Objective: To evaluate the effect of body mass index (BMI, calculated as weight in kilograms divided by height in meters squared) and medical comorbidities on outcomes after lingual tonsillectomy and supraglottoplasty performed for obstructive sleep apnea syndrome (OSAS) caused by lingual tonsillar hypertrophy and occult laryngomalacia.

Design: Retrospective case review series

Setting: Academic tertiary referral center

Patients: Children with persistent OSAS after adenotonsillectomy who underwent surgery to correct obstruction at the level of the lingual tonsils and/or supraglottis identified on sleep endoscopy.

Interventions: All children underwent lingual tonsillectomy, supraglottoplasty, or both.

Main Outcome Measures: Change in polysomnographic parameters, including apnea-hypopnea index (AHI), number of nighttime apneas, and lowest oxygen saturation level.

Results: We analyzed the medical records of 84 children with persistent OSAS after adenotonsillectomy who underwent either lingual tonsillectomy (n=68), supraglottoplasty (n=24) or both (n=8). Compared with children with lingual tonsillar hypertrophy, children with occult laryngomalacia were younger, had lower BMI, and were more likely to have a medical comorbidity. Overall, both operations significantly improved the AHI; however, children with comorbidities had significantly higher postoperative AHIs after supraglottoplasty than those without, and overweight children had significantly higher postoperative AHIs after lingual tonsillectomy than those of normal weight. The BMI z-score and age had direct, though weak, correlations with postoperative AHI among all children undergoing either technique of adjunct airway surgery.

Conclusions: Lingual tonsillar hypertrophy and occult laryngomalacia are 2 important causes of residual OSAS after adenotonsillectomy. However, they tend to affect distinct populations of children, and though appropriate surgical correction can improve AHI, cure rates are significantly worse for overweight children undergoing lingual tonsillectomy and for children with medical comorbidities undergoing supraglottoplasty.


Adenotonsillectomy for obstructive sleep apnea syndrome (OSAS) is among the most common operations performed in children in the United States. The most widely used objective measure of the severity of OSAS is the apnea-hypopnea index (AHI). Though greater than 90% of children undergoing adenotonsillectomy have improvement in their AHI, up to 75% have at least mild OSAS even after surgery. Advanced age, asthma, and especially obesity have been shown to be poor prognostic factors for outcomes after pediatric adenotonsillectomy. Likewise, medical comorbidities, particularly those associated with hypotonia and neuromuscular dysfunction, have been implicated in adenotonsillectomy failures.

Children who have persistent OSAS after adenotonsillectomy often undergo second-stage procedures to further improve their sleep-disordered breathing. In previous studies, we and others have examined the efficacy of 2 procedures—supraglottoplasty and lingual tonsillectomy—in correcting residual OSAS after adenotonsillectomy. Although AHI is decreased in almost all children undergoing these adjunct procedures, a significant proportion still has OSAS even after this second stage of surgery. In the present study, we sought to determine the patient factors that contribute most significantly to the failure of

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adjunct surgical procedures to cure OSAS. In particular, we have focused on the effect of obesity and medical comorbidities—2 of the most significant contributors to failure in primary adenotonsillectomy—on polysomnographic outcomes after either lingual tonsillectomy performed for lingual tonsillar hypertrophy or supraglottoplasty performed for occult laryngomalacia.

### METHODS

#### PATIENT SELECTION

This study was approved by the institutional review board of the Lucile Packard Children's Hospital and was conducted in compliance with the Healthcare Information Portability and Accountability Act. Children who underwent either lingual tonsillectomy or supraglottoplasty by attending pediatric otolaryngologists at the Lucile Packard Children's Hospital from 2005 through 2011 were identified by Current Procedural Terminology (CPT) code search of the electronic medical record for either lingual tonsillectomy (42870) or supraglottoplasty (microdirect laryngoscopy [31520] with epiglottidectomy [31420] or excision [31541]). Medical charts of patients thus identified were examined. Inclusion criteria were as follows:

1. Documentation of either laryngomalacia or lingual tonsillar hypertrophy on flexible fiberoptic laryngoscopy performed either in clinic with patient awake, or in the operating room with patient under anesthesia;
2. Subsequent performance of either supraglottoplasty or lingual tonsillectomy or both;
3. Patient age between 2 and 18 years;
4. Presence of pretreatment polysomnogram indicating OSAS; and
5. Presence of a posttreatment polysomnogram.

These criteria yielded a patient population with OSAS attributable to either laryngomalacia or lingual tonsillar hypertrophy or both. Most patients had undergone adenotonsillectomy prior to the preoperative polysomnogram (86%), and either supraglottoplasty or lingual tonsillectomy reflected the only surgical intervention between the preoperative and postoperative polysomnograms. In a subset of patients (14%), additional surgical interventions were performed in a staged manner to address multiple levels of obstruction without intervening polysomnographic evaluation. Formal assessment of OSAS with polysomnography was only undertaken before and after this set of staged procedures. In all of these cases, however, sleep apnea was clinically persistent after each stage. Prior analysis of a similar patient population showed no significant difference in outcomes when these 2 cohorts were compared; therefore, they were pooled for purposes of the present analysis.

Obesity and medical comorbidities were assessed for each patient in the following manner. Body mass index (BMI, calculated as weight in kilograms divided by height in meters squared) was calculated for each patient, and each was categorized as either overweight (>85th percentile BMI for age) or not. For comparisons of BMI between individuals and groups, BMI z-scores were calculated using the Centers for Disease Control and Prevention 2000 growth standards (www.cdc.gov/growthcharts). Thorough medical chart review, with particular attention to multidisciplinary evaluation by pediatric neurologists, pulmonologists, cardiologists, and medical geneticists, revealed whether significant medical comorbidities were involved. Children were included in the medical comorbidity group if one or more of the following terms was included in the diagnosis: “hypotonia,” “Down syndrome,” “neuromuscular dysfunction,” “cerebral palsy,” or “severe or moderate persistent asthma.” These inclusion terms were used to select specifically for children with syndromic and nonsyndromic neuromuscular and respiratory dysfunction, which have been shown previously to portend worse prognoses after primary adenotonsillectomy. Isolated comorbidities with minimal effect on these systems were excluded, such as pervasive developmental delay, mild asthma, diabetes, isolated cardiac anomalies, and seizure disorders.

#### SLEEP ENDOSCOPY

Children who present to our practice with persistent OSAS after adenotonsillectomy. OSAS with small tonsils (0 or 1+), or OSAS complicated by other medical comorbidities are routinely subjected to flexible fiberoptic drug-induced sleep endoscopy in the operating room. Levels of anatomic obstruction are noted, and in cases of lingual tonsillar hypertrophy or laryngomalacia, lingual tonsillectomy or carbon-dioxide laser supraglottoplasty are performed, respectively, as described previously. In cases where obstruction at both sites are noted synchronously, the site subjectively judged to be more severe is addressed first. If the patient does not exhibit significant clinical improvement, the second procedure is then performed in a staged manner.

#### POLYSOMNOGRAPHY

Apnea-hypopnea indices and minimum oxygen saturations were obtained from the primary reports of polysomnograms obtained at 2 centers: the Stanford Sleep Disorders Clinic and the Lucile Packard Children's Hospital Sleep Laboratory. Obstructive sleep apnea syndrome is categorized as absent (AHI, <1), mild (AHI, 1-5), moderate (AHI, 5-10), or severe (AHI, >10). The minimum oxygen desaturation during an obstructive event is reported as the oxygen nadir. When possible, all polysomnograms for an individual patient were obtained from a single center; in 22 of 92 cases (24%), studies from the 2 different centers were compared. As seen in our group's prior study, subgroup analysis of children who had polysomnograms at the same institution throughout their treatment yielded statistically indistinguishable results from the pooled group (data not shown).

#### STATISTICAL ANALYSIS

Comparisons of paired nonparametric data, including comparisons of preoperative and postoperative AHIs and minimum oxygen saturations, were performed with the Wilcoxon rank-sum test. Nonpaired nonparametric data were compared with the Mann-Whitney rank-sum test. Parametric data were compared with the t test. Binomial data were compared with the Fisher exact test. Statistical significance was set at P<.05.

#### RESULTS

#### PATIENT POPULATION

A list of demographic characteristics of the patient population is given in the Table. Twenty-four children underwent supraglottoplasty, and 68 underwent lingual tonsillectomy, for a total of 92 procedures. Eight children underwent both, for a total of 84 children. There was an overall male preponderance in both groups. Children undergoing lingual tonsillectomy were significantly older and had significantly higher BMI z-scores than those who...
underwent supraglottoplasty, although the percentage of children classified as overweight was not significantly different. A higher percentage of children in the supraglottoplasty group had medical comorbidities. Overall, 23 children had neuromuscular dysfunction, including 5 with cerebral palsy; 13 were syndromic, including 5 with Down syndrome; and 6 had primary respiratory disease. These comorbidities were not significantly different between the lingual tonsillectomy and supraglottoplasty groups. Specific disease entities carried by children in the comorbidity group included the following: chromosomal abnormalities (trisomy 21, trisomy 15q), Beckwith-Wiedemann syndrome, Noonan syndrome, Opitz syndrome, fetal hydantoin syndrome, Prader-Willi syndrome, Sanfilippo type II, lissencephaly, dermatomyositis with respiratory muscle weakness, Duchenne muscular dystrophy, and severe asthma.

Polysomnography was performed before and after surgery in all patients. No significant differences in preoperative or postoperative AHI, number of obstructive apneas, or minimum oxygen saturation were found between the lingual tonsillectomy and supraglottoplasty groups (Table). Consistent with our prior findings, both lingual tonsillectomy and supraglottoplasty resulted in significant improvement in AHI and number of nighttime obstructive apneas, but not in minimum oxygen saturation. The large majority of patients in both groups, approximately 90%, had improvement in AHI after surgery. Overall, 53 of 92 procedures performed among the 84 children (58%) resulted in postoperative AHI lower than 5, indicating either absent or mild residual OSAS. This percentage was not significantly different among children undergoing lingual tonsillectomy (n = 68; 57%) or supraglottoplasty (n = 24; 58%).

**MEDICAL COMORBIDITIES**

Medical comorbidities known to affect outcomes after adenotonsillectomy were not found to be associated with significantly different preoperative or postoperative AHI outcomes among children undergoing lingual tonsillectomy for OSAS. In this group, children with medical comorbidities had similar outcomes to those without, but that after SGP, children with medical comorbidities had significantly worse outcomes than those without.* P < .01. B, Distribution of obstructive sleep apnea (OSA) severity in children with (+) or without (-) medical comorbidities.
On the other hand, though preoperative AHI was also similar among children undergoing supraglottoplasty (16.1 with comorbidities vs 13.8 without), postoperative AHI was significantly higher in those with medical comorbidities (7.3) compared with those without (2.6). Likewise, OSAS tended to be more severe for those children with comorbidities compared with their noncomorbid counterparts after supraglottoplasty but not after lingual tonsillectomy (Figure 1). Within this group, children with comorbidities had many more apneas than those without comorbidities both before (22.5 vs 6.9) and after (3.8 vs 0.7) surgery, but this difference was not statistically significant. Overall, these data suggest that medical comorbidities are a risk factor for poor outcomes after supraglottoplasty but not after lingual tonsillectomy.

**BODY MASS INDEX**

Outcomes were examined after lingual tonsillectomy and supraglottoplasty in both overweight and nonoverweight children. Overweight children undergoing lingual tonsillectomy had significantly higher preoperative and postoperative AHIs (15.4 and 8.6, respectively) compared with nonoverweight children (9.9 and 4.2) (Figure 2A). This was also true for the number of apneas: overweight children had more apneas before (15.3) and after (3.6) lingual tonsillectomy than their nonoverweight counterparts (7.0 and 1.2, respectively). Although a quantitatively similar difference in postoperative AHI between overweight and nonoverweight children was seen after supraglottoplasty (7.2 vs 4.0, respectively), this difference was not statistically significant, perhaps owing to the 3-fold smaller sample size of this group.

Furthermore, after both supraglottoplasty and lingual tonsillectomy, overweight children tended to have a more severe distribution of OSAS (Figure 2B). In addition, while nonoverweight children undergoing supraglottoplasty had an overall decrease in the number of nighttime apneas, from 18.4 to 0.6, overweight children undergoing supraglottoplasty had an overall increase, from 2.9 to 6.1. These results suggest that higher BMI is a risk factor for poorer outcomes after lingual tonsillectomy, and probably for supraglottoplasty as well. Overall, postoperative AHI had a weak but positive correlation with BMI z-score; children with higher BMI z-scores tended to have higher postoperative AHIs. A similar correlation was noted between age and postoperative AHI (Figure 3).

**COMMENT**

Adenotonsillectomy is an effective primary treatment for OSAS and has been shown to improve quality of life, behavior, and polysomnographic indices of sleep-disordered breathing.11,12 A significant number of children, however, have either minimal improvement or persistent OSAS after adenotonsillectomy.1 Drug-induced sleep endoscopy can reveal additional sites of anatomic obstruction,13 and our group and others have previously described methods of surgical correction of obstruction at the level of the supraglottis and lingual tonsils.6–9
Previous findings of improvement in AHI after supraglottoplasty and lingual tonsillectomy are confirmed herein. Overall, results are comparable with either intervention, and these outcomes are similar to those seen with primary adenotonsillectomy. Some differences, however, are notable between the preoperative characteristics of the 2 surgical groups. Children undergoing lingual tonsillectomy were generally older and more likely to be obese; these findings suggest that these children are more akin to their adult counterparts, in whom obesity is a significant contributor to tongue-base obstruction. In contrast, children undergoing supraglottoplasty were younger and more likely to have medical comorbidities. Laryngomalacia, the pathophysiologic phenomenon underlying obstructed breathing in these children, is most often seen in infancy. Infants with congenital laryngomalacia are known to have worsened outcomes in the presence of medical comorbidities. The children in this study, however, do not have typical infantile laryngomalacia; rather, they presented later in childhood with occult laryngomalacia that manifested as OSAS that was only revealed to be attributable to dynamic supraglottic collapse on evaluation by sleep endoscopy. Though this population is distinct from the cohort of infants with congenital laryngomalacia, the overall younger age of the children in the supraglottoplasty intervention group compared with the lingual tonsillectomy group in this study is consistent with the notion that laryngomalacia is a disorder of the immature larynx. Furthermore, the high percentage of children in this group with a medical comorbidity characterized by generalized hypotonia supports the notion that the underlying pathophysiologic presentation of laryngomalacia is a deficiency in supraglottic tone.

Obesity is more common among children undergoing lingual tonsillectomy, and medical comorbidities are more common among those undergoing supraglottoplasty. Our results suggest that these combinations portend worsened outcomes. After lingual tonsillectomy, the AHI of overweight children is twice as high as the AHI of children of normal weight, and children with medical comorbidities have almost 3-fold higher AHI after supraglottoplasty than their noncomorbid counterparts. In contrast, BMI did not significantly affect outcomes after supraglottoplasty, nor did the presence of a medical comorbidity indicate poor prognosis after lingual tonsillectomy. These findings illustrate the limitations of these surgical interventions. Hypotonia attributable to medical comorbidities is associated with clinically apparent laryngomalacia that manifests as obstructive sleep apnea; supraglottoplasty can improve this, but the underlying hypotonia renders intervention less effective. Likewise, obesity exacerbates the obstruction attributed to lingual tonsillar hypertrophy (and perhaps makes it more apparent on preoperative endoscopy) and also contributes to persistent sleep apnea even after removal of the lingual tonsillar tissue.

Limitations of our study are its retrospective nature, and, accordingly, the nonuniform treatment in the 2 groups. In a minority of cases, interventions were staged without intervening polysomnography, so the precise attribution of outcome to isolated procedures cannot be made. The patient population may be subject to selection bias; because it is a mixture of tertiary referrals and children seen originally in our clinic as primary referrals prior to adenotonsillectomy, it may not be reflective of all practice situations. The apnea analysis, though corroborative of the analysis done based solely on AHI, is limited by the large number of sleep studies (both preoperative and postoperative) with no apneas noted. Finally, although the presence of medical comorbidities pertaining to generalized hypotonia and neuromuscular dysfunction was identified in each case by thorough multidisciplinary evaluation, the specific relationship of the comorbid conditions to OSAS in general, and to the specific anatomic abnormalities addressed surgically, was not fully developed. Although evidence has been presented that particular conditions—including Down syndrome, Prader-Willi syndrome, and cerebral palsy, all of which were represented in this study—put children at higher risk for obstructive sleep apnea, the specific pathophysiologic mechanisms have not yet been identified, nor have improved methods of addressing it beyond standard interventions.

Obstructive sleep apnea syndrome in children is primarily treated surgically with a high success rate. Understanding the patient populations where both primary and secondary surgical interventions tend to fail—in this case, children who are overweight and/or have medical comorbidities—is crucial for the development of effective treatment strategies and counseling of these at-risk patients and their parents. Identification of patient factors that contribute to poor outcomes also highlights the need for interdisciplinary collaboration to correct underlying systemic abnormalities to most effectively treat children with refractory sleep apnea.

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REFERENCES


