Supraglottoplasty for Occult Laryngomalacia to Improve Obstructive Sleep Apnea Syndrome

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Objective: To evaluate the polysomnographic outcomes after supraglottoplasty (SGP) performed for obstructive sleep apnea syndrome (OSAS) associated with occult laryngomalacia.

Design: Retrospective case series with medical chart review.

Setting: Tertiary pediatric medical center.

Patients: Twenty-two patients aged 2 to 17 years met the inclusion criteria of polysomnography-proven OSAS and occult laryngomalacia seen on flexible fiber-optic sleep endoscopy. Infants with congenital laryngomalacia were excluded.

Intervention: Carbon dioxide laser SGP was performed either alone or in conjunction with other operations for OSAS.

Main Outcome Measure: Preoperative and postoperative nocturnal polysomnographic data were paired and analyzed statistically.

Results: Supraglottoplasty for occult laryngomalacia resulted in statistically significant reduction in the apnea-hypopnea index (AHI) (from 15.4 to 5.4) ($P<.001$). Subgroup analysis of children who underwent either SGP alone or in combination with other interventions showed comparable reductions in AHI. Medical comorbidities were associated with worsened postoperative outcomes, although still significantly improved compared with baseline. Overall, 91% of children had an improvement in AHI, and 64% had only mild or no residual OSAS after SGP.

Conclusion: Supraglottoplasty is an effective technique for the treatment of OSAS associated with occult laryngomalacia.

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O BSTRUCTIVE SLEEP APNEA syndrome (OSAS), a constellation of findings including snoring, daytime behavioral disturbances, and polysomnographic evidence of airway obstruction at night, is among the most prevalent health problems in the pediatric population, affecting 1% to 4% of all children in the United States.[1] Adenotonsillectomy is considered the primary treatment for obstructive sleep apnea (OSA) in children; it reliably improves both subjective behavior and quality of life,[2,3] as well as objective polysomnographic measures of airway obstruction, particularly the apnea-hypopnea index (AHI).[4] Nevertheless, up to 75% of children undergoing adenotonsillectomy have some degree of residual sleep-disordered breathing after adenotonsillectomy.[5] A number of comorbidities, including obesity, asthma, and preoperative severe OSAS, are known to increase the chance of incomplete resolution of OSAS after adenotonsillectomy. Many of these children will have additional, surgically correctible, anatomical causes of OSAS.

Sleep endoscopy, the use of flexible fiber-optic laryngoscopy under sedation in the operating room with the patient spontaneously ventilating, is useful in revealing the underlying anatomic pathophysiologic mechanisms of OSAS. In particular, it can be used to identify potential surgical targets in cases refractory to initial intervention. Hypertrophy of the lingual tonsils is one such example; reduction in their size is associated with objective improvements in OSAS by polysomnography.[1]

Laryngomalacia, the most common form of airway distress in infancy,[6] is characterized by prolapse of supraglottic structures into the glottic airway on inspiration, leading to inspiratory stridor, feeding difficulties, and failure to thrive. In its congenital form, laryngomalacia can be associated with OSA; supraglottoplasty performed to address laryngomalacia has been shown to significantly improve mean AHI in infants.[7,8] In older children with OSAS, laryngomalacia has also been recognized as a cause of airway obstruction, with a reported incidence among children with OSAS of 3.9%.[9] Different symptomatic patterns have been described, with laryngomalacia associated with feeding, sleep, and exercise.[10] Some, but not all, patients have stridor among their presenting symptoms; thus, we have termed this late-onset form "occult laryngomalacia." Supraglottoplasty in these patients has been suggested to improve OSAS[10,11]; however, objective improvement in the cohort

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of patients with sleep-disordered breathing has not been demonstrated by polysomnography.

In this study, a group of 22 children with PSG-proven OSAS was identified by sleep endoscopy to have occult laryngomalacia subsequently treated with supraglottoplasty. All patients underwent preoperative and postoperative polysomnograms, which objectively demonstrate the efficacy of this procedure. These findings suggest that laryngomalacia should be considered together with hypertrophy of the turbinates, adenoids, palatine tonsil, and lingual tonsils in the constellation of anatomic, surgically correctable causes of OSAS in children.

METHODS

PATIENT SELECTION

This study was approved by the institutional review board of the Lucile Packard Children's Hospital (LPCH) (Palo Alto, California) and was conducted in compliance with the Healthcare Information Portability and Accountability Act. All patients who had undergone supraglottoplasty performed by the senior author (P.J.K.) over a 6-year period (2005-2011) at LPCH were identified by CPT code search of the electronic medical record for microdirect laryngoscopy (31 526) with epiglottidectomy (31 420) or excision (31 541). Individual medical charts thus identified were examined. Inclusion criteria were the following:

1. The presence of an operative note documenting laryngomalacia as dynamic collapse of the arytenoids into the glottis on inspiration during sleep endoscopy
2. The presence of an operative note indicating the performance of a supraglottoplasty
3. Age of 2 to 18 years
4. The presence of presupraglottoplasty polysomnogram indicating OSAS
5. The presence of a postsupraglottoplasty polysomnogram

These criteria yielded a patient population with OSAS and concurrent laryngomalacia, excluding any infants with congenital laryngomalacia. We refer to this population as having "occult laryngomalacia" as a cause of their OSAS, because they did not typically display symptoms usually associated with laryngomalacia, that is, inspiratory stridor, feeding difficulty, or failure to thrive. Occult laryngomalacia, instead, presents with snoring and OSAS.

Patients thus defined were separated into 2 groups. In the first, patients were determined on sleep endoscopy and clinical examination to have multiple levels of obstruction, including adenotonsillar hypertrophy, lingual tonsil hypertrophy, or inferior turbinate hypertrophy as well as occult laryngomalacia. These patients were treated with initial surgical interventions (tonsillectomy and adenoidectomy, lingual tonsillectomy, and inferior turbinate reduction, respectively) as appropriate in a staged manner prior to supraglottoplasty. Formal assessment of OSA with polysomnography was only undertaken before and after this set of staged procedures. In the second group, supraglottoplasty was the only surgical intervention for OSAS, with polysomnography performed before and after the supraglottoplasty, with no other interval surgical procedures. These patients included both children who had prior surgery (most often tonsillectomy and adenoidectomy) and presented with persistent OSAS, as well as children who did not have any other anatomical causes of OSAS and were treated with supraglottoplasty alone.

Because obesity and tonsillar hypertrophy are well known to be associated with OSA, they were quantified for this population. Body mass index (BMI), calculated as weight in kilograms divided by height in meters squared, was calculated, and patients were classified as underweight (<5th percentile for BMI), normal (5th-85th percentile), overweight (85th to 95th percentile), or obese (>95th percentile). Palatine tonsil size was graded as 0 (surgically absent), 1+ (present but not extending beyond the pillars), 2+ (extending beyond the pillars but filling less than 50% of the distance between them), 3+ (filling more than 50% of the distance between the pillars), and 4+ (touching in midline).

OPERATIVE TECHNIQUE

Laryngomalacia was identified using flexible fiber-optic sleep endoscopy, which was performed in children with persistent OSA after adenotonsillectomy, OSA with small (0 or 1+) tonsils, or OSA complicated by other medical comorbidities. Supraglottoplasty was performed in all children under spontaneous ventilation with total intravenous anesthesia with remifentanyl and propofol. Patients were placed in suspension with a Lindholm laryngoscope inserted into the vallecula. Vocal cords were anesthetized with topical plain 1% lidocaine hydrochloride, and the procedure was performed under binocular microscopy. Using the microscope-mounted carbon dioxide laser, the aryepiglottic folds were divided and redundant mucosa excised from the accessory cartilages, taking care not to demucosalize the medial aspects of the arytenoids. Patients were observed overnight in the pediatric intensive care unit, given 3 doses of postoperative decadron (0.5 mg/kg) over 24 hours, and typically discharged the next day on a proton-pump inhibitor for 1 month to prevent laryngopharyngeal reflux.

POLYSOMNOGRAPHY

Apnea-hypopnea indices and minimum oxygen saturation levels were obtained from the primary reports of polysomnograms obtained at 2 centers—the Stanford Sleep Disorders Clinic and the LPCH Sleep Laboratory. Both centers define an obstructive apnea in children as any complete cessation of oronasal airflow for 2 respiratory cycles with persistent respiratory effort. The AHI as used in this study is defined as the number of obstructive apneas plus obstructive hypopneas per hour of sleep. OSAS was defined 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.

Polysomnograms were performed at 3 time points: (1) before any treatment for OSA, (2) after surgical intervention not including supraglottoplasty, and (3) after supraglottoplasty. When possible, all polysomnograms for an individual patient were obtained from a single center; in several cases, studies from the 2 different centers were compared.

STATISTICAL ANALYSIS

Comparisons of paired nonparametric data, including comparison of preoperative and postoperative AHI and minimum oxygen saturations, were performed with the Wilcoxon rank-sum test. Parametric data were compared with t test. $P<.05$ was considered statistically significant.

RESULTS

PATIENT DEMOGRAPHICS

Patient demographics are summarized in the **Table**. A total of 22 patients met the inclusion criteria, with a mean age of 7.4 years (range, 2.2-17.0 years) at time of supraglottoplasty. Six of 22 (27%) were female, and the mean BMI for the entire population was 17.9 (range, 13.2-29.1). Three

Table. Patient Demographics[a]

Characteristic	All Patients	Concurrent SGP	Isolated SGP
Age, mean, y	7.4	7.9	7.1
Patients, No.	22	13	9
Sex, No.			
Male	16	10	6
Female	6	3	3
BMI	17.9	18.3	16.9
Overweight or obese, No. (%)	6 (27)	4 (30)	2 (22)
Comorbid conditions, No. (%)	10 (45)	8 (62)	2 (22)

Abbreviations: BMI, body mass index (calculated as weight in kilograms divided by height in meters squared); SGP, supraglottoplasty.

[a]Demographics, weight, and comorbidity status are presented for all patients, those who underwent SGP together with another procedure for obstructive sleep apnea (concurrent SGP) and those who underwent SGP alone (isolated SGP).

children were overweight (85th-95th percentiles for BMI), 3 were obese (>95th percentile), 2 were underweight (<5th percentile), and 15 had normal BMI for age. Reflective of the tertiary referral patterns that characterize our practice, 10 of 22 patients (45%) had significant medical comorbidities, including trisomy 21 (2 patients); cerebral palsy (2); and Sanfilippo syndrome, Duchenne muscular dystrophy, Prader-Willi syndrome, lissencephaly, microcephaly, and neurofibromatosis type I (1 each). Palatine tonsil size ranged from 0 to 3+, with all children with 2+ or 3+ tonsils having a significant comorbidity to justify the performance of sleep endoscopy.

SURGICAL PROCEDURES

All 22 children underwent flexible fiber-optic sleep endoscopy, due to persistent OSAS after adenotonsillectomy (12 patients), small (1+) tonsils on clinical examination (5 patients), severe hypotonia due to medical comorbidities (4 patients), or laryngomalacia noted on clinic flexible fiber-optic examination (1 patient). Thirteen of 22 patients underwent supraglottoplasty staged with a second operation for OSAS (adenotonsillectomy, lingual tonsillectomy, adenoidectomy, uvulopalatopharyngoplasty, and/or inferior turbinate reduction) between polysomnographic evaluations, with a mean time between procedures of 2.2 months (range, 0-7.2 months). Nine of 22 patients underwent supraglottoplasty alone between polysomnographic evaluations. Six of these children had undergone prior surgical treatment for OSAS (adenotonsillectomy and/or inferior turbinate reduction), with a mean time between procedures of 22.5 months (range, 7.9-83.6 months). Three children underwent supraglottoplasty as a sole treatment. The mean ages, sex distributions, BMIs, and incidence of comorbidities of the concurrent and isolated groups were not significantly different (Table).

Intraoperative findings were consistent for supraglottic collapse on inspiration (**Figure 1**). The epiglottis was typically normally shaped, yet slightly retroflexed due to shortened aryepiglottic folds. There was invariably a con-
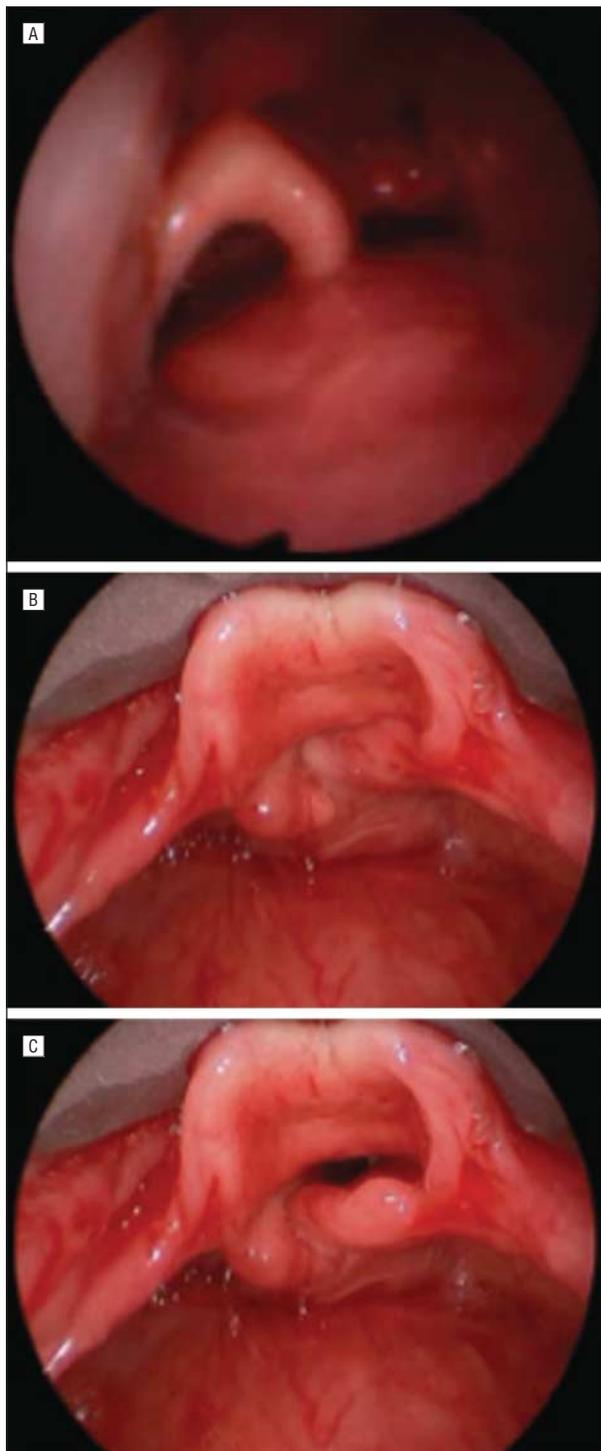
Figure 1. Occult laryngomalacia. A, Sleep endoscopy demonstrates posterior displacement and curled shape of the epiglottis. B and C, Suspension laryngoscopy demonstrates prolapse of the arytenoids into the glottis on inspiration and opening up of the glottis during expiration, respectively.

siderable amount of redundant mucosa overlying the arytenoids that prolapsed into the glottis. In all cases, the carbon dioxide laser was used to divide the aryepiglottic folds and ablate the redundant mucosa. All patients tolerated the procedure well, with no major complications encountered or surgical revisions required.
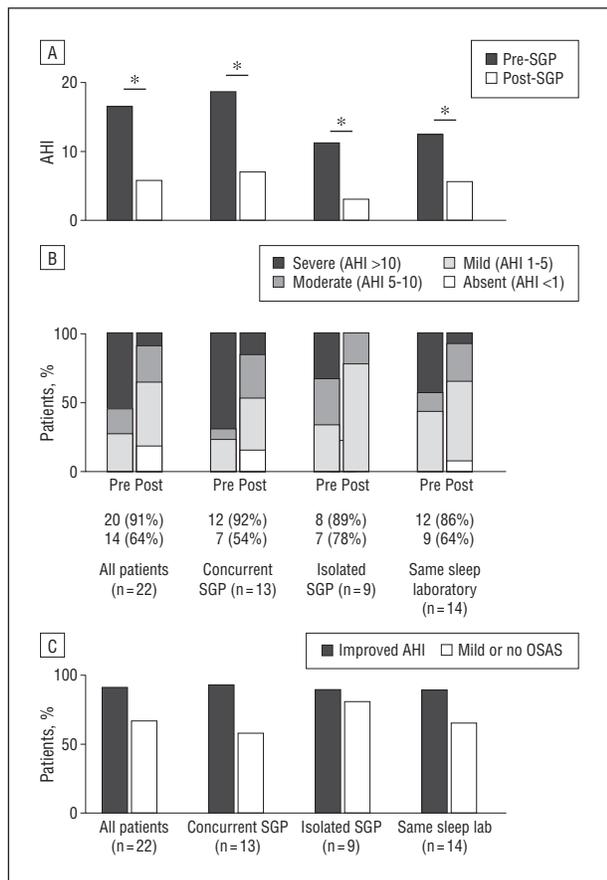
Figure 2. Polysomnographic evaluation before (pre) and after (post) supraglottoplasty (SGP). Four patient populations were evaluated: (1) all patients, (2) children who underwent SGP together with another surgical intervention for obstructive sleep apnea syndrome (OSAS) (concurrent SGP), (3) children who underwent SGP alone (isolated SGP), and (4) the subset of children who had both preoperative and postoperative polysomnograms performed at the same sleep laboratory. A, Comparison of the apnea-hypopnea index (AHI) before (black) and after (white) SGP. *$P < .01$. B, Distribution of patients with severe (black), moderate (dark gray), mild (light gray), and absent (white) OSA before (preop) and after (postop) SGP. C, Overall percentage of children with improved AHI (black) or either no residual OSAS or only mild residual OSAS (white) after SGP.

POLYSOMNOGRAPHIC OUTCOMES

All children underwent polysomnography prior to supraglottoplasty. In 9 children, preoperative and postoperative polysomnograms were obtained before and after adenotonsillectomy performed prior to supraglottoplasty. The mean AHI in this group worsened, from 7.1 to 11.5, whereas minimum oxygen saturation levels stayed stable at 91%, demonstrating the inefficacy of adenotonsillectomy in this population.

Polysomnographic findings are summarized in **Figure 2**. Taken together, the 22 children had a significant improvement in mean AHI from 15.4 (range, 1.4-48.4) to 5.4 (range, 0-24.7) ($P < .001$), with 20 of 22 (91%) having some improvement in AHI and 14 of 22 (64%) improving to either mild or no residual OSAS. There was no change noted in minimum oxygen saturation.

Among the 13 children who underwent supraglottoplasty together with a second staged procedure, AHI also improved significantly, from 18.8 (range, 1.4-48.4) to 7.2 (range, 0-24.7) ($P < .001$). Twelve of 13 (92%) showed
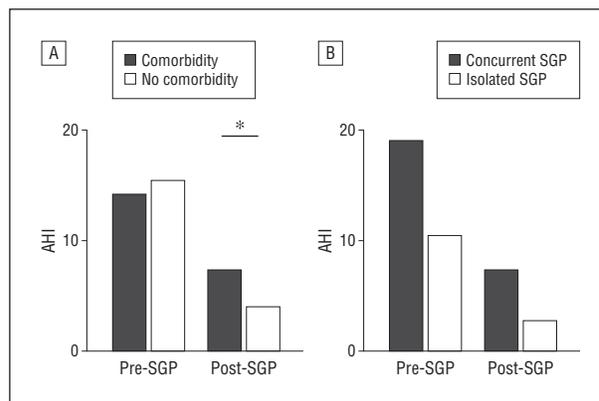

Figure 3. Subgroup analysis. Comparison of presupraglottoplasty (pre-SGP) and postsupraglottoplasty (post-SGP) apnea-hypopnea indices (AHIs) for patients with or without medical comorbidities (A) and for patients undergoing other concurrent surgical interventions for OSA or SGP alone (B). *$P < .05$ by Mann-Whitney test.

improvement in AHI, with 7 of 13 (54%) having either mild or no residual OSAS. As in the whole group, minimum oxygen desaturation did not change.

The 9 patients who underwent supraglottoplasty in isolation, without additional surgery for OSAS, had similar results: mean AHI improved significantly from 10.4 (range, 3.0-28.4) to 2.9 (range, 0-8.7; $P < .01$), without change in minimum oxygen saturation. Eight of 9 children (89%) had improved AHI, and 7 of 9 (78%) had only mild or no residual OSAS after surgery.

Preoperative and postoperative AHIs were not significantly different between the concurrent and isolated intervention groups (**Figure 3**). However, children with medically comorbid conditions, though not noted to have worse preoperative AHI compared with children without such comorbidities, did have significantly worse postoperative AHI, indicating that the presence of medical comorbidities diminished the efficacy of the surgical intervention.

One potential confounding variable in our cohort was that some patients had preoperative and postoperative polysomnography performed at different sleep laboratories. When considering only those children who had polysomnograms at the same institution throughout their treatment, we observed statistically indistinguishable results from those obtain for the group as a whole; AHI improved significantly from 11.6 to 5.2 ($P < .005$), and minimum oxygen desaturation did not change. Twelve of 14 (86%) showed improvement in AHI, and 9 of 14 (64%) achieved either mild or no residual OSAS.

COMMENT

Adenotonsillar hypertrophy is the most common cause of OSAS in children, and removal of the palatine tonsils and adenoids is a highly effective initial treatment in these patients. Other anatomic sources of airway obstruction are not addressed with adenotonsillectomy and may manifest as persistent OSAS. Polysomnography is crucial in providing objective measurement of the severity of OSAS and is recommended by the American Academy of Pediatrics prior to adenotonsillectomy.[12] Performed before surgery, it provides confirmation of disease and in-

tervention, is an important criterion in perioperative risk assessment, and acts as a valuable baseline against which to compare postoperative outcomes. When children continue to have clinical signs and symptoms after adenotonsillectomy, polysomnography becomes even more important to determine whether further, more invasive surgical treatment is warranted.

Under such circumstances, sleep endoscopy is an important tool for identifying potentially addressable sites of persistent obstruction, including lingual tonsillar hypertrophy[13] and occult laryngomalacia. We do not routinely perform sleep endoscopy in all children referred for OSAS; rather, we use it for children with persistent OSAS after adenotonsillectomy, in those with small tonsillar size in whom a second airway lesion is suspected, and in children with such significant hypotonia in whom a comprehensive airway evaluation is warranted.

In this study, we examined the outcomes after supraglottoplasty in children with OSAS and laryngomalacia. None of these children had the stridor, feeding difficulties, or failure to thrive characteristic of congenital infantile laryngomalacia; the noise of this disorder is snoring, and their diagnosis was only recognized on sleep endoscopy. Our intraoperative findings largely corroborated those noted previously[10,11]: compared with congenital laryngomalacia, epiglottic findings were less striking, whereas redundant, prolapsing mucosa overlying the arytenoids was a relatively more significant contributor to supraglottic collapse.

Similar to previous findings, 45% of the children in this study had a medical comorbid condition involving some component of motor delay or hypotonia.[9] Congenital laryngomalacia has been associated with poor laryngeal tone[14]; similar forces may be at play in occult laryngomalacia in older children, which may explain the high prevalence of motor disorders in children with hypotonia, in whom OSAS is notoriously difficult to treat. Children with such comorbidities have worse outcomes in congenital laryngomalacia[15]; they are more likely to need revision surgery or tracheostomy. Our findings corroborate this idea; although children with medical comorbidities had comparable preoperative AHIs, they fared more poorly after surgery, having significantly higher postoperative AHIs than children without comorbid conditions.

In this study, children were segregated into 1 of 2 categories: those who had an additional site of potential airway obstruction identified during clinical examination and sleep endoscopy, such as the palatine tonsils and adenoids, lingual tonsils, or inferior turbinates; and those in whom the only significant site of obstruction was the supraglottis, due to either prior adenotonsillectomy or native small tonsillar size. Ninety-one percent of all children had an improvement in AHI, and 64% had only mild or no residual OSA after surgery; outcomes were not significantly different whether or not additional procedures were performed together with supraglottoplasty. Overall, these findings suggest that occult laryngomalacia is an important contributor to OSA and that its correction can significantly improve sleep in children.

In conclusion, we have found that an occult form of noninfantile laryngomalacia can be a cause of OSA in children. Unlike classic infantile laryngomalacia, which presents with stridor, this form of laryngomalacia is manifest by snoring during sleep. This recently recognized lesion can be readily detected during sleep endoscopy and safely and effectively treated with supraglottoplasty.

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Author Contributions: Dr Chan had full access to all the data in the study and takes responsibility for the integrity of the data and the accuracy of the data analysis. *Study concept and design:* Chan, Truong, and Koltai. *Acquisition of data:* Chan. *Analysis and interpretation of data:* Chan. *Drafting of the manuscript:* Chan. *Critical revision of the manuscript for important intellectual content:* Chan, Truong, and Koltai. *Statistical analysis:* Chan. *Administrative, technical, and material support:* Truong and Koltai. *Study supervision:* Koltai.

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