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RESEARCH ARTICLE

Sleep Disordered Breathing May Signal Laryngomalacia

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Abstract:

Background:

Pediatric anesthesiologists are often confronted with children with sleep-disordered breathing (SDB) presenting for tonsillectomy and/or anesthesia. The typical patient has symptoms of obstructive sleep apnea (OSA) with enlarged tonsils; however, a subset of patients may have underlying laryngomalacia (LM) without tonsillar hypertrophy. Both OSA and LM significantly increase the risk of intra- and postoperative airway obstruction and sensitivity to narcotics. The prevalence of LM may be underestimated, because direct laryngoscopy (DL) is not routinely performed in the diagnostic evaluation of patients with SDB who lack tonsillar hypertrophy.

Aim:

To identify the prevalence and DL findings in pediatric patients with SDB without tonsillar hypertrophy.

Methods:

Retrospective chart review of 108 patients with SDB who underwent general anesthesia for adenotonsillectomy (TA) or adenoidectomy with concomitant DL. The following data were collected: demographic information, medical comorbidities, polysomnography results, anesthetic techniques, and postoperative complications.

Results:

94.5% of children had DL findings consistent with LM, including a retropositioned epiglottis and short aryepiglottic folds. Postglottic edema was observed in 42.2%, and these patients were significantly more likely to have a diagnosis of gastroesophageal reflux (P=0.023). 57.8% had vocal cord edema. 75.3% of children who received routine postoperative follow-up care experienced complete symptom resolution. Postoperative complications following discharge from hospital occurred in 12.4% of patients, and 15.7% underwent supraglottoplasty for continued SDB symptoms after TA or adenoidectomy.

Conclusion:

A substantial proportion of patients with SDB who lacked tonsillar hypertrophy had findings consistent with LM, suggesting that the larynx may be the primary site of upper airway obstruction in these patients. This has significant implications in terms of perioperative management. The majority of patients with SDB had symptomatic improvement following TA or adenoidectomy; however, a subset required further surgical intervention with supraglottoplasty.

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1. INTRODUCTION

Sleep-disordered breathing (SDB) is a general term that encompasses a broad range of disorders featuring aberrant respiratory patterns during sleep. Best known among SDB is obstructive sleep apnea (OSA), in which repeated episodes of upper airway obstruction result in oxygen desaturations and sleep fragmentation [1]. The overall prevalence of OSA in the pediatric population is estimated to range from 1 to 6% [2]. Obese children are disproportionately affected, with over 50% showing symptoms of this condition [3, 4]. The underlying pathogenesis has not been fully elucidated but is believed to be multifactorial with potential causes including excessive adiposity, upper airway abnormalities such as adenotonsillar hypertrophy, and sleep-dependent reductions in neuromuscular tone that lead to airway obstructions [5]. Intra- and postoperative airway obstruction and sensitivity to opioid pain medication are well described in these patients [2]. OSA confers an increased risk of cardio- and cerebrovascular morbidity and mortality, metabolic sequelae, neurocognitive impairments, and perioperative complications [6, 7]. Adenotonsillectomy (TA) remains the surgical treatment of choice for children with SDB and is curative in up to 80% of cases [8].

Laryngomalacia (LM), another breathing-related disorder that stems from abnormalities in the airway, is the most common congenital laryngeal disease of infancy. It results from the collapse of supraglottic structures during inspiratory phase, which leads to obstructed airflow and resultant stridor [9]. Symptoms of LM are classically less pronounced during sleep [10]. Patients may experience feeding difficulties with subsequent failure to thrive [11]. LM was traditionally believed to occur as a consequence of intrinsic abnormalities in the anatomy of the laryngeal cartilage; however, recent evidence has suggested a possible neurologic component [12 - 14]. Most cases are self-limiting and can be managed without surgical intervention; however, nearly 20% of patients require surgical treatment with supraglottoplasty [15]. Airway collapse can occur after extubation, due to the loss of the airway stent effect created by the endotracheal tube and positive pressure ventilation, leading to increased risk for airway complications [16].

Sleep-dependent laryngomalacia (SDL) is a variant of LM, first described by Amin in 1997 [17]. Patients with this form of LM exhibit more prominent obstructive symptoms during sleep and may be asymptomatic while awake. This condition tends to present later in life than typical LM [10, 17]. Its prevalence has not been described [10].

Recent literature has suggested that a small subset of children with symptoms of OSA may have underlying LM, or at least laryngeal collapse that has the same pattern, although etiology and natural history are different [18, 19]. Little attention has been paid to laryngeal abnormalities in patients with SDB who lack tonsillar hypertrophy, and thus may have obstruction elsewhere in the airway. Accordingly, the purpose of this study was to evaluate the frequency of direct laryngoscopy (DL) findings consistent with LM – and thus potential airway obstruction at the laryngeal level – among children without tonsillar hypertrophy who underwent TA or adenoidectomy.

2. METHODS

Following approval from our Institutional Review Board, we conducted a retrospective chart review of pediatric patients without tonsillar hypertrophy (tonsils 1-2+ in size as determined by the Brodsky scale) [20] on preoperative physical examination, who had undergone TA or adenoidectomy with concomitant DL for SDB. All surgical procedures were performed by the senior author at our institution between 2011 and 2016. A database query was performed in order to identify potentially eligible patients. The tonsil size was recorded before and during surgery. All patients received general anesthesia with mask induction, with Sevoflurane, nitrous oxide and oxygen, followed with IV insertion. Spontaneous ventilation was maintained during the direct DL. A Parsons laryngoscope was used to visualize the larynx including the subglottis, and a 0-degree telescope used for photo documentation. In all cases, the laryngoscope was withdrawn along the tongue base to evaluate whether the epiglottis fell backward into the airway. There was no jaw thrust performed. Signs of potential laryngeal obstruction, including postglottic mucosal redundancy, length of aryepiglottic folds and epiglottic position were evaluated. After the DL, the patients were intubated with an appropriate size oral RAE tube. All patients received 1 mcg/kg fentanyl, ondansetron and dexamethasone. DL findings were documented via a standardized template.

After surgery, the patients were extubated when extubation criteria were met and were subsequently transported to the post anesthesia care unit (PACU) for recovery. Exclusion criteria were as follows: patients for whom preoperative tonsil size was not recorded in the electronic medical record, patients without sufficient data available in the electronic medical record, and patients who did not undergo DL simultaneously with TA or adenoidectomy.

The following data were reviewed for each patient: age, sex, body mass index (BMI), comorbidities (including a history of asthma/bronchopulmonary dysplasia, gastroesophageal reflux, developmental delay, prematurity, craniofacial anomalies, or syndromic conditions), method of diagnosis for SDB; polysomnography (PSG) data, including apnea-hypopnea index (if available, number of obstructive events, oxygen saturation (nadir), tonsil and adenoid size, date of TA or adenoidectomy and DL, procedures performed, DL findings (including retropositioned epiglottis/short aryepiglottic (AE) folds, vocal cord edema, postglottic edema, tracheal cobblestoning, and subglottic stenosis), time to emergence from anesthesia, airway obstruction in PACU, PACU stay time, SDB symptom resolution, visits to the Emergency Department (ED) within the first two weeks following surgery, complications following the procedure (including hemorrhage, upper airway obstruction, dehydration, infection, and velopharyngeal insufficiency), and need for supraglottoplasty.

Patients were classified as underweight if their BMI was less than or equal to the 5th percentile, overweight if their BMI was greater than or equal to the 85th percentile but less than the 95th percentile, and obese if their BMI was greater than or equal to the 95th percentile.

Patients' AHI was defined as normal if there was less than 1 event/hour, mild SDB if there was greater than or equal to 1 event/hour but less than or equal to 5 events/hour, and severe SDB if greater than 5 events/hour but less than 10 events/hour, and severe SDB if greater than or equal to 10 events/hour [2].

Data were entered into a Microsoft Excel spreadsheet and imported into SPSS 22 (IBM, Armonk, NY) for analysis. Descriptive statistics were computed to evaluate the frequency of demographic variables, comorbid conditions, preoperative tonsil and adenoid size, and DL findings.

Mann-Whitney U and Kruskal-Wallis tests were performed to evaluate for associations between BMI percentile category (normal weight and overweight/obese) and tonsil and adenoid size, AHI, medical comorbidities, DL findings, symptom resolution, postoperative complications, ED visits within two weeks postoperatively, and need for supraglottoplasty. These comparisons were also made for age, the DL finding of a retropositioned epiglottis/short AE folds, and the DL finding of postglottic edema.

3. RESULTS

A total of 108 patients met inclusion criteria for this study. Mean patient age was 6 ± 0.4 years (mean ± standard error) with a range from 0.9 years to 15.5 years, and gender distribution 70% (*n* = 76) male and 30% (*n* = 32) female. 8.3% (*n* = 9) of children were classified as overweight, and 29.4% (*n* = 32) were obese. 33.9% (*n* = 37) of patients had a comorbid diagnosis of asthma/bronchopulmonary dysplasia, and 25.7% (*n* = 28) had comorbid reflux. Additional patient characteristics are reported in Table **1**.

Table 1. Patient characteristics.

Parameters	Characteristic	Number of Subjects	Percentage of Subjects (%)
Gender	Male	76	70.4
	Female	32	29.6
BMI percentile category	Underweight	6	5.5
	Normal weight	62	56.9
	Overweight	9	8.3
	Obese	32	29.4
Age	< 2 years	16	14.7
	2 to < 5	39	35.8
	5 to < 10	32	29.4
	10 and older	22	20.2
Comorbid conditions	Asthma / bronchopulmonary dysplasia	37	33.9
	Gastroesophageal reflux	28	25.7
	Developmental delay	12	11.0
	History of prematurity	8	7.4
	Craniofacial anomaly	5	4.6
	Syndromic condition	3	2.8
Procedure performed	Adenotonsillectomy	84	77.1
	Adenoidectomy only	24	22.0

No significant differences were encountered between overweight or obese children (*n* = 41) and children of normal weight (*n* = 62) for age, sex, tonsil and adenoid size, PSG results, medical comorbidities, DL findings, symptom resolution, postoperative complications, ED visits within the first two weeks postoperatively, or need for supraglottoplasty. PSG was performed in 61.5% (*n* = 67) of children, which revealed an AHI of 10.8 ± 1.2 as well as 59.9 ± 7.8 obstructive events with an average oxygen saturation nadir of 83% ± 10%. 38.5% (*n* = 42) of children did not undergo PSG and were identified as surgical candidates based on clinical history only.

With respect to age-related differences, younger children (age < 5 years) were significantly more likely to have a larger adenoids (*P* = 0.001). There were no significant differences between age groups for tonsil size, PSG results, medical comorbidities, DL findings, symptom resolution, postoperative complications, ED visits within the first two weeks postoperatively, or need for supraglottoplasty.

94.5% (*n* = 103) of patients were found to have a retropositioned epiglottis/short, AE folds consistent with LM on DL. Postglottic edema was observed in 42.2% (*n* = 46) of children. Additional DL findings are reported in Table **2**. Patients with postglottic edema were significantly more likely to have a diagnosis of gastroesophageal reflux (*P* = 0.023). No other significant differences were encountered for this variable. There were no significant differences between patients with a retropositioned epiglottis/short AE folds and sex, tonsil and adenoid size, PSG results, medical comorbidities, symptom resolution, postoperative complications, ED visits within the first two weeks postoperatively, or need for supraglottoplasty.

Table 2. Direct laryngoscopy findings at time of TA or adenoidectomy.

DL Finding	Number of subjects	Percentage of subjects (%)
Retropositioned epiglottis / short aryepiglottic folds	103	94.5
Vocal cord edema	63	57.8
Postglottic edema	46	42.2
Tracheal cobblestoning	32	29.4
Subglottic stenosis	3	2.8

Average time from surgery end to exit from the OR was 16 minutes (*n* = 42), and PACU time was on average 1 hour and 1 minute (*n* = 42). There were no Intensive Care admissions required post-op. Twelve point eight percent (*n* = 14) sought treatment in the ED during the first two weeks after surgery. Ten percent (*n* = 11) reported postoperative complications: 2 patients experienced hemorrhage, 5 patients reported dyspneic symptoms consistent with upper airway obstruction, and 4 patients were dehydrated.

Complete resolution of SDB symptoms (as determined by clinical history elicited from parent/guardian) occurred in 75.3% (*n* = 67) of patients who received follow-up care. Outcomes following TA or adenoidectomy with DL are summarized in Table **3**. 12.8% (*n* = 14) of patients ultimately underwent supraglottoplasty for persistent SDB complaints, with resolution in 64.3% (*n* = 9) of patients. Post-supraglottoplasty PSGs were available for only two of these patients. In one patient, a preoperative PSG had not been performed; however, the postoperative AHI was 3.6, showing evidence of mild persistent SDB. In the other patient, the preoperative AHI was 30.6 and decreased to 20.5 postoperatively showing improved but persistent severe SDB.

Table 3. Outcomes after TA or adenoidectomy with DL.

Outcome	Number of subjects	Percentage of subjects (%)
Visited emergency department in first 2 weeks postoperatively	14	12.8
Lost to follow-up	20	18.3
Postoperative complications*	11	12.4
Complete resolution of sleep-disordered breathing symptoms*	67	75.3
Underwent supraglottoplasty*	14	15.7
Symptomatic improvement following supraglottoplasty	9	64.3

*Expressed as a percentage of the 89 total patients that received routine follow-up care in our outpatient otolaryngology clinic.

4. DISCUSSION

The most important finding from this study is that an overwhelming majority (94.5%) of pediatric patients with symptoms of SDB who lacked tonsillar hypertrophy on physical examination had findings consistent with potential laryngeal obstruction. This suggests that the larynx can be the primary site of airway obstruction in children with a history of SDB symptoms without enlarged tonsils, and could account for airway obstruction in the immediate post-operative period. This can impact post-anesthesia care and result in difficulty managing the airway.

The frequency of diagnostic findings consistent with LM in children with SDB has been variable in the literature. Thevasagayam *et al.* reported a prevalence of 3.9% of LM findings as determined by sleep nasopharyngoscopy among 358 children presenting with SDB symptoms [19]. The substantial difference in prevalence between this study and ours is likely attributable to their inclusion of patients with tonsillar hypertrophy. Revell *et al.* also examined the presence of airway findings consistent with LM using airway endoscopy for 19 patients with coincident diagnoses of LM and OSA. 89% of their patients were found to have tonsillar hypertrophy, and the most commonly encountered LM finding was redundant mucosa of the aryepiglottic folds [18].

Supraglottoplasty is the preferred intervention for LM causing SDB, and significant improvements in PSG parameters have been observed in children with LM following this procedure [21, 22].

Surprisingly, our finding showed that TA actually appeared to be effective despite the presence of LM, as 75.3% of our patients who received routine outpatient follow-up reported a subjective resolution in symptoms following surgery. This is consistent with the range of previously reported success rates of 27-83% for TA in the treatment of SDB [17].

Our primary finding calls into question the optimal treatment approach for these patients, however, patients with upper airway abnormalities such as LM and no tonsillar hypertrophy may be exposed to the risks of tonsillectomy without strong expectation of improvement in their airway obstruction following surgery [8].

Among the 22 children for whom TA or adenoidectomy was unsuccessful at SDB symptom alleviation, 14 underwent subsequent supraglottoplasty, with 9 patients reporting symptomatic improvement following this procedure. The most commonly cited reason for opting against supraglottoplasty in patients who failed TA or adenoidectomy was parental preference to avoid further surgical intervention.

The underlying airway abnormalities in patients with SDB who lack tonsillar hypertrophy pose important considerations in terms of perioperative management. The risk of perioperative complications is greatly increased in patients with SDB undergoing all surgical procedures [23]. Previous studies have demonstrated that primary care providers lack satisfactory knowledge of pediatric sleep disorders and do not regularly screen patients for OSA symptoms [24 - 26]; therefore, it is crucial for pediatric anesthesiologists to include these questions as part of their preoperative assessment to identify patients at high risk of postoperative airway obstruction. Anesthetic technique and airway management must be tailored to reducing the potential for airway obstruction during and after anesthesia (e.g., upright or lateral decubitus positioning, jaw thrust maneuvers, positive pressure administration, and placement of a nasal or oral airway). Awareness of SDB symptoms can also influence postoperative analgesia choices, as these children may be more prone to respiratory depression from opioids [27].

We also found two additional statistically significant associations worth mentioning. First, we encountered a correlation between postglottic edema and a diagnosis of gastroesophageal reflux. This finding is consistent with prior study by Carr *et al.* in which 69% of pediatric patients diagnosed with reflux were observed to have postglottic edema on DL [28]. We also observed a statistically significant link between increased adenoid size in younger children, which coincides with prior literature demonstrating atrophy of the adenoid with age [29].

Our study has inherent limitations worth noting due to its retrospective nature. Additionally, although the timeframe during which patients were sampled was relatively long, the methodology was standardized by using the same anesthesia technique, equipment, perioperative routines, operative approaches, and documentation methods throughout the study's duration. Typically, diagnosis of an upper airway extra-laryngeal obstruction is done with flexible transnasal laryngoscopy – evaluating the nose until the vocal cords. We used DL to identify findings in children with sleep disordered breathing so that anesthesiologists would be aware that post-op obstruction could be related to collapse at the larynx. This could result in more difficulty intubating the patients post-operatively should it become necessary.

CONCLUSION

A substantial proportion of pediatric patients with sleep-disordered breathing without significant tonsillar hypertrophy have DL findings consistent with potential laryngeal obstruction. Thus, the larynx may be the primary site of upper airway obstruction in children with this presentation, and this obstruction will persist after surgery. It is crucial for pediatric anesthesiologists to be aware of this association, as recognition of upper airway obstruction preferably prior to surgery, but also in the perioperative period may ensure timely and appropriately management of these patients.

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ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was approved by the Institutional Review Board at the Penn State Milton S. Hershey Center.

HUMAN AND ANIMAL RIGHTS

No animals were used for the studies that are base of this research. All clinical investigations were conducted in accordence with the declaration of Helsinki principles.

CONSENT FOR PUBLICATION

Not applicable.

CONFLICT OF INTEREST

The authors declare no conflict of interest, financial or otherwise.

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