Evaluation of the Efficacy of Supraglottoplasty in Obstructive Sleep Apnea Syndrome Associated With Severe Laryngomalacia

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Objective: To evaluate the clinical and polysomnographic evolution of patients with severe laryngomalacia who underwent supraglottoplasty.

Design: Retrospective study.

Setting: University hospital.

Patients: Seven children with severe laryngomalacia.

Main Outcome Measures: The patients were evaluated with a questionnaire given to their parents and with polysomnographic examination before and 3 months after surgery. The clinical data of respiratory and swallowing symptoms as well as the parameters of minimum oxygen saturation and respiratory disturbance index were evaluated after bilateral supraglottoplasty and compared with the preoperative data.

Results: Two patients had pharyngolaryngomalacia and required tracheotomy. Four patients had a marked improvement of respiratory and deglutition symptoms. Polysomnographic data showed a significant improvement in the respiratory disturbance index after surgery ($P<.05$) but not in the minimum oxygen saturation level. However, this improvement was only partially achieved in 3 patients, in whom there were associated airway or neurologic changes. No serious surgical complications were observed.

Conclusion: Supraglottoplasty led to a marked improvement in all 5 patients without pharyngolaryngomalacia, but the 2 patients with pharyngolaryngomalacia required tracheotomy.


LARYNGOMALACIA (LM) IS characterized by obstruction of the upper airways, especially during inspiration, owing to collapse of the arytenoid mucosa and of the epiglottis toward the glottic lumen, which is associated with shortening of the aryepiglottic wall. These changes occur in different degrees and in different combinations. Laryngomalacia is the most common laryngeal malformation (60%-70% of cases), and it is the main cause of stridor in neonates and children. The cause of LM is not fully understood, but this condition is believed to occur mainly because of lack of neuromotor coordination and because of anatomical alterations. Gastroesophageal reflux is an important cofactor in causing edema of the laryngeal mucosa and in increasing the obstruction of the laryngeal lumen. In turn, because of an increase in intrathoracic pressure, LM can facilitate the presence of gastroesophageal reflux.

The predominant symptom of LM is stridor, which starts at birth or during the first 15 days of life and becomes more marked when the infant is agitated, cries, feeds, or is in the supine position. In general, stridor worsens during the first 6 to 8 months of life and then decreases, with spontaneous resolution occurring by 2 years of age. Some symptoms, such as dyspnea, apnea, costal or suprasternal retractions, difficulty in swallowing, failure to thrive, and delay in neuropsychomotor development, may also eventually be associated with LM.

The diagnosis, which is confirmed by flexible fibroscopy in the outpatient setting, reveals an omega-shaped epiglottis, a shortened aryepiglottic wall, and redundant mucosa in arytenoid regions, with prolapse of these structures toward the glottic lumen. These changes may occur separately or in combination, the most common being the association of shortening of the aryepiglottic wall and a redundant arytenoid mucosa.

Laryngomalacia may occur separately or in association with other malformations (in 17%-47% of cases), either in the airways or in other organs, such as the caen-
Central nervous system and the heart. Among the most common airway malformations are pharyngolaryngomalacia (PLM), tracheomalacia, vocal fold paralysis, and subglottic stenosis. Pharyngolaryngomalacia is defined by the lack of neuromotor coordination in the entire pharynx and larynx, with collapse of all the structures involved. The predominant symptoms are respiratory difficulty and marked difficulty in swallowing, with frequent choking and aspiration. If PLM is diagnosed, in view of the lack of success in cases in which supraglottoplasty is performed, tracheotomy may be indicated.

In 10% to 15% of LM cases, the symptoms may be more intense, with the condition being called severe LM, and surgery is necessary. The severity of LM is not related to the intensity of stridor or to its frequency but rather to the presence of associated symptoms. Therefore, the indications for surgical treatment are (1) presence of resting dyspnea or intense effort dyspnea; (2) obstructive sleep apnea syndrome (OSAS); (3) hypoxia or hypercapnia; (4) pulmonary hypertension or cor pulmonale; (5) considerable difficulty in swallowing; (6) marked difficulty in gaining weight; (7) failure to thrive; and (8) delay in neuropsychomotor development.

At present, in cases of severe LM, the surgical procedure of choice is supraglottoplasty or aryepiglottoplasty. In most cases, the findings of outpatient endoscopic examination direct the surgery to the site that is predominantly involved. Thus, the most common procedures, often performed in combination, are incision of the aryepiglottic wall, resection of redundant arytenoid mucosa (at times with resection of the cuneiform cartilages), and suture of the lingual surface of the epiglottis to the base of the tongue (epiglottopexy). These procedures can be performed either with microscissors, carbon dioxide laser, or microdebriders, and no differences in results have been reported between these techniques.

Despite its highly encouraging results, supraglottoplasty may present some complications, among them synchiae, granulomas, aspirations, and supraglottic stenoses. In view of the severity of this last condition, which may be very difficult to treat, some authors recommend only the incision of the aryepiglottic wall, while others propose unilateral supraglottoplasty alone. The disadvantage of these less invasive procedures is the greater chance that a second endoscopic procedure may be needed. The objective of the present study was to evaluate retrospectively in an objective manner the improvement of symptoms in patients with severe LM who underwent supraglottoplasty, with special emphasis on the respiratory symptoms determined by polysomnography (PSG).

**METHODS**

The study was conducted on patients with LM who were followed up at University Hospital, Faculty of Medicine of Ribeirão Preto, University of São Paulo, Ribeirão Preto, Brazil, from June 2002 to December 2004. The patients underwent a clinical examination and flexible fibroscopy for the diagnostic confirmation of severe LM. The patients then underwent PSG, which was performed throughout the nocturnal period (a 7-hour examination on average). During this examination, specific emphasis was placed on changes related to OSAS, especially the score on the obstructive respiratory disturbance index (RDI), including apnea and hypopnea, the mean and minimum oxygen saturation during sleep, and signs of paradoxical respiration, in addition to changes in cardiac rhythm. Obstructive sleep apnea syndrome was diagnosed and graded as severe, moderate, or mild, according to Katz et al. After confirmation of OSAS, the patients underwent direct laryngoscopy to determine the degree of collapse and the structures involved, as well as eventual associated glottic and subglottic changes. Next, supraglottoplasty was performed bilaterally, with emphasis on the anatomical alterations observed by flexible outpatient fibroscopy. The procedure was performed with cold material, with a bilateral incision of the aryepiglottic wall, followed by bilateral exeresis of excess mucosa in the lateral arytenoid region. After this procedure, the epiglottis was examined and, if it was posteriorized, which occurred in 1 case, epiglottopexy was performed; in this case, an incision was made on the lingual surface of the epiglottis, and then the epiglottis was attached to the base of the tongue with absorbable polyglactin 910 sutures (3-0 Vicryl; Ethicon Inc, Somerville, NJ). After surgery, the patients were taken to the pediatric intensive care unit and remained intubated for 24 hours on average. During the postoperative period, they received amoxicillin, bromopride, and prednisolone for an average period of 1 week.

Body weight was measured at the time of surgery and compared with the ideal weight for age and sex according to the z score (measured weight minus reference weight divided by the SD of the reference weight for age and sex). The z score is the method recommended by the World Health Organization for the nutritional evaluation of children. Approximately 3 months after surgery, the patients again underwent PSG for comparison of the indices with preoperative values. The Wilcoxon test was used for comparison of preoperative and postoperative PSG data, with the level of significance set at P<.05.

Seven patients, 4 boys and 3 girls, were evaluated. The mean age at diagnosis was 6.82 months (range, 1-15 months), and the mean age at surgery was 7.14 months (range, 1-17 months). Four of the 7 patients had a history of stridor, and in the 3 cases without stridor the predominant obstructive symptom was snoring. All children had a history of cyanosis on effort and nocturnal dyspnea or apnea (Table). Three of the 7 patients had evident symptoms of aspiration, with 2 of them requiring a nasogastric tube for feeding. Three patients had associated neurologic alterations (neuropsychomotor retardation [n=3] and agyria [n=1]). No cardiac changes were observed. All patients had a marked weight deficit, with 6 having a z score of less than –2 and 4 of the 6 having a z score of less than –3.

Flexible fibroscopy revealed an omega-shaped epiglottis, a shortened aryepiglottic wall, and redundant arytenoid mucosa in all cases. Associated conditions were marked posteriorization of the epiglottis (n=1), intra-arytenoid stenosis (n=1), tracheomalacia (n=1), and marked collapse of the entire pharynx and larynx (PLM) (n=2). Preoperative PSG diagnosed moderate (n=1) or severe (n=6) OSAS in all patients, with paradoxical respiration, an RDI ranging from 5.4 to 22.8 (mean±SD, 11.66±7.51), and minimum oxygen saturation ranging

**RESULTS**

Preoperative PSG diagnosed moderate (n=1) or severe (n=6) OSAS in all patients, with paradoxical respiration, an RDI ranging from 5.4 to 22.8 (mean±SD, 11.66±7.51), and minimum oxygen saturation ranging...
from 70% to 94% (mean ± SD, 81.71% ± 8.47%). No changes in cardiac rhythm were observed during the examination in any of the patients. After confirmation of OSAS, the patients underwent supraglottoplasty. The extubation was unsuccessful in the 2 girls with PLM, and both patients required tracheotomy.

Of the 5 patients who were successfully extubated, 4 showed marked clinical improvement of the respiratory symptoms and 1 showed only partial improvement of apnea and stridor. The 2 patients with associated feeding difficulties showed marked improvement, with the nasogastric tube being successfully removed during the postoperative period. These 5 patients were submitted to postoperative PSG an average of 82 days after surgery, showing marked improvement of RDI (a mean of 10.0 during the preoperative period vs 2.2 during the postoperative period, P < .05) (Figure 1) and a tendency to improved minimum oxygen saturation (83.2 during the preoperative period vs 86.4 during the postoperative period, P = .07) (Figure 2).

In 3 patients, the postoperative RDI did not completely normalize and remained above 1, although a significant improvement was seen when compared with the preoperative RDI. Patient 3 had associated tracheomalacia; patient 4 had a marked neurologic deficit; and patient 2 had hypertrophy of the pharyngeal and palatine tonsils. In all 3 cases, fibroscopy revealed considerable improvement of the laryngeal changes. The first patient, the only one who still had respiratory symptoms, underwent treatment with continuous positive airway pressure, while the other two, who had no symptoms, were simply kept under clinical observation. The other patients, even though they were asymptomatic, underwent postoperative fibroscopy, which demonstrated an improved laryngeal lumen in all cases. One of the children developed discrete supraglottic stenosis in the intra-arytenoid region, but her symptoms improved, and she had no respiratory or digestive repercussions. She has been periodically examined by outpatient fibroscopy for 2 years, and her stenosis has remained unchanged.

The main symptom of LM is stridor, being present in as many as 100% of cases in some reports. In the present study, all 7 children had cyanosis and apnea, 4 had stridor, and 3 had a history of snoring. Indeed, in 1 case, there was a delayed diagnosis of LM because hypertrophy of the pharyngeal tonsils had been diagnosed at an outside institution. The supraglottic site was the main point of obstruction observed in our group, which may explain the high frequency of snoring, instead of stridor, among these children.

Severe LM may be associated with a failure to thrive, retarded neuropsychomotor development, or feeding problems such as aspiration and difficulty in swallowing. At least 1 of these symptoms was present in all pa-
tients in our study, and the association between them was common. The z score was less than –2 in 6 of the 7 children, confirming the marked weight deficit usually observed in this group of patients. There was an association with other airway malformations in 4 children, and 3 of them had neurologic changes. No cardiac changes were seen in this group. According to the literature, the most common associated alterations are airway and neurologic malformations.4,11,12,15,19

Some authors advocate the use of bronchoscopy in all children with LM to determine possible associated airway malformations. We, as well as Onley et al,20 believe that this procedure should be performed only in children who have more severe symptoms or whose symptoms are incompatible with the endoscopic findings. The fibroscopic findings (omega-shaped epiglottis, shortened aryepiglottic wall, and redundant arytenoid mucosa) were similar to those described in the literature7,8,11 and were systematically detected in all patients. In 1 child, narrowing of the larynx occurred mainly in an anterior region, and she underwent epiglottoplasty1 in conjunction with supraglottoplasty. In the other cases, the narrowing was mainly posterior, and the simple incision of the aryepiglottic wall and bilateral resection of the mucosa in an arytenoid region was sufficient to provide larger laryngeal lumina.

All of our patients had apnea confirmed by PSG, which was performed at night during spontaneous sleep for a minimum of 7 hours. On the basis of the RDI and oxygen saturation findings, the apnea was considered to be moderate to severe in all patients. Respiratory disorders detected on the basis of PSG findings or changes in gasometry were also reported by others.11,16,17,21

Of the 7 patients who underwent surgery, 2 (29%) had PLM and required tracheotomy. This percentage is similar to that reported by Froehlich et al,13 who observed PLM in 33% of their patients with severe LM. Our 2 patients have been followed up for approximately 1 year, and they still need tracheotomy for adequate ventilation. Froehlich et al13 and Roger et al11 also reported a lack of success with supraglottoplasty in PLM, with their patients requiring tracheotomy or bilevel positive airway pressure for improvement of respiratory symptoms.

Four of the 5 patients in our study who underwent successful intubation showed marked improvement of respiratory symptoms; 1 child continued to have occasional complaints of stridor and apnea. This favorable outcome was similar to that reported by others.17,19,21 In contrast, there was a marked but partial improvement in the PSG findings in the 3 patients with associated tracheomalacia and severe hypoxic encephalopathy, stressing the importance of multifactorial causes for the occurrence of symptoms in these patients.

Regarding complications, discrete interarytenoid stenosis was seen in 1 case, with no respiratory or digestive repercussions and no other complications or aspirations being observed. In the case with persistent symptoms, the lack of clinical improvement was attributable to the associated alterations rather than to surgical failure. We believe that this solely minor complication is the result of exhaustive care on the part of the surgeon not to remove an excessive amount of mucosa, especially in areas of contact, and to avoid extensive manipulation of the larynx.

Denoyelle et al12 and Reddy and Matt18 reported the occurrence of supraglottic stenosis in approximately 4% of their patients. Other minor complications (such as granulation tissue and synechiae) have also been reported. These case reports involving bilateral surgery were essential for greater care in the management of these procedures. Reddy and Matt18 advocated the advantage of unilateral surgery for a reduction of the rate of complications, especially supraglottic stenosis. However, we believe that bilateral surgery, if performed with extreme care, may present a low risk of complications, with a high rate of resolution or an marked improvement of symptoms.

Postoperative PSG confirmed a marked improvement of symptoms, with a significant decrease in RDI and a tendency to an increased minimum oxygen saturation. Marcus et al21 observed a marked improvement of hypoxia and hypercapnia in 6 patients who underwent epiglottoplasty. However, their PSG study was performed for a short period of only 2 hours during the daytime, which might have impaired their findings.

Denoyelle et al12 reported that partial or complete failure of surgery is related to the presence of associated alterations, especially neurologic and syndromic ones. Our cases with an unsatisfactory outcome had associated tra-
cheomalacia or neurologic changes. Therefore, supraglottoplasty seems to be an effective surgical procedure, with low morbidity, for the treatment of OSAS associated with severe LM.

CONCLUSIONS

A marked improvement of respiratory symptoms and PSG parameters may be achieved with the use of supraglottoplasty in children with severe LM. Nevertheless, when PLM is diagnosed, supraglottoplasty seems to be ineffective, and tracheotomy may be considered as a treatment option in such cases.

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