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Original Article

Increased prevalence of obstructive sleep apnea in patients with pectus excavatum: A pilot study

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Abstract

Objective: Laryngomalacia is the most common congenital laryngeal anomaly and is associated with pectus excavatum (PE). Patients with laryngomalacia and patients with obstructive sleep apnea (OSA) both experience upper airway obstruction, and patients with laryngomalacia had been found to have a higher prevalence of PE. However, no studies have established the prevalence of OSA in patients with PE. We conducted this pilot study to evaluate the prevalence of OSA in patients with PE. **Materials and Methods:** A total of 42 patients ≥20 years old with PE who were admitted for Nuss surgery to correct PE in Taipei Tzu Chi Hospital between October 2015 and September 2016 were invited to participate in the study; 31 of the 42 patients agreed. All 31 patients completed an Epworth sleepiness scale questionnaire to evaluate excessive daytime sleepiness (EDS) and underwent overnight polysomnography to evaluate OSA before Nuss surgery. **Results:** The prevalence of snoring in the study participants was 100%. Ten of 31 patients (32.3%) reported EDS. The overall prevalence of OSA with an apnea/hypopnea index ≥5/h was 25.8%, and all patients with OSA were men. **Conclusions:** The prevalence of OSA in patients with PE seemed to be higher than that previously reported in the general population, implying that OSA might be a potential etiology or, at least, an aggravating factor for the development or progression of PE or might be responsible for the postoperative recurrence of PE in some patients. Further studies are needed to clarify this relationship.

Keywords: *Obstructive sleep apnea, Pectus excavatum, Prevalence*

Introduction

Laryngomalacia is the most common congenital laryngeal anomaly and has been found to be associated with both obstructive sleep apnea (OSA) [1,2] and pectus excavatum (PE) [3,4]. Similar to patients with laryngomalacia, patients with OSA experience upper airway obstruction that results in chest retraction during inspiration. However, only limited studies have evaluated the association between OSA and PE. In 1992, Castiglione *et al*. reported that 82% of children with OSA had PE [5]. In 2016, Ma *et al*. reported that a 5-year-old child with previously known persistent snoring during sleep for more than 4 years experienced aggravated very severe sternum depression mimicking PE during an episode of upper airway infection. Polysomnography (PSG) revealed that he had severe OSA. His severe OSA and sternal depression improved after an adenotonsillectomy which was done for correction of the OSA but not for correction of PE [6]. The authors suggested that OSA might aggravate sternal depression. Sternal depression might persist in mimicking PE if OSA is not treated for a long time. However, not all OSA patients exhibit PE clinically. No previous studies have established the prevalence of OSA in patients with PE, and it is not known if patients with PE have a higher incidence of OSA than those without PE. In conducting this pilot study, we aimed to evaluate the prevalence of OSA in patients with PE.

Materials and methods

This prospective observational cohort study was conducted from October 2015 to September 2016 at Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation. The study was approved by the hospital's Institutional Review Board (protocol No. 04-XD15-056) and was supported by a grant from the Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation (TCRD-TPE-105-36). All participants provided informed consent.

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Participants

Forty-two consecutive patients ≥20 years old with PE were invited to participate in the study. Inclusion criteria were as follows: no previously established diagnosis of OSA, no known lung disease (asthma or chronic obstructive pulmonary disease), no known cardiovascular disease, no known psychological disease, and no current use of any hypnotics. Among the 42 patients invited to participate, 31 patients agreed to participate and completed the validated Chinese version of the Epworth sleepiness scale (ESS) questionnaire [7] and underwent a full-night PSG the night before Nuss surgery.

Assessment of clinical characteristics

Data regarding patient medical history, surgical indications, disease severity assessed by the Haller index, body height (BH), body weight (BW), body mass index (BMI), neck circumference (NC), waist circumference (WC), hip circumference (HC), waist-to-hip ratio (WC/HC ratio), smoking status, occupation, pulmonary function test results, ESS results, and snoring habits were collected.

Overnight polysomnography

Standard full-night PSG studies were performed on all 31 study participants the night before Nuss surgery. All participants had a recording of ≥6 h. The standard monitoring included simultaneous electroencephalography, electrooculography, chin and bilateral anterior tibialis surface electromyography, electrocardiography, measurement of airflow through the nose and mouth with a thermistor, measurement of thoracoabdominal movements with respiratory inductive plethysmography, position sensing with respiratory inductive plethysmography, snore sensing, and measurement of oxygen saturation with pulse oximetry. All sleep technicians had received appropriate training from the Taiwan Society of Sleep Medicine and had at least 1 year of experience. PSG results were analyzed by manual scoring of every 30-s epoch. Sleep stage was scored by trained sleep technicians according to the standard criteria of Rechtschaffen and Kales [8]. Respiratory events were scored according to the standard criteria of the American Academy of Sleep Medicine [9]. The apnea/hypopnea index (AHI) was calculated as the total number of apnea and hypopnea events per hour of sleep. Snoring was subjectively recorded by trained sleep technicians as mild, moderate, or severe.

Definition of obstructive sleep apnea

OSA was defined as an AHI ≥5 events/h. The severity of OSA was classified as follows: mild with an AHI of 5.0–14.9 events/h, moderate with an AHI of 15.0–29.9 events/h, and severe with an AHI ≥30 events/h.

Definition of the severity of snoring

The severity of snoring was subjectively graded by trained sleep technicians as mild, moderate, or severe according to routine practice.

Definition of excessive daytime sleepiness

Excessive daytime sleepiness (EDS) was defined as an ESS score ≥10.

Statistical analysis

The continuous variables of age, BH, BW, BMI, NC, WC, HC, WC/HC ratio, and pulmonary function test results were normally distributed and are presented as the mean ± standard deviation; the correlation of disease severity of OSA (AHI) and severity of PE (Haller index) was evaluated using the Pearson correlation coefficient using the 22nd edition of SPSS software (IBM Corporation, New York, United States). Multivariate analysis was performed to evaluate the correlations between the Haller index, age, sex, NC, WC/HC ratio, BMI, forced vital capacity (FVC) (percentage of predicted), and AHI.

Results

Table 1 shows the clinical characteristics of the 31 study patients with PE. The indications for Nuss surgery for PE correction were noncosmetic in 87.1% of patients and cosmetic in the other 12.9%. Most patients were male (90.3%), nonstudents (71.0%), and nonsmokers (90.3%). The mean patient age was 26.1 ± 5.7 years with a range of 20–42 years. The mean BH was 173.6 ± 7.1 cm, the mean BW was 61.7 ± 7.6 kg, and the mean BMI was 20.6 ± 2.5 kg/m^2. Pulmonary function testing showed a mean percent of predicted FVC of 83.7 ± 13.9%, a mean forced expiratory volume in 1 s (FEV1) of 85.5 ± 13.8%, and a mean FEV1/FVC of 86.6 ± 7.2%.

Table 2 shows that all patients snored; 35.5% had mild, 38.8% had moderate, and 25.8% had severe snoring. Ten of the 31 patients (32.3%) reported EDS with an ESS score ≥10. The overall prevalence of OSA in the 31 study patients was

Table 1: Clinical characteristics of 31 patients with pectus excavatum

Haller index, mean±SD	4.1±1.4
Surgical indication	
Cosmetic, *n* (%)	4 (12.9)
Noncosmetic, *n* (%)	27 (87.1)
Gender, *n* (%)	
Male	28 (90.3)
Female	3 (9.7)
Occupation, *n* (%)	
Student	9 (29.0)
Nonstudent	22 (71.0)
Age (years), mean±SD	26.1±5.7
Smoker, *n* (%)	3 (9.7)
BH, cm, mean±SD	173.6±7.1
BW, kg, mean±SD	61.7±7.6
BMI, kg/m^2, mean±SD	20.6±2.5
NC	35.1±2.2
WC	77.2±7.9
HC	91.9±5.5
WC/HC	0.8±0.1
Pulmonary function tests	
FEV1/FVC, %, mean±SD	86.6±7.2
FEV1, percentage of predicted, mean±SD	85.5±13.8
FVC, percentage of predicted, mean±SD	83.7±13.9

SD: Standard deviation, FEV1: Forced expiratory volume in 1 s, FVC: Forced vital capacity, BMI: Body mass index, BH: Body height, BW: Body weight, NC: Neck circumference, WC: Waist circumference, HC: Hip circumference, WC/HC: Waist-to-hip ratio

Table 2: Sleep-related parameters in 31 patients with pectus excavatum

	n (%)
Excessive daytime sleepiness	
Epworth sleepiness scale ≥10	10 (32.3)
Snoring, *n* (%)	31 (100)
Mild	11 (35.5)
Moderate	12 (38.7)
Severe	8 (25.8)
Obstructive sleep apnea	
Prevalence, overall, *n* (%)	8 (25.8)
Prevalence in men, *n* (%)	8 (100)
Prevalence in women, *n* (%)	0
Mild	3 (37.5)
Moderate	2 (25.0)
Severe	3 (37.5)

25.5% (8 of 31), and all the 8 patients with OSA were men. Among the 8 patients with PE and OSA, 3 had mild, 2 had moderate, and 3 had severe OSA.

The Pearson correlation coefficient showed no correlation between the AHI (severity of OSA) and Haller index (severity of PE) ($r = -0.068$, $P = 0.715$). Multivariate analysis showed that there were no statically significant differences in any variances (Haller index, age, sex, NC, WC/HC ratio, BMI, and FVC percentage of predicted) compared with the AHI (the F value of Wilks' lambda = 1.247, $P = 0.379$).

Discussion

The indications for Nuss surgery for correction of PE are mainly cosmetic in children and adolescents under 20 years old [10]. However, an increasing number of adult patients (>20 years old) with PE are undergoing Nuss surgery for noncosmetic reasons [11]. Our patients were primarily nonstudent adults with noncosmetic indications for Nuss surgery.

Known risk factors for OSA are age, high BMI, large NC, male gender, retrognathia or micrognathia, macroglossia, alcoholism, the use of sedatives or hypnotic medications, and craniofacial abnormalities. Our PE patients were young, tall, and thin; none used alcohol, sedatives, or hypnotic medications, and they were not at a high risk for developing OSA. Pulmonary function tests showed only mild restrictive impairment, which likely had no effect on saturation during sleep and did not interfere with the diagnosis of OSA. However, we did not perform cephalometry or assess other risk factors for OSA such as upper airway or craniofacial abnormalities. Therefore, it is unknown whether our patients had other risk factors associated with the development of OSA or whether PE *per se* could cause OSA.

The prevalence of snoring was 13.5% in 20–23-year-old Korean soldiers [12] and 14.1% in the Chinese general population over 20 years old [13]. In adults 30–60 years old, the snoring prevalence ranges from 15.0% to 35.7% [14-16]. Therefore, the 100% snoring prevalence in our study patients with PE was remarkably elevated. Since snoring is highly associated with OSA [17], the prevalence of OSA in our PE patients might be higher than we observed. One possible explanation for this might be the "first-night effect, " a combination of poor sleep quality and less deep sleep during the first night of PSG, which has been reported to affect PSG results and might cause underestimation of the AHI [18]. Our patients might have experienced poor sleep and a lower AHI because their PSG studies were done the night before Nuss surgery, making them more anxious and less likely to attain deep sleep during the study. However, some investigators have reported that the first-night effect does not affect sleep study results [19,20].

The previously reported overall prevalence of OSA among young- and middle-aged adults 39.6 ± 17.5 years old was 4.3% in the Chinese general population [21], with a prevalence of 4.0%–8.1% in men and 2.0%–2.5% in women [Table 3] [12,14-16,21-23]. In 1998, Bixler *et al*. reported a 7.9% prevalence of OSA among men 20–44 years old with an unknown BMI [23]. In 2013, Lee *et al*. reported an OSA prevalence of 8.1% among 20–23-year-old male soldiers in Korea who had a higher BMI than our patients [12]. Among our 14 male participants with the same age distribution as Lee's male soldiers, however, with a relatively lower BMI (19.9 ± 2.0), there was still a 7.1% (1 out of 14) prevalence of OSA. Our patients seemed to have a higher prevalence of OSA than that reported in these previous studies. In 2007, Liu *et al*. reported a 4.3% prevalence of OSA among 14–60-year-old individuals with a mean age of 39.6 ± 17.5 years and BMI of 21.6 ± 6.4 kg/m^2 in the general population [21]. Even though our patients seemed to have BMIs similar to that seen in their patients, the prevalence of OSA was still higher in our study. Therefore, even though our study lacked a control group for comparison, we believe that our findings demonstrated a possibly higher prevalence of OSA in patients with PE.

Severe chest wall deformity may lead to restrictive ventilatory impairment, resulting in hypoxemia and hypercapnia by altering chest wall mechanics and producing ineffective respiratory muscle mechanics, particularly in rapid eye movement sleep and the supine position. Patients with chest wall deformity often complain of disrupted nocturnal sleep with reduced deep sleep and rapid eye movement sleep, headaches, and EDS, mimicking the typical symptoms of OSA [24]. This could explain why PSG has not been done in clinical practice to evaluate these patients for OSA. In addition, it is worth mentioning that pulmonary function tests in our participants showed only mild restrictive ventilatory impairment which would not induce the above sleep problems. However, 32.3% our participants had EDS, but only 25.8% had confirmed OSA on PSG, suggesting that the prevalence of OSA in our participants might be higher than the rate of 25.8% we obtained.

OSA might aggravate sternal depression [6]; however, it is unclear if OSA can contribute to PE since we showed no correlation between the severity of OSA and severity of PE. Further studies are needed to evaluate if OSA contributes to PE.

Using the Pearson correlation coefficient and multivariate analysis, we found no correlation between the Haller index and known risk factors (NC, BMI, age, and WC/HC ratio) for OSA and the AHI. This might be due to the relatively small sample size. Since the Haller index (severity of PE) was not correlated

Table 3: Prevalence of snoring and obstructive sleep apnea in previous studies and the present study

Study	Country	Type of PSG	Population studied	Age, years/old mean±SD	BMI, kg/m² mean±SD	Snoring prevalence (%)	OSA prevalence		
							Overall (%)	Among men (%)	Among women (%)
Liu *et al.*, 2007 [21]	China	Type 1	14-60 year olds	39.6±17.5	21.6±6.4	27.3	4.3	5.9	2.5
Young *et al.*, 1993 [14]	America	Type 1	30-60 year olds	N/A	N/A	35.7	-	4.0	2.0
Ip *et al.*, 2001 [15]	Hong Kong	Type 1	30-60-year-old men	41.2±6.4	23.9±3.5	23.0	-	4.1	-
Ip *et al.*, 2004 [16]	Hong Kong	Type 1	30-60-year-old women	41.6±7.4	22.4±3.2	15.0	-	-	2.1
Lee *et al.*, 2013 [12]	Korea	Type 3	20-23-year-old men, soldiers	21.6±1.0 (snorers) 21.8±1.2 (nonsnorers)	23.9±3.0 (snorers) 22.9±2.7 (nonsnorers)	13.5	-	8.1	-
Present study, 2017	Taiwan	Type 1	20-42 year olds	26.1±5.7	20.6±2.5	100	25.8	25.8	0

BMI: Body mass index, PSG: Polysomnography, OSA: Obstructive sleep apnea, SD: Standard deviation, N/A: Not available

with the AHI (severity of OSA) in our study, there might be more risk factors than OSA alone in the development of PE. Further larger studies are needed to evaluate this.

There were several limitations in the present study. First, the total number of study patients was really too small to be representative of the general population. Second, our study lacked a healthy control group for comparison. Previous studies of the prevalence of OSA in the general population focused primarily on middle-aged or older adults. We found only a few studies of OSA in young adults. The prevalence and severity of OSA are highly affected by age and the screening tools employed [25], and we were not able to precisely compare OSA prevalence among different ages, BMIs, and ethnicities. Third, it has been found that 80% of middle-aged adults with OSA are undiagnosed [26]. Therefore, the true prevalence of OSA among middle-aged adults in the general population might have been underestimated in previous studies, and the 25.8% prevalence of OSA in our patients with PE might not be higher than that seen in the general population. Larger multicenter studies should be conducted to determine if untreated OSA in patients with PE results in a higher recurrence of PE after Nuss surgery, if treatment of OSA in patients with PE can reduce the recurrence rate after Nuss surgery, and to determine the optimal timing and type of treatment for OSA in patients with PE who have undergone Nuss surgery.

Conclusions

The prevalence of OSA in patients with PE seemed higher than that previously reported in the general population, suggesting that OSA might be a potential etiology or aggravating factor for the development of PE, or might be responsible for the postoperative recurrence of PE in some patients. PSG study is recommended before Nuss surgery for PE correction. If patients have both PE and OSA, they should be closely monitored after Nuss surgery correction to check if the PE will recur because of OSA related sternal depression. PSG should be followed up after removal of the bars. Further larger studies are needed to evaluate the prevalence of OSA in patients with PE, to determine whether adequate treatment of OSA is needed for patients with PE before or after Nuss surgery, and to address the optimal timing of OSA treatment for patients with PE.

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Conflicts of interest

There are no conflicts of interest.

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