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# Comparative outcomes of severe obstructive sleep apnea in pediatric patients with Trisomy 21

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ABSTRACT

*Objectives:* To analyze the outcomes of severe obstructive sleep apnea (OSA) in pediatric patients with Trisomy 21 compared with non-syndromic patients.
*Methods:* A retrospective chart review was performed for patients with a diagnosis of severe obstructive sleep apnea, (defined as, Apnea–Hypopnea index (AHI) of $\geq$10) in a tertiary children's hospital. Data were analyzed for subjective and objective outcomes along with perioperative care and health care utilization. Patients with Trisomy 21 were compared with non-syndromic patients.
*Results:* A total of 230 patients with severe OSA were included in the study. Eighteen of these patients had Trisomy 21. Adenotonsillectomy was the most common surgical intervention in both groups. There was no statistical difference in the preoperative AHI between groups. Post treatment AHI in the Trisomy 21 group changed from an average of 26.6 to an average of 11.6 as compared with 24.5 to 3.6 in the non-syndromic group. The average perioperative hospital stay was 3.8 days in Trisomy 21 group compared to 1.7 days for the non-syndromic group ($p < 0.001$, Mann–Whitney *U* test). Complete resolution was seen in 35% of the Trisomy 21 group versus 75% in the non-syndromic group.
*Conclusions:* A majority of Trisomy 21 patients with severe OSA had residual symptoms following surgical intervention. There is also an increased risk of post-operative airway intervention and increased length of hospital stay in these patients.



## 1. Introduction

Obstructive sleep apnea (OSA) is a common condition affecting 2–5% of the general pediatric population [1]. The American Academy of Pediatrics defines OSA as disorder of breathing during sleep characterized by prolonged partial upper airway obstruction and/or intermittent complete obstruction that disrupts normal ventilation and sleep patterns [2]. The causes of this condition are numerous; the most common of these are adenotonsillar hypertrophy and obesity [3]. Hypotonic neuromuscular conditions, dental problems and congenital cranio-facial abnormalities which cause reduced airway size or increased airway collapsibility, can additionally contribute to the pathogenesis of OSA [4,5]. Snoring and poor sleep quality are the most common symptoms of OSA however, the clinical presentation can vary depending on the patients' age [6]. Symptoms common in younger children include, enuresis, somnambulism and hyperactivity. Poor academic achievement, emotional instability and cognitive defects are more apparent in older children [7]. Physical sequelae, though rare, can include mild pulmonary hypertension and cardiovascular complications as well as failure to thrive and delayed development in untreated patients [8]. Compared with the general population, children with Trisomy 21 have a significantly higher prevalence of OSA [9,10]. A recent study showed some degree of obstructive sleep apnea in 28/29 8-year-old children with Trisomy 21, and a 59% prevalence of moderate–severe OSA in that cohort of patients, highlighting the prevalence of OSA in this subset of patients [11]. The pathophysiological reasons for this are related to both the cranio-facial features of this syndrome; mid-face hypoplasia and relative macroglossia, as well as generalized hypotonia, lymphoid hyperplasia and preponderance towards obesity [12,13]. A raised BMI appears to correlate strongly with OSA in both groups of patients [14].

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Adenotonsillectomy (AT) is the most common treatment for OSA in patients with and without Trisomy 21 [15]. Thus far, studies reporting outcomes of treatments for patients with Trisomy 21 have not been encouraging, with some reports describing rates of successful treatment, as measured by polysomnography (PSG), at 50% following AT [16,17]. The reasons for poorer outcomes in Trisomy 21 patients after AT have been primarily associated with higher frequency of co-morbidities and synchronous airway pathology [15]. With the above in mind, the specific post-operative findings with regards to post-operative airway intervention, hospital length of stay, and symptomatology have not been examined in depth. Below we describe the results of a retrospective, case controlled study examining the outcomes following surgical treatment for severe OSA in pediatric patients with Trisomy 21 compared to non-syndromic patients.

## 2. Materials and methods

### 2.1. Study design

A retrospective chart review was performed for patients with a diagnosis of severe obstructive sleep apnea (defined as, obstructive Apnea–Hypopnea index (AHI) of >9.9) identified between January 2007 and June 2012. Data regarding patient demographics, perioperative hospitalizations and post-operative outcomes were collected from patient charts. Data concerning pre and post-operative symptoms were obtained from responses to routinely asked questions during patient consultations, documented in patient charts. These data were analyzed for subjective and objective outcomes along with perioperative care and health care utilization. Patients with Trisomy 21 were then compared with non-Trisomy 21 patients.

### 2.2. Subjects

Patient and surgical information of children with and without the diagnosis of Trisomy 21 were collected. A total of 4972 non-Trisomy 21 and 103 Trisomy 21 patients who underwent AT were initially reviewed. Inclusion criteria included children with severe obstructive sleep apnea (obstructive AHI ≥10) documented on polysomnogram who underwent adenotonsillectomy. In our practice, all children with documented severe obstructive sleep apnea undergo post-operative polysomnogram 3–4 months post-operatively. Children who did not have both a pre-operative and post-operative polysomnogram were excluded. Children suffering from developmental delays or syndromes other than Trisomy 21 were also excluded.

### 2.3. Polysomnography

Polysomnography was conducted within an overnight sleep laboratory at the Children's Hospital of Pittsburgh. As per AAO-HNS clinical practice guidelines on polysomnography for sleep-disordered breathing prior to tonsillectomy in children, the physiologic parameters measured included gas exchange, respiratory effort, airflow, snoring, sleep stage, body position, limb movement, and heart rhythm [18]. Apneic events were measured and interpreted as per the recommendations of the AASM Manual for the Scoring of Sleep and Associated Events [19]. AHI was graded as follows, mild (AHI 1–4.9), moderate (AHI 5–9.9) and severe (AHI ≥10). Children suffering from primarily central sleep apnea were excluded. The presence of some central apneas in addition to severe obstructive sleep apnea was not an exclusion criteria, however the obstructive AHI rather than the total (obstructive + central) AHI was used for our statistical analysis.

### 2.4. Data analysis

Comparative analysis was performed both pre and post-operative findings between both groups of patients. This was also conducted on post-operative airway interventions required. Wilcoxon signed rank tests were utilized to examine pre and post-surgical obstructive AHI's between the two groups. While post-operative hospitalization days were analyzed using Mann–Whitney *U* testing.

The University of Pittsburgh Medical Center institutional review board reviewed the protocol summary and full approval was granted for the collection and reporting of data in this study.

## 3. Results

### 3.1. Patient demographics

Two hundred-thirty patients met criteria for this study and of these, 18 had a diagnosis of Trisomy 21. There were approximately equal percentages of male and female patients in each group, 53% male and 47% female in the non-Trisomy 21 group and 56% male, 44% female in the Trisomy 21 group. The mean age of children in the non-Trisomy 21 group, at the time of surgery was 72 months, the Trisomy 21 group mean age was 88 months ($p = 0.2$). Pre-operatively, patients in the non-Trisomy 21 group had a mean BMI of 20, the Trisomy 21 group had a mean BMI of 19.7 ($p = 0.9$). There was an increased incidence of overall previous airway surgery prior to adenotonsillectomy in the Trisomy-21 group compared with non-Trisomy 21, 22% compared with 12%. Previous airway surgeries included supraglottoplasty, septal surgery, partial adenoidectomy, modified palatoplasty.

Polysomnogram data was collected and analyzed for Obstructive-Apnea and Hypopnea Indices (O-AHI) for both groups. As demonstrated in Fig. 1, the post treatment O-AHI in the Trisomy 21 group decreased from an average of 26.6 to an average of 11.6 ($p < 0.015$) while in the non-syndromic group the average O-AHI decreased from 24.5 to 3.6 ($p < 0.001$).

Overall, patients with Trisomy 21 experienced increased rates of immediate post-operative airway interventions while in the recovery room. These included the use of oxygen delivered by face-mask, heliox, nasal trumpet, and patients who remained intubated or required re-intubation. The incidence of flash pulmonary edema was also examined. Presence of pulmonary edema was diagnoses based on chest X-ray. Fig. 2 is a graphical representation of the percent of patients in each group who experienced post-operative airway intervention following surgery. Twenty-six percent ($n = 55$) of non-Trisomy 21 patients required oxygen delivered by face

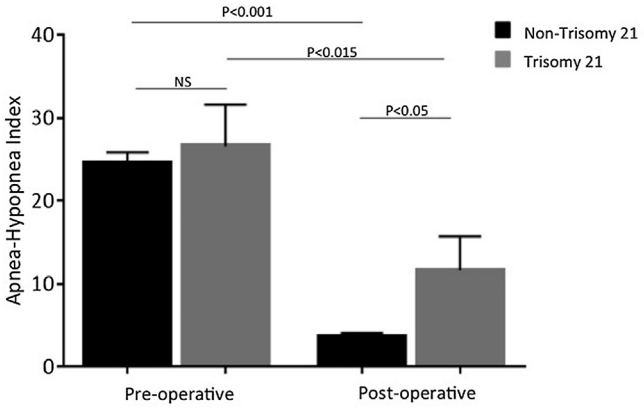


**Fig. 1.** Pre and post-operative obstructive-AHI of Trisomy 21 and non-Trisomy 21 children.