Characterization of Sleep Architecture in Down Syndrome Patients Pre and Post Airway Surgery

Mark Mims, MD[1]; Prasad Thottam, DO [1,2]; Dennis Kitsko, MD[1]; Amber Shaffer, PhD[1]; Sukgi Choi, MD[1]
[1]University of Pittsburgh Medical Center, [2]Children's Hospital of Michigan

Abstract

Objectives: To define obstructive sleep architecture patterns in Down Syndrome (DS) children as well as changes post-operatively.
Study Design: Retrospective review
Methods: 45 Pediatric DS patients that underwent airway surgery between 2003 and 2014 at a tertiary children's hospital for obstructive sleep apnea (OSA) were investigated. Postoperative changes in respiratory parameters and sleep architecture (SA) were assessed and compared to general pediatric normative data using paired t-tests and Wilcoxon signed-rank test.
Results: 22 participants were male. 30 participants underwent tonsillectomy and adenoidectomy, 4 adenoidectomy, 10 tonsillectomy, and 1 base of tongue reduction. Patients were divided into two groups based on age (<6 years & >6 years) and compared to previously published age matched normative SA data. DS children in both age groups spent significantly less time than controls in REM and N1 ($p<0.02$). Children younger than 6 spent significantly less time in N2 than previously published healthy controls ($p<0.0001$). Children 6 years of age or older spent more time than controls in N3 ($p=0.003$). Airway surgery did not significantly alter SA except for an increase in time spent in N1 ($p=0.007$). Surgery did significantly reduce median AHI ($p=0.004$), OAHI ($p=0.006$), hypopneas ($p=0.005$), total apneas ($p<0.001$), and central apneas ($p=0.02$), and increased the lowest oxygen saturation ($p=0.028$).
Conclusions: DS children are a unique population with different SA patterns than the general pediatric population. Airway surgery has little impact on their SA.

Introduction

Down Syndrome (DS) is a common genetic disorder, affecting approximately 8 per 100,000 people in the United States[1]. DS patients are at increased risk for the development of obstructive sleep apnea (OSA), which can affect up to 50% of the pediatric DS population and >90% of DS adults[2]. OSA has been found to have multiple detrimental effects on health and leads to an overall decreased quality of life for children. Surgical and nonsurgical interventions have been shown to improve OSA, and DS patients often require a multimodal approach. Nonsurgical options include environmental modification, weight loss, dental appliance, and CPAP or BiPAP[2]. Airway surgery, such as adenotonsillectomy, can lead to both short and long term improvement in this population[3]. Although it is often the first line surgical treatment for the management of OSA, it is not always curative as a unimodal therapy[2,4]. And though sleep architecture alterations with airway surgery have been described for non-syndromic children[3], there is little available literature addressing the changes that occur in DS patients after airway surgery.

Methods and Materials

A retrospective review was performed of DS children with OSA that underwent airway surgery over 11 years at a tertiary pediatric center. Only patients under the age of 18 at the time of surgery with pre- and post-operative polysomnograms (PSG) within 1 year of airway surgery were included. PSG data gathered included the apnea-hypopnea index (AHI), obstructive apnea-hypopnea index (OAHI), total hypopneas, total apneas, percentage of apneas that were central, obstructive, or mixed in origin, O2 nadir, rapid eye movement (REM) onset, and total sleep time (%TST) spent in stages N1, N2, N3, and REM. Baseline patient data including age, sex, BMI, tonsil size, and percent obstruction from adenoids at time of surgery were also recorded. Respiratory parameters, sleep architecture, and post-operative changes were compared to non-DS pediatric normative data using t-tests and Wilcoxon signed-rank tests. Normally distributed data are presented as mean ± SD. Non-normally distributed data are presented as median (range).

Results

In total, 121 patient charts were reviewed with 45 patients meeting the inclusion criteria. 23 females and 22 males were included. The median age at time of surgery was 5.2 years old (range 1.6-16.6 years) and median BMI was 17.5 (range 10.8 to 43.7). 30 patients (67%) underwent adenotonsillectomy as their primary airway surgery, 4 (9%) underwent adenoidectomy only, 10 (22%) only had tonsillectomy, and 1 (2%) underwent base of tongue reduction. 3 patients received pharyngoplasty as a secondary airway surgery including 1 patient who also received an inferior turbinate reduction. Pre- and post-operative PSG parameters are shown in Table 1. Sleep architecture was then evaluated in two different age categories, <6 years old and ≥6 years old. Montgomery-Downs et al. established normative data for children and distinguished distinct polysomnography profiles for children <6 years old and ≥6 years old[5]. This data was used to compare sleep data with our cohort (Table 2).

Table 1. Respiratory Parameters.

	Pre-operative	Post-operative	Δ (Pre-Post)	p-value
AHI	10.1 (0.2, 83.2)	4.2 (0.4, 37.7)	-4.7 (-58.2, 15.8)	0.004
OAHI	9.3 (0.2, 74.4)	3.4 (0.4, 37.7)	-4.3 (-58.8, 15.7)	0.006
Hypopneas	40 (1, 343)	12.5 (3, 261)	-17 (-340, 142)	0.005
Apneas	13.5 (0, 267)	4 (0, 65)	-9.5 (-254, 38)	<0.001
Central	3 (0, 44)	2 (0, 28)	-1 (-44, 20)	0.020
Mixed	0 (0, 13)	0 (0, 25)	0 (-13, 15)	0.738
Lowest O_2	86 (42, 94)	88 (60, 94)	2 (-13, 42)	0.028

Table 2. Sleep Architecture by Age

	Age (Yr)	Control[5]	Pre-operative	Pre v. control p-value	Post-operative	Post v. control p-value	Pre v. post p-value
REM (%TST)	<6	23.6	19.3 ± 8.0	0.011	20.2 ± 5.0	0.001	0.651
	≥6	22.6	15.4 ± 7.4	0.002	14.6 ± 7.1	<0.001	0.743
REM Onset (min)	<6	87.8	94.5 (18, 442.5)	0.280	102.5 (45.5, 239.5)	0.107	0.929
	≥6	132	127.75 (35.5, 326)	0.900	147.25 (59.5, 274.5)	0.272	0.802
N1 (%TST)	<6	6.6	0.1 (0, 4.6)	<0.001	1.1 (0, 8.1)	<0.001	0.007
	≥6	7.1	1.85 (0.2, 9.4)	0.006	3.0 (0.8, 5.5)	0.002	0.875
N2 (%TST)	<6	41.6	53.5 ± 8.2	<0.001	50.5 ± 8.4	<0.001	0.186
	≥6	46.1	50.2 ± 9.7	0.171	55.5 ± 12.0	0.020	0.083
N3 (%TST)	<6	28.2	26.0 ± 6.4	0.079	28.5 ± 7.6	0.837	0.133
	≥6	24	30.4 ± 7.1	0.004	25.5 ± 8.9	0.529	0.085

For our patients, n=12-27 depending on information available in patient records.
A total of 27 patients were <6 years old. A total of 18 patients were ≥6 years old.

Discussion

Children with DS have previously been shown to have unique sleep architecture when compared with non-syndromic controls[6]. However, few studies have investigated the effects of airway surgery for this patient population and none have specifically examined whether these interventions provide a more "normal" sleep architecture for this population. Our data confirms that DS patients have altered sleep architecture. Decreased time spent in REM and N1 was noted in both age groups. Increased time was spent in N2 in patients younger than 6, and increased time spent was spent N3 in patients 6 or older.

Our DS population demonstrated post-interventional decreases in expected markers of OSA, confirming that DS patients benefit from airway surgery with regard to OSA symptoms. In addition, patients 6 or older no longer spent increased time in N3 after surgery. Churchill et al. found DS patients with SDB were at risk for additional functional impairments compared to non-SDB DS patients[7]. Improvement in both sleep architecture and sleep apnea could significantly improve the daily lives of DS patients, in addition to the longer-term health benefits. Additionally, Breslin et al. demonstrated that DS patients with disordered sleep architecture have significantly lower IQ than DS patients without disordered sleep architecture[8]. This further demonstrates the need for optimization of DS sleep architecture and the importance of the potential improvement provided by airway surgery.

Conclusions

Down Syndrome patients have a significantly different sleep architecture compared with their non-syndromic peers. The origin of this difference is likely multifactorial and in-part related to their OSA. Airway intervention assists in reducing both central and obstructive events and in normalizing sleep architecture. This can lead to both short and long term improvements in health for the DS population.

Contact

Mark Mims, MD
Children's Hospital of Pittsburgh, University of Pittsburgh Medical Center
Email: mimsmm2@upmc.edu
Website: http://www.chp.edu/our-services/ent
Phone: 412-692-6220

References

1. Presson AP, Partyka G, Jensen KM, *et al*. *J Pediatr*, 2013: 163(4):1163-8.
2. Lal C, White DR, Joseph JE, *et al*. *Chest*, 2015: 147(2):570-9.
3. Lee CH, Kang KT, Weng WC, *et al*. Int J Pediatr Otorhinolaryngol, 2015: 79(2):210-5.
4. Thottam PJ, Choi S, Simons JP, Kitsko DJ. Otolaryngol Head Neck Surg, 2015: 153(4):644-8.
5. Montgomery-Downs HE, O'Brien LM, Gulliver TE, *et al*. Pediatrics, 2006: 117(3):741-53.
6. Nisbet LC, Phillips NN, Hoban TF, *et al*. Sleep Breath, 2015: 19(3):1065-71.
7. Churchill SS, Kieckhefer GM, Bjornson KF, *et al*. Sleep, 2015: 38(1): 61–71.
8. Breslin J, Spanò G, Bootzin R, *et al*. Dev Med Child Neurol, 2014: 56(7):657-64.