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Polysomnography outcomes of sleep endoscopy–directed intervention in surgically naïve children at risk for persistent obstructive sleep apnea

Erin Kirkham[1,2] · Cheng-Cheng Ma[3] · Natalia Filipek[3] · David L. Horn[3,4] · Kaalan Johnson[3,4] · Maida L. Chen[4,5] · Sanjay R. Parikh[3,4]

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Abstract

Purpose Drug-induced sleep endoscopy (DISE) is useful in children with obstructive sleep apnea (OSA) that persists after adenotonsillectomy (AT), but its utility in surgically naïve children is unclear. We report polysomnography outcomes of surgically naïve children who underwent DISE-directed intervention because they were considered high risk for persistent OSA after adenotonsillectomy.

Methods This study is a case series of 62 surgically naïve children with OSA who were considered high risk for persistence after AT and underwent DISE-directed intervention with pre- and postoperative polysomnography between 2012 and 2016. Analysis was performed with the paired *t* test.

Results Children were on average 5.9 (± 5.5, 0.2–18.6) years old at the time of surgery, 68% male, 18% obese, and 60% white. Thirty-eight percent had a syndromic diagnosis: 19% trisomy 21, 11% hypotonic neuromuscular disorder, and 8% craniofacial condition. The remaining 62% were non-syndromic but underwent DISE because they had at least one risk factor for OSA persistence after AT (age > 7 years, black race, 1+ tonsils, obesity, and/or severe OSA). Forty-two percent underwent AT, while 58% underwent treatment other than AT, including 18% who had multilevel surgery. Children improved significantly in 4 out of 5 polysomnography parameters tested, including obstructive apnea-hypopnea index (oAHI; 22.2 to 7.2, $p < 0.01$) and oxygen nadir (82 to 87, $p < 0.01$). Thirty-eight (61%) had a postoperative oAHI < 5; 16 (21%) had a postoperative oAHI < 2.

Conclusion DISE resulted in intervention other than AT in 58% of surgically naïve children at high risk for persistent OSA after AT. DISE-directed intervention resulted in significant mean improvement in postoperative OSA.

Keywords Sleep endoscopy · DISE · Pediatric · Sleep apnea

Introduction

Pediatric obstructive sleep apnea (OSA) syndrome is characterized by repetitive breathing pauses during sleep, resulting in sleep disruption and negative neurodevelopmental, cardiovascular, and behavioral consequences [1–4].

Adenotonsillectomy (AT) is the first-line treatment for children with OSA [5]. The Childhood Adenotonsillectomy Trial (CHAT) demonstrated that 79% of normal, healthy children ages 5–9 years will have polysomnographic resolution of their OSA after adenotonsillectomy [6]. However, on average, 40% of all children who undergo the procedure will have persistent sleep apnea, with rates of residual OSA up to 65% in children with comorbidities [7]. Persistent OSA after AT occurs in greater proportions in those with risk factors including age > 7 years [2, 8], severe baseline OSA (oAHI > 10) [2, 8–10],

✉ Erin Kirkham
ekirkham@umich.edu

1 Department of Otolaryngology: Head and Neck Surgery, The University of Michigan, Ann Arbor, MI, USA

2 Mott Children's Hospital, University of Michigan Medical Center, 1540 E. Hospital Dr. CW 5-702, SPC 4241, Ann Arbor, MI 48109, USA

3 Department of Otolaryngology: Head and Neck Surgery, The University of Washington School of Medicine, Seattle, WA, USA

4 Seattle Children's Hospital, Seattle, WA, USA

5 Department of Pulmonary and Sleep Medicine, Seattle Children's Hospital, Seattle, WA, USA

obesity [2, 7, 8, 11–15], trisomy 21 [16–18], craniofacial syndromes [19–21], and hypotonic neuromuscular disorders [10, 20]. Persistence frequently occurs when the tonsils and adenoids are not the only source of obstruction [22, 23]. Drug-induced sleep endoscopy (DISE) is a flexible fiber-optic assessment of the upper airway under a sleep-like state that facilitates targeted surgical intervention at specific sites of obstruction. DISE adds diagnostic benefit to awake endoscopy and clinical exam, and has been shown to influence surgical decision-making in many patients [24–27].

Previous study of DISE-directed intervention in *otherwise healthy* surgically naïve patients suggests that tonsillectomy with or without adenoidectomy is the DISE-directed intervention performed in the majority of these patients with a high percentage of success [28–30]. We sought to evaluate the utility of DISE in surgically naïve children with *risk factors for persistent OSA after AT* as there is little published on procedures performed and polysomnography outcomes in this often heterogeneous group of patients. We report the frequency with which the use of DISE resulted in a treatment plan that differed from standard-of-care AT. The primary aim was to evaluate the effectiveness of DISE-directed surgery in this series of surgically naïve children with OSA who underwent DISE prior to AT due to pre-identified risk factors for persistent OSA after AT. We hypothesized that there would be a significant improvement in five preselected polysomnography outcome measures, i.e., obstructive apnea-hypopnea index (oAHI, primary measure), apnea-hypopnea index (AHI), oxygen desaturation index (ODI), oxygen nadir (O_2 nadir), and percent of sleep time with end tidal $CO_2 > 50$ mmHg (%TST $CO_2 > 50$).

Methods

Data collection

Following approval by the Seattle Children's Hospital review board, institutional data was queried to identify patients who had undergone DISE-directed surgery along with pre- and postoperative polysomnography from February 2012 to September 2016. Chart review was performed on those identified to collect polysomnography parameters, DISE findings, DISE-directed surgeries performed, and demographic data including age at surgery, sex, race, preoperative tonsil size, previous airway surgery, sleep-related comorbidities, and perioperative body mass index (percentile for age). Data was stored in REDCap, a secure, web-based research database application [31].

All children included in the analysis underwent standard overnight, monitored pre- and postoperative polysomnography in the Seattle Children's sleep laboratory within 12 months before and within 24 months after DISE-directed surgery. No child underwent any additional surgery or continuous positive airway pressure (CPAP) treatment in the time between DISE-directed surgery and postoperative polysomnography. Polysomnography included electroencephalography, electromyography, electrooculography, electrocardiogram, and measurement of oronasal airflow by a nasal pressure cannula and thermistor, chest/abdominal excursion, capnography/transcutaneous CO^2, and pulse oximetry. Studies were scored by board-certified sleep medicine physicians using the pediatric scoring guidelines of the American Academy of Sleep Medicine [32]. Obstructive apnea was defined as ≥ 90% reduction in airflow lasting ≥ 2 breath cycles. Hypopnea was defined as a ≥ 30% reduction in airflow lasting ≥ 2 breath cycles, associated with an arterial desaturation ≥ 3% from baseline and/or a cortical arousal. AHI was defined as the number of apneas (obstructive and central) and hypopneas per hour of sleep. oAHI was defined as the number of obstructive apneas and hypopneas per hour of sleep. Oxyhemoglobin desaturation index was defined as the number of desaturations ≥ 3% from baseline per hour of sleep. Oxygen nadir (O_2) was the lowest level of oxygen saturation detected on pulse oximetry, and percent of total sleep time with end tidal carbon dioxide > 50 mmHg (%TST $CO_2 > 50$) was recorded by capnography. Mild OSA was defined as oAHI > 1 to < 5, moderate ≥ 5 to < 10, and severe ≥ 10 events per hour [33].

Children included in this analysis underwent DISE if they had an oAHI > 1, an AHI > 2, and significant risk factors for persistent OSA after AT. Children considered for DISE prior to AT fell into two groups: syndromic and non-syndromic. The syndromic group included children with trisomy 21, craniofacial conditions, and neuromuscular disorders with hypotonia as a prominent feature (referred to as "hypotonia" throughout). The non-syndromic group included otherwise healthy children that underwent DISE due to factors that increased their risk for persistent OSA after AT including the following: age greater than 7 years old, small tonsils (Brodsky 1+), obesity (> 95th percentile for age), and/or severe baseline OSA (oAHI ≥ 10 events/h) [8–10, 13, 16, 20]. DISE was performed by three surgeons (SP, KJ, DH) with the patient supine in the operating room under spontaneous breathing conditions. Patients underwent inhaled induction followed by maintenance propofol anesthetic without the use of topical anesthetics or decongestants. A validated scoring system was employed to grade obstruction at five levels, i.e., the adenoid, palate, lateral pharyngeal wall, tongue base, and supraglottis. Obstruction is scored at each level on a scale as follows: 0 (none), 1 (< 50% obstructed), 2 (> 50% obstructed), and 3 (complete) with a total score ranging from 0 to 15 [34].

Where appropriate, surgery was performed at the level of the adenoid, palatine tonsil, lingual tonsil, and/or supraglottis directly following sleep endoscopy. Surgical decision-making

was based on DISE findings in combination with individual patient factors and family preferences.

Analysis

Statistical analysis was performed with the STATA/MP 15 software (StataCorp LP, College Station, TX). Descriptive summaries for continuous variables are reported as ranges and means ± standard deviations. Frequencies are reported for categorical variables. Univariate associations between intervention type (AT vs non-AT) and baseline patient characteristics were tested with the χ^2 test, Fisher's exact test, or Wilcoxon rank-sum test, where appropriate. Univariate associations between intervention type (AT vs non-AT) and obstruction scores at each level and overall were compared using the Wilcoxon rank-sum test. Pre- and postoperative polysomnography parameters were statistically compared with the paired *t* test. The null hypothesis was rejected at $p < 0.05$.

Results

Of 117 children who underwent DISE from 2012 to 2016, 86 (74%) were surgically naïve. Of those, 62 (72%) had pre- and postoperative polysomnography data and were included in the analysis. Forty-two percent underwent adenotonsillectomy, while 58% underwent treatment other than adenotonsillectomy.

Baseline demographic characteristics are shown in Table 1. Children were on average 5.9 (± 5.5, 0.2–18.6) years old at the time of DISE, 68% percent male, 18% obese (≥ 95th percentile for age), and 60% percent white. The majority (82%) had Brodsky 2+ or smaller tonsils on preoperative examination; 58% had 1+ tonsils. The distribution of tonsil size was significantly different in the two intervention groups; children who underwent non-AT interventions had a significantly higher proportion of 1+ tonsils than those who underwent AT. Children who underwent AT had a significantly higher proportion of severe baseline OSA ($p = 0.01$) than those who underwent non-AT intervention. Twenty-four subjects (38%)

Table 1 Characteristics of children overall and by adenotonsillectomy (AT) vs non-AT intervention

	All subjects ($n = 62$)	Intervention		
		Non-AT ($n = 36$)	AT ($n = 26$)	
Characteristics	Mean (± standard deviation, range) or *n* (%)			*p*
Age at surgery (years)	5.9 (± 5.5, 0.2–18.6)	5.7 (± 5.3, 0.2–17.5)	6.2 (± 5.8, 1.0–18.6)	0.6
Follow-up time (months)	4.0 (± 2.1, 0.8–11)	3.9 (± 1.9, 1.2–7.9)	4.1 (± 2.5, 0.8–11)	0.8
Male	42 (68)	27 (75)	15 (58)	0.2
Race				0.5
White	37 (60)	21 (58)	16 (62)	
Black	2 (3)	2 (6)	0 (0)	
Asian/Pacific Islander	7 (11)	3 (8)	4 (15)	
Other/unknown	16 (26)	10 (28)	6 (23)	
Tonsil size				< 0.01
1+	36 (58)	30 (83)	6 (23)	
2+	15 (24)	2 (6)	13 (50)	
3+	10 (16)	4 (11)	6 (23)	
4+	1 (2)	0 (0)	1 (4)	
Body mass index				0.3
< 85th percentile	41 (66)	26 (72)	14 (58)	
85th–94th percentile	10 (16)	6 (17)	4 (15)	
≥ 95th percentile	11 (18)	4 (11)	7 (27)	
Baseline OSA				0.01
Mild (oAHI < 5)	15 (24)	9 (25)	6 (23)	
Moderate (oAHI ≥ 5, < 10)	21 (34)	17 (47)	4 (15)	
Severe (oAHI ≥ 10)	26 (42)	10 (28)	16 (62)	
Syndromic diagnosis	24 (38)	13 (36)	11 (42)	0.6
Trisomy 21	12 (19)	6 (17)	6 (23)	0.5
Hypotonia	7 (11)	6 (17)	1 (4)	0.1
Craniofacial condition	5 (8)	1 (2)	4 (15)	0.1

OSA obstructive sleep apnea syndrome, *oAHI* obstructive AHI

had an underlying syndromic diagnosis, including trisomy 21 (19%), hypotonic neuromuscular disorder (11%), or a craniofacial condition (8%). The proportion of children with a syndromic diagnosis did not differ significantly in the two intervention groups. A total of 8 subjects (12.9%) had a diagnosis of gastroesophageal reflux disease (GERD). Of those, 6 were on an antacid medication and 2 were not.

Mean sleep endoscopy scores are presented in Table 2. Children who underwent AT had significantly higher mean obstruction scores at the adenoid and lateral pharyngeal wall (tonsil) levels than those who underwent non-AT intervention (all $p < 0.01$). Children who underwent non-AT intervention had significantly higher obstruction scores at the supraglottis ($p < 0.001$), but total obstruction scores and scores at the velum and tongue base were not significantly different between the two groups ($p > 0.05$).

Frequencies of DISE-directed surgery performed are presented in Table 3. While a substantial proportion (42%) underwent AT, more patients (58%) underwent surgery that differed from standard-of-care adenotonsillectomy (non-AT intervention). Overall, 40% had single-level intervention at either the adenoid (7%), tonsil (3%), supraglottis (26%), or lingual tonsil (3%). Of 62 total patients, 11 (18%) had non-AT multilevel surgery. A substantial portion (43%) underwent supraglottoplasty for sleep-state laryngomalacia, either in isolation or in combination with other procedures. Supraglottoplasty was significantly associated with younger age and smaller tonsils (both $p < 0.001$). The mean age of children who underwent supraglottoplasty was 4.6 ± 5.0 years old (range 0.2–17.5 years) compared with a mean age of 6.9 ± 5.6 years (range 1.0–18.6 years; $p = 0.03$). There were no significant differences in any other baseline parameters between children who did and did not undergo supraglottoplasty (all $p > 0.05$).

DISE-directed surgery was successful in reducing the total number of obstructive breathing events (oAHI) in 79% of subjects, though not all of those children improved in their categorical OSA severity. Figure 1 displays the distribution of pre- and postsurgical OSA severity. Baseline OSA severity was defined by obstructive AHI, with 24% of patients having mild (oAHI < 5), 34% moderate (oAHI ≥ 5 to < 10), and 42% severe OSA (oAHI ≥ 10). None of the children met the criteria for sleep hypoventilation. After DISE-directed surgery, nearly two-thirds (61%) had mild or no OSA, 18% had moderate OSA, and 21% had severe OSA. Overall, 44% of subjects had the same degree of OSA before and after surgery, 48% improved by at least one category (i.e., moderate to mild or severe to either moderate or mild), and 8% worsened by one category. Of the children whose OSA category worsened or did not improve, 47% were mild at baseline, 19% moderate, and 34% severe. Of those who improved, 50% had moderate and 50% severe OSA. This difference in distribution of baseline OSA was significant ($p < 0.01$). There was a trend for older age in those whose OSA category worsened or failed to improve (7.2 years vs 4.5 years, $p = 0.53$). There were no other systematic or significant differences between the two groups (all $p > 0.05$). Furthermore, the five children who worsened did not differ significantly from the remainder with respect to age, sex, race, obesity status, or comorbidity status (all $p > 0.05$).

At baseline, 23% of children in the AT group had mild OSA; 15% had moderate OSA, and 62% had severe OSA. After surgery, the AT group had 38% with mild, 31% with moderate, and 31% with severe OSA. In the group who underwent non-AT intervention, 25% had mild baseline OSA, 47% moderate OSA, and 28% severe OSA. After DISE-directed surgery, the group had 78% with mild, 8% moderate, and 14% severe OSA. The distribution of OSA severity was significantly different between the non-AT and AT groups, with a larger proportion of children in the non-AT than the AT group having mild postoperative OSA (78% vs 38%, $\chi^2 = 10.1$; < 0.01).

Sleep test results are displayed in Table 4. The series as a whole improved from severe baseline OSA with a mean

Table 2 Mean sleep endoscopy scores (total and subsite) overall and for those who underwent AT or non-AT intervention

	All subjects ($n = 62$)	Intervention		
		Non-AT ($n = 36$)	AT ($n = 26$)	
Variable	Mean (standard deviation)			p
Subsite scores (range 0–3)				
Adenoid	1.4 (1.1)	1.0 (1.1)	1.8 (1.1)	< 0.01
Velum	1.2 (1.2)	1.2 (1.3)	1.2 (1.2)	0.8
Lateral pharyngeal wall	1.6 (1.2)	1.0 (1.0)	2.5 (0.8)	< 0.001
Tongue base	1.4 (1.2)	1.3 (1.3)	1.5 (1.2)	0.5
Supraglottis	1.6 (1.4)	2.2 (1.2)	0.8 (1.1)	< 0.001
Total score (range 0–15)	7.2 (2.6)	6.7 (2.8)	7.8 (2.2)	0.1

Table 3 Drug-induced sleep endoscopy–directed interventions ($n = 62$)

	n (%)
Adenoidectomy	4 (7)
Tonsillectomy	2 (3)
Adenoidectomy + tonsillectomy	26 (42)
Lingual tonsillectomy	2 (3)
Supraglottoplasty	17 (27)
Adenotonsillectomy + supraglottoplasty	5 (8)
Adenoid + supraglottoplasty	3 (5)
Lingual tonsillectomy + supraglottoplasty	2 (3)
Tonsillectomy + lingual tonsillectomy	1 (2)

preoperative oAHI of 22.2 events per hour to moderate OSA with a mean postoperative oAHI of 7.2 events per hour ($p < 0.01$). Additionally, there were overall statistically significant improvements in AHI, ODI, and oxygen saturation nadir. Though there was an improvement in percent total sleep time with $CO_2 > 50$ mmHg, this change was not statistically significant.

Discussion

We report improved polysomnography outcomes after DISE-directed surgery in a series of 62 surgically naïve patients with preoperative risk factors for adenotonsillectomy failure. Fifty-eight percent of patients underwent an intervention other than standard-of-care adenotonsillectomy. DISE-directed surgery was successful in reducing the total number of obstructive respiratory events in 79% of subjects. The mean severity of sleep apnea improved from severe to moderate, and postoperative oAHI was mild (< 5) in 61% of patients in this high-risk group. Children who underwent AT did not experience as much improvement overall as those who underwent non-AT intervention.

This study and the series that have come before it are important steps in better understanding DISE as a tool to direct surgical intervention. While some authors argue that DISE should be reserved for cases of OSA that do not improve after AT [27], others suggest that DISE be used routinely in all patients prior to adenotonsillectomy [30]. Studies by Boudewyns et al. and Gazzaz et al. have demonstrated that

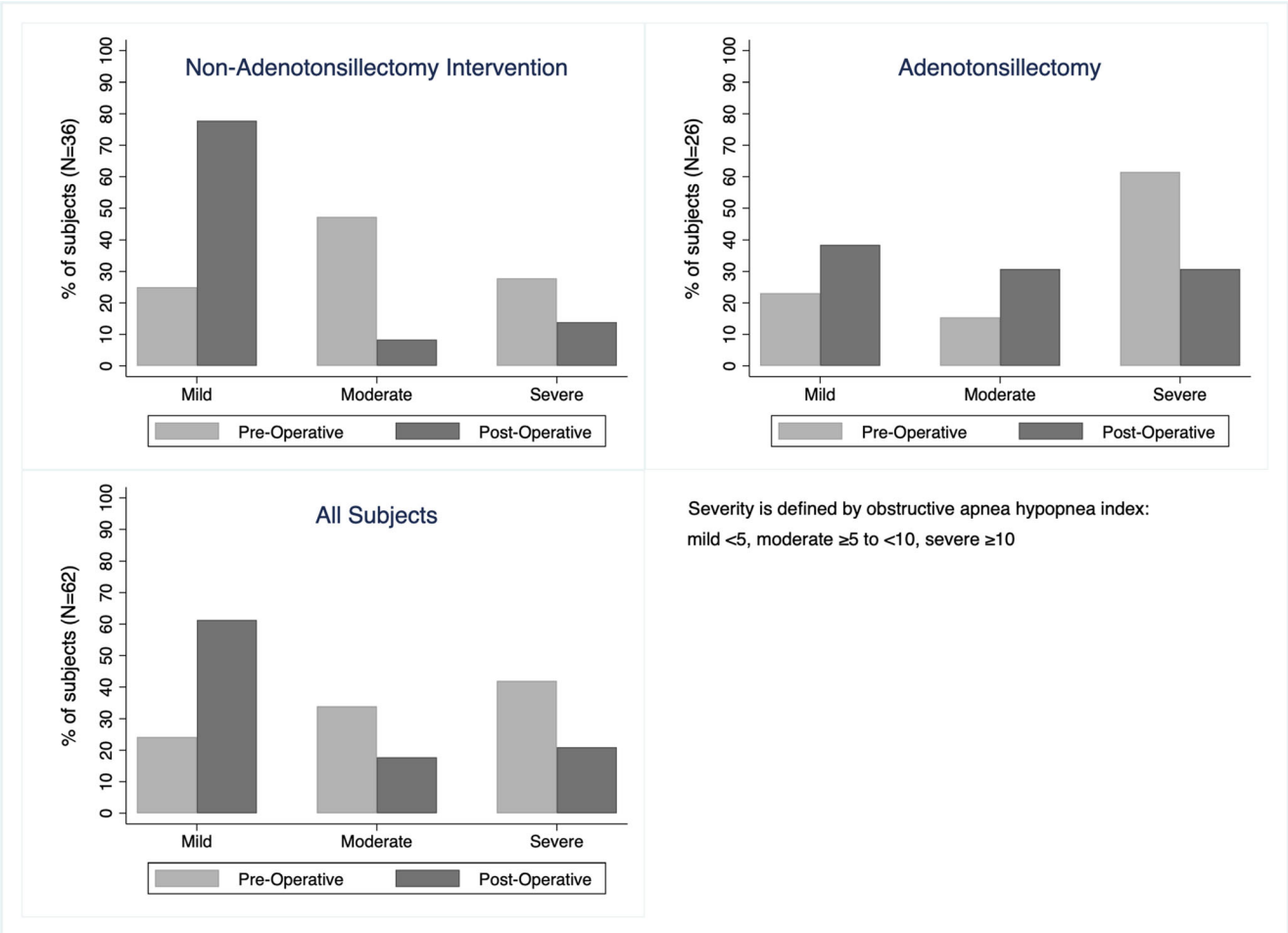

Fig. 1 Pre- and postoperative severity of sleep apnea overall and by adenotonsillectomy vs non-adenotonsillectomy intervention

Table 4 Changes in polysomnography parameters after drug-induced sleep endoscopy–directed intervention in surgically naïve children ($n = 62$)

	Preoperative mean (SD)	Postoperative mean (SD)	Mean difference[a]	p
Obstructive apnea-hypopnea index	22.2 (42.1)	7.2 (8.2)	15.0 (39.5)	< 0.01
Apnea-hypopnea index	24.8 (41.4)	9.7 (8.5)	15.1 (39.5)	< 0.01
Oxygen desaturation index	14.5 (28.7)	7.2 (8.0)	7.2 (25.8)	0.04
Oxygen nadir (% saturation)	82 (17)	87 (7)	5 (15)	< 0.01
% total sleep time $CO_2 > 50$ mmHg	3.4 (4.4)	3.3 (11.6)	0.1 (12.2)	0.9

CO_2 end tidal carbon dioxide interval, *SD* standard deviation

[a] Positive values indicate improvement

DISE changes the surgical decision-making in healthy surgically naïve patients in 25–35% of cases [26, 30]. It stands to reason that DISE may be of even greater benefit in patients at high risk for adenotonsillectomy failure than in healthy children.

Polysomnography outcomes after DISE-directed surgery in patients at risk for OSA persistence have been previously reported in two studies; however, even after the use of DISE, the majority underwent intervention at the tonsils, adenoids, or both. Maris et al. reported polysomnography outcomes in 25 surgically naïve children with trisomy 21 [35]. Sixty-four percent underwent adenotonsillectomy, 28% tonsillectomy, and 8% adenoidectomy. Mean oAHI decreased from 11.4 to 5.6 events/h. Truong et al. reported a decrease in AHI from 13.8 to 8.0 events per hour in 39 patients, 28% of whom had a coexisting syndromic diagnosis and the majority (90%) of whom underwent adenotonsillectomy [36]. In addition to the above, He et al. [37] reported a decrease in oAHI from 16.3 to 6.5 events per hour in 21 surgically naïve infants, though 70% underwent tracheostomy for treatment, making it difficult to compare with outcomes of studies reporting primarily on upper airway interventions.

Children in the present series experienced overall similar levels of improvement from severe to moderate OSA. In contrast to the above studies, however, the majority of children in this study did not undergo adenotonsillectomy. An equal proportion of subjects in this study underwent single-level intervention and adenotonsillectomy (each ~ 40%), with a smaller proportion undergoing non-AT multilevel intervention (18%). This may reflect differences in the samples studied, or changes in practice patterns over time or by region.

We performed a substantial number of supraglottoplasties (42%) in isolation or combination with other airway levels to treat sleep-state laryngomalacia. Many infants with awake inspiratory stridor diagnosed with classic congenital laryngomalacia can also have sleep-state laryngeal obstruction and can benefit from polysomnography to evaluate for OSA and use of DISE to confirm the site of obstruction prior to supraglottoplasty [38]. A meta-analysis pooled four studies of 38 infants with laryngomalacia and coincident OSA and found that the AHI improved significantly from 21 to 3.8 events per hour, though 88% still had residual disease [39].

State-dependent laryngomalacia was first described in five patients by Amin and Isaacson [40]. Digoy et al. [41] later reported polysomnography on outcomes of supraglottoplasty in 36 patients > 12 months of age with sleep-state laryngomalacia. They found an overall decrease in AHI from 13.3 to 4.1 events per hour in this series. However, in contrast to our study, 75% of those subjects had undergone previous AT at the time of their supraglottoplasty. A recent meta-analysis of isolated supraglottoplasty for laryngomalacia with obstructive sleep apnea pooled 64 patients with sleep-state laryngomalacia and found that the mean AHI decreased from 14 to 3.3 events per hour [42]. It is still unknown whether relieving obstruction at higher levels of the airway (by performing AT for example) has an impact on sleep-state laryngomalacia. Our decision to perform supraglottoplasty either in isolation or in conjunction with additional levels was driven primarily by the amount of obstruction seen at the supraglottis; 79% of children who underwent supraglottoplasty had maximal obstruction, and an additional 13% had > 50% obstruction at the supraglottic level.

This study has several limitations. This was a retrospective series from a single center. The high degree of comorbidity and the racial breakdown in the series limit the generalizability of results. However, the medical complexity of the series reflects the population seen at a large tertiary pediatric referral center. For many subjects, not all sites of maximal obstruction were addressed simultaneously, due in some cases to parental preference to minimize procedures and in others to the clinical practice of staging upper airway surgery to avoid complications. This series did not capture all stages of sleep surgery for all patients and thus may underrepresent final surgical response after all stages are complete. We did not evaluate for subglottic pathology in conjunction with DISE in this series; however, recent work by Bliss et al. suggests that this is not routinely necessary as subglottic lesions are rare [43]. Undertreatment would bias results toward the null, and despite this bias, the group still experienced significant improvement across respiratory parameters. As this was a case series without a control group, it is unclear how much benefit DISE adds

beyond adenotonsillectomy alone in high-risk patients, which warrants further study. Finally, we did not collect data on quality-of-life measures or specific sleep apnea–related symptoms, which are important clinical measures that are not always well-correlated with sleep test parameters [9, 44].

Conclusion

DISE-directed surgery in a surgically naïve population resulted in a surgical plan that differed from adenotonsillectomy in 58% of patients. Intervention was successful in reducing oAHI in 79% of subjects, and significant improvement was seen in 4 out of 5 polysomnography parameters tested. The mean severity of sleep apnea improved overall from severe to moderate, and postoperative oAHI was mild (< 5) in 61% of patients in this high-risk group. Children who underwent non-AT intervention experienced overall greater improvement than children who underwent AT. Our results highlight the severity of baseline disease in this challenging population as well as the potential utility of DISE in directing surgical intervention that may differ from standard-of-care adenotonsillectomy. They also underscore the challenge in completely eliminating obstruction in a highly comorbid population and the need for prospective, controlled studies to identify how much benefit DISE affords over adenotonsillectomy in patients at high risk for persistent OSA after AT.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflicts of interest.

Ethical approval All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from all individual participants included in the study.

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