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Sleep Apnea in Children with Laryngomalacia: Diagnosis via Sedated Endoscopy and Objective Outcomes after Supraglottoplasty

G. Paul Digoy, MD¹, Mohanad Shukry, MD², and Julie A. Stoner, PhD³

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Abstract

Objective. The authors study the contribution of laryngomalacia to obstructive sleep apnea syndrome (OSAS) in children older than 12 months. The clinical and polysomnographic outcomes in patients with OSAS who underwent a supraglottoplasty were also studied.


Study Design. A case series with chart review.

Subjects and Methods. A review of consecutive pediatric patients diagnosed with both OSAS and state-dependant laryngomalacia (SDL) between 2005 and 2008. The diagnosis of SDL was made via laryngoscopy under light general anesthesia (sleep endoscopy). All subjects underwent a supraglottoplasty.

Results. A total of 43 patients met inclusion criteria, and 36 patients had complete pre- and postoperative data available for review. The apnea-hypopnea index (AHI) score decreased following supraglottoplasty for 33 (92%; 95% confidence interval [CI], 78%-98%) of the 36 patients. The mean (SD) change in AHI score (calculated as the postoperative minus the preoperative measure) was −9.2 (11.2), representing a statistically significant reduction (95% CI, −13.0 to −5.5; \( P < .0001 \)). The mean (SD) preoperative AHI was 13.3 (12.9). The minimum oxygen saturation increased following supraglottoplasty for 21 (58%; 95% CI, 41%-74%). The mean (SD) change in the minimum oxygen saturation was 3.5 (8.3), which was a statistically significant increase (95% CI, 0.7-6.3; \( P = .015 \)).

Conclusion. Laryngomalacia may contribute significantly to OSAS in some children who are 12 months and older. Sleep endoscopy appears to be an effective method in the diagnosis of SDL. When present, a supraglottoplasty can be an effective procedure and may significantly improve symptoms of OSAS.

Keywords

laryngomalacia, sleep apnea, sleep endoscopy, polysomnography, supraglottoplasty, pediatric

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Obstructive sleep apnea syndrome (OSAS) is a common condition in childhood and can lead to significant behavioral and cognitive impairment.⁰⁻³ Although adenotonsillectomy is considered the first line of therapy in children with OSAS, recent reports have suggested that the persistence of OSAS after adenotonsillectomy may be significant, ranging from 20% to 75% of cases.⁴⁻⁸

The laryngeal contribution to sleep apnea is poorly understood, and few studies have attempted to investigate the larynx as a primary source of sleep apnea. A condition known as state-dependant laryngomalacia (SDL) was coined by Amin and Isaacson in 1997 and refers to a child with normal breathing while awake but stridor and increased work of breathing during sleep.⁹ As the pediatric airway develops, it is unclear if this is a congenital condition that resolves as the child grows vs a neuromuscular and/or anatomic abnormality that independently continues to contribute to OSAS in older children. The diagnosis and management of SDL are poorly understood, and objective evidence such as pre- and postoperative polysomnography after supraglottoplasty is limited, especially in older children.

This study examines the clinical and polysomnographic outcomes after supraglottoplasty in children older than 12 months with findings suggestive of SDL on measures of

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OSAS—namely, the number of obstructive events per hour, apnea-hypopnea index (AHI), and minimum oxygen saturation. This study also seeks to determine if the use of pediatric sleep endoscopy is an effective diagnostic tool for SDL.

Methods

This retrospective case series was approved by the institutional review board of the University of Oklahoma Health Sciences Center. Only data from consecutive subjects with findings suggestive of state-dependent laryngomalacia on sedated endoscopy who underwent supraglottoplasty were included in this analysis. The study period is from August 2005 through December 2008. Patients were identified who met the following criteria: the presence of clinically significant sleep-disordered breathing, a sleep study with an AHI greater than 2, and a sedated laryngoscopy with findings suggestive of state-dependent laryngomalacia. A child was not considered for surgery based on sleep study findings or clinical symptoms alone. When mild OSAS was present, surgery was considered only when clinical symptoms were felt to be significantly affecting his or her quality of life.

In this series and in the practice of the primary author (G.P.D.), sleep endoscopy (as described below) was part of the workup to determine the primary source of airway obstruction in children who had either failed adenotonsillectomy or had very small tonsils that did not appear to significantly contribute to OSAS. It was also used in younger children (age <24 months) who had a history of laryngomalacia as infants. Prior to undergoing sleep endoscopy, children underwent a thorough assessment for other sources of obstructive breathing, including nasal obstruction. When other sources of obstruction, such as allergic rhinitis, were suspected from the history and physical examination, children underwent testing and treatment (such as topical nasal steroids) prior to considering other airways procedures.

Only children who were older than 12 months were included in this study. Those younger were excluded for 2 fundamental reasons. First, the infant develops rapidly during the first year of life and is generally more prone to laryngomalacia rather than the more common tongue base and tonsillar obstruction seen later in childhood. The second reason is that polysomnographic characteristics also evolve significantly during the first year of life, and these values may not be comparable with older children.

Clinical outcome was further assessed by asking the caregivers to complete a postoperative telephone questionnaire (see Table 1).

Table 1. Postoperative Telephone Questionnaire

<table>
<thead>
<tr>
<th>Question</th>
<th>Response Options</th>
</tr>
</thead>
<tbody>
<tr>
<td>1a. Did your child experience any difficulty with swallowing after surgery? (ie, choking on liquids or increased coughing when eating)?</td>
<td>Y es or no</td>
</tr>
<tr>
<td>b. If yes, when did these symptoms resolve?</td>
<td></td>
</tr>
<tr>
<td>2. Was there a general increase in coughing and/or throat clearing noticed after surgery (even when not related to eating)? Did this resolve?</td>
<td></td>
</tr>
<tr>
<td>3. Did your child experience any difficulty with breathing postoperatively?</td>
<td></td>
</tr>
<tr>
<td>4. How would you describe your child's sleep after surgery? much improved / improved / no change / worse</td>
<td></td>
</tr>
<tr>
<td>5. If your child snored preoperatively, how did this snoring change?</td>
<td>snoring resolved / snoring decreased / snoring unchanged / snoring worse</td>
</tr>
<tr>
<td>6. Are you glad your child underwent this procedure? Yes or no</td>
<td></td>
</tr>
</tbody>
</table>

Sedated Endoscopy (“Sleep Endoscopy”)

The diagnosis of a laryngeal contribution to sleep apnea in this study was made under general anesthesia, which was induced with sevoflurane (8%) in 100% oxygen. While the patient was spontaneously breathing, intravenous (IV) access was obtained. The administration of sevoflurane was then reduced or stopped as the surgeon performed either indirect laryngoscopy or direct laryngoscopy. Earlier cases were evaluated via direct laryngoscopy alone. Occasionally, small doses of IV propofol were needed for the child to tolerate direct laryngoscopy. As the child exhaled sevoflurane, the concentration of the gas decreased, as so the depth of anesthesia. As the child then neared stage II anesthesia, dynamic breathing and pharyngolaryngeal muscle movements were examined. Figures 1 and 2 illustrate a typical case of a patent airway during expiration and laryngeal obstruction during inspiration. Our practice has evolved to favor flexible endoscopy over rigid as this method can be done at a lighter level of anesthesia and without the use of a shoulder role and may provide a more valid assessment of airway dynamics.

Supraglottoplasty

The procedure began by first dividing the aryepiglottic folds using laryngeal scissors. The carbon dioxide laser was then used at the lowest setting possible (typically at 1.5 watts—superpulse) to remove the redundant supraglottic mucosa along with the superior portion of the cuneiform cartilage (see Figure 3). The amount of supraglottic tissue removed was guided by the findings during sleep endoscopy. Care was taken to avoid injury to intra-arytenoid space. In all cases, the procedure was performed bilaterally. This laser-assisted method was used rather than a completely cold technique because it provided better precision and decreased bleeding. In the primary author’s (G.P.D.) experience, a cold technique in older children generally results in increased bleeding and decreased surgical precision.
**Statistical Analysis**

Descriptive statistics, including the median, 25th and 75th percentiles, mean, and standard deviation, were used to summarize the distribution of continuous measures, and proportions, with exact 95% confidence intervals, were used to summarize the distribution of dichotomous measures. The distribution of characteristics was compared between independent groups using a Wilcoxon rank sum test for continuous measures and a Fisher exact test for categorical measures. The preoperative and postoperative measures were compared using a paired t test to account for the paired structure of the data. The association between changes in the OSAS measures and age at the time of surgery or the time lag between surgery and the postoperative sleep study was investigated using scatter plots, and Spearman rank correlation coefficients were calculated. A 2-sided α level of 0.05 was used to define statistical significance.

**Results**

Forty-three children had an AHI >2, had clinically significant sleep-disordered breathing, were diagnosed via sleep endoscopy of having state-dependant laryngomalacia, and underwent a laser supraglottoplasty. Thirty-six patients had pre- and postoperative sleep studies available for comparison, whereas postoperative studies were not available for 7 subjects. A summary of demographic and preoperative characteristics is provided in Table 2 for subjects with and without postoperative sleep study data available. Subjects without postoperative data tended to be younger, were less likely to have Down’s syndrome, were more likely to be male, and had a lower number of obstructive events and lower AHI values compared with subjects with postoperative data, although none of the differences were statistically significant. Among those with postoperative sleep study data available, 22 (61%) were male.

Nine of 43 patients were diagnosed with a syndrome (Down’s, n = 6; DiGeorge, n = 2; CHARGE, n = 1). Two children were suspected of having a syndrome but had not been officially diagnosed. Five children had cerebral palsy. Twenty-seven patients were nonsyndromic and did not have other major comorbidities. Twenty-two of these 27 had postoperative sleep study data available for review.

Thirty-two of 43 patients had OSAS despite having previously undergone an adenotonsillectomy. Polysomnography in this population was performed a minimum of 3 months following adenotonsillectomy. Of the 11 children who did not undergo adenotonsillectomy prior to supraglottoplasty, 7 were younger than 24 months and had very small to average tonsils (1-2+ in size), and 4 were older than 24 months with tonsils considered to be very small (1+).

The time lag between surgery and the postoperative sleep study ranged from 16 to 483 days with a median time lag of 56 days (25th-75th percentile, 37-93 days). A summary of the preoperative and postoperative OSAS and polysomnographic measures, as well as the change calculated as the
postoperative measures minus the preoperative measures, is provided in Table 3. The time lag between surgery and the postoperative sleep study was not significantly associated with the postoperative changes in the number of obstructive sleep events (Spearman correlation coefficient $r = -0.18$, $P = .30$), the change in AHI score ($r = -0.094$, $P = .58$), or the change in minimum oxygen saturation ($r = 0.27$, $P = .11$). The number of obstructive events per hour decreased following supraglottoplasty for 31 (86%; 95% confidence interval [CI], 71%-95%) of the 36 patients, and the average reduction was statistically significant (Figure 4). Thirty-three patients (92%; 95% CI, 78%-98%) experienced a decrease in the AHI score, where the average reduction was statistically significant (Figure 5). The minimum oxygen saturation increased following supraglottoplasty for 21 (58%; 95% CI, 41%-74%) of the patients, and the average increase was statistically significant (Figure 6). Similar results were seen among the subset of 22 subjects who were nonsyndromic and did not have other major comorbidities, where the number of obstructive events per hour decreased following supraglottoplasty for 17 (77%; 95% CI, 55%-92%), 20 patients (91%; 71%-99%) experienced a decrease in the AHI score, and the minimum oxygen saturation increased following supraglottoplasty for 12 (55%; 32%-76%).

**Table 3. Preoperative and Postoperative Polysomnographic Measures**

<table>
<thead>
<tr>
<th>Outcome Measure</th>
<th>Preoperative Data (n = 36)</th>
<th>Postoperative Data (n = 36)</th>
<th>Change (Postoperative – Preoperative) (n = 36)</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>Mean (SD)</td>
<td>Mean (SD)</td>
<td>Mean (SD) 95% CI Chemical</td>
</tr>
<tr>
<td>Obstructive events per hour</td>
<td>9.5 (8.6)</td>
<td>2.8 (4.3)</td>
<td>−6.7 (7.7) −9.3 to −4.1 &lt;.0001</td>
</tr>
<tr>
<td>Apnea-hypopnea index</td>
<td>13.3 (12.9)</td>
<td>4.1 (5.0)</td>
<td>−9.2 (11.2) −13.0 to −5.5 &lt;.0001</td>
</tr>
<tr>
<td>Minimum oxygen saturation</td>
<td>83.0 (8.6)</td>
<td>86.5 (4.9)</td>
<td>3.5 (8.3) 0.7 to 6.3 .015</td>
</tr>
</tbody>
</table>

Abbreviations: AHI, apnea-hypopnea index; CI, confidence interval.

**Figure 4.** Change in the number of obstructive sleep apnea events per hour following supraglottoplasty.

**Figure 5.** Change in the apnea-hypopnea index following supraglottoplasty.
Thirty-three of 43 patients completed a postoperative telephone questionnaire (Tables 1 and 4). Twenty of these 33 patients were described by caregivers as having “much improved” sleep postoperatively. Ten had “improved” sleep, and 3 did not experience a noticeable change, resulting in 30 (91%) reporting at least some improvement in sleep (95% CI, 76%-98%). Twenty-nine of 33 reported to snore preoperatively, and 15 of these experienced a complete resolution of snoring, 10 had a reduction in snoring, and 4 described no change in snoring. Overall, 25 of 33 (86%) patients experienced a reduction or complete resolution of snoring (95% CI, 68%-96%).

If a patient had residual symptoms of dysphagia 1 month postoperatively, a modified barium swallow study was performed. Of the 33 children who participated in this study’s postoperative assessment for dysphagia, 6 patients were primarily fed via a gastrostomy tube and did not report any noticeable change postoperatively. Of the remaining 27 patients, 7 (26%) reported postoperative dysphagia (Table 4; 95% CI, 11%-46%). Most of these 7 patients described symptoms of choking or coughing on clear liquids, especially when drinking rapidly. Four of 7 resolved within a few days after surgery, and 1 patient had resolution of dysphagia by 1 month postoperatively. Two patients described having dysphagia that did not resolve after 6 months, and both had normal modified barium swallow studies. One of the 2 children with continued dysphagia had some uncertainty as to when the symptoms began and may not have been related to the surgery.

Three (9%) children of 33 surveyed had a noticeable increase in coughing and/or throat clearing postoperatively that was not necessarily associated with feeding (95% CI, 2%-24%). One resolved during the first month after surgery. In the other 2 patients, this did not seem to resolve, although caregivers did report a gradual improvement over time.

Overall, 31 (94%) of 33 caregivers reported they were glad their child underwent a supraglottoplasty (95% CI, 80%-99%). One caregiver reported he was “not sure” if he was pleased with having had surgery. This child had Down’s syndrome and experienced a significant improvement in AHI (from 8.6 to 0.9) but did not experience a noticeable change in snoring or sleep patterns. One caregiver stated she was not satisfied with the surgery, and her child’s postoperative sleep study did not improve.

**Discussion**

Few studies have attempted to link laryngomalacia and OSAS. Most series are small (<10 patients) and focus on sleep apnea in the setting of infantile laryngomalacia (younger than 12 months).13-15 Fewer articles have looked at laryngomalacia in older children (>12 months). Smith et al16 in 2005 were the first to report a small series of 4 patients who were 3 to 4 years of age and who experienced a subjective improvement in their sleep-disordered breathing after supraglottoplasty. In 2008, Richter et al17 reported 7 patients who averaged 6.3 years of age and who were diagnosed with OSA and laryngomalacia and experienced clinical resolution of symptoms following supraglottoplasty. Neither group used postoperative sleep studies to assess outcomes.

More recently, Revell et al18 presented their experience assessing for laryngomalacia via direct laryngoscopy in children with symptoms of OSA who were older and younger than 3 years. The younger group included 7 patients who were all younger than 10 months. In the older group, they found some form of laryngomalacia (mild to severe) via direct laryngoscopy in 19 of 70 patients. In this older group, 5 children underwent a supraglottoplasty as the only surgical intervention, and 3 of them had PSG data showing an improvement in AHI. Thavasagayam et al19 diagnosed twelve children in their series with SDL, and 6 underwent a supraglottoplasty. Only 3 of the 6 children were reported to have improved clinically.

In our series of children older than 12 months, we use pre- and post-supraglottoplasty PSG data to demonstrate a

![Figure 6. Change in the minimum oxygen saturation level following supraglottoplasty.](image-url)
achieve sedation. Other reported methods have included propofol, a serious potential side effect to this procedure.

We found that postoperatively, the complication of transient dysphagia is significant (7/27) yet transient, resolving in most children. We presented 2 of 27 patients in this series who reported postoperative dysphagia beyond 1 month. Although these dysphagia symptoms were described as mild and continued to improve over time, it still presents a serious potential side effect to this procedure.

A great challenge in the diagnosis of SDL is the need for an examination under sedation. In this study, we report the first series using sevoflurane as the primary agent used to achieve sedation. Other reported methods have included propofol alone, propofol/narcotic combination, ketamine/dexmedetomidine, ketamine/dexametomidine, and midazolam/narcotic. A significant benefit of using sevoflurane is that it is commonly used to induce anesthesia in children and can be used as a single agent, reducing drug-drug interactions, and may make our examination more reproducible. Sevoflurane can also be exhaled quickly to decrease any airway suppressant effects it may have during the examination.

Thevasagayam et al reported a prevalence of SDL to be 3.9% among 358 children presenting with sleep-disordered breathing who underwent sedated nasopharyngoscopy. They used remifentanyl, propofol, and, in some children, “an inhaled agent.” In our experience, the use of multiple agents (especially propofol with a narcotic) can lead to decreased reliability in achieving stage II airway dynamics, and this may have underestimated the prevalence of SDL.

There are many limitations in our current understanding of “sleep endoscopy” in children. First is that we are lacking sufficient articles that compare the various agents used in children for sleep endoscopy, and we lack consistency in the methodologies proposed. The second obstacle is that as we find an agent or agents that seem to satisfactorily and safely sedate a child in the workup of OSAS, more studies like this one will be needed that objectively validate clinical improvement after an intervention (such as supraglottoplasty).

This study has a number of limitations. First, there is no control group. Ideally, a control group of children who have OSAS and SDL but have not undergone a supraglottoplasty would be needed to properly compare the effect of this surgery on PSG data. Second is the lack of long-term follow-up. The time lag between surgery and the postoperative sleep study in our series ranged from 16 to 483 days with a median time lag of 56 days (25th-75th percentile, 37-93 days). The sustainability of these results over time would require a repeat sleep study 1 or more years after surgery. Another limitation is the use of a nonvalidated postoperative caregiver questionnaire. A validated outcome measure specific for laryngomalacia (considering side effects such as aspiration) would give greater authority to our results. Finally, it would have been revealing to study whether subclinical silent aspiration develops and whether this leads to a potential increase in pneumonia over time. Future studies could use pre- and postoperative nuclear salivagrams to rule out silent aspiration.

### Conclusion

Laryngomalacia may contribute significantly to OSA in children older than 12 months. We report the largest series of children who were diagnosed with SDL and sleep apnea who underwent a laser supraglottoplasty and experienced a statistically significant improvement in AHI/OAI. We found that sleep endoscopy appeared to be effective in diagnosing this condition. We also report that the primary complication after supraglottoplasty was dysphagia and postoperative coughing/throat clearing, which was transient in most children but not in all. In this series, the diagnosis and management of SDL led to high patient/caregiver satisfaction.

### Author Contributions

G. Paul Digoy, primary surgeon, primary researcher, carefully reviewed and participated in all aspects of this study, including review of research data and primary manuscript composition; Mohanad Shukry, primary responsibility of composing the manuscript from the perspective of a pediatric anesthesiologist; Julie A. Stoner, primary responsibility of data assessment and statistical analysis, assisting in study design and gathering of data.

### Disclosures

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