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Research Article

Prevalence of Sleep Disruption and Obstructive Sleep Apnea in Children with Cerebral Palsy Using Diagnostic Polysomnography

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Abstract

Introduction: Sleep disruption and sleep disordered breathing have been extensively reported in patients with cerebral palsy (CP) by studies conducted using questionnaires. Polysomnography (PSG) is the gold standard for objectively determining sleep architecture and sleep disorder breathing (SDB) in children and in adults. In this study, we aim to evaluate sleep architecture abnormalities and estimate prevalence of obstructive sleep apnea (OSA) in CP patients using PSG. **Methods:** All patients (0-18 year) with physician confirmed diagnosis of cerebral palsy who underwent diagnostic PSG study between September 2019 and June 2023 at a tertiary pediatric hospital were included in the study. Clinical and PSG data were retrospectively collected, summarized and reported. Sleep staging and sleep related respiratory events were scored following AASM criteria. OSA diagnosis was considered present if apnea hypopnea index (AHI) was ≥1.5 events per hour of total sleep time. Comparison between patients with OSA and patients with no OSA was also conducted. Student t test or Qui square analysis were used as appropriate. P value >0.05 was considered significant. Results: A total of 65 patients (31 male and 34 females) were included in the study. The mean (range) age was 7.2 (0.9 - 18) years. Majority of patients had quadriplegic CP. Only 12 (18.4%) patients had history of snoring or sleep apnea. Mean (range) sleep efficiency was 69.9% (44%-95%). Mean (range) sleep latency was 52.7 (1-270) minutes. Mean (range) REM latency was 118.8 (2.5-341) minutes. Mean (range) wakefulness after sleep onset was 102.6 (0.5-273.7) minutes. Duration of all sleep stages (REM, N1, N2, N3) expressed as percentages of total sleep time was within normal limits. The mean (range) AHI was 4.2 (0-38) events/hour. The prevalence of OSA was estimated at 54%. However, only 12 patients had documented symptoms related to OSA. The OSA group was younger compared to the non-OSA group and less likely to have seizure disorders. There was no significant difference in the frequency of reported symptoms between both groups. **Conclusion:** We found normal sleep architecture in patients with CP but decreased sleep efficiency and high prevalence of OSA using PSG. History of sleep related respiratory symptoms was not frequent and did not correlate with PSG detected OSA. Therefore, screening for sleep disruption and OSA using PSG in these patients should be considered.

Keywords: Cerebral Palsy; Sleep Disorder Breathing; Obstructive Sleep Apnea; Polysomnography

Introduction

Cerebral palsy (CP) is a heterogeneous group of non-progressive neurological disorders caused by brain insult and results in permanent motor dysfunction affecting muscle tone, posture, and movement. Cerebral palsy is the most common cause of motor abnormalities seen in children [1]. As we learn more about CP pathophysiology, we understand that it's not an unchanging disorder caused by an early insult to a developing brain. CP is an evolving condition with genetic susceptibility to subsequent insults leading to multi-organ dysfunction associated with CP [2]. The prevalence of CP ranges from 1.5 to 3 per 1,000 live births and varies between countries [3].

Respiratory complications are the leading cause of morbidity and mortality in patients with CP [4]. Sleep related breathing disorders such as obstructive sleep apnea (OSA), central sleep apnea (CSA) and hypoventilation are common respiratory complications in these patients. Patients with CP are at increased risk for sleep disordered breathing (SDB) and is related to multiple structural and functional factors including pharyngeal and glossal hypotonia, retrognathia, maxillary hypoplasia, laryngomalacia and decreased central response to hypoxemia and hypercapnia [5]. Other CP related comorbidities such as scoliosis and seizure disorders can also contribute to sleep disordered breathing [2]. Sleep disruption not related to sleep disordered breathing has also been frequently reported including irregular sleep-wake cycle, abnormal initiation and maintenance of sleep,parasomnias and frequent arousals [6].

Sleep disruption and sleep disordered breathing in patients with CP have significant impact on the quality of their lives and the life of their care givers. Therefore, early recognition and early intervention of both are essential. Polysomnography (PSG) is the gold standard method for the diagnosis of both disruption in sleep architecture and for the diagnosis of SDB. Unfortunately, all studies evaluating sleep abnormalities in patients with CP have been questionnaire based. The Sleep Disturbance Scale for Children (SDSC) [7] and Pediatric Sleep Questionnaire (PSQ) [8] are used to screen sleep problems in pediatrics and are frequently used in pediatrics CP patients. However, these tools were not validated in CP patients against PSG gold standard. Also, studies evaluating prevalence of SDB among patients with CP using PSG is severely lacking. The purpose of this study is to characterize sleep architecture and to evaluate prevalence of SDB in pediatric patients with CP using polysomnography.

Methods

The study was designed as a retrospective evaluation of the prevalence of sleep architecture abnormalities and sleep discorded breathing in children with CP. Patients were identified from the electronic medical records by diagnosis code of cerebral palsy. All pediatric patients (age 0-18 year) who were diagnosed with CP by a physician and completed a diagnostic overnight polysomnography (PSG) at Sidra Hospital (Doha, Qatar) between September 2019 and June 2023 were included in the study. The following demographic and clinical data were obtained: age, gender, BMI, Gross Motor Function Classification System (GMFCS), sleep related respiratory symptoms (snoring, witnessed sleep apnea), comorbidities (i.e., seizure disorder, gastroesophageal reflux, asthma, allergic rhinitis, congenital heart disease, pulmonary hypertension), etiology of cerebral palsy, history of adenoid/tonsil hypertrophy, and anti-epileptic treatments.

The following PSG results were reviewed and collected: sleep efficiency, total sleep time, sleep latency, REM latency, wakefulness of sleep onset, arousal index, sleep stage percentages (REM%, N1%, N2%, N3%), Apnea Hypopnea Index (AHI), O_2 saturation, transcutaneous/end tidal CO_2, and oxygen desaturation index (ODI). Sleep staging and sleep related respiratory events including obstructive apneas, central apneas, obstructive hypopneas, central hypopneas as well as mixed apneas were scored according to the AASM published criteria [9]. OSA was defined as AHI ≥ 1.5 events/hour. OSA severity was defined as mild if AHI 1.5-4.99 events/hour, moderate if AHI 5-9.99 events/hour and severe if AHI ≥10 events/hour. Comparison between patients with OSA (OSA group) and patients without OSA (non-OSA group) was made in terms of clinical and demographic variables. Treatments of different sleep related breathing disorder including oxygen supplements, adenotonsillectomy, non-invasive ventilation support and tracheostomy were also reviewed.

Comparison between continuous variables was conducted using non-paired student t-test and comparison between categorical variables was conducted using Chi square. P value < 0.05 was considered statistically significant. The study was approved by the institutional review board at Sidra Medicine (IRB project # 2073906-1)

Results

A total of 65 patients (31 male and 34 females) with a physician-confirmed diagnosis of cerebral palsy (CP) were included in the study. The mean (range) age was 7.2 years (0.9 - 218). Mean (range) BMI z- score was -0.4 (-6.3 – 3.2). Most of the patients were diagnosed with quadriplegic CP (78.4%) and 86% of patients were non-ambulatory. Almost half of the patients had seizure (66.2%), and 22 (33.8%) patients did not have history of seizure disorder. The following comorbidities were documented: dysphagia in 81.5%, scoliosis in 58.4% and GERD in 47% of patients. Detailed demographic and clinical data are shown in (Table 1).

Characteristics	Total n=65	Non-OSA group n=30	OSA group N=35	P value
Age (years)	7.2(0.9-18)	8.2(2.1-18)	6.3(0.9-16.8)	0.02
Gender (M/F), n (%)	31/34 (48.5%/51.5%)	12/18	19/16	0.2
BMI (kg/m^2)	16.2(9.5-25.1)	16.4(11.9-25.1)	16(9.5-25.1)	0.2
BMI z- score, mean (range)	-0.4(-6.3-3.2)	-0.3(-3.5-3.2)	-0.4(-6.3-3)	0.7
Type of CP, n (%) • Quadriplegic • Diplegic • Others	 51/65(78.4%) 7/65(10.8%) 7/65(10.8%)	 26/30 (86.6%) 2/30 (6.7%) 2/30 (6.7%)	 25/35(71.4%) 5/35(14.3%) 5/35(14.3%)	0.2
Etiology, n (%) • Genetic mutation • Hypoxic ischemic encephalopathy • Post meningitis	 31/65(47.7%) 32/65(49.2%) 2/65(3%)	 13/30 (43.3%) 16/30 (53.3%) 1/30 (3.3%)	 18/35(51.4%) 16/35(45.7%) 1/35(2.9%)	0.5
Non-ambulatory, n (%)	56/65(86%)	29/31(93.5%)	28/35(80%)	0.04
Symptomatic, n (%) • Symptomatic • Non-symptomatic	 12/65(18.4%) 53/65(81.6%)	 3/30 (10%) 27/30 (90%)	 9/35 (25.7%) 26/35(74.3%)	0.1
Epilepsy, n (%) • None • Seizure	22/65(33.8) 43/65(66.2%)	 6/30 (20%) 24/30 (80%)	 16/35 (45.7%) 19/35 (54.3%)	0.02
Antiepileptic treatment, n (%) • None • Anti-epileptic treatment	 21/65(32.3%) 44/65(67.7%)	 6/30 (20%) 24/30 (80%)	 15/35 (42.9%) 20/35 (57.1%)	0.04

Comorbidities, n (%) • GERD • Dysphagia • Aspiration • Recurrent chest infections • Scoliosis • Cardiac	 31/65(47.69%) 53/65(81.5%) 19/65(29.2%) 17/65(26.1%) 38/65(58.4%) 3/65(4.6%)	 15/30 (50%) 26/30 (86.6%) 10/30 (33.3%) 10/30 (33.3%) 15/30 (50%) 1/30 (3.3%)	 16/35 (45.7%) 27/35 (77.1%) 9/35 (25.7%) 7/35 (20%) 23/35 (65.7%) 2/35 (5.7%)	 0.7 0.3 0.5 0.2 0.1 0.6
Feeding modality, n (%) • Oral • NG tube • G-tube • G-tube/Fundoplication • GJ tube	 17/65 (26%) 1/65(1.5%) 33/65(50.7%) 9/65(13.8) 4/65(6.15%)	7/30 (23.3%) 1/30 (3.3%) 17/30 (56.7%) 4/30 (13.3%) 1/30 (3.3%)	 10/35 (28.6%) 0/35 (0%) 17/35 (48.6%) 5/35 (14.3%) 3/35 (8.6%)	0.6
History of adenotonsillectomy, n (%)	3/65 (4.6%)	1/30	2/35	
Tracheostomy, n (%)	2/65(3%)	1/30	1/35	

Table 1: Baseline demographic and clinical characteristics of in all CP patients, CP patients without OSA and CP patients with OSA.

The mean (range) sleep efficiency defined as total sleep time divided by total recording time was 69.9% (44%-95%). Mean (range) sleep latency was 52.7 (1-270) minutes. Mean (range) REM latency was 118.8 (2.5-341) minutes. Mean (range) wakefulness after sleep onset was 102.6 (0.5-273.7) minutes. Mean (range) REM percentage of total sleep time was 19% (2.4-34.5), mean N1 percentage was 4.5% (0-19.6), mean N2 percentage was 47.8% (14.7-90.0) and mean N3 percentage was 28.6% (0-67.8). The Arousal index mean (range) was 9 (1.3-29.5).

AHI mean (range) was 4.2 (0-38) events/hour, REM AHI was 9.2(0 – 102.9) events/hour, and NREM AHI was 3.1(0 – 40) events/hour. Thirty-five patients were diagnosed with OSA based on AHI ≥ 1.5 events/hour, with prevalence rate of 54%. Only 12 patients had documented symptoms related to OSA. OSA was mild in 20 (57%) patients, moderate in 6 (17%) patients and severe in 9 (26%) patients. Average oxygen saturation mean was 96% and oxygen saturation nadir mean was 86%. The ODI mean (range) was 7.3(0-36) events/hour.

Patients with OSA were younger (P=0.02), more likely to be ambulatory (P= 0.04) and less likely to have seizure disorder (P=0.02) compared to patients with no OSA. Patients with OSA were also less likely to be diagnosed with seizure disorder. There was no significant difference in the sleep architecture between the OSA and the non-OSA group. However, there was significant statistical difference between both groups in the following SDB related PSG parameters: AHI, REM AHI, NREM AHI, OAHI, CAHI, average O_2 saturation, average REM O_2 saturation, average NREM O_2 saturation, O_2 nadir, ODI, Peak $ETCO_2$ and the arousal index. PSG data are detailed in (Table 2).

PSG findings	Total population (n=65)	Non-OSA group n=(30)	OSA group n=(35)	P value
Sleep efficiency	69.9(44-95)	70.1(44-97.9)	70.5(44-94)	0.7
Sleep latency	52.7(1-270)	45.9(1-247.5)	57.4(1-270)	0.5
REM latency	118.8(2.5-341)	130(2.5-341)	108.5(9-327.5)	0.2
WASO	102.6(0.5-273.7)	111.8(7-248)	91.8(0.5-273.7)	0.2
REM%	19(2.4-34.5)	18.4(5.6-33.7)	19.7(2.4-34.5)	0.4
N1%	4.5(0-19.6)	4.7(0.3-13.6)	4.3(0-19.6)	0.6
N2%	47.8(14.7-90.9)	46.2(19.2-73.9)	48.9(14.7-90.9)	0.5
N3%	28.6(0-67.8)	30.8(0-67.8)	27.1(0-62.7)	0.3
Sleep position, n (%) • Supine • Lateral	45/65(69.2%) 20/65(30.8%)	22/30(73.3%) 8/30(26.7%)	23/35(65.7%) 12/35(34.3%)	0.5
Snoring events, (%)	3.5(0-72.3)	1(0-26.5)	6.5(0-72.3)	0.02
Patients with OSA, n (%) • Mild OSA • Moderate OSA • Severe OSA	35/65 (54%) 20/35(57%) 6/35(17%) 9/35(26%)			
AHI	4.2(0-38)	0.5(0-1.4)	7.4(1.6-38)	0.000004
REM AHI	9.2(0-102.9)	1.7(0-11)	15.6(2.1-102.9)	0.0001
NREM AHI	3.1(0-40)	0.3(0-1.5)	5.6(0-40)	0.0001
OAHI	3.1(0-37)	0.3(0-1.26)	5.6(0-37)	0.0001
CAHI	1(0-9.4)	0.2(0-1.82)	1.6(0-9.37)	0.002
Average O_2 sat	96(83-99)	97(92-99)	95(83-99)	0.01
REM average O_2 sat	96(90-100)	97(92-100)	95(90-99)	0.002
NREM average O_2 sat	96(92-99)	97(92-99)	95(92-99)	0.002
O_2 nadir	86(60-97)	90(70/97)	83(60-94)	0.00006
% of total sleep time with SpO_2<90%	3(0-99)	0.7(0-9.8)	5.1(0-99)	0.12
ODI	7.3(0-36)	2.5(0-13.3)	11.4(0.1-36)	0.000003
Peak $ETCO_2$	45(32-56)	44(33-55)	46.9(32-65)	0.04
Arousal index	9(1.3-29.5)	6.2(1.3-16.3)	11.4(2.1-29.5)	0.0001

Nocturnal respiratory support, n (%)	13/65(20%)	3/31	11/35	
O_2 supplement	4/13(31)	1/31	3/35	
• CPAP support	2/13(15)	0/31	2/35	
• Bipap support	7/13(54)	2/31	6/35	

Table 2: Polysomnography data in all CP patients, CP patients without OSA and CP patients with OSA

Based on the PSG results, 13 (20%) patients were started on respiratory support, including 4 patients who required nighttime nasal O_2, 2 patients who required CPAP and 7 patients who were initiated on BiPAP.

Discussion

In this study we evaluated sleep architecture and prevalence of sleep disordered breathing, mainly OSA, in patients with severe quadriplegic CP using PSG. We found high prevalence of OSA (54%). Multiple previous studies have examined sleep disturbances and sleep disordered breathing in patients with CP using different types of validated sleep questionnaires. However, sleep questionnaires can potentially overestimate or underestimate the sleep disruption and sleep disordered breathing in these patients when compared with PSG criteria which is the gold standard. Furthermore, most of these sleep questionnaires have not been validated in children with CP specifically.

In a recent systemic review and meta-analysis of all previous studies conducted in patients with CP, the prevalence of sleep disordered breathing using the Sleep Disturbance Scale for Children (SDSC) ranged widely from 9.6% to 25.6%, and the prevalence using Pediatric Sleep Questionnaire (PSQ) also ranged widely from 7.3% to 88.5% [6].

To our knowledge, no previous studies have evaluated sleep architecture in patients with CP using PSG data. Only few studies used PSG to asses OSA in CP patients as part of a larger cohort of patients with developmental disability{Citation} [10]. Another small study used PSG to examine the effect of postural devices on worsening OSA in CP patients [11]. The discrepancies between the prevalence rate found in our study compared with previously reported prevalence rates in CP patients again reflect the fact all previous studies were questionnaire based while our study was based on PSG data.

Sleep architecture in our population, defines as percentage of different sleep stages (N1, N2, N3, and REM) was within normal limits. However, sleep efficiency was found low which was due to either delayed sleep onset, prolonged wakefulness after sleep onset (WASO) or due to frequent arousals. Such findings appeared to be not related sleep disordered breathing such as OSA, since we did not find any significant differences in sleep efficiency or sleep architecture between the OSA and non-OSA groups. Patients with severe CP usually have multiple comorbidities that can contribute to poor sleep efficiency, insomnia and frequent arousals. Such comorbidities include epilepsy, anti-epileptic medications, visual impairment and musculoskeletal aches and pains due to muscle spasm and contractures. Furthermore, children with developmental delay in general and CP patients in particular can have dysfunctional release of hormones that affects circadian regulation such as Melatonin. Previously, Santos et al showed that patients with CP had absence of day/night rhythmicity and lower nocturnal content of salivary melatonin compared to controls [12].

Despite the high prevalence of OSA in our patient population, symptoms of snoring and apnea were very low and did not correlate with OSA diagnosis which strongly suggests that symptoms-based questionnaires are not a reliable method to predict SDB in CP patients and that PSG remains the gold standard.

When comparing patients with OSA versus patients with no OSA, patients with OSA were younger and less likely to have seizure disorders. Results of previous questionnaire-based studies correlating seizure disorders and OSA in CP patients were inconsistent. Some studies showed positive correlation, others showed no significant effect of seizure disorder on SDB [13,14]. The differences between studies could be related to differences in other cofounding variables such as age, severity of CP and the effect of anti-epileptic treatments. Also, we found that the average O_2 saturation and the nadir O_2 saturation were significantly lower in the OSA group compared to non OSA group, which makes overnight pulse oximetry potentially as a more reliable alternative to questionnaires in screening CP patients for OSA. Future studies validating home O_2 monitoring for OSA in CP are needed.

Our study is a retrospective review of clinical and PSG data in patients with severe CP (i.e., with quadriplegia) and with

multiple comorbidities which could explain the high rate of OSA in this population. Future prospective studies of OSA prevalence among all CP patients regardless of their disease severity and presence of comorbidities are needed. Further studies are also necessary to validate the different non-PSG assessment tools in predicting sleep disruption and SDB in CP patients.

Author Contributions

Amal Alnaimi contributed to the study design, study proposal and data collection. Amal Alnaimi and Mutasim Abu-Hasan contributed to data analysis, data summarization and writing the manuscript. Ahmed Abushahin contributed to manuscript editing and critically reviewing the article. Ibrahim Janahi monitored the overall the progress of the project and provided key input through the process of study design, data analysis and manuscript writing.

Conflict Of Interest

The authors declare no conflict of interest.

Data Availability Statement

The data for this study are available from the corresponding author upon request.

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