

Review of Pediatric Polysomnographic Studies over a 2-year Period in a Tertiary Care Hospital in Bengaluru, India

Ilin Kinimi¹, Neha Mohan Rao²

ABSTRACT

Introduction: Pediatric polysomnography is the gold standard in diagnosing sleep-related breathing disorders, the most common cause of this being obstructive sleep apnea (OSA). Instituting good sleep hygiene measures, early recognition of symptoms and signs of sleep-disordered breathing, and timely referral for polysomnography play a vital role in improving a child's quality of life. We report our experience with sleep disorders in children and the challenges faced during diagnosis.

Aims: To review indications for pediatric sleep studies done over 2 years and their outcomes in a tertiary care center in Bengaluru, India.

Methods: Retrospective descriptive analysis of 65 pediatric sleep studies done between 2018 and 2020 (2-year period) for various indications.

Results: The total numbers of studies included in the analysis were 65, of which complete studies were done in 58 children and portable studies in 7 children. The most common indication for performing a sleep study was suspected OSA, followed by referrals for children with spinal muscular atrophy and Duchenne muscular dystrophy, respectively. Average age of the patient was 93.56 months (range 3–216 months). The number of boys and girls included in the study was 45 and 20, respectively.

The average duration of sleep was 375.35 minutes, of which rapid eye movement (REM) sleep contributed to 45.33 minutes (12.13%) (range, 0–90 minutes). The average sleep latency time was 21.36 minutes (range, 0.5–138 minutes). The total apnea–hypopnea index (AHI) was 12.64/hour with 26 (40%) children having mild OSA, defined as an AHI of less than 5/hour; 17 (26.1%) having moderate OSA, defined as an AHI of 5 to 10/hour; 22 (33.8%) having severe OSA defined as an AHI of 15 to 30/hour. The mean REM AHI was 29.67/hour. Thirty-four children were started on bilevel positive airway pressure (BiPAP) following the study and four children had their BiPAP setting titrated following the study with one child whose high flow oxygen was continued.

Conclusion: Of the 65 pediatric sleep studies included, 39 children (60%) underwent some form of intervention following the study, indicating the clear benefit of performing polysomnography and indicating a high prevalence of sleep disorders in children. Mouth breathing was the most common symptom and OSA was the most common diagnosis.

Keywords: Polysomnograph, Sleep disorders, Sleep study.

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INTRODUCTION

Sleep is important for the growth and development of children and is vital for a child's developing brain. Assessing and advocating good sleep hygiene measures are an essential part of the evaluation of every well-child, as important as nutrition and/or immunization practices. Pediatricians, during their routine practice, should look out for symptoms and signs that may indicate sleep-disordered breathing, thereby ensuring that a greater number of children with sleep-related disturbances are identified early and referred for polysomnography.

Over the last decade, the importance of sleep in the overall well-being of a child has been increasingly recognized; however, awareness among parents and pediatricians is often lacking. Lack of or poor quality of sleep is associated with poor cognitive development and school performance.¹ Sleep deprivation contributes to several short-term effects such as poor attention, hyperactivity and other behavioral disturbances, excessive daytime sleepiness, and emotional problems. Short sleep duration during early childhood also has significant long-term effects like poor receptive vocabulary during middle childhood,¹ lower cognitive performance in neurodevelopmental tests,² and increased risk of excess weight or obesity in childhood.³ In addition, long-term effects of untreated obstructive sleep apnea (OSA) which contribute to mortality include failure to thrive, right-sided heart failure, and metabolic complications.

^{1,2}Department of Pediatrics, Manipal Hospital, Bengaluru, Karnataka, India

Corresponding Author: Ilin Kinimi, Department of Pediatrics, Manipal Hospitals, Bengaluru, Karnataka, India, Phone: +91 9980306721, e-mail: ilinkinimi@gmail.com

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Parents of children should routinely be asked for symptoms such as habitual snoring, mouth breathing, labored breathing, sleeping in odd positions (for instance, with an extended neck), restless sleep, behavioral concerns, excessive daytime sleepiness, and/or daytime hyperactivity—all of which could indicate OSA. OSA is characterized by prolonged episodes of increased upper airway resistance associated with partial or complete obstruction of the upper airway, resulting in disruption of normal gas exchange and sleep patterns.⁴ Signs such as overcrowded or maloccluded teeth, high arched palate, stertor at rest, or adenotonsillar hypertrophy suggest OSA. Risk factors for OSA

include obesity, adenotonsillar hypertrophy, and presence of allergic rhinitis or chronic nasal obstruction.⁵ In addition to OSA, children with underlying risk factors such as neurological, genetic, or craniofacial syndromes should be formally evaluated with a pediatric polysomnograph as a part of their routine screening.

In order to obtain an objective diagnosis of obstructive sleep-related breathing disorders, the gold standard is a polysomnograph or a formal sleep study. Pediatric scoring guidelines for polysomnographs in children have been established and are commonly used to score sleep studies.⁶ In India, there are a handful of centers where sleep studies are performed because of various challenges such as the lack of awareness of its importance, lack of equipment that is specifically manufactured for pediatric use, lack of sleep labs, and lack of technicians skilled in performing these studies.

The incidence of sleep-related disorders in Indian children is very high and it has been estimated that approximately 47.5% of Indian school-going children suffer from some form of sleep disorder.³ Sleep disorders affect children and their families, schools, pediatricians, and therefore the community at large. We aim to sensitize pediatricians about the importance of sleep hygiene, help in early recognition of symptoms and signs indicating sleep-related disorders, aid timely referral, in addition to highlighting possible timely interventions to improve the child's quality of life.

MATERIALS AND METHODS

In this observational study, we retrospectively analyzed records of pediatric sleep studies done in our center over 2 years (2018–2020) in children below the age of 18 years. Our sleep study laboratory in Manipal hospitals, Old Airport Road, Bengaluru is one of few centers in the country doing level 1 polysomnographs. A total of 67 pediatric sleep studies were done between the years 2018 and 2020. Of these, 65 were included in the analysis. The two patients excluded were either above the age limit (patient with Duchenne muscular dystrophy (DMD) aged 26 years) or had a portable study done for a duration of only 30 minutes in the pediatric intensive care unit (ICU) before the child became unwell (child with critical illness myopathy—post-bone marrow transplant).

Details about each patient including demographics, date of study, indication for the study were obtained and details of each study were entered into Microsoft Excel and were analyzed. Of the total 65 studies included, complete polysomnography was performed for 58 children, which included an electroencephalogram, electrocardiogram, electrooculogram, respiratory movements (thoracic and abdominal bands), leg movements, airflow using nasal and oral canisters, microphone, oxygen saturations, heart rate monitoring, and a video recording. We included 7 patients who underwent a portable study since they were admitted in the pediatric ICU and were too unwell to be shifted to the sleep lab for a complete sleep study.

Overnight complete sleep studies were conducted in a dedicated pediatric sleep laboratory by a qualified on-site sleep technician with experience in pediatric polysomnography. Based on the modified American Academy of Sleep Medicine scoring manual (pediatric criteria), scoring was done by a qualified pediatric pulmonologist. Patients and parents of patients were explained about the study and all measures to make the sleep lab as child-friendly as possible were taken into account.

RESULTS

Of the 65 sleep studies, 58 were complete studies and 7 were portable studies. The portable studies were done in the pediatric ICU under strict hemodynamic monitoring. There were 12 studies done in 2018, 35 in 2019, and 18 done until June 2020.

Of these, four studies were performed on children who were previously on bilevel positive airway pressure (BiPAP) in order to titrate their settings and one was performed on a child who was receiving high-flow oxygen.

From the data available, symptoms that patients presented with were mouth breathing (*n* = 6), snoring (*n* = 4), decreased activity (*n* = 1), fatigue (*n* = 1), or excessive drowsiness during the day (*n* = 1), obesity (*n* = 3) or poor weight gain (*n* = 4), decreased appetite (*n* = 1), poor school performance (*n* = 1), speech delay (*n* = 1), recurrent upper airway infections (*n* = 2), restlessness (*n* = 1), jerky movements during sleep (*n* = 1), allergic rhinitis and wheeze (*n* = 1), and headache (*n* = 1). On examination, five patients had adenotonsillar hypertrophy and two had evidence of pulmonary hypertension on echocardiography.

We received referrals for sleep studies from the ENT team, the neurological team, the neurosurgical team and the general pediatric team. The most common indication for referral for a sleep study was suspected OSA and this was present in 25 patients. After OSA, the commonest reason for referral was neurological disorders such as spinal muscular atrophy (SMA) (*n* = 25) and DMD (*n* = 8). The other indications for sleep studies included congenital syndromes such as Down syndrome (*n* = 1), Sanjad–Sakati syndrome (*n* = 1), Apert syndrome (*n* = 1), and prune belly syndrome (*n* = 1). In addition, one child each with congenital heart disease and pulmonary hypertension, myopathy under evaluation, limb girdle muscular dystrophy, myotonic dystrophy, and stroke under evaluation underwent sleep studies. Lastly, one child with postventilation subglottic stenosis and one with chronic lung disease also underwent polysomnography.

Table 1: Causes of OSA in our center

Grading of OSA	AHI	Causes
Mild	<5/hour	<ul style="list-style-type: none"> • SMA (5) • Adenotonsillar hypertrophy (2) • Limb girdle muscular dystrophy (1) • Tuberosous sclerosis (1) • Prune belly syndrome (1) • Chronic lung disease with pulmonary hypertension (1)
Moderate	5-10/hour	<ul style="list-style-type: none"> • SMA (5) • DMD (2) • Adenotonsillar hypertrophy (2) • Down syndrome (1) • Congenital heart disease (1) • Sanjad–Sakati syndrome (1) • Stroke (1) • Postventilation subglottic stenosis (1)
Severe	>10/hour	<ul style="list-style-type: none"> • SMA (9) • DMD (5) • Apert syndrome (1) • Myopathy (1) • Myotonic dystrophy (1)

The average age of patients in the study was 93.56 months or 7 years and 9 months. Of the 65 children, there were 45 boys and 20 girls. The average duration of sleep was 375.35 minutes with average duration of rapid eye movement (REM) sleep being 45.33 minutes (range, 0–90 minutes), accounting for 12.13% of total sleep duration. The mean sleep latency time was 21.36 minutes (range, 0.5–138 minutes).

Data for distorted architecture of sleep were available in 62 patients, of which 61 had evidence of distorted sleep. Of 60 patients for whom data on awakening episodes during sleep were available, 21 had episodes of awakenings. 59 studies had data on sleep associated respiratory disturbances and arousals, and 54 and 57 studies had evidence of each of these, respectively. The average arousal and awakening index (Ar + Aw index) was 10.85.

Of the 65 patients, 63 had evidence of hypopnea and 45 had apnoeic episodes. 41 had obstructive apneas, 13 had central apnea and 17 had mixed apneas which included 18 children having a combination of two types of apneas. The average apnea–hypopnea index (AHI)(total) was 12.64, with an average REM AHI of 29.67 (average of 60 patients). Grading of OSA based on AHI showed 26 (40%) children having mild OSA, defined as an AHI of less than 5/hour; 17 (26.1%) having moderate OSA, defined as an AHI of 5 to 10/hour; 22 (33.8%) having severe OSA defined as an AHI of 15 to 30/hour.⁷

Percentage of studies with desaturations was 86% (56 of 65 studies).

Of the 65 patients, 4 patients were already on BiPAP and their settings were titrated; an additional 34 patients were started on BiPAP following their sleep study, and 1 child was continued on high-flow oxygen support.

DISCUSSION

Problems with sleep are grossly under-recognized in the pediatric age group. A recent cross-sectional study done in Chennai, India estimated that 34% of children visiting the pediatric OPD had some form of sleep disorder.⁸ Statistically significant risk factors for childhood sleep disorders were described to be history of sleep problems in infancy, absence of siblings, and presence of parents while sleeping. However, in the same study, only 4.5% of patients with sleep problems were referred for formal polysomnography.

In our cohort, the most common indication for performing a sleep study was children with symptoms suggestive of OSA (38.4% of patients) which has been described as the most common indication for pediatric polysomnography in the past.²

Our data showed that almost all children had some form of respiratory disturbance during sleep, either in the form of distortion of sleep architecture or awakenings and arousals. However not all children had complaints related to sleep and were referred as a part of routine screening in complex disorders, indicating that performing a polysomnograph is an important part of the screening routine of such patients, in order to formally assess the quality and quantity of sleep-related breathing disorders. Our data also highlighted about one-fourth of patients who underwent polysomnography to have moderate or severe OSA. In addition, more than 50% of patients were started on BiPAP following their sleep study in order to relieve their symptoms and are being followed up.

Parents of all children should be educated about instituting routine sleep hygiene measures including sleep-promoting practices such as quiet, dark bedroom, regular bed/wake times, and a consistent routine, and practices that interfere with sleep such as alcohol intake or smoking, excessive daytime napping or

excessive screen time must be avoided. In a systematic review and meta-analysis done in 2016, access to and use of a media device at bedtime was significantly associated with inadequate sleep quantity, poor sleep quality, and excessive daytime sleepiness.⁷ In addition, parental education and socioeconomic status have also been described as factors that influence the incidence of sleep disorders.⁸

Reasons for under-recognition of sleep-related disorders in children are multifactorial. Children are unable to vocalize their problem; warning signs are considered as normal and often ignored by parents and pediatricians, possible reasons being that they do not visibly seem to harm the child. Poor school performance is generally attributed to a lack of interest. In addition, there is a lot of anxiety noticed in parents and children to perform a sleep study due to several reasons such as cost of the test, the process of sleeping in a new environment, and the requirement of hospitalization. This may also alter sleep study findings and must be taken into account before planning an intervention. Several studies have been done to assess the efficacy of home unattended polysomnography;^{9–11} however, further studies are required to understand the feasibility of these in India. A few measures that can be instituted in order to make the sleep lab more child-friendly include allowing an accompanying parent in the room, age-appropriate bedding (crib/cot/bed), and explaining the procedure beforehand to children who are old enough to help with psychological preparedness.²

In addition to OSA, pediatric polysomnographs are used to assess patients with central apneas and hypoventilation syndromes. In children with neuromuscular disorders such as SMA or DMD, who may not present with classical symptoms such as snoring due to respiratory muscle weakness, polysomnograph is used as a screening tool in order to assess the quality of sleep and if needed, intervene to provide adequate positive end-expiratory pressure, thereby helping decrease the frequency of lower respiratory tract infections. In addition, PSGs are used in children with chronic lung diseases such as bronchopulmonary dysplasia to titrate the oxygen requirement and for guiding decannulation of tracheostomy. Children with parasomnias, restless leg syndrome, and periodic limb movement disorders may also benefit from polysomnographs.

OSA, which is the commonest cause of sleep-disordered breathing is the most common indication for performing a pediatric polysomnograph. Brockman et al., in their systematic review, compared the diagnostic test accuracy of different tests to PSG for the diagnosis of OSA, they found sleep-lab based polygraphy, anterior rhinomanometry, and urinary biomarkers to have comparative diagnostic testing accuracy to PSG, but with limited evidence.¹² Specific features of OSA such as arousals, apneas, hypopneas, and desaturation events can be analyzed and classification of disease severity based on AHI can be done using a PSG. However, PSG may not be used for predicting consequences of OSA and occasionally clinical manifestations and PSG measurements may be difficult to correlate.¹² While an AHI of greater than 1 is considered abnormal, the clinical picture of the child and impact on the child's quality of life must be taken into consideration before planning an intervention. There is also increasing evidence of OSA being associated with monosymptomatic nocturnal enuresis (Table 1).¹³

Mild OSA can be treated medically with intranasal corticosteroids and/or leukotriene antagonists. Other interventions for OSA, usually caused by adenoid–tonsillar hypertrophy, include tonsillectomy and adenoidectomy (T and A) in those with moderate to severe symptoms. Additionally, weight management, use of traction

devices in a select population, and other forms of surgical intervention may be considered. In patients with neuromuscular disorders or severe OSA despite T and A, respiratory support in order to provide positive pressure ventilation such as BiPAP at night may be initiated. PSG may also be useful in titrating pressures for children on BiPAP. Children with craniofacial malformations undergoing surgical correction may require a PSG both preprocedure and postprocedure as part of their evaluation.

Interventions such as BiPAP/T and A require good teamwork between pediatricians, pulmonologists, orthodontists, ENT, and pediatric surgery teams. It also requires good communication with the patient's family and adherence to sleep hygiene measures despite these interventions. Following treatment, it is imperative that these children are followed up for residual symptoms to identify persistent disease and take remedial action.

Limitations of this report include the absence of measurement of carbon dioxide (CO₂) levels during polysomnography. However in patients with central apnea hypoventilation syndromes or those in whom CO₂ toxicity was a concern, an arterial blood gas was done after the study to assess the severity of CO₂ toxicity. Additionally, since the design of the study was retrospective, although all effort was taken to collate complete data, some data variables were found to be missing due to lack of entry by the physician at the time of consultation.

CONCLUSION

In this report, we highlight the need for formal evaluation of sleep-related disorders in children and discuss challenges faced while performing polysomnographic studies. In addition to describing the variety of clinical indications for PSG, we wish to highlight that more than half the patients needed BiPAP as an intervention to help their symptoms.

CLINICAL SIGNIFICANCE

A greater number of child-oriented sleep laboratories with qualified personnel conducting the study can reduce the cost of these studies. Standardized interpretation of studies and timely interventions can significantly improve the child's quality of life.

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