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Late-onset laryngomalacia: A cause of pediatric obstructive sleep apnea☆

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ABSTRACT

Objective: To describe the presentation, diagnosis, and treatment of late-onset laryngomalacia in children with obstructive sleep apnea syndrome (OSAS).
Design: Retrospective study.
Setting: Tertiary care children's hospital.
Patients: Seventy-seven children were identified who had OSAS diagnosed by polysomnography and underwent airway endoscopy to evaluate for laryngomalacia between July 2006 and December 2008. Children with significant neurologic disease or craniofacial malformations were excluded. Seven children under 3 years of age had laryngomalacia and OSAS (Group A), 19 children 3–18 years of age had laryngomalacia and OSAS (Group B), and 51 children 3–18 years of age had OSAS but not laryngomalacia (Group C).
Main outcome measures: Comparison of pre-operative findings, intra-operative findings, interventions, and outcomes between the 3 groups.
Results: Group A was consistent with previous reports of congenital laryngomalacia with respect to presentation, diagnosis, and treatment. Groups B and C had similar pre-operative findings, including a high incidence of adenotonsillar hypertrophy, and the only significant difference was the intra-operative finding of laryngomalacia in Group B. Treatments were individualized to include supraglottoplasty, adenoidectomy, tonsillectomy, adenotonsillectomy, or a combination of the above. Of the 52 patients who returned in follow-up, 44 noted improvement, but this was rarely confirmed by polysomnogram.
Conclusions: Late-onset laryngomalacia may act alone or in concert with additional dynamic or fixed lesions to cause pediatric OSAS. Although there is no specific pre-operative indicator to diagnose late-onset laryngomalacia, it can be readily identified intra-operatively and effectively treated with supraglottoplasty, with or without concurrent adenotonsillectomy.
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1. Introduction

Obstructive sleep apnea syndrome (OSAS) is a syndrome of partial or complete upper airway obstruction occurring during sleep and causing a disruption of normal ventilation and sleep patterns. While this may occur in patients of any age, the classic description of pediatric OSAS is in a child 2–8 years old with symptoms of loud snoring, witnessed apneas, frequent nighttime arousals, and chronic mouth breathing [1]. Ten percent of children have some form of sleep disordered breathing (including primary snoring) and 1–3% of children have obstructive sleep apnea [2]. These numbers are important because pediatric OSAS is associated with neurocognitive, cardiovascular, and metabolic sequelae that can have lasting consequences throughout the child's life. School performance suffers due to behavioral problems, developmental delay, and inattention, while overall health is damaged by systemic inflammation and metabolic derangements, all of which contribute to a poor quality of life for the child. Major risk factors for OSAS in children include adenotonsillar hypertrophy, obesity, neuromuscular disease, and craniofacial anomalies [3–5].

Pediatric OSAS is often attributed to adenotonsillar hypertrophy, yet research suggests the cause is often multifactorial in nature, with decreased neuromuscular tone during REM sleep also playing a large role [1,6,7]. Despite a recognition that the cause of obstruction is multifactorial, adenotonsillectomy continues to be the most common intervention, supported by studies citing improvements in behavior [8] and quality of life [9] and overall cure rates of 75–100% after adenotonsillectomy [10]. As larger studies with better levels of evidence emerge, however, it is now clear that adenotonsillectomy is not a cure-all for pediatric OSAS. Tauman et al. [11] studied 110 patients and found that while all showed improvement in sleep parameters after adenotonsillectomy, only 25% had complete cure of OSAS (defined as an apnea

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hypopnea index (AHI) < 1). Mitchell [12] reported on 79 non-obese, otherwise healthy children with OSAS, all of whom underwent pre- and post-operative polysomnograms. Sixteen percent of children had persistent disease when defined by an AHI greater than or equal to 5 and 27% of children had persistent disease after adenotonsillectomy when defined by a respiratory distress index (RDI) of greater than or equal to 1. The largest and most recent study by Bhattacharjee et al. [13] retrospectively evaluated the efficacy of adenotonsillectomy in 548 children with OSAS. Although 90.1% of patients showed a reduction in AHI, only 27.2% had complete normalization of their breathing patterns during sleep after adenotonsillectomy. Patients more likely to have persistent disease were greater than 7 years old, had a higher body mass index (BMI), had a higher pre-operative AHI, and had asthma. Looking specifically at obese patients, Mitchell and Kelly [14] found that 76% of obese patients had persistent disease after adenotonsillectomy. The results of these studies support the notion of OSAS as being multi-factorial in nature and raise the question of what additional upper airway pathology, whether fixed or dynamic, is contributing to OSAS in children.

Congenital laryngomalacia is a well-described dynamic lesion of the larynx, causing collapse of the supraglottic structures during inspiration. It is the most common congenital laryngeal abnormality, presenting with inspiratory stridor during the first 2 weeks of life, worsening over the next 6–8 months, and resolving by 2 years of age [15]. In this patient population, only 10% of affected patients require intervention due to complications such as failure to thrive, obstructive sleep apnea sydrome, resting dyspnea, hypoxia or hypercapnea, pulmonary hypertension, or cor pulmonale [16]. More recently, variations of this typical presentation have been reported, including state-dependent and late-onset laryngomalacia. State-dependent laryngomalacia was described by Smith et al. [17] after identifying 5 patients between the ages of 3 and 4 years old who were asymptomatic while awake, but experienced stridor and airway collapse during sleep. This was thought to be secondary to decreased neuromuscular tone in the sleeping child, causing it to be missed on an office exam including flexible fiberoptic laryngoscopy. Late-onset laryngomalacia was described by Richter et al. [18] in patients older than 2 years of age with sub-categories of feeding-disordered, sleep-disordered, and exercise-induced laryngomalacia. Seven school-aged children presented with sleep disordered breathing, five of whom had already undergone adenotonsillectomy without resolution of symptoms. They were all identified as having sleep-disordered laryngomalacia and improved after treatment with supraglottoplasty.

As more data emerges about pediatric obstructive sleep apnea syndrome, more questions arise regarding the pathophysiology of this disease and the appropriate work-up, intervention, and follow-up of these patients. Airway endoscopy has not historically been a part of the initial work-up for pediatric OSAS, so it is possible that late-onset laryngomalacia is present in a number of these patients but is not being recognized. Using the results of routine airway endoscopy in children with OSAS, this study aims to increase awareness of late-onset laryngomalacia as a cause of pediatric OSAS and to compare the presentation, diagnosis, and management of these patients to that of younger children with congenital laryngomalacia and to age-matched children with obstructive sleep apnea but not laryngomalacia.

2. Materials and methods

With approval of the institutional review board, a retrospective study was conducted on patients treated in the pediatric otolaryngology clinic at Christus Santa Rosa Children's Hospital (San Antonio, TX) for laryngomalacia and/or obstructive sleep apnea between July 2006 and December 2008. Only patients with obstructive sleep apnea diagnosed by polysomnogram and airway endoscopy performed by the senior author to confirm or deny the diagnosis of laryngomalacia were included. Patients with significant craniofacial malformations or neurologic disorders were excluded. Data was collected from inpatient and outpatient electronic and paper charts to include history and physical exam findings, polysomnogram results, operative reports, operative photos and videos, and post-operative course.

Seventy-seven patients met inclusion criteria for the study. For the purposes of this study, a cut-off of 3 years of age was used to separate congenital from late-onset laryngomalacia, allowing for division of the patients into three distinct groups. Group A (congenital laryngomalacia) contained patients ages 0–3 years old with obstructive sleep apnea and laryngomalacia, Group B (late-onset laryngomalacia) contained patients ages 3–18 years old with obstructive sleep apnea and laryngomalacia, and Group C (classic pediatric OSAS) contained patients ages 3–18 years old with obstructive sleep apnea but without laryngomalacia.

Pre-operative data points included sleep symptoms, medical comorbidities, weight, presence of stridor and/or retractions, and tonsil and adenoid size. Patient weights were plotted on CDC growth curves and standard formulas were used to calculate the growth percentile for each patient. For patients under 24 months of age, weight to length percentiles were used to determine if the patient was underweight (<5th percentile), normal weight (5th to 95th percentile), or overweight (>95th percentile). Patients over 24 months of age were assessed based on body mass index percentiles and divided into underweight (<5th percentile), normal weight (5th to 85th percentile), overweight (85th to 95th percentile), and obese (>95th percentile) categories [19]. Tonsil size was graded from 1 to 4 with size 1 tonsils hidden within the pillars, size 2 tonsils extending to the edge of the pillars, size 3 tonsils extending beyond the pillars, and size 4 tonsils meeting in the midline. Adenoids were simply described as obstructing or non-obstructing. Mild laryngomalacia was diagnosed if the supraglottic collapse was intermittent or led to incomplete obstruction of the laryngeal inlet while severe laryngomalacia was diagnosed if the collapse was persistent with complete obstruction. All patients underwent pre-operative full-night polysomnogram, most of which were conducted by a pediatric pulmonologist at the same children's hospital. Respiratory events were scored based on American Academy of Sleep Medicine guidelines [20] and disease was categorized as mild (AHI 1–5), moderate (AHI 5–10), or severe (AHI > 10) for each patient.

Direct laryngoscopy and tracheobroncoscopy were performed with each patient spontaneously ventilating under intravenous anesthesia. A propofol drip was started at 200–250 mcg/kg/min and titrated to achieve deep sedation. Direct laryngoscopy was then performed with a Parson's laryngoscope, a weight-appropriate dose of 2% lidocaine was applied to the vocal cords, and then tracheobronchoscopy was performed with a Hopkin's rod telescope. During this period, observations were made regarding adenotonsillar size, cobblestoning of the tracheal mucosa, and presence of any other airway pathology. Once this had been completed, the laryngoscope was kept in the vallecula while the anesthesiologist turned off the propofol, allowing for evaluation of dynamic laryngeal pathology as the patient began to awaken from anesthesia and produced vigorous inspiratory efforts. During this period, the aryepiglottic folds were observed for collapse with inspiration allowing for diagnosis of mild or severe laryngomalacia or no laryngomalacia.

Indications for supraglottoplasty in a child with polysomnogram-proven OSAS included severe laryngomalacia with or without adenotonsillar hypertrophy or mild laryngomalacia

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