Improvement in Polysomnographic Objective Sleep Quality in Adults with Pectus Excavatum After the Nuss Procedure

Mei-Chen Yang^{1,2} · Chou-Chin Lan^{1,2} · Yao-Kuang Wu^{1,2} · Min-Shiau Hsieh^{2,3} · Bo-Chun Wei³ ·
Yeung-Leung Cheng^{2,3}

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Abstract

Background Patients with pectus excavatum have a poorer subjective sleep quality and quality of life than the general population. The Nuss procedure has been shown to improve these patients' quality of life, but data regarding their postoperative sleep quality are lacking. We aimed to evaluate the objective sleep quality of adults with pectus excavatum before and after the Nuss procedure.

Methods Twenty-eight participants completed this study. Epworth Sleepiness Scale (ESS) scores for daytime sleepiness, Pittsburgh Sleep Quality Index (PSQI) scores for subjective sleep quality, and overnight polysomnography for objective sleep quality were evaluated before and 6 months after the Nuss procedure.

Results Subjective sleep quality improved after the Nuss procedure. The median PSQI score decreased from 7 [interquartile range (IQR): 5; 9] to 5 (IQR: 4; 7, $p = 0.029$); the median percentage of poor PSQI sleep quality decreased from 64.3 to 35.7% ($p = 0.048$). The median percentage of rapid eye movement sleep significantly increased after surgery [15.6% (IQR: 12.2%; 19.8%) vs. 20.4% (IQR: 14.5%; 24.9%), $p = 0.016$]. Sleep interruptions also improved, with the median arousal index decreasing from 9.5 (IQR: 4.8; 18.2) to 8.2 (IQR: 4.3; 12.1; $p = 0.045$). However, there was no significant change in ESS scores after surgery ($p = 0.955$).

Conclusions Pectus excavatum may be associated with poor subjective and objective sleep quality in adults, and the condition may improve after the Nuss procedure. For adults with pectus excavatum who report poor subjective sleep quality, polysomnography should be considered to assess their preoperative and postoperative sleep condition.

Introduction

Pectus excavatum (PEx) is the most common congenital chest deformity caused by overgrowth of costal cartilages. As the deformed chest wall worsens, the compression of the heart and lungs may become serious, resulting in clinical symptoms and affecting the patient's quality of life (QOL) [1–6].

Surgical correction is the main treatment for PEx. A minimally invasive technique for PEx repair was introduced by Dr. Donald Nuss in 1998 [7]. The Nuss procedure uses steel plate(s) under the depressed thoracic wall with the aid of a thoracoscope. The popularity of this procedure has increased rapidly [3, 8, 9]. The procedure provides

✉ Yeung-Leung Cheng
ndmc0928@yahoo.com.tw

¹ Division of Pulmonary Medicine, Department of Internal Medicine, Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, New Taipei, Taiwan

² School of Medicine, Tzu Chi University, Hualien, Taiwan

³ Division of Thoracic Surgery, Department of Surgery, Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, No. 289, Jianguo Rd., Xindian Dist., New Taipei 23143, Taiwan

support to the depressed thoracic wall, thereby reducing the compression on cardiopulmonary organs.

Previous studies have shown that QOL improved after the Nuss procedure [3, 10–13]. However, sleep quality is an important aspect of QOL and should be included in QOL evaluations [14, 15]. The Pittsburgh Sleep Quality Index (PSQI) is widely used to evaluate subjective sleep quality, and the Epworth Sleepiness Scale (ESS) questionnaire is used to assess daytime sleepiness [16, 17]. Our team conducted a study to examine the subjective sleep quality of adult PEx patients using the PSQI questionnaire. We found that PEx may be associated with poor subjective sleep quality, which may improve after Nuss surgery [18]. To date, objective sleep quality has not been investigated in PEx adults. We aimed to evaluate objective sleep quality using polysomnography in PEx adults before and after Nuss surgery.

Materials and methods

Participants

The study was approved by the institutional review board of Taipei Tzu Chi Medical Foundation in New Taipei City, Taiwan (Protocol No.: 04-XD15-056). All adults with PEx scheduled for Nuss surgery at the Division of Thoracic Surgery in Taipei Tzu Chi Hospital from October 2015 to June 2017 were recruited. The inclusion criteria were ≥ 20 years of age with a Haller index ≥ 3.2 [19], no psychiatric or medical illness (heart, renal, and/or liver disease or chronic airway obstruction), and no use of psychoactive, soporific, or illegal drugs. Participation was completely voluntary, and written informed consent was obtained from each participant.

Baseline clinical characteristics, including age, gender, body height (BH), body weight (BW), body mass index (BMI, kg/m^2), pulmonary function evaluated by spirometry, and postoperative data, were collected. The indications for surgical repair followed the criteria proposed by Dr. Nuss [20].

The surgical techniques, which employed a bilateral thoracoscopy-assisted Nuss procedure, were performed by Cheng as previously described and demonstrated on video [18]. All patients were interviewed at baseline, with a follow-up about 6 months after Nuss surgery. The patients were encouraged to return to normal daily activity and normal exercise 3 months after the operation.

A total of 108 patients were scheduled for a Nuss procedure during the study interval. Sixty-eight patients met the inclusion criteria and were invited to participate in the study; 32 patients agreed. These patients underwent the Nuss procedure as scheduled without perioperative complications and

recovered uneventfully during hospitalization. These patients were asked to complete the PSQI questionnaire, ESS questionnaire, and overnight polysomnography evaluations at baseline and about 6 months after the Nuss procedure.

Assessment of daytime sleepiness

Daytime sleepiness was subjectively evaluated using the Chinese version of the ESS questionnaire [17].

Assessment of subjective sleep quality

The validated Chinese version of the PSQI questionnaire was used to assess subjective sleep quality [16, 21]. The scale consists of 19 items with 7 components: sleep quality (PSQI-1), sleep latency (PSQI-2), sleep duration (PSQI-3), sleep efficiency (PSQI-4), sleep disturbance (PSQI-5), frequency of sleeping-medication use (PSQI-6), and daytime functional impairment (PSQI-7). Each PSQI item was rated from 0–3 (0 = no difficulty, 3 = extreme difficulty). The PSQI sleep quality was calculated using total PSQI scores, which ranged from 0 to 21 (higher total PSQI scores indicated poorer sleep quality). Good sleep quality was defined as total PSQI scores ≤ 5 , while poor sleep quality was defined as total PSQI scores ≥ 6 .

Assessment of objective sleep quality

Standard overnight polysomnography was performed at the sleep center. During polysomnography, electroencephalography (EEG), electrooculography, chin and bilateral anterior tibialis surface electromyography, electrocardiography, and assessments of airflow through the nose and mouth (registered by a thermistor and nasal pressure sensor), thoracoabdominal movements (registered by respiratory inductive plethysmography), position (by a sensor on the respiratory inductive plethysmography unit), snoring, and oxygen saturation (by pulse oximetry) were performed. The polysomnography lasted for at least 6 h and was analyzed via manual scoring of every 30 s epoch by trained sleep technicians and reviewed by a sleep specialist. Sleep stage was manually scored using the standard criteria proposed by Rechtschaffen and Kales [22]. An apnea event was defined as an 80–100% reduction in airflow lasting for at least 10 s. A hypopnea event was defined as a reduction in airflow of at least 50% for at least 10 s or of at least 30% for at least 10 s in comparison with the baseline and associated with at least 3% oxygen desaturation or an EEG arousal. The apnea/hypopnea index (AHI) was calculated as the ratio of the number of apnea and hypopnea events per hour. The desaturation index (DI) was defined as $>3\%$ oxygen desaturation per hour. The arousal index (AI) was defined in terms of the number of arousal

episodes per hour [23]. Sleep efficiency was defined as the fraction of total sleep time to total recording time. Sleep latency was defined as the time from lights off to the first identifiable sleep stage. Rapid eye movement (REM) latency was defined as the time from the first identifiable sleep stage to the first REM sleep.

Statistical analysis

Continuous variables were presented as the median and interquartile range (IQR: 25th and 75th percentiles) and all displayed non-normal distribution. Categorical variables were summarized as a count (%). Continuous variables before and after the Nuss procedure were compared using the Wilcoxon signed-rank test. Categorical variables before and after the Nuss procedure were compared using the Fisher exact test. p values < 0.05 were considered statistically significant. Statistical analyses were performed using SPSS software (version 24; SPSS; Chicago, IL, USA).

Results

Among the 32 participants, four withdrew from the study after operation (one studied in Germany, one worked in the Republic of China, one was excluded due to a surgical complication [flipping of the bar found one month after surgery], and one was lost to follow-up). Thus, 28 patients were included in the final analysis. The mean hospital stay was 6.5 ± 1.1 days. The mean postoperative follow-up period was 16.5 months (range: 12–20 months).

Table 1 shows the demographic characteristics of the 28 participants. The median age was 24.0 years (IQR: 22.0; 27.5). The majority of the patients were male (25/28 patients: 89.3%). The median BMI was 20.2 kg/m^2 (IQR: 18.9; 21.9). None of the patients smoked cigarettes. The median Haller index was 3.6 (IQR: 3.3; 4.3). The median forced expiratory volume in 1 s/forced vital capacity (FEV1/FVC) was 87.5% (IQR: 83.3; 91.0), with the median FEV1 being 83.5% (IQR: 73.5; 91.3) and median FVC being 80% (IQR: 69.5; 91.3). Only two patients (7.1%) had restrictive ventilatory impairment. The median postoperative interval for performing the overnight polysomnography examination was 6.54 months (range: 5.8–10.6 months).

Table 2 shows the ESS scores, PSQI, and polysomnography results before and after the Nuss procedure. The median ESS scores showed no significant difference after surgery (6.5 vs. 6.0, $p = 0.955$). Total PSQI scores decreased from 7 to 5 ($p = 0.029$) (Fig. 1a). The median percentage of poor PSQI sleep quality significantly decreased from 64.3 to 35.7% ($p = 0.048$). There was no

Table 1 Demographic characteristics of 28 adult patients with pectus excavatum

Characteristics	Cases ($n = 28$)
Age, years (IQR)	24.0 (22.0; 27.5)
Gender	
Male, n (%)	25 (89.3%)
Female, n (%)	3 (10.7%)
Body height, cm (IQR)	173 (167; 177.8)
Body weight, kg (IQR)	62.5 (55.0; 65.8)
Body mass index, kg/m^2 (IQR)	20.2 (18.9; 21.9)
Haller index (IQR)	3.6 (3.3; 4.3)
Pulmonary function rest	
FVC, % of predicted value (IQR)	80.0 (69.5; 91.3)
FEV1, % of predicted value (IQR)	83.5 (73.5; 91.3)
FEV1/FVC, % (IQR)	87.5 (83.3; 91.0)
TLC, % of predicted value (IQR)	88.5 (84.0; 96.0)
Restrictive lung defect, n (%)	2 (7.1%)

Continuous data (age, body height, body weight, body mass index, Haller index) are shown as the median and interquartile range, and categorical data (gender) are shown as a count (%)

IQR interquartile range, FVC forced vital capacity, FEV1 forced expiratory volume in 1 s, TLC total lung capacity

statistical difference in subjective PSQI sleep efficiency after surgery (91.3% vs. 90.5%, $p = 0.988$). For polysomnography results, the objective sleep efficiency showed no difference before and after the Nuss procedure (77% vs. 78.6%, $p = 0.284$). N1 and N2 sleep did not differ after surgery (8.1% vs. 9.5%, $p = 0.716$ and 56.4% vs. 59.9%, $p = 0.829$, respectively). Non-rapid eye movement (NREM) sleep significantly decreased after surgery (84.4% vs. 79.6%, $p = 0.015$), which was mainly due to the decrease in N3 sleep (18.1% vs. 12.7%, $p = 0.019$). REM sleep significantly increased after the Nuss procedure (15.6% vs. 20.4%, $p = 0.016$) (Figs. 1b and 2). The AI significantly improved after the Nuss procedure (9.5 vs. 8.2, $p = 0.045$). There were no differences in sleep latency, REM latency, mean and nadir SaO_2 , mean heart rate, DI, or AHI after the Nuss procedure.

Discussion

Our results showed that adults with PEx may be associated with both poor subjective and objective sleep quality (less REM sleep and more interrupted sleep), which could be improved after the Nuss procedure. To our knowledge, this is the first study using polysomnography to evaluate objective sleep quality in PEx patients. Although our study sample was small, the gender distribution and demographic characteristics of our patients were similar to those in

Table 2 Comparison of the subjective sleep quality, daytime sleepiness, and objective sleep quality of 28 adults with pectus excavatum before and after Nuss procedure

Characteristics	Before Nuss procedure	After Nuss procedure	<i>p</i> value
Daytime sleepiness (ESS) (IQR)	6.5 (4.3; 8.8)	6 (4; 10.5)	0.955
Subjective sleep quality (PSQI)			
Total PSQI score	7 (5; 9)	5 (4; 7)	0.029*
PSQI sleep quality			0.048*
Good sleep quality (PSQI $\leq$ 5), <i>n</i> (%)	10 (35.7%)	18 (64.3%)	
Poor sleep quality (PSQI $\geq$ 6), <i>n</i> (%)	18 (64.3%)	10 (35.7%)	
Sleep efficiency, % (IQR)	91.3 (85.7; 100)	90.5 (86.0; 100)	0.988
Objective sleep quality (polysomnography variables)			
Sleep efficiency, % (IQR)	77.0 (65.6; 83.9)	78.6 (72.5; 87.2)	0.284
N1 stage, % (IQR)	8.1 (5.5; 14.6)	9.5 (6.6; 12.2)	0.716
N2 stage, % (IQR)	56.4 (51.7; 63)	59.9 (48; 65.0)	0.829
N3 stage, % (IQR)	18.1 (10.7; 22.1)	12.7 (7.3; 17.2)	0.019*
NREM stage (N1 + N2 + N3), % (IQR)	84.4 (80.2; 87.8)	79.6 (75.1; 85.4)	0.015*
REM stage, % (IQR)	15.6 (12.2; 19.8)	20.4 (14.5; 24.9)	0.016*
Sleep latency, min (IQR)	42.5 (23.0; 76.3)	42.5 (21.5; 88.1)	0.767
REM latency, min (IQR)	82.5 (66.8; 148.0)	87.3 (69.4; 119.9)	0.936
Mean SaO ₂ , % (IQR)	95.0 (95.0; 96.0)	95.0 (95.0; 96.8)	0.134
Nadir SaO ₂ , % (IQR)	91.0 (89.0; 92.0)	91.5 (88.0; 93.0)	0.767
Mean heart rate, beats/min (IQR)	64.0 (55.8; 69.5)	65.0 (60.0; 68.5)	0.213
Desaturation index, events/h (IQR)	1.9 (0.4; 2.9)	0.7 (0.2; 3.1)	0.143
Apnea–hypopnea index, events/h (IQR)	2.8 (0.9; 6.4)	2.1 (0.3; 4.9)	0.194
Arousal index, events/h (IQR)	9.5 (4.8; 18.2)	8.2 (4.3; 12.1)	0.045*

Continuous variables are shown as the median (interquartile range) and were analyzed using the Wilcoxon signed-rank tests. Categorical variables are shown as the count (%) and were analyzed using Fisher's exact test. $p < 0.05$ indicated significant difference before and after the Nuss procedure

ESS Epworth sleepiness scale, IQR interquartile range, PSQI Pittsburgh sleep quality index, NREM non-rapid eye movement, REM rapid eye movement, SaO₂ oxygen saturation

Fig. 1 a, b Comparison of total Pittsburgh Sleep Quality Index (PSQI) scores among adults with PEX before and after Nuss procedure. **a** PSQI scores significantly decreased after surgery. **b** REM sleep significantly increased after the Nuss procedure. (Minimum, maximum, median, *Q*1, and *Q*3 values are shown in box and whisker plots, and the measurement results are shown in **a** and **b**)

Fig. 2 Polysomnography results of a 25-year-old male with pectus excavatum (body height 168 cm; body weight 56 kg, body mass index 19.8) before (upper part) and after Nuss procedure (lower part). Rapid eye movement sleep increased after surgery (14.2% vs. 20.0%). Sleep fragmentation improved after surgery

previous studies [24, 25]. As such, our sample may represent the actual situation of PEx patients; our results are valuable because they provide an additional surgical indication for the Nuss procedure in PEx adults.

The QOL of PEx patients was often evaluated in previous investigations. Our previous study showed the subjective sleep quality of adults with PEx was poorer than that of the general population and could be improved after the Nuss procedure [18]. In this study, we obtained similar results. The PSQI questionnaire is a relatively quick and simple way to evaluate subjective sleep quality. We believe the PSQI questionnaire is valuable for evaluating sleep quality as a part of the QOL of adults with PEx. Shen et al. found PSQI score positively correlated with age in hemodialysis patients (elderly patients had higher PSQI scores, poorer subjective sleep quality) [26]. Our patients were all aged over 20 years. A future study focusing on individuals younger than 20 years of age is needed.

In 1981, Guilleminault et al. studied the polysomnography findings of five patients with severe restrictive lung disease (severe kyphoscoliosis-related musculoskeletal deformities) and found these patients often complained of disrupted sleep with frequent arousal because of sleep-associated hypoxemia and hypercapnia [27]. We also observed interrupted sleep in our patients; however, we did not observe hypoxemia or hypercapnia, and the pulmonary function tests yielded relatively normal results. However, the interrupted sleep improved after the Nuss procedure, suggesting there might be other factors beyond hypoxemia and hypercapnia disrupting their sleep. The physiological thoracic wall compression caused by PEx or its

psychological impact might have a negative impact on sleep continuity. Further studies are needed to address this issue.

Patients with restrictive lung disease also had excessive daytime sleepiness, as thoracic cage deformity alters chest wall mechanics [28]. In our study, the ESS scores did not change after operation because the ESS questionnaire only assessed patients' attention ability while performing daily activities such as watching television, talking with others, or waiting for a traffic light. The ESS questionnaire is used clinically to evaluate the severity of sleep-disordered breathing (SDB) and might be not suitable to evaluate the degree of fatigue in dynamic daily activities such as housework or exercise. Other methods, such as the 6-minute walking test for submaximal exercise capacity or a comprehensive cardiopulmonary test for maximal exercise capacity, might be more useful for daytime fatigue evaluation in PEx patients.

Although polysomnography remains the gold standard in sleep investigations, there is some debate regarding the use of single-night recordings because of the first-night effect and the night-to-night variability in AHI [29]. The normal range of sleep efficiency is 85–95%, and the normal ranges of N1, N2, N3, and REM sleep for young adults are 2–5%, 45–55%, 13–23%, and 20–25%, respectively [30]. The first-night effect, mainly characterized by lower sleep efficiency, longer sleep and REM latency, more N2 sleep, and less N3 and REM sleep during the first night, has been reported on the first two consecutive nights of polysomnography rest [29, 31]. In the present study, the patients received the second polysomnography examination more than 6 months after the first. In our data, only

REM sleep showed significant improvement after operation. Even so, in previous studies, elderly patients, especially those aged more than 40 years, showed a greater first-night effect [31, 32]. Most of our patients were aged 20–30 years. Therefore, our results were not entirely influenced by the first-night effect.

There are many factors affecting sleep quality, including age, BMI, upper respiratory tract variation, brain injury, alcohol, and drug use. The median age of our patients was 24 years, and the median BMI was 20.2. None of the patients had other special medical circumstances or histories of long-term medication use, making this study population at low risk of poor sleep quality. The subjective PSQI questionnaire and the polysomnography in this study showed meaningful improvement after surgery. As such, it can be preliminarily conceived that the chest depression itself might be an important factor affecting the sleep quality of PEx patients. Therefore, the improvements in REM sleep and sleep continuity in our patients after the Nuss procedure might be attributed to the effects of surgical correction of the pectus deformity. The median AI in our patients was 9.5 per hour, similar to that in the general population [23]; the AI improved after the Nuss procedure, indicating that correction of the pectus deformity may have a beneficial effect on sleep continuity. Since one of the characteristics of SDB is sleep fragmentation with increasing AI, we believed that the improvement in AI of our patients might be the result of the operation.

Indications for the Nuss procedure for PEx include two or more of the following criteria: (1) cardiopulmonary compression symptoms, (2) Haller index > 3.2, (3) restrictive/obstructive lung impairment, (4) mitral valve prolapse, and (5) cardiac compression [7, 18]. The indications for surgery in our patients were based on these criteria. Our present study and previous study [18] revealed that subjective and objective sleep quality in PEx adults was poor and could be improved after the Nuss procedure. In terms of clinical application, if an adult with PEx reports poor subjective sleep quality, the practitioner might consider polysomnography to further evaluate their objective sleep quality, to exclude other sleep-disordered breathing, and to evaluate the effect of the Nuss procedure.

There were several limitations to our study. First, the sample size was small and might not represent the entire population. Second, we only focused on PEx adults. There might be age-related differences between adults and children with PEx. Third, we did not include normal Taiwanese subjects for comparison. This approach may have overlooked racial differences. Larger multicenter (even international) studies are warranted.

In conclusion, we found PEx in adults may be associated with poor subjective and objective sleep quality, which

could be improved after the Nuss procedure. For adults who report poor subjective sleep quality, polysomnography should be considered as an auxiliary method to evaluate their sleep quality and surgical outcome.

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Compliance with ethical standards

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

Ethical approval The Institutional Review Board of Taipei Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation approved this study on August 20, 2015 (Protocol No: 04-XD15-056).

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

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