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Utility of polysomnography in determination of laryngomalacia severity☆

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ABSTRACT

Objective: To examine the efficacy of polysomnography in determining the severity of laryngomalacia in pediatric patients.
Methods: Prospective cohort study. Pediatric patients referred to our pediatric otolaryngology department with a polysomnogram already performed for a presumptive diagnosis of laryngomalacia were enrolled in the study. Patients with concurrent airway lesions or neuromuscular disorders were excluded. Patients underwent history, physical exam, and flexible fiberoptic laryngoscopy. These results were used to calculate a total laryngomalacia severity score.
Results: 25 pediatric patients ($n = 25$) with an average age of 3.9 months at time of initial evaluation met criteria for enrollment in our study. 100% of patients had obstructive sleep apnea by definition. 80% of these patients underwent supraglottoplasty. The average AHI of those who underwent surgery (57.26) was not significantly different in those who underwent surgery vs. those that did not (55.43) ($p = 0.41$). In comparison, the average laryngomalacia severity score based from history, physical exam and flexible laryngoscopy was significantly greater in the patients that required supraglottoplasty (11.16) vs. those who did not (5.33) ($p = 0.03$). In addition a higher laryngomalacia severity score was not correlated with a higher AHI ($p = 0.81$, $r = 0.08$, CI: -0.5197 to 0.6235).
Conclusion: In our cohort, polysomnography was not useful in determining the severity of laryngomalacia, did not correlate with the clinical evaluation, and alone was not predictive of the patients that would require surgical intervention. History, physical exam, and endoscopic findings remain reliable predictors of disease severity and need for operative intervention.

1. Introduction

Laryngomalacia represents the single most common cause of stridor in infants. Flexible fiberoptic laryngoscopic examination remains the cornerstone for diagnosis, revealing characteristic findings of collapse of supraglottic structures. Holinger et al. further defined these features, including one or more of the following: 1) complete or near-complete collapse of supraglottis concomitant with stridor during inspiration, which may obstruct glottis visualization with short aryepiglottic folds; 2) anterior prolapse of arytenoid and possibly corniculate cartilages; and 3) Posterior prolapse of epiglottis which may assume a tubular structure; an omega shaped epiglottis in isolation is not necessarily pathologic [1].

Numerous factors have been identified which contribute to the clinical presentation of laryngomalacia. One finding is edema, which may result from gastroesophageal or pharyngolaryngeal reflux disease. Edema increases turbulent airflow, and subsequent increased negative intrathoracic pressure promotes worsening GERD, thereby perpetuating the cycle [2]. Concurrent airway abnormalities including laryngeal dyskinesia, vocal cord paralysis, subglottic stenosis, and tracheomalacia are present in approximately 79% and 28% of cases of “severe” and “mild” laryngomalacia, respectively [3].

Though the single greatest etiological factor in pathogenesis of laryngomalacia has been long disputed, many authors have argued in favor of the role of neuromuscular hypotonia [4]. In short, competent laryngeal function represents an interplay between

☆ Original prospective data collected at Ochsner Clinic Foundation, Department of Pediatric Otolaryngology.

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peripheral sensory afferent reflexes, brainstem function, and motor efferent responses to the larynx. It has been demonstrated that in children with laryngomalacia, the laryngopharyngeal sensory testing thresholds, whose input results from sensory stimulation of the mechano- and chemoreceptors in the region of the aryepiglottic folds, are elevated compared with those of controls. This decreased sensitivity, likely resulting from neurological immaturity, results in less input to the brainstem, less motor output, and subsequently diminished tone of the upper aerodigestive tract [5].

Given the role of poor neuromuscular tone in the pathogenesis of obstructive sleep apnea (OSA), it is conceivable that patients with severe laryngomalacia demonstrate some degree of sleep-disordered breathing. The laryngeal contribution to obstructive sleep apnea has been the subject of increasing investigation. Powitzky et al. examined pre- and post-supraglottoplasty values for apnea-hypopnea index (AHI), obstructive apnea index, obstructive AHI, O_2 saturation nadir, and percentage of sleep spent with <90% O_2 saturation in 20 infants initially diagnosed with moderate to severe laryngomalacia. The authors found that supraglottoplasty created statistically significant improvements in AHI. Obstructive apnea index, O_2 saturation nadir, and percentage of sleep spent with <90% O_2 saturation also improved, though not to a statistically significant degree. When subjects were stratified according to concurrent reflux treatment, age at supraglottoplasty, and lag time between supraglottoplasty and postoperative polysomnography (PSG), these findings were upheld. In addition, postoperative AHI scores correlated with improvements in stridor in 80% of patients [6].

In a retrospective case series of 10 infants who underwent PSG before and after supraglottoplasty for moderate laryngomalacia, Zafereo et al. documented statistically significant improvement in all PSG parameters, including obstructive AHI, respiratory disturbance index (RDI), and O_2 saturation nadir. Moreover, caregivers reported mild improvement (10%), significant improvement (70%), and complete resolution (20%) of stridor and nocturnal snoring at four-week post-operative visits [7].

Taken collectively, these findings underscore the fact that obstructive sleep apnea may contribute significantly to laryngomalacia severity and may help guide decisions for surgical intervention. Conversely, comprehensive evaluation of obstructive sleep apnea should take possible laryngeal contribution into consideration. While existing literature has examined the efficacy of supraglottoplasty in the treatment of laryngomalacia in patients with coexisting obstructive sleep apnea, no study, to our knowledge, has examined the efficacy of PSG for the determination of laryngomalacia severity. The present study seeks to examine this relationship.

2. Material and methods

2.1. Study design

A prospective cohort study was conducted at Ochsner Clinics Foundation with approval of the Institutional Review Board. Inclusion criteria were patients who underwent PSG prior to referral for evaluation for presumptive diagnosis of laryngomalacia between January 2012 and October 2014. Patient demographics can be found in Table 1. Indications for polysomnography including snoring, stridor, sleep-disordered breathing, and were often multifactorial; many patients with abnormal PSGs were referred to our center, only those patients with stridor were included in this study. PSG interpretation was performed by pediatric pulmonologists. Subjects were subsequently evaluated for presence and severity of laryngomalacia using history, physical exam, and flexible fiberoptic laryngoscopic exam. Direct laryngoscopy/bronchoscopy was performed if the child ultimately underwent supraglottoplasty. Subjects with synchronous airway lesions, neuromuscular lesions, or history of tracheostomy dependence for other indications were excluded. History, physical exam, and laryngoscopic findings were used to score laryngomalacia severity, as described by Sivan et al. and detailed in Tables 2 and 3 [12]. The laryngomalacia severity score (LSS) was calculated by adding the scores from the history (Table 2) and physical exam (Table 3) together.

Table 1
Patient demographics.

Patient Characteristics		
Total	**(n)**	**(%)**
	23	100
Sex		
Male	16	69.5
Female	7	30.4
Age at initial evaluation	**Months**	
Mean	7.08	
Median	4.0	
Surgical intervention		
Supraglottoplasty	16	69.57
None	7	30.43
Obstructive sleep apnea classification		
Mild	1	4.35
Moderate	2	8.70
Severe	20	86.96

2.2. Patients

23 patients met inclusion criteria consisting of 7 females and 16 males with average age at time of initial evaluation of 7.1 months and range of 1.5–54 months (Table 1). The vast majority (21 out of 23) patients were under 9 months of age, with a median age of 4 months. All patients underwent complete PSG prior to evaluation and severity scoring for laryngomalacia.

2.3. Statistical methods

Patient data, including demographic information, history and physical, PSG, and operative reports were kept on a password-protected laptop. Statistical analyses were performed using GraphPad Software Prism 7(La Jolla, CA 2016). Mean polysomnographic findings between those who underwent supraglottoplasty for laryngomalacia and those who did not were compared using a chi-squared test. Average laryngomalacia severity scores for those who did and did not undergo surgery were also compared using unpaired t-tests. Linear regression analysis was used to determine correlation between AHI and laryngomalacia severity scores.

3. Results

The pediatric patients included in the study ($n = 23$) had an average age of 7.1 months at time of initial evaluation. 95.7% (22/23) of study participants had moderate to severe obstructive sleep apnea, by definition [8]. 69.6% (16/23) of study participants subsequently underwent supraglottoplasty. Recommendation for supraglottoplasty was made in those patients with cyanosis, failure to thrive/inability to feed, severe stridor necessitating multiple hospitalizations, as well as documentation of classic flexible fiberoptic laryngoscopy findings of supraglottic collapse, arytenoid prolapse, and/or posterior prolapse of the epiglottis. While all children with severe obstructive sleep apnea were offered supraglottoplasty and counseled on adverse long term effects of