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Supraglottoplasty for sleep endoscopy diagnosed sleep dependent laryngomalacia

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ABSTRACT

Objectives: To evaluate the polysomnographic outcomes of supraglottoplasty (SGP) performed for sleep endoscopy diagnosed sleep dependent laryngomalacia as treatment for obstructive sleep apnea syndrome (OSAS).
Methods: Nine subjects aged 6–55 months underwent supraglottoplasty for sleep dependent laryngomalacia. All subjects underwent both pre- and post-procedural polysomnograms.
Results: Supraglottoplasty for sleep dependent laryngomalacia resulted in improvement of OSAS as measured by collective improvements in 8 different primary polysomnogram parameters: apnea–hypopnea index (AHI), minimum (nadir) and mean oxygen saturation, mean and maximum carbon dioxide, total sleep time, sleep efficiency, arousal index, as well as improvement in weight for length percentiles. Subjects had a significant 80% decrease in percentage change in AHI ($p < 0.005$), with decrease in mean AHI from 23.4 to 4.8 following supraglottoplasty. Seven of 9 subjects demonstrated improvement in nadir saturations, 6 of 9 subjects had improvement in sleep efficiency, and 7 of 8 subjects under 4 years of age had improvement in weight for length percentile.
Conclusions: Supraglottoplasty for sleep dependent laryngomalacia is an effective treatment of OSAS, and can be readily diagnosed using sleep endoscopy. Further investigation is warranted to increase awareness and outcomes related to sleep dependent laryngomalacia.

1. Introduction

Laryngomalacia, the prolapse of supraglottic structures into the glottic airway, is the most common laryngeal disease of infancy [1]. Laryngomalacia often presents as snoring, stridor, and difficulty feeding; in most cases the condition self-resolves as the child develops. Classically, the airway obstruction associated with laryngomalacia is less pronounced during sleep. However, some infants may develop sleep disturbance related to a certain variant of laryngomalacia [2–4]. This condition, termed sleep dependent laryngomalacia, is characterized by an increased work of breathing related to supraglottic collapse, most acute during sleep, and occasionally by its relatively late onset. The prevalence of sleep dependent laryngomalacia is currently unknown. Thus children with sleep dependent laryngomalacia often go on to have frank airway obstruction during sleep, consistent with Obstructive Sleep Apnea Syndrome (OSAS). OSAS in infants has been associated with failure to thrive, cognitive and behavioral deficits, and sudden infant death [1,5,6]. The etiology of laryngomalacia is typically determined via clinical history and exam, flexible fiber-optic laryngoscopy in clinic, or sleep endoscopy. The gold-standard diagnosis of OSAS is via overnight polysomnography (PSG) [1]. Surgical treatment of laryngomalacia is warranted only in severe cases (i.e. failed medical, dietary and behavioral modifications), and is indicated when symptoms include poor weight gain, episodes of respiratory distress, and OSAS [1]. Supraglottoplasty is the standard surgical intervention, and is defined as any procedure that modifies the supraglottis to alleviate the obstruction [1,7]. Supraglottoplasty in patients with sleep dependent laryngomalacia has been suggested to improve OSAS; however more investigations utilizing objective polysomnographic metrics are necessary to stratify and optimize patient outcomes [1,3,8,9]. The aim of this study was to review the impact of supraglottoplasty for children with sleep dependent laryngomalacia and OSAS, with the

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hypothesis that supraglottoplasty resulted in marked improvement in the severity of OSAS.

2. Methods

2.1. Subjects

Subjects from ages 6 to 55 months at the time of supraglottoplasty were identified retrospectively in one of two ways: by extraction from an institutional sleep endoscopy database from 2011 to 2013, or identified by Current Procedure Terminology coding (31540, 31541, and 31599). Of the subjects who had laryngomalacia diagnosed on direct airway visualization, those who subsequently underwent supraglottoplasty and who had both pre- and postoperative PSG were ultimately included in this review. Subjects' medical records were reviewed, and data from pertinent demographic, medical and operative histories, as well as PSG results, were entered into an Excel database. This study was approved by the institutional review board of Seattle Children's Hospital (SCH) (Seattle, Washington) and was conducted in compliance with the Healthcare Information Portability and Accountability Act.

2.2. Sleep endoscopy and supraglottoplasty

These procedures were done as part of routine clinical care. Sleep endoscopy was performed using a Pentax FNL-RP3 nasopharyngolaryngoscope under general anesthesia with "total intravenous anesthesia" technique. Patients were maintained in spontaneous ventilation in supine position on continuous propofol intravenous drip. Laryngomalacia was diagnosed if during inspiratory phase, supraglottic structures were noted to prolapse into the laryngeal introitus, obstruct respiratory flow and generate perturbation. Once diagnosed, all subjects underwent standard supraglottoplasty with division of aryepiglottic folds, redundant arytenoid mucosa reduction, and sparing of inter-arytenoid groove mucosa. Supraglottoplasty was performed using carbon dioxide laser, microdebrider, or micro-scissor technique by one of four fellowship trained pediatric otolaryngologists.

2.3. Polysomnography

PSGs were also obtained as part of routine clinical care, and performed at the American Academy of Sleep Medicine (AASM) accredited Pediatric Sleep Disorders Center at Seattle Children's Hospital. PSG data were collected on the Rembrandt® (Buffalo, New York) or XLTEK® (Oakville, Ontario, Canada) systems. All studies were interpreted by board certified sleep physicians using AASM scoring criteria [10]. Both thermistors and nasal pressure transducers were used. End-tidal carbon dioxide was used interchangeably with transcutaneous carbon dioxide monitoring for the purpose of measuring ventilation. The AHI was calculated using the total number of obstructive apneas plus obstructive hypopneas per hour of sleep. The minimum oxygen saturation is reported as the oxygen nadir. The arousal index was defined as the number of 3-s cortical arousals per hour of sleep. For subjects who underwent split night titration studies, only the baseline diagnostic portions of the PSG are included in the results.

2.4. Data analysis

Analyses were largely descriptive, and correction for multiple comparisons was not done on this relatively small sample of retrospective data. Paired *t*-tests were used to compare preoperative and postoperative metrics within subjects, including AHI, nadir oxygen saturations, mean oxygen saturations, mean and maximum carbon dioxide, total sleep time, sleep efficiency, arousal index, and weight for length percentile. One sample *t*-tests were used to assess whether the percent change in each outcome metric was significantly different than the null hypothesis that there was no change between pre- and postoperative states. Significance was set using a two-tailed $p < 0.05$. Logistic regression was run between continuous variables and correlations were examined to see if any preoperative characteristics were predictive of postoperative change. Statistical analyses were done on STATA 12® (College Station, TX).

3. Results

Initial search using CPT codes, as well as institutional sleep endoscopy database from 2011 to 2013, yielded 183 subjects. Of these 183 subjects who were between ages 6–55 months, 28 underwent supraglottoplasty, 18 of which were for sleep dependent laryngomalacia. Ultimately, nine subjects aged 6–55 months met the inclusion criteria of supraglottoplasty for sleep dependent laryngomalacia with pre- and postoperative polysomnograms; 7 of these subjects were diagnosed via flexible fiber-optic endoscopy. Two subjects were diagnosed via rigid telescope. Four out of 9 subjects underwent adenotonsillectomy immediately prior to supraglottoplasty.

Five of the 9 subjects were male and median age was 17 months (range, 6–55 months) at time of supraglottoplasty. Seven of 9 subjects (78%) had significant medical comorbidities, including agenesis corpus callosum (1), dysmyelination disorder (1), vocal cord paresis (1), and autism (1); 6 of 9 subjects were noted to have developmental delay, and 8 of the 9 subjects were documented to have some form of hypotonia. A summary of demographic and operative characteristics is provided in Table 1.

All children underwent PSG prior to supraglottoplasty, with the study occurring a mean of 91 days prior to surgery. All children also underwent polysomnography following surgery, with a mean of 155 days following the procedure. Collectively, all nine subjects had evidence of OSAS on preoperative study, as defined by an AHI of >1.5 [11]. If a more conservative definitions of OSAS is used of an AHI >5, then 8 out of 9 fell into this range.

As a group, subjects demonstrated improvement following PSG in each of the eight polysomnographic metrics: AHI, nadir saturation, mean oxygen saturation, mean and maximum carbon dioxide, total sleep time, sleep efficiency, and arousal index, though only comparison of AHI reached statistical significance on paired *t*-test. Subjects also improved comparing preoperative and postoperative weight to length percentiles. Subjects had a significant 80% decrease in percentage change in AHI ($p < 0.005$), with decrease in mean AHI from 23.4 (range, 2.3–109.7) to 4.8 (range, 2.2–9.2), as demonstrated in Fig. 1. Furthermore, 6 of 8 subjects transitioned from an elevated AHI into a commonly accepted "normal range" of AHI ≤ 5, with the ninth subject having no change (started and ended with an AHI < 5). Other sleep metric improvements are outlined in Table 2. The 8 subjects under the age of 4 (cutoff age for CDC weight for length percentile) demonstrated a 115% improvement in weight

Table 1
Summary of patient demographic and operative characteristics.

Characteristic	All subjects
Median age at time of surgery, m	17
Subjects, no	9
Male:female ratio	5:4
Comorbid conditions, no. (%)	7 (78%)
Sleep endoscopy performed at time of surgery, no. (%)	7 (78%)
Days between pre-op polysomnography and SGP, mean	70
Days between post-op polysomnography and SGP, mean	184