologic impairment; 12 showed significant improvement after combination T&A with UPPP. However, AHI was recorded in only 2 patients. Another study by Kerschner et al[4] showed that T&A with UPPP improved OSN from 65% preoperatively to 85% postoperatively in neurologically impaired children. Shott and Cunningham[8] demonstrated that uvulectomy subjectively improved apnea in 4 pediatric patients. Another study involving children with CP demonstrated that some patients had improved OSA with a combination of tonsillectomy and/or adenoidectomy with uvulectomy. UPPP was performed on a few patients with limited success. They concluded that uvulectomy and UPPP are options for patients

who fail T&A. However, objective measures of improvement (ie, AHI) were not used.[9]

Our study demonstrated that palate surgery as a part of multilevel sleep surgery in children produced an over 4-fold improvement in mean AHI. Palate surgery as a part of multilevel sleep surgery also improved mean OSN, but it was unclear to which degree. In the main cohort of 12 patients, mean OSN did not significantly improve. Palate surgery as a part of multilevel sleep surgery also improved OSA severity in 50% of the 12-patient cohort. In patients in whom the site of obstruction is unclear, sleep endoscopy may help direct surgical management. Sleep endoscopy allows otolaryngologists to identify particular anatomical levels of collapse, aiding with the diagnosis of OSA.[11] The main areas to examine are the nose and nasopharynx, the posterior oropharynx, the lateral pharyngeal walls, the hypopharynx (primarily the base of tongue), and the supraglottic larynx.[12] In our cohort, sleep endoscopy assisted with identifying the level of obstruction in 5 patients. In our opinion, palate procedures as a part of multilevel sleep surgery are underused in the pediatric population. Our study demonstrates that palate procedures play a role in the management of pediatric OSA, particularly in patients with residual OSA after other sleep procedures. More important, the addition of palate surgery as a part of multilevel sleep surgery has shown to be a safe option in our cohort. Zero patients experienced acute surgical complications and only 1 patient developed

velopharyngeal insufficiency (VPI), which resolved with speech and swallowing therapy.

The results of this study should be interpreted within the context of certain limitations. Although the results are in a small sample size, there have been no previous efforts to identify outcomes in children with isolated OSA. To our knowledge, this is the largest pediatric OSA cohort treated with palate surgery evaluated with objective measures. However, a major limitation in our study was our preoperative evaluation of the patients. Prior to 2013, our institution did not use DISE. Therefore, surgical planning was determined by relatively subjective physical examination findings, including soft palate and uvular redundancy, adenoid hypertrophy, and posterior inferior turbinate hypertrophy. Another limitation was our inability to measure the individual success of palate surgery. All of the patients with the exception of 1 underwent additional sleep procedures. In addition, no patient-centered quality-of-life measures were assessed. There were not enough patients to compare uvulectomy (n = 4) and UPPP (n = 8) head-to-head. The patients were studied over a time interval, allowing for dynamic factors to contribute to worsening OSA severity. Therefore, obesity and advanced age can potentially serve as a confounding variable; however, multivariate analysis did not suggest this. It should be mentioned that selection bias was present because children without improvement after T&A were chosen for multilevel sleep surgery, including palate procedures. Finally, this cohort contained only children of Hispanic and African American descent. Therefore, the results of this study may not be translatable to other populations with different socioeconomics.

In conclusion, there is limited information about the use of palate surgery as a part of multilevel sleep surgery in children with OSA. This study includes one of the largest populations of children in whom palatal surgery as a part of multilevel sleep surgery has been performed safely with no major complications and a low rate of velopharyngeal insufficiency. Therefore, palatal surgery as a part of multilevel sleep surgery is not necessarily the pariah that we have traditionally thought it is in pediatric otolaryngology.

Author Contributions

Jason E. Cohn, substantial contributions to the conception and design of the work; the acquisition, analysis, and interpretation of data for the work; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **George E. Relyea**, acquisition, analysis, and interpretation of data for the work; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Srihari Daggumati**, substantial contributions to the conception and design of the work; the acquisition, analysis, and interpretation of data for the work; drafting the work and revising it critically for

important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved; **Brian J. McKinnon**, substantial contributions to the conception and design of the work; drafting the work and revising it critically for important intellectual content; final approval of the version to be published; agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Disclosures

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