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Original Article

Management and outcome of extreme pediatric obstructive sleep apnea☆

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A B S T R A C T

Objectives: Classify post-adenotonsillectomy (AT) respiratory support, identify variables that predict these interventions, and evaluate outcomes in children with extreme obstructive sleep apnea (OSA).
Methods: Retrospective chart analysis was performed on patients found to have apnea/hypopnea index (AHI) > 100 events/h. Patients with chronic diseases other than obesity were excluded.
Results: Forty-one subjects were studied, average age of 11.4 ± 4.3 years, majority (73.1%) were Hispanic, with a mean total AHI (TAHI) of 128.1 ± 22.9/h. Twenty-eight (68.3%) patients underwent AT. Lower age ($P < 0.001$), lower BMI Z-score ($P < 0.01$), higher OAHI ($P < 0.05$) were associated with having surgery. Eleven out of 28 (39.3%) surgical patients required respiratory support (oxygen or positive airway pressure) postoperatively. Longer % total sleep time S_pO_2 <90% during PSG ($P < 0.05$) and lower S_pO_2 nadir ($P < 0.05$) were associated with requiring airway support. No patients experienced mortality, reintubation, or hospital readmission following AT, with majority (71.4%) discharged 1 day post-operatively. Eleven (57.9%) of the 19 patients who had a postoperative PSG had residual OSA, defined as AHI >5 events/h, but there was a significant improvement in TAHI ($P < 0.01$).
Conclusion: Our findings confirm the need for postoperative observation in a controlled setting for patients with extreme OSA undergoing AT. Although at higher risk of needing respiratory support, those patients undergoing AT for extreme OSA did not require re-intubation post-operatively or suffer serious harm. Barring contraindications to AT, surgery may still be a first-line therapy for some children with extreme OSA.

1. Introduction

Obstructive sleep apnea (OSA) is characterized by partial or complete upper airway obstruction that disrupts normal sleep and respiration. Pediatric OSA leads to episodes of oxyhemoglobin desaturation, hypercapnia, and frequent arousals during sleep, leading to daytime sleepiness, neurobehavioral and mood problems, nocturnal enuresis, and systemic or pulmonary hypertension [1,2]. Prevalence varies based on age, sex, and ethnicity, ranging from 1 to 5%, and is significantly higher in children with obesity (13%–59%) [3,4].

For children with comorbidity, polysomnography (PSG) is often used to diagnose OSA, using the apnea/hypopnea index (AHI) as a marker [5]. Severe OSA is defined differently throughout the literature, with most studies using an AHI > 10 events/h [6]. Hypertrophy of adenotonsillar tissue often contributes to the pathophysiology of OSA in children. Therefore, the first line therapy for treatment of severe OSA remains adenotonsillectomy (AT) [2]. Treatment with positive airway pressure (PAP) has also been shown to effectively treat pediatric OSA and should be considered in patients in whom AT is contraindicated or residual OSA is identified following AT [7]. However, efficacy can be limited due to suboptimal adherence [8]. Children with very severe OSA are at increased

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risk of complications post-operatively. Both the American Academy of Pediatrics (AAP) and Academy of Otolaryngology-Head and Neck Surgery (AAO-HNC) recommend inpatient observation in this population post-operatively, but no specific level of care has been identified [7,9].

Children with the highest severity of OSA have not been widely studied, and limited data exists to guide treatment. This project characterizes children diagnosed with extreme OSA, defined as a total AHI >100 events/h. We aimed to classify treatment decisions, assess post-AT respiratory support requirements, and evaluate the frequency of residual OSA after AT. We hypothesized that PSG variables assessing gas exchange, age, and BMI z-score predict postoperative respiratory support requirements and postoperative OSA resolution.

2. Materials and methods

We performed a retrospective study which was approved by the University of California, San Diego Institutional Review Board, and a waiver of consent was granted. The charts of children who underwent PSG at Rady Children's Hospital San Diego between August 2015 and March 2020 were retrospectively reviewed. The earliest time point for enrolling study participants was chosen based on when the sleep center at Rady Children's Hospital instituted the current polysomnography interpretation software database. Inclusion to the study included patients ages 0–21 and those with extreme OSA defined as total apnea/hypopnea index (TAHI) > 100 events/h. Patients with comorbidities such as Down syndrome, cerebral palsy, and craniofacial, genetic, cardiovascular, or neuromuscular disease were excluded.

Data for each patient were extracted from both the electronic medical record and sleep software database. Demographic data included age, sex, race, and ethnicity. Sex, race, and ethnicity were family-identified. BMI percentile and BMI Z-score were collected using Center of Disease Control and Prevention (CDC) clinical growth charts, at the time of the initial PSG. Obesity was defined as BMI Z-score of greater than 1.64. The PSG interpretation software database was used to collect PSG specific variables including total AHI (TAHI), obstructive AHI (OAHI), and central AHI (CAHI). PSG variables assessing gas exchange included time spent with oxygen saturation below 90% (%S_pO_2 < 90%), oxygen saturation nadir (Nadir S_pO_2), time spent with end-tidal CO_2 greater than 50 mmHg (% $etCO_2$ > 50 mmHg), and peak $etCO_2$. Tonsillar size was defined on a 1–4 scale according to the Brodsky grading scale [10]. Tonsillar size was averaged if one tonsil size varied from the other. Tonsillar grading was done by either a pediatric pulmonologist or a pediatric otolaryngologist at primary evaluation for sleep disordered breathing. Tonsillar hypertrophy was defined as having a grade greater than or equal to 3.

Surgical indication was determined by the pediatric otolaryngologist, including AT or adenoid revision in patients who have had previous AT. All patients were admitted for inpatient post-operative observation. Post-operative respiratory support was defined as requiring respiratory support of any kind including nasal cannula, high flow nasal cannula, or positive airway pressure (PAP). The respiratory support interventions were further subdivided into major, defined as a new requirement for positive airway pressure (PAP) therapy and high flow oxygen, and minor, defined as a new requirement for low flow oxygen. When available, data from the post-operative PSG was collected. Adherence to PAP therapy was evaluated in the groups treated with PAP therapy alone and in those who were prescribed chronic PAP therapy after surgery. Adherence was defined as using PAP therapy for >50% of the time in a week or month period. Clinical report of PAP adherence based on caretaker report was used when objective adherence reports were not available.

PSG was conducted and analyzed in accordance with the AASM guidelines of sleep and associated events with a computer software system [11]. All PSG studies were performed at the Rady Center for Healthy Sleep as per institutional protocols as previously published for Rady Children's Hospital [12]. PSG were interpreted by sleep physicians.

Variables were reported using mean with standard deviation or percentage based on frequency. Normality tests were conducted to determine what analyses to run. Two sample t-tests were utilized for continuous, normal data, while the Wilcoxon rank sum test was utilized for continuous non-normal data. Additionally, chi-squared analyses were utilized for categorical variables. The following effect sizes were calculated: Cohen's d for parametric tests and rank–biserial correlation r for non-parametric tests. To assess which variables were associated with hospital stay, a logistical regression model was fitted using clinically and statistically significant pre-operative variables. Analysis of variance (ANOVA) was used to compare postoperative PSG results between levels of OSA severity. Statistical significance was set at P value < 0.05.

3. Results

Forty-six patients with TAHI >100 were identified. Five of the patients had exclusionary comorbidities and were excluded from the data analysis. Most of the children were male (80.5%), Hispanic (73.2%), and obese (92.7%) with an average age of 11.4 ± 4.3 years (Table 1). Baseline PSG characteristics are summarized in Table 1, with a mean TAHI of 128.1 ± 22.9/h.

Twenty-eight (68.3%) patients underwent surgery, the majority (27 of 28 or 96.3%) having AT and one patient had adenoid revision. Univariate analysis demonstrated that lower age (P < 0.001; d = 0.74), lower BMI Z-score (P < 0.01; r = 0.42), higher OAHI (P < 0.05; r = 0.38), and larger tonsil size (P < 0.05; d = 0.74) were

Table 1
Description of all patients included in the study (n = 41) in terms of demographics and initial polysomnography (PSG) data. Tonsil size from initial evaluation by either a pediatric pulmonologist or otolaryngologist.

Characteristics	Value
Age, mean (SD)	11.39 (4.32)
Sex, no. (%)	
Male	33 (80.5)
Female	8 (19.5)
Ethnicity, no. (%)	
Hispanic	30 (73.2)
Non-Hispanic	11 (26.8)
Race, no. (%)	
White	13 (31.2)
Black	2 (4.9)
Asian	1 (2.4)
Native American	1 (2.4)
Other (did not identify)	24 (58.5)
Body mass index (BMI) Z-score, mean (SD)	2.52 (0.89)
Obesity, no. (%)	38 (92.7)
TAHI, mean, (SD)	128.1 (22.9)
OAHI, mean, (SD)	121.9 (24.2)
CAHI, mean, (SD)	5.6 (10.3)
%S_pO_2 < 90%, mean (%), (SD)	32.8 (23)
Nadir S_pO_2, mean (%), (SD)	66.8 (11)
%$etCO_2$ > 50 mmHg, mean (%), (SD)	17.6 (21)
Peak $etCO_2$, mean, (SD)	59.3 (8.84)
Tonsil size ≥ 3, number, (%)	23 (56%)

PSG: polysomnography; TAHI: total apnea/hypopnea index; OAHI: obstructive apnea/hypopnea index; CAHI: central apnea/hypopnea index; %S_pO_2 <90%: time spent with oxygen saturation below 90%; Nadir S_pO_2: oxygen saturation nadir; % $etCO_2$ > 50 mmHg: time spent with end-tidal CO_2 greater than 50 mmHg.

associated with having surgery (Table 2). Eleven out of 28 (39.3%) patients required respiratory support postoperatively. Longer $\%S_pO_2$ <90% during PSG ($P < 0.05$; $d = 0.58$) and lower nadir S_pO_2 ($P < 0.05$; $d = 0.63$) were associated with needing respiratory support, whereas higher OAHI was not. When further divided, major respiratory support (urgent need for PAP therapy or high flow) was provided in 17.9% (5/28) of all surgical patients and minor respiratory support (low flow nasal cannula) was needed in 21.4% (6/28) of patients who had surgery (Table 3). No statistical significance was seen between the major and minor interventions in any of the variables.

Requiring respiratory support, minor or major, was associated with a longer hospital stay, defined as longer than one day. In addition, a longer hospital stay was associated with a higher BMI z-score ($P < 0.05$, $r = 0.34$) and longer $\%S_pO_2$ <90% on PSG ($P < 0.01$; $d = 0.50$). OAHI did not predict a longer hospital stay but CAI did show statistical significance (Table 4). Logistic regression was used to assess which variables amongst BMI z-score, $\%S_pO_2 < 90\%$, and OAHI are associated with hospital stay. $\%S_pO_2 < 90\%$ was positively associated with longer hospital stay (OR = 3.85, $P = 0.046$, CI [-0.22, 12.12]) while OAHI and BMI z-score were not significantly associated with hospital stay. No mortality, reintubation, or hospital readmission occurred in AT patients.

Nineteen of the 28 patients who had surgery had a post-operative PSG. One (5.3%) had complete resolution of OSA (AHI < 1.5 event/h), six (31.6%) had mild OSA (TAHI 1.5–5 events/h) and twelve (63.2%) were found to have moderate or severe OSA (TAHI >5 events/h). All but one (TAHI = 5.3 events/hour) patient with residual moderate or severe OSA were prescribed PAP therapy post-operatively. Two of the patients were discharged post-operatively with PAP therapy and had a PAP titration study post-op, therefore their postoperative PSG data were excluded. There was significant improvement in TAHI, OAHI, peak $etCO_2$, and PSG gas exchange variables on post-op PSG (Table 5). ANOVA was performed to identify whether age, BMI z-score, postoperative OAHI, CAHI, or PSG gas exchange variables were associated with the three different groups, TAHI < 5 events/h (resolved/mild OSA), TAHI 5–10 events/h (moderate OSA), and TAHI > 10 events/h (severe OSA). No statistically significant variables were identified between the 3 groups.

Adherence was assessed in patients who were treated with PAP therapy only (ie did not undergo AT) and in those requiring PAP therapy after surgery, with adherence found to be 57.1% and 40% respectively. There was a higher loss to follow up post-operatively (6/13, 46%) compared to patients who were treated with PAP therapy alone (0/13, 0%). We then subdivided the patients into two groups that had OSA successfully treated. Group 1 being patients treated solely with PAP therapy (no surgery) and were adherent and group 2 being the group that had a post-op study with TAHI <5 events/h or were adherent to post-op PAP therapy. Older ($P < 0.05$) patients and those with a longer $\%S_pO_2$ <90% ($P < 0.05$) were more successfully treated with PAP therapy alone. Group 1 and group 2 included patients that had appropriate treatment of OSA, with or without surgery, and our findings demonstrated that patients that were older and had worse oxygenation during PSG had better success when treated with PAP therapy alone rather than surgery.

Tonsil size was divided into two groups, tonsils <3+ and tonsils ≥3+. Of those who had both pre and post-operative PSG, having larger tonsils was associated with having surgery, but was not significantly associated with resolution of OSA postoperatively nor did it predict a larger change from pre-op TAHI and post-op TAHI.

4. Discussion

To our knowledge this is the first case series examining children with extreme OSA (TAHI > 100 events/h). We found that a significant number of those undergoing AT required respiratory airway support but none experienced mortality, respiratory failure, permanent disability, or prolonged hospitalization. While OAHI improved after surgery, a significant number of patients had residual OSA post-AT and required PAP therapy.

The male predominance in this population is similar to findings in previous studies [5,13]. Patients with comorbidities such as Down syndrome, cerebral palsy, craniofacial, genetic, cardiovascular, and neuromuscular disorders were excluded. Severity of OSA is defined variably in the literature, with most studies using AHI as the main marker of disease [9]. The AAO-HNC recommends patients who are younger than 3, with AHI >10 and oxygen saturation nadir less than 80% to be monitored inpatient post-adenotonsillectomy [9]. Similar to others, our study suggests that abnormal oxygenation (nadir S_pO_2 and time spent $\%S_pO_2$ <90%), but not OAHI is associated with postoperative oxygen use and longer hospital stay [14,15]. In our study, the logistic regression model demonstrated that time spent S_pO_2 <90%, but not OAHI, was associated with length of stay. This brings up the consideration whether the severity of OSA should be based on AHI rather than hypoxemia when determining post-operative care and if hypoxemia should be the primary determinant for inpatient observation.

Once severe pediatric OSA is diagnosed, management decisions focus on the decision to treat with surgery or non-invasive PAP therapy [2,16]. While these children likely incur the most severe neurocognitive, cardiovascular, and metabolic complications due to OSA, they are also at higher operative risk compared to those with mild to moderate OSA. Our study shows that younger patients, those with lower BMI z-scores, larger tonsils, and higher OAHI were more likely to have AT compared to PAP treatment, reflective of our

Table 2
Identification of variables that are associated with AT in patients with extreme OSA. Variables that were associated with having AT include lower age, lower BMI Z-score, higher OAHI, and larger tonsil size.

Characteristics	PAP Therapy Only	Surgery Only	P-value
	n = 13	n = 28	
Age, mean, (SD)	15.5 (2.5)	9.5 (3.8)	$P < 0.001$
BMI Z-score, mean (SD)	3.0 (0.4)	2.4 (0.9)	$P < 0.05$
TAHI, mean, (SD)	118.4 (10.7)	132.14 (26.9)	$P = 0.13$
OAHI, mean, (SD)	108.5 (13.6)	126.2 (27.5)	$P = 0.03$
$\%S_pO_2 < 90\%$, mean (%), (SD)	33.8 (21.3)	32.5 (22.8)	$P = 0.87$
Nadir S_pO_2, mean (%), (SD)	69.0 (12.0)	66.2 (11.3)	$P = 0.52$
$\%etCO_2 > 50$ mmHg, mean (%), (SD)	16.5 (20.8)	21.0 (22.3)	$P = 0.58$
Tonsil size ≥ 3, number	3 (23%)	20 (71%)	$P = 0.03$

AT: adenotonsillectomy; TAHI: total apnea/hypopnea index; OAHI: obstructive apnea/hypopnea index; CAHI: central apnea/hypopnea index; $\%S_pO_2$ <90%: time spent with oxygen saturation below 90%; nadir S_pO_2: oxygen saturation nadir; $\%etCO_2 > 50$ mmHg: time spent with end-tidal CO_2 greater than 50 mmHg; OSA: obstructive sleep apnea.

Table 3
Identification of predictors for requiring post-adenotonsillectomy respiratory support in patients with extreme OSA. Respiratory support is further divided into major (new PAP therapy or high flow oxygen) and minor (low flow oxygen). PSG oxygenation parameters (% $S_pO_2 < 90\%$ and Nadir S_pO_2) are associated with requiring post-operative respiratory support. No statistically significant variables identified to predict requiring major respiratory support.

Variable	Post-op respiratory support Mean ± SD n = 11	No post-op respiratory support Mean ± SD n = 17	*P*-value	Major respiratory support Mean ± SD n = 5	Minor respiratory support Mean ± SD n = 6	*P*-value
Age	10.6 ± 3.3	9.9 ± 4.4	$P = 0.71$	9.77 ± 3.4	11.25 ± 3.4	$P = 0.49$
BMI, Z-score	2.7 ± 0.2	2.3 ± 1.0	$P = 0.13$	2.81 ± 0.3	2.59 ± 0.2	$P = 0.12$
OAHI	132.6 ± 31.3	119.9 ± 21.3	$P = 0.22$	140.1 ± 45.6	126.4 ± 14.0	$P = 0.50$
% $S_pO_2 < 90\%$	45.2% ± 22.0%	23.7% ± 21.0%	$P < 0.05$	55% ± 17.1%	37.0% ± 23.7%	$P = 0.19$
Nadir S_pO_2	60.0% ± 8.7%	70.2% ± 10.7%	$P < 0.05$	59.8% ± 8.2%	60.2% ± 9.8%	$P = 0.95$
%$etCO_2 > 50$ mmHg	17.7% ± 24.8%	22.1% ± 15.1%	$P = 0.16$	30.8% ± 16.5%	14.8% ± 9.9%	$P = 0.08$

Post-op: post-operative; OAHI: obstructive apnea/hypopnea index; %$S_pO_2 < 90\%$: time spent with oxygen saturation below 90%; nadir S_pO_2: oxygen saturation nadir; % $etCO_2 > 50$ mmHg: time spent with end-tidal CO_2 greater than 50 mmHg.

Table 4
Prolonged length of stay was defined as length of stay longer than 1 day. BMI Z-score, CAHI, and %$S_pO_2 < 90\%$ were associated with a prolonged length of stay.

Characteristics	LOS, 1 day n = 20	LOS, >1 day n = 8	*P*-value
Age, mean, (SD)	10.1 (3.4)	10.2 (4.2)	$P = 0.97$
BMI Z-score, mean (SD)	2.3 (0.9)	2.8 (0.3)	$P < 0.05$
TAHI, mean, (SD)	126.8 (21.2)	138.5 (34.8)	$P = 0.27$
OAHI, mean, (SD)	120.2 (20.3)	136.7 (35.3)	$P = 0.16$
CAHI, mean, (SD)	5.6 (11.9)	1.8 (4.4)	$P = 0.049$
%$S_pO_2 < 90\%$, mean (%), (SD)	25.2 (21.0)	49.8 (21.2)	$P < 0.05$
Nadir S_pO_2, mean (%), (SD)	68.1 (11.1)	61.6 (9.9)	$P = 0.17$
%$etCO_2 > 50$ mmHg, mean (%), (SD)	18.4 (22.9)	22.1 (17.8)	$P = 0.39$

LOS: length of stay; TAHI: total apnea/hypopnea index; OAHI: obstructive apnea/hypopnea index; CAHI: central apnea/hypopnea index; %$S_pO_2 < 90\%$: time spent with oxygen saturation below 90%; nadir S_pO_2: oxygen saturation nadir; % $etCO_2 > 50$ mmHg: time spent with end-tidal CO_2 greater than 50 mmHg; OSA: obstructive sleep apnea.

Table 5
Comparing pre-operative and post-operative PSG characteristics in those patients that completed a post-operative PSG. Statistically significant improvement in all but one variable (%$etCO_2 > 50$ mmHg) was seen.

Characteristics	Pre-op PSG	Post-op PSG	*P*-value
TAHI, mean, (SD)	128.1 (21.4)	9.3 (11.2)	$P < 0.001$
OAHI, mean, (SD)	120.7 (20.7)	8.1 (11.3)	$P < 0.001$
CAHI, mean, (SD)	6.3 (12.3)	1.13 (1.3)	$P = 0.07$
%$S_pO_2 < 90\%$, mean (%), (SD)	27.0 (24.9)	2.14 (6.8)	$P < 0.001$
Nadir S_pO_2, mean (%), (SD)	68.2 (11.7)	85.7 (9.9)	$P < 0.001$
%$etCO_2 > 50$ mmHg, mean (%), (SD)	13.2 (13.8)	7.3 (18.05)	$P = 0.17$
Peak $etCO_2$, mean, (SD)	57.6 (7.0)	53.2 (4.6)	$P < 0.01$

PSG: polysomnography; TAHI: total apnea/hypopnea index; OAHI: obstructive apnea/hypopnea index; CAHI: central apnea/hypopnea index; %$S_pO_2 < 90\%$: time spent with oxygen saturation below 90%; nadir S_pO_2: oxygen saturation nadir; % $etCO_2 > 50$ mmHg: time spent with end-tidal CO_2 greater than 50 mmHg; peak $etCO_2$: peak end tidal CO_2.

center's practice pattern. Tonsil size has been used as a metric to help determine if surgery was an appropriate treatment for OSA [2]. Similar to the study by Isaiah A et al., our findings show that larger tonsil size did predict having AT, but it did not predict a greater change in AHI from preoperative PSG to postoperative PSG, and it did not predict complete resolution of OSA [13]. These findings suggest that multiple factors should be evaluated and considered when deciding on surgery in these patients to reduce risk of postoperative complications and optimize treatment outcomes.

Overall, our patient population tolerated AT well with no patients suffering intraoperative morbidity, prolonged ICU or hospital stay, or death. Thirty-nine percent of patients did require new oxygen or PAP therapy post operatively. Although our study is limited by a small population size, the frequency of need for postoperative respiratory support confirms the need for inpatient admission. Since 18% required new PAP therapy, these findings could suggest that these patients should be admitted specifically to a higher level of care where PAP can be provided urgently if needed. No variables were found to predict the need for PAP therapy over supplemental oxygen, suggesting that patients with this severity of OSA should be considered for ICU admission for initiation of PAP or high flow nasal cannula.

The high frequency of residual OSA post-AT highlights the importance of longitudinal care in this population. We found that only 67.8% of patients had completed a postoperative PSG, and a quality improvement project is underway at our institution to increase this frequency. While our practice has been to recommend post-operative PSG for patients with severe OSA, children can often be lost to follow up. No variables were found to be associated with OSA resolution post-operatively, possibly due to the small sample size of our population. Imanguli et al. demonstrated that teenagers with obesity were at high risk of residual OSA [17]. Although our findings did not show statistically significant differences in patients with higher BMI z-scores, patients with extreme OSA and significantly elevated BMI should be counseled that there is a high likelihood of PAP requirement post-AT.

PAP adherence likely exists as a significant barrier to optimal OSA treatment in this population. Adherence varies widely throughout literature and our definition of adherence included both the subjective reports of parents along with electronic downloads of hours of usage. Our data did show that the percentage of patients who were adherent to therapy was higher in the group that were treated with PAP alone compared to those who required PAP after surgery. This goes along with a recent paper by Katz et al. published in the Journal of Clinical Sleep Medicine, that showed that patients with lower oxygen saturation nadir were more adherent to outpatient PAP therapy, and in our study those were the patients in the PAP therapy only group compared to those who had residual OSA post-operatively [18]. It's also possible to consider that although they have residual OSA after surgery, those patients have significant improvement in the level of their OSA and are not as motivated to be adherent. These findings could also be skewed due to higher loss to follow up post-operatively in the group that underwent AT. When evaluating overall success of treatment of extreme OSA (postoperative resolution of OSA and/or adherence to prescribed PAP), our data suggests that for older patients and those with worse oxygenation (longer %S_pO_2 <90%) PAP therapy alone may be preferable to AT. Since longer %S_pO_2 <90% is also associated with requiring airway support postoperatively, these findings could potentially help physicians identify patients who could be recommended PAP therapy before surgery.

This study provides a description of demographics, treatment options, and outcomes in a very specific population. While severe pediatric OSA has been studied, we chose to focus on this highest risk group (ie extreme OSA) because of the challenges of weighing the risks and benefits of surgery vs. waiting to institute PAP therapy in the setting of ongoing significant gas exchange abnormality. While we did not find severe morbidity associated with AT in our population, we suggest extreme caution with anesthesia and surgery.

Specific limitations to our study deserve comment. The generalizability of our study should be evaluated with caution since our cohort was older, primarily male and Hispanic. Our data might not be as helpful or accurate in areas with younger children with a different ethnicity population. This was a retrospective study and there was likely variability in decision to proceed with surgery based on individual practice styles and consulting physician's specialty (pulmonary vs. otolaryngology). Due to the variability in treatment and limited nature of the retrospective study, our cohort is heterogenous in terms of interventions and portions of the data analysis did contain children who had both AT and PAP therapy. The goal was to report data on treatment outcomes of the population as a whole, regardless of type of treatment. Post-operatively, the definition of hypoxemia was not controlled and the decision to treat with supplemental oxygen was likely variable based on the practice of the inpatient treatment team. Similarly, the decision to treat with oxygen vs. PAP therapy postoperatively was subjective. Generally, PAP therapy was started if there was obvious clinical airway obstruction or severe hypoxemia recalcitrant to supplemental oxygen therapy. Finally, we did not have objective adherence data on all patients prescribed PAP, although this was not the primary aim of this study, future studies should assess adherence using objective PAP-based download data to assess adherence in children with extreme OSA.

5. Conclusion

In summary, in patients who underwent AT, we found surgery to be a reasonably safe treatment option in this population when postoperative care occurs in the ICU setting. However, families should be counseled that while PSG parameters will likely improve, PAP therapy will often be recommended post-operatively. Longer term studies of this cohort will serve to further define the efficacy of the treatment options.

Disclosure

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Credit author statement

Tatyana G. Mills MD: Investigation, Writing – original draft; **Rakesh Bhattacharjee MD:** Supervision, Conceptualization, Writing – Reviewing and Editing; **Javan Nation MD:** Supervision, Conceptualization, Writing – Reviewing and Editing; **Emily Ewing, MA:** Formal analysis, Data curation; **Daniel J. Lesser MD:** Supervision, Conceptualization, Methodology, Writing – Reviewing and Editing.

Abbreviations definition

$\%S_pO_2 < 90\%$	time spent with oxygen saturation below 90%
$\%etCO_2 > 50$ mmHg	time spent with end-tidal CO_2 greater than 50 mmHg
AHI	apnea/hypopnea index
ANOVA	analysis of variance
AT	adenotonsillectomy
CAHI	central apnea/hypopnea index
CDC	center of disease control
Nadir S_pO_2	oxygen saturation nadir
OAHI	obstructive apnea/hypopnea index
PAP	positive airway pressure
PSG	polysomnography

Conflict of interest

None declared.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2021.09.006.

References

[1] Capdevila OS, Kheirandish-Gozal L, Dayyat E, et al. Pediatric obstructive sleep apnea: complications, management, and long-term outcomes. Proc Am Thorac Soc 2008;5(2):274–82. https://doi.org/10.1513/pats.200708-138MG.
[2] Marcus CL, Brooks LJ, Draper KA, et al. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics 2012;130(3):576–84. https://doi.org/10.1542/peds.2012-1671.
[3] Lumeng JC, Chervin RD. Epidemiology of pediatric obstructive sleep apnea. Proc Am Thorac Soc 2008;5(2):242–52. https://doi.org/10.1513/pats.200708-135MG.
[4] Verhulst SL, Van Gaal L, De Backer W, et al. The prevalence, anatomical correlates and treatment of sleep-disordered breathing in obese children and adolescents. Sleep Med Rev 2008;12(5):339–46. https://doi.org/10.1016/j.smrv.2007.11.002.
[5] Kushida CA, Littner MR, Morgenthaler T, et al. Practice parameters for the indications for polysomnography and related procedures: an update for 2005. Sleep 2005;28(4):499–521. https://doi.org/10.1093/sleep/28.4.499.
[6] Iber C, Ancoli-Israel S, Chesson AL, et al. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. Westchester, IL: American Academy of Sleep Medicine; 2007.
[7] "Clinical Practice Guideline. Diagnosis and management of childhood obstructive sleep apnea syndrome. Pediatrics 2002;109(4):704–12. https://doi.org/10.1542/peds.109.4.704.
[8] Bhattacharjee R, Benjafield AV, Armitstead J, et al. Adherence in children using positive airway pressure therapy: a big-data analysis. Lancet Digit Health 2020;2(2). https://doi.org/10.1016/s2589-7500(19)30214-6.
[9] Roland PS, Rosenfeld RM, Brooks LJ, et al. Clinical practice guideline: polysomnography for sleep-disordered breathing prior to tonsillectomy in children. Otolaryngol Head Neck Surg 2011;145(1 Suppl.):S1–15. https://doi.org/10.1177/0194599811409837.
[10] Brodsky L. Modern assessment of tonsils and adenoids. Pediatr Clin N Am 1989;36(6):1551–69. https://doi.org/10.1016/s0031-3955(16)36806-7.
[11] Berry RBQS, Abreu AR, Bibbs ML, et al. The AASM manual for the scoring of sleep and associated events: rules, terminology and technical specifications. Darien, Illinois: American Academy of Sleep Medicine; 2020., version 2.6.
[12] Hurvitz MS, Lesser DJ, Dever G, et al. Findings of routine nocturnal polysomnography in children with Down syndrome: a retrospective cohort study. Sleep Med 2020;76:58–64. https://doi.org/10.1016/j.sleep.2020.10.003.
[13] Isaiah A, Hamdan H, Johnson RF, et al. Very severe obstructive sleep apnea in children: outcomes of adenotonsillectomy and risk factors for persistence. Otolaryngol Head Neck Surg 2017;157(1):128–34. https://doi.org/10.1177/0194599817700370.
[14] Keamy DG, Chhabra KR, Hartnick CJ. Predictors of complications following adenotonsillectomy in children with severe obstructive sleep apnea. Int J Pediatr Otorhinolaryngol 2015;79(11):1838–41. https://doi.org/10.1016/j.ijporl.2015.08.021.
[15] Molero-Ramirez H, Tamae Kakazu M, Baroody F, et al. Polysomnography parameters assessing gas exchange best predict postoperative respiratory complications following adenotonsillectomy in children with severe OSA. J Clin Sleep Med 2019;15(9):1251–9. https://doi.org/10.5664/jcsm.7914.
[16] Gozal D, Tan HL, Kheirandish-Gozal L. Treatment of obstructive sleep apnea in children: handling the unknown with precision. J Clin Med 2020;9(3):888. https://doi.org/10.3390/jcm9030888. Published 2020 Mar 24.
[17] Imanguli M, Ulualp SO. Risk factors for residual obstructive sleep apnea after adenotonsillectomy in children. Laryngoscope 2016;126(11):2624–9. https://doi.org/10.1002/lary.25979.
[18] Katz SL, Kirk VG, MacLean JE, et al. Factors related to positive airway pressure therapy adherence in children with obesity and sleep-disordered breathing. J Clin Sleep Med 2020 May 15;16(5):733–41. https://doi.org/10.5664/jcsm.8336.