Supraglottoplasty for Laryngomalacia with Obstructive Sleep Apnea

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Abstract

Objective: To determine if supraglottoplasty is effective in reversing abnormal respiratory patterns in children with laryngomalacia and obstructive sleep apnea.

Study Design: Retrospective case series at a tertiary referral children’s hospital.

Methods: Ten patients with laryngomalacia and obstructive sleep apnea as documented by polysomnography underwent supraglottoplasty between 2005 and 2007. Data collected included age, findings on flexible and rigid endoscopy, type of procedure performed, and postoperative course. The preoperative polysomnograms were reevaluated to identify which patients had episodes of obstructive apnea index (OAi), obstructive apneas/hypopneas index (OAIH), respiratory disturbance index (RDI), and low arterial oxygen saturation (O2 sat) after supraglottoplasty.

Results: All ten patients were successfully extubated following supraglottoplasty. There were no postoperative complications, and no patient required a subsequent intubation procedure. Each patient had a postoperative nocturnal polysomnogram performed following supraglottoplasty at 11 weeks (range 2-29 weeks). Caregivers reported mild improvement (70%), significant improvement (20%), and complete resolution (10%) of stridor and nocturnal snoring at a follow-up visit four weeks following discharge. Associated improvements were observed in OAi, OAIH, RDI, and O2 sat nadir, of which all were statistically significant (P < 0.05).

Conclusions: Polysomnography should be considered in the initial evaluation of infants with moderate laryngomalacia to rule out obstructive sleep apnea. Supraglottoplasty is an effective treatment for infants with laryngomalacia and obstructive sleep apnea. The improvements in the procedure outweigh the low morbidity. Improvement after surgery was reliably confirmed by polysomnography in this study.

Introduction

Laryngomalacia is the most common cause of stridor in infants.1,2 It is caused by intermittent upper airway obstruction secondary to supraglottic collapse. The disorder presents in the first two weeks of life with stridor which increases with agitation or when the child is placed supine. The symptoms generally worsen during the first six to eight months, after which they gradually resolve spontaneously.3 Most children with laryngomalacia are symptom-free by two years of age.

Obstructive Sleep Apnea (OSA), affecting approximately 1% of children, is characterized by periodic upper airway obstruction and associated apneas and hypopneas.4,5 Previous studies have described sleep-disordered breathing and polysomnographic (PSG) abnormalities in children with laryngomalacia.6 It would seem obvious that patients with laryngomalacia would have varying degrees of obstruction, some of which may require active intervention. However, to the best of our knowledge there are currently no definitive recommendations regarding the role of PSG in children with laryngomalacia. We present our initial experience investigating apneas seen and treating with PSG in infants with moderate laryngomalacia performing SGP for those with OSA. Success was objectively measured by improvement in postoperative respiratory parameters.

Methods

A retrospective study was conducted of infants seen at Children’s Memorial Hermann Hospital between March 2005 and March 2007 who had surgical intervention for laryngomalacia. Inclusion criteria were a diagnosis of moderate laryngomalacia, polysomnographic confirmation of OSA, surgical intervention with SGP, and a postoperative PSG. Moderate laryngomalacia was diagnosed by the degree of stridor, associated cough and expectoration, and findings on flexible endoscopy in the absence of failure to thrive, cyanotic spells, and sign of severe airway obstruction at rest. The surgical procedure consisted of a telescopic laryngoscopy, bronchoscopy, and supraglottoplasty to eliminate the supraglottic fuses, either alone or with unilateral excision of redundant mucosa and the cartilage framework. The postoperative PSG was generally scheduled at the first follow-up visit, approximately 4 weeks after the procedure.

Results

Ten patients met criteria for inclusion in the study. The mean age of the patients at presentation was 4.3 months (range 1-6 months) and there were 8 males and 2 females. All 10 patients underwent an uncomplicated SGP and had complete pre- and postoperative PSGs. Comparison of pre- and postoperative PSGs are included in Table I. All patients had evidence of residual obstructive apneas, T1, and severe obstructive apneas judged by the degree of oxygen desaturation, and 5 patients had evidence of residual obstructive apneas along with unilateral excisions of the conus cartilage and redundant mucosa. All 10 patients were successfully extubated following SGP. There were no perioperative or postoperative complications, and no patient required a subsequent intubation procedure. Additionally, no patient required postoperative positive pressure ventilation.

Each patient had a postoperative nocturnal PSG following SGP at 11 weeks (range 2-29 weeks). Caregivers reported mild improvement (70%), significant improvement (20%), and complete resolution (10%) of stridor and nocturnal snoring at a follow-up visit four weeks following discharge. Associated improvements were observed in OAi, OAIH, RDI, and O2 sat nadir, of which all were statistically significant (P < 0.05) (Table II, Figure II).

Table I: Preoperative Polysomnography

Table II: Postoperative Polysomnography

Table III: Comparison of Pre- and Postoperative Polysomnography

Figure I: Postoperative Stridor

Figure II: Comparison of Pre- and Postoperative Polysomnography

Discussion

Indications for surgical intervention in severe laryngomalacia have been previously reported.1 However, to the best of our knowledge, there have been no recommendations for limited SGP in a treatment for moderate laryngomalacia with OSA. Furthermore, there are no definitive recommendations regarding the role of PSG in children with laryngomalacia.

Looking at preoperative sleep studies in the present study (Table I), 90% of patients would be categorized as severe or moderate OSA according to this classification system. Improvement of OSA documented by PSG has been previously described in a small number of patients after SGP for severe laryngomalacia.6-8 In the current investigation, both subjective and objective measurements confirmed significant improvement in sleep disordered breathing following limited SGP. Comparison of pre- and postoperative PSG revealed a 72% decrease in OAi, a 66% decrease in OAIH, and a 74% decrease in RDI (Table III). O2 sat increased by 10% (P > 0.001), and 16% of apneas reported significant or complete resolution of stridor four weeks postoperatively.

While complications of SGP including synchiae, granuloses, aspiration, and supraglottic stenosis have been reported,9-11 simple supraglottic resection and unilateral excision of redundant mucosa and cartilage has been associated with significant complications.11 There were no perioperative or postoperative complications in the current study, with 49% of SGP’s performed by simple division of the supraglottic fuses and 66% with unilateral excision of redundant mucosa and the cartilage framework. Limitations of the current report are that retrospective design, small sample size, and absence of a control group.

However, it can be argued that the differences between pre and postoperative measurements may be attributable to known spontaneous resolution of most cases of laryngomalacia by 12 to 24 months of age.12 However, it seems unlikely that such dramatic resolution of symptoms would occur over an average 16-week period without intervention. We feel that the low morbidity of a limited SGP along with the significant improvement in respiratory parameters observed after surgery justifies its role in laryngomalacia with OSA.

While there are no standard criteria for defining severe laryngomalacia that requires surgical intervention, dysphagia at rest and/or severe dyspnea with exertion, poor feeding, failure to thrive, and uncontrollable gastroesophageal reflux have been previously described as indications for supraglottoplasty.5 Infants with characteristic anatomical findings of moderate laryngomalacia to rule out obstructive sleep apnea. Supraglottoplasty is an effective treatment for infants with moderate laryngomalacia and obstructive sleep apnea. The improvements in the procedure outweigh the low morbidity. Improvement after surgery was reliably confirmed by polysomnography in this study.

Conclusion

OSA represents an overlooked component of laryngomalacia. Moderate laryngomalacia associated with OSA has potentially detrimental consequences with respect to infant growth and development. The current study highlights the potential benefits of PSG to be part of the initial diagnostic workup of moderate laryngomalacia. If PSG confirms OSA, it could potentially influence or modify the surgical management of moderate laryngomalacia and its associated respiratory patterns in infants with laryngomalacia and OSA. Based on our initial experience with a limited number of patients, we conclude that infants with laryngomalacia and OSA diagnosed by PSG benefit significantly from a limited SGP.

References