Polysomnography is an Important Method for Diagnosing Pediatric Sleep Problems: Experience of One Children's Hospital

Chien-Heng Lin[1], Chieh-Ho Chen[1], Syuan-Yu Hong[1], I-Ching Chou[1], Shinn-Jye Liang[1], and Liang-Wen Hang[1]

[1]China Medical University

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Abstract

In this study, we collected and analyzed PSG data to investigate that value of polysomnography (PSG) in diagnosing sleep problems in children. The results of PSG studies of children (< 18 years old) with sleep problems conducted from April 2015 to May 2017 at a children's hospital in Taiwan were collected and analyzed retrospectively. Data for 310 patients (209 males and 101 females) who underwent PSG were collected. The final diagnoses were as follows: obstructive sleep apnea in 159 (51.3%), snoring in 81 (26.4%), limb movement sleep disorder in 25 (8.1%), hypersomnias in 12 (3.9%), central apnea in 8 (2.9%), enuresis in 7 (2.3%), bruxism in 5 (1.6%), sleep terrors in 5 (1.6%), narcolepsy in 3 (1.0%), sleep seizures in 3 (1.0%), sleep walking in 1 (0.3%), and insomnia in 1 (0.3%). PSG may help detect significant sleep-related problems in children and is useful for making therapeutic decisions regarding children. Obstructive sleep apnea syndrome (OSAS) was the primarily sleep problem for most of the children (51.3%); however, only 7.4% of them underwent surgery for OSAS, even though those with OSAS underwent surgery without undergoing PSG. We therefore suggest that children with sleep problems should undergo PSG.

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Running head: Pediatric sleep problems and polysomnography

Chien-Heng Lin, MD[1], Chieh-Ho Chen, MD[1], Syuan-Yu Hong, MD[2], I-Ching Chou, MD[2], Shinn-Jye Liang, MD[3], Liang-Wen Hang, MD[3]*

[1]Division of Pediatric Pulmonology, China Medical University Children's Hospital, Taiwan

[2]Divison of Pediatric Neurology, China Medical University Children's Hospital, Taiwan

[3]Sleep Medicine Center, Department of Pulmonary and Critical Care Medicine, China Medical University Hospital, Taiwan

***Corresponding Author:** Liang-Wen Hang, MD, Sleep Medicine Center, Department of Pulmonary and Critical Care Medicine, China Medical University Hospital

No. 2, Yuh-Der Road, Taichung 404, Taiwan.

Tel: +886-2-2194-5179

Fax: +886-2-2194-746

E-mail: sub179090@gmail.com

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Abstract

In this study, we collected and analyzed PSG data to investigate that value of polysomnography (PSG) in diagnosing sleep problems in children. The results of PSG studies of children (< 18 years old) with sleep problems conducted from April 2015 to May 2017 at a children's hospital in Taiwan were collected and analyzed retrospectively. Data for 310 patients (209 males and 101 females) who underwent PSG were collected. The final diagnoses were as follows: obstructive sleep apnea in 159 (51.3%), snoring in 81 (26.4%), limb movement sleep disorder in 25 (8.1%), hypersomnias in 12 (3.9%), central apnea in 8 (2.9%), enuresis in 7 (2.3%), bruxism in 5 (1.6%), sleep terrors in 5 (1.6%), narcolepsy in 3 (1.0%), sleep seizures in 3 (1.0%), sleep walking in 1 (0.3%), and insomnia in 1 (0.3%). PSG may help detect significant sleep-related problems in children and is useful for making therapeutic decisions regarding children. Obstructive sleep apnea syndrome (OSAS) was the primarily sleep problem for most of the children (51.3%); however, only 7.4% of them underwent surgery for OSAS, even though those with OSAS underwent surgery without undergoing PSG. We therefore suggest that children with sleep problems should undergo PSG.

Introduction

Sleep constitutes an opportunity for the body to conserve energy, restore its normal processes, promote physical growth, and support mental development. It also plays a vital and often underestimated role in the growth and development of children. Sleep problems have been reported to have high prevalence rates throughout childhood, affecting 25% to 50% of preschoolers and up to 40% of adolescents[1-3].

The taking of a detailed medical history is the first method that should be applied to screen for and identify pediatric sleep problems, followed by polysomnography (PSG), which is a powerful diagnostic sleep medicine tool that can be used to continuously and simultaneously record multiple different physiological parameters of a sleeping individual.

In this study, we retrospectively collected the data of children who underwent PSG in our hospital due to sleep problems. The indications of these PSG studies and the final diagnoses of these patients were then analyzed and evaluated.

Materials and Methods

Ethical approval was provided by the ethical review board of China Medical University Hospital (CMUH103-REC2-082). Patient consent was not required for this research. The data for children (< 18 years old) with sleep problems who came to the outpatient department (OPD) of China Medical University Children's Hospital for help and underwent PSG from April 2015 to May 2017 were collected and analyzed retrospectively. However, data for those who underwent follow-up PSG studies after adenotonsillectomy or for other reasons were excluded in this study.

The indications for PSG include snoring, sleep apnea, other sleep-related breathing disorders, excessive daytime sleepiness, limb movements while sleeping, bruxism, enuresis, sleep walking, insomnia, difficulty falling sleeping, and others.

A level 1 PSG study is performed in our sleep laboratory with a sleep technologist present, and 12 channels are recorded, including eight channels of electroencephalography (EEG), 2 channels of electrooculography (EOG), 1 channel of submentalis (chin) electromyography (EMG), and 1 channel of electrocardiogram (ECG)/heart rate, and 1 channel of pulse oximetry (SpO_2). A multiple sleep latency test (MSLT) was also arranged for the day after any PSG study for those children who had a history of more than 3 months of excessive daytime sleepiness.

Obstructive sleep apnea syndrome (OSAS) was diagnosed based on the PSG data when obstructive events were noted and the apnea-hypopnea index (AHI) was one or more per hour, with 1< AHI [?]5 taken to indicate mild OSA, 5< AHI [?]10 taken to indicate moderate OSAS, and an AHI >10/hr of total sleep time (TST) taken to indicate severe OSAS.

The periodic limb movements (PLMs) during sleep were scored if there were at least four movements of 0.5–5/sec' duration that occurred between 5 and 90s apart. A PLM index of >5 per hour of sleep is generally considered to be rare in normal children, and therefore this threshold was used to define the presence of periodic limb movement disorder (PLMD).

Central sleep apnea (CSA) was defined as the absence of both inspiratory effort and chest wall movement lasting longer than 20 seconds when accompanied by a central apnea index greater than 1.

Furthermore, the presence of two or more sleep-onset rapid eye movement (REM) periods (SOREMPs) and a mean sleep latency of <8 minutes on the MSLT was regarded as being diagnostic of narcolepsy.

Results

Data for a total of 325 patients with sleep problems who came to the OPD for help from April 2015 to May 2017 and underwent PSG were collected in our study. Among these patients, a total of 242 patients had problems of snoring, sleep apnea, and other sleep-related breathing disorders. Forty of the patients had involuntary limb movements during sleep. Twenty-seven of the patients had symptoms suggestive of parasomnias such as bruxism, enuresis, and sleep walking. Twenty-two patients had a history of excessive daytime sleepiness, and one had difficulty falling asleep.

A total of 310 of the children (209 males and 101 females aged from 5-17 years old, with a mean age of 8.5 years and median age of 10 years) had positive findings in the PSG and/or MSLT studies.

The final diagnoses in the 209 male patients were as follows: OSAS in 109 (52.2%), primary snoring in 57 (27.3%), PLMD in 19 (9.1%), idiopathic hypersomnia in 8 (3.8%), central apnea in 6 (2.9%), enuresis in 6 (2.9%), sleep terrors in 3 (1.4%), narcolepsy in 3 (1.4%), sleep seizures in 1 (0.5%), 1 bruxism in 1 (0.5%). There were 5 male children who were diagnosed with both OSAS and PLMD. In the 109 male children with OSAS, 80 had mild OSAS, 24 had moderate OSAS, and 5 had severe OSAS. Furthermore, 27 of them had asthma or allergic rhinitis, 5 had obesity, 2 had Duchenne muscular dystrophy, 1 had prematurity, and 1 had mucopolysaccharidosis (MPS) type 2. In the 19 male children with PLMD, 4 had underlying diseases: 2 had developmental delays, 1 had Tourette syndrome, and 1 had hydrocephalus. In the 6 male children with central apnea, 4 had congenital heart disease after operation, 1 had ganglioglioma post operation, and the other did not have any underlying diseases.

The final diagnoses in the 101 female patients were as follows: OSAS in 50 (49.5%), primary snoring in 24 (23.8%), limb movement sleep disorder in 13 (12.9%), idiopathic hypersomnia in 4 (4.0%), bruxism in 4 (4.0%), sleep terrors in 2 (2.0%), sleep seizures in 2 (2.0%), enuresis in 1 (1.0%), and sleep walking in 1 (1.0%). There were 2 female children who had both OSAS and PLMD. In 101 female children with OSAS, 66 had mild OSAS, 31 had moderate OSAS, and 4 had severe OSAS. Furthermore, 10 of them had allergic rhinitis, 2 had diabetes mellitus, 1 had Down syndrome, and 1 had Prader-Willi syndrome. In the 13 female children with PLMD, 4 had underlying diseases: 2 had a history of febrile convulsion, 1 had a history of syncope history, and 1 had cerebral palsy. In the 2 female children with central apnea, 1 of them had congenital heart disease after operation, and the other did not have any underlying diseases. All of the patients' final diagnoses are shown in **Table 1** .

The methods used to manage the 270 of the 310 total patients with positive findings (40 patients did not come back to the OPD for follow-up visits) were as follows: operation with adenoidectomy and tonsillectomy was suggested for 37 patients with moderate or severe OSAS, and 23 patients (15 males and 8 females) (23/37=62%, or 7.4% of all the children enrolled in this study) underwent the operation; continuous positive airway pressure (CPAP) was used in 2 patients with central apnea (0.6%); medical treatment was used in 190 patients (61.3%); and the remaining patients were only placed under observation (n=99, 31.9%) (**Figure 1**). For the 14 patients with moderate or severe OSAS who did not undergo an operation, a repeated PSG study 6-12 months later was suggested, but only 3 of them returned for follow-up visits, and their AHI results were decreased at those follow-ups.

Discussion

As with adults, there are all sorts of reasons why children do not sleep well. Some of those reasons are more serious than others. In our study, the sleep problem of the majority of the children (51.3%) was OSAS; however, only 7.4% of the total pediatric patients in the study underwent surgery for OSAS, even though some of the children with OSAS underwent surgery without undergoing preoperative PSG.

The etiologies of pediatric OSAS are multiple, and can be classified into intrinsic upper airway narrowing or increased upper airway collapsibility [4]. Adenotonsillar hypertrophy is currently the most common cause of intrinsic upper airway narrowing, with other anatomical features resulting in upper airway narrowing including craniofacial syndrome, achondroplasia, Down syndrome, Beckwith Wiedemann syndrome, and MPS. In our study, three patients had underlying diseases with intrinsic upper airway narrowing: one had Prader-Willi syndrome, one had Down syndrome, and one had MPS. Adenotonsillectomy was suggested for these three patients, but their families refused this surgical intervention.

A decrease in muscle tone in the upper airway can cause increased upper airway collapsibility, with potential cause of such decreases including cerebral palsy, neuromuscular disorders, or inflammatory conditions such as allergic rhinitis and asthma. In our study, 2 OSAS patients had the underlying disease of Duchenne muscular dystrophy, and 24.5% (37/159) of the OSAS patients had allergic rhinitis or asthma.

Obesity appears to facilitate the emergency of OSAS; therefore, there is a high prevalence of OSAS among obese children. However, there is a higher proportion of children with OSAS who are obese. Thusm it appears that both OSAS and obesity can coexist and potentiate the adverse impacts of one another [5]. The prevalence of OSAS is increasing globally due to the growing occurrence of obesity in society. In obese children, the fat deposits in the upper respiratory tract cause breathing difficulties during sleep, thus causing OSAS[6,7], with reports of the prevalence of OSAS in obese children ranging from 13 to 59% [8]. In our study, 3.1% (5/159) of the children with OSAS were also obese, and all of them were male patients. One of these 3 patients had severe OSAS and underwent an adenotonsillectomy, while it was recommended that the other 2 patients achieve body weight reductions through an increase in their dialy intense physical activities. A recent cross-sectional, prospective multicenter study, the NANOS study, assessed the contribution of obesity and adenotonsillar hypertrophy to pediatric OSAS and found that 46.6% of obese children in the community had OSAS [9].

Adenotonsillectomy is generally considered the first-line therapy in children with moderate or severe OSAS. In our study, there were 65 children with moderate OSAS and 9 with severe OSAS, and it was suggested that 26 of the moderate cases and all 9 of the severe cases undergo an operation. Of those 37 patients, 62% (23/37) underwent the operation. Some of the patients did not receive the operation because they their family members wanted to further observe their symptoms for a period of time. For these fourteen patients who did not undergo the operation, repeated PSG studies were performed for 3 of them who come back for follow-up visits, and their AHI results were found to be decreased in those follow-ups.

PSG prior to adenotonsillectomy is indicated for children with some conditions that increase the risk of perioperative respiratory complications. These conditions include obesity (especially if severe), Down syndrome, craniofacial abnormalities, neuromuscular disorders, sickle cell disease, or MPS [10]. The purpose of the PSG in these high-risk children is to improve diagnostic accuracy and define the severity of OSAS to optimize perioperative planning. In our study, the OSAS children for whom the operation was suggested did have at least one of the aforementioned conditions, but some, including 3 with obesity, 2 with Duchenne muscular dystrophy, 1 with Down syndrome, 1 with MPS, and 1 with Prader-Willi syndrome did not undergo the operation.

Several authors have recommended that a clinical reevaluation be given to all children several months after adenotonsillectomy to determine whether snoring and the symptoms of OSAS have been resolved, especially in those children with higher risk of persistent disease, such as those with severe obesity or craniofacial syndromes. Furthermore, a postoperative PSG should be considered even in the absence of snoring or other symptoms in order to determine whether additional treatment is necessary for residual OSAS[4]. However, no studies to date have evaluated the timing of postoperative PSG evaluations, and this issue is not specifically

addressed in the practice guidelines regarding the management of pediatric OSA. In our study, we did not collect postoperative PSG data, so further studies should be designed for evaluating this issue.

Medical therapies such as anti-inflammatory agents or CPAP are used as alternatives to adenotonsillectomy for children with OSAS, depending on the severity and specific locations of airway obstruction in the individual patient, and on associated comorbidities.

The second most common sleep problem among the children in our study was primary snoring, and the management of pediatric primary snoring consists of treating any upper airway obstructions or observation in cases in which there is no upper airway obstruction.

The third most common sleep problem in our study was PLMD. Thirty-one children (10%) had PLMD proven by PSG, and 7 of those 31 also had OSAS. In the large clinical case series reported by Gingras JL et al., PLMD was found to be common, affecting 14% of the 468 referred children[11]. It is relevant to note that there are no Food and Drug Administration-approved treatments for PLMD in children. However, most children with PLMD have low iron storage; therefore, iron therapy should be considered as the first line of treatment in children with PLMD whose iron levels are low. In our study, the iron level was checked in 9 children, and only one child with PLMD had a low iron level and was thus treated with iron therapy. Therefore, our management for PLMD consisted non-pharmacologic treatments, such as education, massage, exercise, or observation only.

The fourth most common sleep problem in our study was idiopathic hypersomnia, which is extremely rare in children. Hypersomnia is present in 4% to 6% of the general population, with only 1% of the population having idiopathic hypersomnia and most of the people with that being adolescents or adults [12]. According to a report by Han F et al., 86% (361/417) of the children presenting with a complaint of primary hypersomnia to a sleep clinic in China met the criteria for narcolepsy with cataplexy [13], while only 20% (3/15) of the children with excessive daytime sleepiness in our study had narcolepsy.

The clinical characteristics and experiences of CSA are very limited in children compared to the adult population, and it is thought to occur in about 1-5% of healthy children [14]. CSA has been noted to occur more commonly in children with underlying diseases, and the presence of CSA may influence the course of those diseases[14]. In our study, there were 8 children who had CSA; 6 of them had secondary CSA and 2 had idiopathic CSA. Idiopathic CSA is really rare in children, and it cannot be reliably identified or diagnosed on the basis of history or a specific set of signs and symptoms [14]. For the 2 children with a PSG-based diagnosis of CSA in this study, we suggested a magnetic resonance imaging (MRI) evaluation to assess for neuroanatomical abnormalities, but neither of the children came back for a follow-up visit.

A study by Felix O et al. reported that 18 of the 441 (4.1%) patients recorded during the study period had CSA, while 8 of the 310 (1.9%) patients in our study had central apnea [15]. In the study by Felix O et al, the underlying disorders were dominated by neurosurgical disorders; however, congenital heart diseases dominated in our study.

Many parasomnias in children can be recognized by history alone, but some require nocturnal PSG for appropriate diagnosis and management. In our study, there were 7 patients who had enuresis, 5 who had bruxism, and 1 who had sleep walking according to the PSG results, with these 13 patients accounting for 4.2% of all the patients who underwent PSG. We believe however that there are still so many children with parasomnias who do not go to pediatric OPDs for help.

There were several limitations in our study. The first was that our study was only a 2-year retrospective study, and some children with sleep problems, especially those suspected of having insomnias and parasomnias, may go to psychiatric OPDs for help, not pediatric OPDs, and thus may not undergo a PSG study. Second, long-term follow-up is necessary for children with sleep problems in order to observe whether their symptoms are relieved or their AHI results are decreased after treatment, especially for those with OSAS after adenotonsillectomy. Third, blood sample tests, such as tests of serum Fe or ferritin levels, are not typically conducted for children with PLMD. Further studies prospectively collecting PSG data from children

with pediatric sleep disorders will thus be required.

Conclusion

The sleep problem of the majority of the children (51.3%) in this study was OSAS; however, only 7.4% of the total pediatric patients in the study underwent surgery for OSAS, even though some of the children with OSAS underwent surgery without undergoing PSG. We thus suggest that children with sleep problems should all undergo PSG studies, as PSG could help to detect significant sleep-related problems, and the application of PSG results is useful for making therapeutic decisions regarding children.

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Figure legend

Figure 1 Management of 310 patients

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Table 1.pdf available at https://authorea.com/users/390224/articles/504622-polysomnography-is-an-important-method-for-diagnosing-pediatric-sleep-problems-experience-of-one-children-s-hospital