Unilateral vs Bilateral Supraglottoplasty for Severe Laryngomalacia in Children

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Objectives: To study the efficacy of unilateral supraglottoplasty in comparison with bilateral supraglottoplasty for the treatment of severe laryngomalacia in children and to study factors that may be predictive of major complications or the need for a subsequent contralateral or revision procedure.

Design: Retrospective medical record review.

Setting: University tertiary care pediatric hospital.

Patients: One hundred six consecutive pediatric patients, aged 9 days to 18 years, who had undergone unilateral or bilateral supraglottoplasty for severe laryngomalacia.

Main Outcome Measures: Resolution of clinically significant laryngomalacia, development of major complications (supraglottic stenosis or aspiration), and an association between study variables (demographics, medical comorbidities, synchronous airway abnormalities, sites of excision, and techniques of excision) and the need for subsequent contralateral or revision supraglottoplasty.

Results: We achieved a high success rate (95.7%), a low complication rate (8.5%), and observed the need for a contralateral procedure in 7 (14.9%) of the 47 patients who underwent initial unilateral supraglottoplasty. Two patients who underwent initial bilateral supraglottoplasty developed supraglottic stenosis. No significant association existed between our study variables and the development of complications or the need for contralateral or revision supraglottoplasty.

Conclusions: Unilateral supraglottoplasty was associated with a high success rate, low complication rate, and the avoidance of supraglottic stenosis in our study population. The percentage of patients requiring a subsequent contralateral procedure was comparable to that reported in the literature, and no major complications were associated with the second operation in these patients. Therefore, unilateral supraglottoplasty seems to be a reasonable option for initial surgical management of pediatric patients with severe laryngomalacia.

Laryngomalacia is the most common congenital laryngeal anomaly and the most common cause of stridor in infants. The condition consists of collapse of any of the major components of the supraglottis, including the epiglottis, aryepiglottic folds (AEFs), and arytenoid mucosa. This collapse leads to varying degrees of upper airway obstruction.

The primary clinical manifestation of laryngomalacia is inspiratory stridor, which can vary in description from the usual high-pitched “hiccuping” or “squeaking” sound to the less frequently described sounds such as “croaking” or “crowing.” This stridor is usually first noticed at or shortly after birth, increasing over the next several months, and gradually resolving by 2 years of age, although it may persist into later childhood. Most cases of laryngomalacia are mild, self-limiting, and usually unassociated with respiratory distress. Such cases do not require surgical intervention and are managed expectantly. However, 10% to 15% of the patients may present with signs of respiratory difficulty and may develop associated medical problems, including failure to thrive, obstructive sleep apnea, and cor pulmonale resulting from chronic hypoxia. In these severe cases, surgical intervention is required. The most common procedure performed consists of excision of floppy supraglottic tissues or supraglottoplasty.

Supraglottoplasty may be unilateral or bilateral; indeed, all of the earlier reports on the subject described bilateral procedures. Controversy has recently arisen about the safety of bilateral supraglottoplasty, specifically with reference to the severe, although rare, risks of postoperative supraglottic stenosis and aspiration.
MATERIALS AND METHODS

We reviewed the medical records of all patients who had undergone endoscopic supraglottoplasty at our pediatric tertiary care institution by one of us (B.H.M.) from July 1, 1989, to September 30, 1997. We studied variables deemed to have a potential influence on the outcome of supraglottoplasty. These variables included general demographics (age at time of initial procedure, sex); medical comorbidities; additional upper airway procedures performed either preoperatively, postoperatively, or concurrently with supraglottoplasty; areas of excision of supraglottic tissue; and operative techniques. We also reviewed outcomes, including the resolution of laryngomalacia; persistent laryngomalacia requiring either revision or contralateral supraglottoplasty; postoperative supraglottic stenosis; and postoperative aspiration.

All patients were referred to the Division of Pediatric Otolaryngology–Head and Neck Surgery, Indiana University School of Medicine, Indianapolis, for evaluation of stridor associated with various disorders, including obstructive sleep apnea, failure to thrive, and episodes of apnea or cyanosis. All patients were diagnosed as having severe laryngomalacia on the basis of flexible and/or rigid endoscopic findings, as well as the presence of associated disorders, as verified by the appropriate diagnostic evaluations (eg, polysomnogram, scintiscan, and others). All patients underwent direct microrigid laryngoscopy and bronchoscopy at the time of supraglottoplasty to evaluate the airway for synchronous lesions. Supraglottoplasty was performed using the apneic or spontaneous respiration technique in most cases, and all procedures were performed with microscissors or the carbon dioxide laser under suspension microlaryngoscopy. Postoperatively, the vast majority of patients were extubated and observed in the intensive care unit for at least 24 hours, and a regimen of antibiotic agents and anti-reflux medications was started for at least 2 weeks. This medical regimen was prophylactic and intended to decrease the risk of supraglottic infection and edema, which could lead to airway obstruction in either group, or scarring and supraglottic stenosis in patients with bilateral excision sites. Postoperative follow-up included fiberoptic laryngoscopy, as well as diagnostic studies for associated conditions, when indicated.

At our institution, many supraglottoplasties have been performed over the past 10 years. During the past few years, we have performed the unilateral procedure in most patients. In our experience, the incidence of severe complications following either bilateral or unilateral supraglottoplasty has been low. However, we recently noted that a few patients who underwent unilateral supraglottoplasty required a contralateral procedure at a later date, due to persistent laryngomalacia. Owing to concerns regarding the need for a second operation in these patients, we decided to study the outcomes of unilateral and bilateral supraglottoplasty in our patient population, including the efficacy of each procedure in eliminating clinically significant laryngomalacia, the need for revision supraglottoplasty, and the development of complications, specifically supraglottic stenosis and aspiration. Ultimately, we sought to determine if unilateral supraglottoplasty is sufficiently efficacious and safe to recommend as the initial procedure of choice in the surgical management of patients with severe laryngomalacia.

## RESULTS

### DEMOGRAPHICS

Data were obtained for a total of 106 consecutive patients. These patients were grouped into 2 main categories: those initially undergoing bilateral (group 1) or unilateral (group 2) supraglottoplasty. Group 1 consisted of 59 patients and group 2 consisted of 47 patients. The mean age at the initial procedure in group 1 was 27.1 months (age range, 2 weeks to 18 years), and in group 2 was 38.3 months (age range, 9 days to 15 years). Sixty-five patients were male and 41 female.

### MAJOR INDICATIONS FOR SUPRAGLOTTOPLASTY

Although most patients had more than 1 contributing medical diagnosis or comorbidity, the decision to proceed to supraglottoplasty was based on 1 of 4 major indications (Table 1). The vast majority of patients had stridor and obstructive symptoms during sleep, and the diagnosis of obstructive sleep apnea was confirmed by polysomnography or oxypneumocardiogram. Other major indications included failure to thrive, episodes of apnea/bradycardia/cyanosis, or cyanosis during feedings.

### COMORBIDITIES

Of the comorbidities associated with laryngomalacia, gastroesophageal reflux was the most common (Table 2).
Others disorders, in decreasing order of frequency, were neuromuscular, respiratory, and cardiovascular.

**AREAS OF EXCISION**

The supraglottis consists of 3 distinct sites: the epiglottis, AEFs, and arytenoid mucosa. From these 3 sites, 7 specific combinations of resection were possible (Table 3). The amount and location of tissue excised depended on preoperative flexible fiberoptic laryngoscopy findings, as well as findings during intraoperative evaluation. Within group 1, 22 patients had excision of all 3 sites, 12 patients had excision of the epiglottis and AEFs together, and 9 patients had excision of the AEFs alone. Other combinations were less common. Within group 2, the most common combinations were AEFs together with arytenoid mucosa (16 patients) and AEFs alone (13 patients).

**OPERATIVE TECHNIQUES**

Most of the earlier procedures were performed using microscissors, with carbon dioxide laser excision becoming the preponderant method in more recent cases. In group 1, 23 patients were treated by microcissor excision and 36 patients by laser. In group 2, only 5 patients were treated by microcissor excision and 42 patients by laser.

**ADDITIONAL UPPER AIRWAY PROCEDURES**

Many patients in both groups underwent procedures for treatment of synchronous upper airway anomalies, preoperatively, postoperatively, or concurrently with initial or revision contralateral supraglottoplasty. Forty patients in group 1 underwent bilateral supraglottoplasty only; 24 patients in group B underwent unilateral supraglottoplasty only.

**MAIN OUTCOME MEASURES**

We defined outcomes in terms of the resolution or persistence of clinically significant laryngomalacia, the development of severe complications (supraglottic stenosis and aspiration), the association between study variables and complications, and between study variables and the need for revision supraglottoplasty. Success rate was defined by resolution or improvement of laryngomalacia, as evidenced by follow-up laryngoscopy. Failure was defined by persistent laryngomalacia requiring tracheostomy or failure to decannulate.

Of the 59 patients in group 1, 55 (93.2%) had clinical resolution of laryngomalacia after either the initial or the revision procedure. Three patients required a revision supraglottoplasty for persistent laryngomalacia after the initial procedure. The revision procedures were unilateral in 2 of these patients and bilateral in the third, who subsequently required tracheostomy for persistent laryngomalacia. Another patient underwent tracheostomy concurrently with supraglottoplasty and was unable to be decannulated owing to persistent laryngomalacia. Two patients developed supraglottic stenosis that required surgical intervention. One was treated with initial carbon dioxide laser release of AEF scarring, followed by tracheostomy for persistent stenosis; and 1 was treated with tracheostomy alone. Two patients in group 1 developed postoperative aspiration that was not documented preoperatively. Tracheostomy was performed for other medical conditions, such as ventilator dependence due to pulmonary abnormalities, in 9 patients.

Of the 47 patients in group 2, 45 (95.7%) had clinical resolution of laryngomalacia after either the initial or the contralateral procedure. Seven patients required a contralateral procedure for persistent laryngomalacia after the initial unilateral procedure. Six of these patients had resolution of laryngomalacia following the contralateral procedure, and 1 required tracheostomy for persistent laryngomalacia. Four patients demonstrated postoperative aspiration not documented preoperatively, including 1 of the 7 patients requiring contralateral supraglottoplasty. One of these patients eventually required a tracheostomy 2 months later for “sudden (near) death” episodes. Overall, 2 patients required tracheostomy related to their laryngomalacia. Four patients required tracheostomy for other medical conditions.

Patients who developed supraglottic stenosis or aspiration or who required a revision or contralateral supraglottoplasty were analyzed using the same study variables as the other patients in groups 1 and 2. Of the 3 patients who required revision supraglottoplasty following an initial bilateral procedure, ages at the time of the initial procedure were 1, 7, and 14 months (mean age, 7.3 months). Of the 7 patients who required contralateral supraglottoplasty following an initial unilateral procedure, the age range was 9 days to 32 months (mean age, 11.4 months). Of the 2 patients who developed supraglottic stenosis, 1 was 3 weeks of age at the time of the initial procedure and had a supraglottic laser release at 6 weeks of age for stenosis; this was followed by tracheostomy at 2 months for persistent stenosis. The other patient was 2 months of age at the time of the initial procedure and had tracheostomy performed at 3 months of age for stenosis. The average age at the time of the initial procedure for these patients in group 2 was 7 months (mean age, 6 months).

### Table 3. Sites of Excision of Supraglottic Tissues

<table>
<thead>
<tr>
<th>Type of Supraglottoplasty</th>
<th>Epi</th>
<th>AEFs</th>
<th>Ary</th>
<th>Epi/AEFs</th>
<th>Epi/Ary</th>
<th>AEFs/Ary</th>
<th>Epi/AEFs/Ary</th>
</tr>
</thead>
<tbody>
<tr>
<td>Unilateral</td>
<td>0</td>
<td>12</td>
<td>1</td>
<td>6</td>
<td>0</td>
<td>16</td>
<td>4</td>
</tr>
<tr>
<td>Bilateral</td>
<td>2</td>
<td>9</td>
<td>5</td>
<td>12</td>
<td>0</td>
<td>5</td>
<td>22</td>
</tr>
<tr>
<td>Contralateral</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
<td>0</td>
<td>2</td>
<td>1</td>
</tr>
<tr>
<td>Revision bilateral</td>
<td>0</td>
<td>1</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>1</td>
</tr>
</tbody>
</table>

*Epi indicates epiglottis; AEFs, aryepiglottic folds; and Ary, arytenoid mucosa.*
2 patients was 5.5 weeks. Of the 2 patients who developed aspiration following a bilateral procedure, ages at the time of supraglottoplasty were 7 weeks and 11 months (mean age, 6.5 months). Of the 4 patients who developed aspiration following a unilateral procedure, ages at the time of supraglottoplasty were 3, 15, 18, and 23 months (mean age, 14.8 months); the youngest of these patients developed aspiration after undergoing a subsequent contralateral procedure at 3½ months of age. No association was found between any of the variables and the development of supraglottic stenosis, aspiration or the need for a revision or contralateral procedure, although patients in this group were younger, in general, than other patients in groups 1 and 2.

The overall success rate (Table 4) for bilateral supraglottoplasty was 93.2% (55 of the 59 patients) and for unilateral supraglottoplasty was 95.7% (45 of the 47 patients). Failures excluded those patients requiring a tracheostomy for associated problems, such as pulmonary disease requiring ventilatory assistance or tracheomalacia. None of the patients in our series developed immediate complications such as airway obstruction, cardiovascular complications, or bleeding. None developed operative site infection. Overall, the complication rate for group 1, which included the 2 patients who developed supraglottic stenosis and the 2 with postoperative aspiration, was 6.8% (4 of the 59 patients). The complication rate for group 2, which included the 4 patients with postoperative aspiration, was 8.5% (4 of the 47 patients). The revision rate for bilateral supraglottoplasty was 5.1% (3 of the 59 patients), and for unilateral supraglottoplasty (contralateral procedure) was 14.9% (7 of the 47 patients).

Success, failure, revision, and complication rates were compared using χ² analysis (α = .05). None of the results were found to be significantly different between groups 1 and 2. Although patients requiring a revision or contralateral procedure were notably younger than those not requiring a procedure, this difference was not found to be statistically significant.

### Table 4. Overall Results in 106 Study Subjects

<table>
<thead>
<tr>
<th>Main Outcome</th>
<th>Unilateral (n = 47)</th>
<th>Bilateral (n = 59)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Success</td>
<td>45 (95.7)</td>
<td>55 (93.2)</td>
</tr>
<tr>
<td>Failure</td>
<td>2 (4.3)</td>
<td>4 (6.8)</td>
</tr>
<tr>
<td>Supraglottic stenosis</td>
<td>0</td>
<td>2 (3.4)</td>
</tr>
<tr>
<td>Aspiration</td>
<td>4 (8.5)</td>
<td>2 (3.4)</td>
</tr>
<tr>
<td>Contralateral procedure</td>
<td>7 (14.9)</td>
<td>0</td>
</tr>
<tr>
<td>Revision bilateral procedure</td>
<td>0</td>
<td>3 (5.1)</td>
</tr>
</tbody>
</table>

### COMMENT

Although laryngomalacia is widely regarded to be the most common cause of congenital infantile stridor, constituting 60% to 75% of the cases, its true incidence is unknown. Indeed, the very definition of the term is not always uniform. Laryngomalacia is rarely diagnosed in the general population and is more commonly diagnosed at tertiary care pediatric hospitals. This is probably because of the frequency of comorbidities that characterize this patient population, as well as an increased familiarity with the diagnosis among physicians at tertiary care centers. For example, Belmont and Grundfast reported that 80% of the patients had associated gastroesophageal reflux, 23% had neurologic disorders, and 23% had obstructive sleep apnea episodes.

The vast majority of cases of laryngomalacia are considered benign, with no need for surgical intervention and complete resolution of symptoms during early childhood, usually by 2 years of age. However, 10% to 15% of the cases are considered severe enough to require surgical intervention owing to serious complications such as upper airway obstruction, cor pulmonale, and failure to thrive. Tracheostomy has been the surgical criterion (“gold”) standard for bypassing the obstruction and allowing unimpeded air-flow. However, tracheostomy is not without risks, such as the development of infection or fatal accidental decannulation. In addition, tracheostomy care requires an enormous amount of commitment and cooperation among family members, and also involves a certain financial cost. For these reasons, alternative procedures have been developed to treat laryngomalacia.

Iglauer, in 1922, was the first to describe partial epiglottectomy for severe laryngomalacia, and several subsequent studies have proposed various techniques of excision of supraglottic tissues for this condition. The functional value of supraglottoplasty was studied prospectively by Marcus et al., who showed polysomnographic evidence that obstructive sleep apnea resolved in 4 of 6 patients following supraglottoplasty. These studies and others described the bilateral excision of supraglottic tissues, with only a few complications reported in the literature, including the description of 2 infants who developed supraglottic stenosis by Solomons and Prescott, and a description of interarytenoid scarring in 1 patient by Jani et al. The first article to systematically analyze results in patients undergoing unilateral supraglottoplasty was put forth by Kelly and Gray in 1995. They had a 94% success rate and no major complications. Three (17%) of their 18 patients required a contralateral supraglottoplasty at a later date owing to persistent laryngomalacia.

The obvious disadvantage of a staged contralateral supraglottoplasty is the need for a second general anesthetic, thereby subjecting the patient to the usual risks of pediatric airway surgery. This scenario would be ideally avoided. However, when deciding on unilateral vs bilateral procedures, one must consider the dire consequences of supraglottic stenosis, which is theoretically much more likely following a bilateral procedure. If the contralateral supraglottoplasty rate is not prohibitively higher than the revision or complication rate for bilateral supraglottoplasty, then the risk of a second airway procedure may be justifiable. Our contralateral supraglottoplasty rate was comparable to that in the study by Kelly and Gray.

Although our overall success rate was high for bilateral supraglottoplasty, and only 2 patients developed supraglottic stenosis, we have largely abandoned the bi-
lateral approach because of the severity of this complication in the few patients in which it occurs. Supraglottic stenosis leads to severe airway compromise that usually requires tracheostomy. The stenosis results from adhesion of opposing areas of excision and the resulting scar formation and can occur even when great care is taken not to excise directly opposing tissue sites or the interarytenoid mucosa. Because of the recalcitrant nature of supraglottic stenosis, decannulation in these patients is difficult, and sometimes impossible. Supraglottic stenosis also leads to impairment of airway protection and an increased potential for aspiration owing to limited motion as well as decreased sensation at the level of the supraglottic tissues. Therefore, supraglottoplasty, whether bilateral or unilateral, should involve excising only the minimal amount of tissue necessary to improve the airway. This is particularly true of the epiglottis, which serves a vital function in airway protection. In addition to the risk of supraglottic stenosis, overexcision of the epiglottis may result in frank aspiration. In our patients in whom the epiglottis was included in the supraglottoplasty, only the lateral aspect of the epiglottis was excised.

Unilateral supraglottoplasty avoids bilaterally opposing excision sites and, therefore, should not lead to supraglottic stenosis. It is a more conservative approach with excision of less overall tissue than the bilateral procedure. A disadvantage of this procedure, therefore, is the potential for excision of too little tissue to effect resolution of laryngomalacia. However, the unilateral procedure has been shown to have a high success rate in our study, and of those patients requiring a contralateral procedure, few complications have resulted. In addition, the risk of a revision procedure also exists for bilateral procedures, as evidenced by 3 patients in group 1. Thus, there is no guarantee of avoiding a second operation with the bilateral procedure.

Our unilateral supraglottoplasty population (group 2) was not without complications. Four patients developed postoperative aspiration not documented preoperatively. One of these patients had undergone a contralateral supraglottoplasty; aspiration in this case could be potentially due to overexcision of tissues. The other 3 patients had mild aspiration that resolved with medical therapy, including thickened feedings. Many patients in both groups had aspiration documented preoperatively but not postoperatively, and some had aspiration both preoperatively and postoperatively. The possibility remains that some patients with postoperative aspiration actually had undiagnosed preoperative aspiration. In any case, only 1 patient in group 2 developed postoperative aspiration severe enough to warrant a tracheostomy.

None of our study variables identified patients at risk of requiring a second operation. There was no indication that specific sites of excision affected the chance of requiring a revision or contralateral procedure. One would assume that a contralateral or revision procedure would be needed more often in cases in which too little tissue was initially excised; however, among the 7 contralateral supraglottoplasties, only 2 involved initial excision of just 1 supraglottic site (AEFs). There was also no correlation between the amount of tissue excised and the chance of supraglottic stenosis or aspiration. Of the 2 patients who developed supraglottic stenosis, one had only the AEFs excised; the other had the lateral epiglottis and AEFs excised.

Comorbidities also did not predict which patients were more likely to require revision or contralateral procedures. In fact, no subset of patients with any combination of comorbidities had an increased rate of revision procedures, and even in patients who had multiple comorbidities, there were no trends. Thus, the number and type of comorbid conditions did not predict which patients would develop complications or would need a revision or contralateral supraglottoplasty.

With regard to surgical technique, all 7 patients requiring a contralateral supraglottoplasty had undergone laser excision. However, this most likely reflects the parallel shift from bilateral to unilateral procedures and from microscissor to carbon dioxide laser excision over time, and probably does not reflect a cause-and-effect relationship between the two.

Finally, our study population consisted of a large number of patients who required surgical intervention for laryngomalacia. Although one criticism of our study may be that the threshold for surgical intervention might have been lower than usual to accrue such a large series of operative cases, our analysis indicates that all of the patients in our series did require supraglottoplasty because of the severity of their condition. All of the patients had signs of physiologic compromise secondary to laryngomalacia and airway obstruction as manifested by their primary presenting report and as supported by diagnostic studies. Most of these patients had already undergone intensive medical or surgical therapy with persistent laryngomalacia. For example, more than half of the patients with gastroesophageal reflux required a fundoplication procedure to control their reflux, with the remainder receiving medical therapy for at least several weeks. Patients with feeding difficulties and failure to thrive had been treated with feeding therapy and failed. Finally, patients with obstructive sleep apnea underwent trials of home monitoring, in some cases with continuous positive airway pressure or oxygen therapy, without success. By definition, all of our study patients had failed medical therapy. Therefore, surgical intervention was indeed a last resort in these patients. In addition, the overall number of patients undergoing supraglottoplasty in our study, on a per-year basis, compares with the numbers in some other studies.

A second potential criticism involves the inclusion of patients with synchronous lesions in our study. Some of these, most commonly hypertrophic adenoids and tonsils, were untreated until after supraglottoplasty had been performed. One could argue that the presence of these synchronous lesions at the time of supraglottoplasty may have contributed to laryngomalacia, and that treatment of the synchronous lesions may have been tried first. However, these patients still represented a minority of cases in our study. A larger subgroup of our study population included those patients who underwent combined surgery, including procedures for synchronous lesions at the same time as supraglottoplasty. Including these patients in the analysis clearly contaminates the data.
This study was designed to review our extensive experience with supraglottoplasty in patients with laryngomalacia and to determine the success, failure, and complication rates of unilateral and bilateral supraglottoplasty, as well as to determine any factors that would be predictive of complications or the need for revision supraglottoplasty for persistent laryngomalacia. We found that none of the study variables could predict the need for revision supraglottoplasty or the development of complications. Our overall success rate was high, and our complication rate was low, for both groups. None of our patients undergoing a contralateral supraglottoplasty experienced complications attributable to the second procedure. Therefore, avoiding the severe consequences of supraglottic stenosis may well be worth the risk of subjecting a patient to a second procedure following unilateral supraglottoplasty. Based on the results of our study, we believe that unilateral supraglottoplasty should be the initial procedure for patients with severe laryngomalacia requiring surgery.

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### REFERENCES