

## RESEARCH ARTICLE

## Sleep Disorder Prevalence and Influencing Factors in Children with Cerebral Palsy

Uni Gamayani,<sup>1</sup> Milda Aryani,<sup>2</sup> Nushrotul Lailiyya,<sup>1</sup> Iin Pusparini<sup>1</sup>

<sup>1</sup>Department of Neurology, Faculty of Medicine, Universitas Padjadjaran/Dr. Hasan Sadikin General Hospital, Bandung, Indonesia, <sup>2</sup>Department of Neurology, Regional General Hospital Cilegon, Cilegon, Indonesia

### Abstract

Children with cerebral palsy are considered a population at risk for sleep disturbance. Various factors can cause sleep disorders in children with cerebral palsy. This study investigates the relationship between endogenous factors and sleep disorders in children with cerebral palsy. It was a cross-sectional analytical study using randomized sampling on children with cerebral palsy who met the inclusion criteria for the period of May–August 2017. The location of the study was special schools in the Bandung area, Indonesia. All participants were screened with the Sleep Disturbances Scale for Children (SDSC) questionnaire to determine the prevalence of period of sleep disorders. Data analysis was then performed using the unpaired t test to compare the characteristics of two variables with a p value  $\leq 0.05$  considered statistically significant. Sixty-six subjects aged 8–14 years were recruited. The results showed that the prevalence of sleep disorders was 67% (32 children), with insomnia as the most common type of sleep disorder (39%). There was a significant association between motor disabilities type and sleep disorders ( $p \leq 0.05$ ). The most common type of sleep disorder in children with cerebral palsy is insomnia. In conclusion, there is a relationship between motor disability type and sleep disorders in cerebral palsy children.

**Keywords:** Cerebral palsy, children, sleep disorders

### Introduction

Sleep is a state of reduced responses and interactions with the environment that is reversible and takes place quickly. Sleep serves as a means to improve the metabolic processes of the body regularly, which are indispensable in the physical growth and intellectual development of a child. Sleep disorders are a collection of symptoms characterized by disturbances in the number, quality, and duration of sleep that a person experiences. Approximately 25% of children suffer from sleep disorders, and it is one of the most common problems faced by parents.<sup>1,2</sup>

Children with neurological developmental disorders such as cerebral palsy are included in the population with a high risk for sleep disorders.<sup>2,3</sup> Cerebral palsy is a group of movement and posture disorders that cause limited movement due to non-progressive disorders in the brains of developing fetuses or infants. In developing countries, including Indonesia, cerebral palsy is estimated to occur in 1.5–2.5 children per 1,000 live births and is the most common cause of physical abnormalities in children.<sup>4</sup> Approximately 23–46% of cerebral

palsy children suffer from sleep disorders.<sup>5-7</sup> In a study on cerebral palsy children, Munyumu et al.<sup>8</sup> stated that 27% of these children suffer from sleep disorders, such as the difficulty of starting and maintaining sleep, sleep-wake transition disorder, sleep hyperhidrosis, and obstructive sleep apnea.

Many factors influence changes in the state of awake and sleep in cerebral palsy children. Factors that can cause sleep disorders may include internal anomalies in sleep regulation and comorbidity disorders due to cerebral palsy, such as spasticity, pain, severe motor disabilities, epilepsy, mental retardation, and primary sensory disorders gastroesophageal reflux disease (GERD), and other comorbidities.<sup>6,9</sup> Other factors include environmental factors, individual/family habits before bed, parental roles, parental education, family economic status, social circumstances, culture, nutrition, and so on.<sup>10,11</sup>

Research on sleep disturbances in children with cerebral palsy in Indonesia is minimal. Based on this issue, the researchers want to study the features and kinds of sleep problems and what factors are related to sleep disorders in children with cerebral palsy aged 8 to 14 years. In addition,

Received: 28 October 2021; Revised: 17 April 2022; Accepted: 20 April 2022; Published: 30 April 2022

**Correspondence:** Dr. Uni Gamayani, dr., Sp.S.(K). Department of Neurology, Faculty of Medicine, Universitas Padjadjaran/Dr. Hasan Sadikin General Hospital. Jln. Pasteur No. 38, Bandung 40161, West Java, Indonesia. E-mail: [gamayani@yahoo.com](mailto:gamayani@yahoo.com)

this study aimed to determine the characteristics of sleep disorders in children of that age. Thus subsequent focused treatments can be carried out to ensure that children's development is optimal.

## Methods

It was an analytical study on cerebral palsy children who attended special schools (*sekolah luar biasa*, SLB) in the Bandung area, Indonesia (State SLB Cileunyi, SLB Yayasan Suryakanti, and Special School for Children with Physical Disability (SLB D) Yayasan Pembinaan Anak Cacat (YPAC)) who met the following inclusion criteria: a) SLB students with cerebral palsy aged 8–14 years with good health, b) had parents/guardians/caregivers living with the child for at least the last six months, and c) parents/guardians and children were willing to participate in the study. In addition, students were excluded if they were identified as having an acute disease during an examination. This study was performed from May to August 2019 and approved by the Faculty of Medicine, Universitas Padjadjaran Health Research Ethics Committee by issuing ethical clearance number: 650/UN6.C.10/PN/2017.

The Sleep Disturbance Scale for Children (SDSC) questionnaire is commonly used to screen for sleep disorders among elementary school-age children.<sup>12</sup> This scale consists of 26 questions, with a 5-point Likert scale of 1=never, 2=rarely (1–2× nights/month), 3=sometimes (1–2× nights/week), 4=often (3–5× nights/week), and 5=always (every day). The total score of this questionnaire ranges from 26 to 130, with a cut-off point of 39, where a score of <39 reflects normal sleep (no sleep disorder) while a score of >39 means that there are abnormalities in the sleep (sleep disorder).<sup>13</sup>

Univariate data analysis was used to characterize the dependent and independent variables to assist in more detailed bivariate analysis. Furthermore, it is utilized to ascertain the features and clinical state of the study subjects. Univariate data statistic depicts the proportions of each variable, which were presented descriptively. Before statistical tests on numerical data, normality was assessed using the Kolmogorov Smirnov. Next, statistical analysis for categorical data was tested with the chi-square at a significance level of 0.05. The unpaired t test was used to compare the characteristics of the two study groups. Finally, multivariable data

analysis was conducted using binary logistic regression analysis to determine the relationship between factors associated with sleep disorders. The analysis tool is the statistical software SPSS 21.0 version for windows.

## Results

This study included 66 children with cerebral palsy, as described in Table 1. The average age of the subjects was 12.8 years, with an almost balanced gender proportion (53%, n=35 male). Most parents graduated from senior high school (47%) with an income of 2.5–5 million/month (moderate socioeconomic level) (58%). The prevalence of cerebral palsy in children with sleep disorders was 67% (44 children). Most subjects in this study had spastic cerebral palsy (73%) and were generally quadriplegic (61%), with a quadriplegic motor disability as the most common type (67%). In terms of mobilization capability, most subjects had Gross Motor Function Classification System (GMFCS) I–II (58%). Subjects suffered from one comorbidity (35%), with mental retardation as the most common comorbid (100%).

The types of sleep disorders experienced by the subjects are listed in Table 2. Difficulties in starting and maintaining sleep are the most prominent type of sleep disorder in this group (39%).

Table 3 presents a significant relationship between motor disabilities and sleep disorders ( $p < 0.05$ ). In the analysis of variable data on types of motor disabilities, the more motor disabilities suffered by a child, the more likely he or she would suffer from sleep disorders. Data analysis of other variables, i.e., cerebral palsy, GMFCS type, and several comorbidities, did not significantly correlate with sleep disorders ( $p > 0.05$ ).

Table 4 reflected the lack of relationship between the type of comorbidity, quadriplegic motor disability status, and sleep disorders ( $p > 0.05$ ).

## Discussion

In this study, the prevalence of sleep disorders among cerebral palsy children was 67%. The average age of subjects who experienced sleep disorders was 12.8 years, with difficulty starting and maintaining sleep as the most common disorder. These results complement the data on

**Table 1 Subject Characteristics**

Variables	n=66 (%)
Average age	
12.8 years old	66 (100)
Gender	
Male	35 (53)
Female	31 (47)
Parents' education	
Elementary school	15 (23)
Junior high school	9 (14)
Senior high school	31 (47)
Diploma	3 (4)
University	8 (12)
Parents' income (million)	
<2.5 (low)	18 (27)
2.5–5 (medium)	38 (58)
>5 (high)	10 (15)
SDSC value	
>39 (sleep disorders)	44 (67)
≤39 (no sleep disorders)	22 (33)
Type of cerebral palsy	
Spastic	48 (73)
Dyskinetic	4 (6)
Ataxic	3 (4)
Mixture	11 (17)
Type of motor disability	
Monoplegia	7 (10)
Diplegia	9 (14)
Hemiplegia	9 (14)
Triplegia	1 (1)
Quadriplegia	40 (61)
Quadriplegic motor disability	
Quadriplegia	44 (67)
Non-quadriplegia	22 (37)
Type of GMFCS	
GMFCS I–II	38 (58)
GMFCS III	28 (42)
Number of comorbidities	
1 comorbid	23 (35)
2 comorbid	20 (30)
3 comorbid	16 (24)
4 comorbid	4 (6)
5 comorbid	3 (5)
Concomitant disorder (comorbidity)	
Eating-drinking disorders	21 (32)
Pain disorders (scale>4)	2 (3)
Mental retardation	66 (100)
Urinary bladder dysfunction	22 (33)
Severe visual disorder	8 (12)
Epilepsy	5 (8)

Note: SDSC: Sleep Disturbance Scale for Children, GMFCS: Gross Motor Function Classification System

the prevalence of sleep disorders in cerebral palsy children based on the age range stated by previous studies. The prevalence of sleep disorders in cerebral palsy children is 96% in a study by Selina

**Table 2 Sleep Disorder Characteristics based on Sleep Disturbance Scale for Children**

Type of Sleep Disorder	n=44 (%)
Disorder of starting and maintaining sleep/Insomnia	17 (39)
Disorder of sleep and awake transition	10 (23)
Hyperhidrosis during sleep	8 (18)
Excessive somnolence disorder/hypersomnia	4 (9)
Respiration disorder during sleep	4 (9)
Consciousness disorder/Parasomnia	1 (2)

et al.<sup>14</sup> and 40% in Romeo et al.'s<sup>5</sup> study. Both studies also suggested that the most common type of sleep disorder in this group is starting and maintaining sleep, which is in line with the result of this study. This difference in prevalence is likely due to differences in research methods. Selina et al.<sup>14</sup> used a descriptive approach on 50 cerebral palsy children in a younger age range of 4–12 years. The instrument used for data collection in their study is the Child's Sleep Habits Questionnaire (CSHQ). Of all subjects in their research, 82% suffered from spastic cerebral palsy. It is different from the study conducted by Newman et al.,<sup>9</sup> conducted on 173 cerebral palsy children aged 6–12 years old in physiotherapy clinics using the SDSC questionnaire. Their finding demonstrated that most subjects had diplegia type of disability (48%), and only 10.4% had a quadriplegic disability.

Nevertheless, all GMFCSs were identified with GMFCS I found in most of the subjects (42.2%). However, this present study involves an older population of children with cerebral palsy. In addition, most had quadriplegic-type of motor disability (67%) and only affected those with GMFCS I, II, and III, which may contribute to the different results.

Table 2 shows that the most frequent type of sleep disorder in this study was difficulties in starting and maintaining sleep or insomnia, seen in 39% (17 children) of the subjects, followed by sleep and awake transition disturbance in 23% (10 children). This result corresponds to the literature that stated that insomnia's prevalence in cerebral palsy children is relatively high.<sup>6,7</sup>

Atmawidjaja et al.<sup>15</sup> said that insomnia's prevalence in children with cerebral palsy in Malaysia is 37%, almost similar to this study's

**Table 3 Relationship between Subject Characteristics and Sleeping Disorder**

Variables	Groups		p Value
	No Sleep Disorder (n=22)	Sleep Disorder (n=44)	
Type of cerebral palsy			0.231
Spastic	15	33	
Dyskinetic	0	4	
Ataxic	2	1	
Mixture	5	6	
Type of motor disability			0.042
Monoplegia	1	6	
Diplegia	2	7	
Hemiplegia	7	2	
Triplegia	0	1	
Quadriplegia	10	30	
Type of GMFCS			0.738
GMFCS I–II	15	23	
GMFCS III	7	21	
Number of comorbidities			0.613
1 comorbid	9	19	
2 comorbid	8	12	
3 comorbid	3	9	
4 comorbid	1	2	
5 comorbid	1	2	

Note: GMFCS: Gross Motor Function Classification System

**Table 4 Relationship between Comorbidity and Sleep Disorder**

Variables	Groups		p Value
	No Sleep Disorder n=48 (%)	Sleep Disorder n=81 (%)	
Comorbidity/concomitant disorder			0.765
Eating-drinking disorders	5 (11)	16 (20)	
Pain disorders (scale>4)	3 (6)	2 (3)	
Mental retardation	26 (54)	40 (49)	
Urinary bladder dysfunction	8 (17)	16 (20)	
Severe visual disorder	2 (4)	6 (7)	
Epilepsy	4 (8)	1 (1)	

finding of 38.6%. In another study by Munyumu et al.,<sup>8</sup> cerebral palsy patients in Uganda also presented starting and maintaining sleep or insomnia (27%) as the most prevalent sleep disorder. External and internal factors influence the etiology of insomnia.<sup>6,16</sup> The environmental factors (exogenous) include the habit of long-duration television watching until late at night, a bright and noisy environment, stress, or poor sleep hygiene. The internal factors include the activity of the hypothalamus-pituitary-adrenal

(HPA) axis, which affects the amount of blood cortisol, causing circadian rhythm disturbances of the sleep and wake cycle, body temperature, craving bedtime and wakefulness time, and the spasticity and severity of motor disabilities.<sup>17–19</sup>

The other sleep disorder widely experienced by subjects in this study was the sleep and awake transition disturbance, which was identified in 23% of the subjects. The prevalence rates of sleep and awake transition disturbance in studies by Newman et al.<sup>9</sup> and Atmawidjaja et

al.<sup>15</sup> were 17.9% and 13.2%, respectively. Based on the 2014 International Classification of Sleep Disorders-3 (ICSD-3), this type of sleep disorder is categorized into the sleep motion disorders, such as restless leg syndrome (RLS), bruxism, and periodic limb movement disorder (PLMD), and head and neck movements during sleep. These disorders can be caused by genetic abnormalities, brain immaturity, and psychological stress. The condition is commonly found in children <6 years of age and decreases with age.<sup>20</sup>

Table 3 describes the relationship between subject characteristics (cerebral palsy type variables, motor disability type, GMFCS type, and several comorbidities) and sleep disorders. The statistical tests showed a significant relationship between types of motor disabilities and sleep disorders. However, no significant relationship was found with sleep disorders for cerebral palsy type, GMFCS type, and several comorbidity variables. The significant relationship between motor disability type and sleep disorders found in this study corroborates with the results of a previous.<sup>20-22</sup> Another study found that motor disabilities involving more body components have a greater risk for sleep disorders.<sup>23</sup> The absence of a significant relationship between the GMFCS type and sleep disorders was also stated by Newman et al.<sup>9</sup>

In contrast, Munyumu et al.<sup>8</sup> discovered that GMFCS V or IV and epilepsy link to sleep disorders. Romeo et al.<sup>5</sup> showed that 48% of subjects with GMFCS V experience sleep disorders. These differences may be due to the difference in the patient population because patients in Uganda have a more severe form of cerebral palsy than those in other countries.<sup>8</sup> In addition, the absence of a significant relationship between the cerebral palsy type, GMFCS type, and the number of comorbidity in this study may also be since this study only involved subjects with GMFCS I, II, III, i.e., subjects who can still move around independently, with or without aids. Therefore, the degree of comorbidity experienced by the subjects was also not assessed in this present study.

Table 4 describes the characteristic relationships of subjects (concomitant disorder/comorbidity variables) between the group without sleep disorders and with sleep disorders. The statistical tests showed no significant or statistically significant relationship between comorbid disorders and the occurrence of sleep

disorders. This contrasts with Munyumu et al.<sup>8</sup>, demonstrating that epileptic comorbidity is associated with sleep disorders. It can be due to unknown differences in the degree of concomitant disorder experienced. The subjects involved in this study are only those with GMFCS I, II, and III, which is different from their research. No cerebral palsy patients with GMFCS IV and V were recruited in this study. Patients with GMFCS IV and V generally have a higher number and more severe comorbidities, which may contribute to the Munyumu et al.'s<sup>8</sup> study. This present study did not assess the degree of eating disorders or mental retardation suffered by the patients, so it is likely to cause no significant relationship between concomitant disorders or comorbidities and sleep disorders. Quadriplegic can describe the presence of bilateral lesions and more extensive lesions than in the non-quadriplegic type.<sup>24</sup> The division of people with quadriplegic and non-quadriplegic disabilities is not related to sleep disorders. Instead, it can be caused by differences in the severity of quadriplegic experienced by the subject. The quadriplegic condition can also occur in cerebral palsy children with independent mobilization abilities that do not require aids. In this study, 58% of subjects had a quadriplegic disability and had independent mobilization capability (GMFCS I–II).

This study has several limitations because no division in the degree of comorbidity severity was applied, and no objective measuring instruments, such as actigraphy or polysomnography were used. Thus, the results should be interpreted accordingly.

## Conclusions

There is a relationship between motor disabilities and sleep disorders in cerebral palsy children. The most common sleep disorder experienced by children with cerebral palsy is difficulties in starting and maintaining sleep (insomnia). Further studies are required to understand sleep disorders in light of external factors that also affect sleep disorders in cerebral palsy children. In addition, more variety in the severity of cerebral palsy as specified with GMFCS I–V should also be included in future studies.

## Conflict of Interest

The authors declared that there is no conflict of

interest.

### Acknowledgments

Thank you to all authors for providing the opportunity to carry out this research.

### References

- Sandella DE, O'Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. *Sleep Med.* 2011;12(3):252–6.
- Wayte S, McCaughey E, Holley S, Annaz D, Hill CM. Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. *Acta Paediatr.* 2012;101(6):618–23.
- Hutson JA, Snow L. Sleep assessments for children with severe cerebral palsy: a scoping review. *Arch Rehabil Res Clin Transl.* 2020;2(4):100087.
- Oskoui M, Shevell MI, Swaiman KF. Cerebral palsy. In: Swaiman KF, Ashwal S, Ferriero DM, Schor NF, Finkel RS, Gropman AL, et al., editors. *Swaiman's pediatric neurology: principles and practices.* 6th Edition. New York: Elsevier; 2017. p. 734–45.
- Romeo DM, Brogna C, Quintiliani M, Baranello G, Pagliano E, Casalino T, et al. Sleep disorders in children with cerebral palsy: neurodevelopmental and behavioral correlates. *Sleep Med.* 2014;15(2):213–8.
- Dutt R, Roduta-Roberts M, Brown CA. Sleep and children with cerebral palsy: a review of current evidence and environmental non-pharmacological interventions. *Children (Basel).* 2015;2(1):78–88.
- Horwood L, Li P, Mok E, Shevell M, Constantin E. A systematic review and meta-analysis of the prevalence of sleep problems in children with cerebral palsy: how do children with cerebral palsy differ from each other and from typically developing children? *Sleep Health.* 2019;5(6):555–71.
- Munyumu K, Idro R, Abbo C, Kaddumukasa M, Katabira E, Mupere E, Kakooza-Mwesige A. Prevalence and factors associated with sleep disorders among children with cerebral palsy in Uganda; a cross-sectional study. *BMC Pediatr.* 2018;18(1):26.
- Newman CJ, O'Regan M, Hensey O. Sleep disorder in children with cerebral palsy. *Dev Med Child Neurol.* 2006;48:564–8.
- Chellappa SL, Steiner R, Oelhafen P, Lang D, Götz T, Cajochen C. Acute exposure to evening blue-enriched light impacts on human sleep. *J Sleep Res.* 2013;22(5):573–80.
- van Kamp I, Persson Waye K, Gidlöf-Gunnarsson A. The effects of noise disturbed sleep on children's health and cognitive development. *J Child Adolesc Behav.* 2015;3(1):179.
- Petersen S, Francis KL, Reddihough DS, Lima S, Harvey A, Newall F. Sleep problems and solution seeking for children with cerebral palsy and their parents. *J Paediatr Child Health.* 2020;56(7):1108–13.
- Bruni O, Ottaviano S, Guidetti V, Romoli M, Innocenzi M, Cortesi F, et al. The Sleep Disturbance Scale for Children (SDSC). Construction and validation of an instrument to evaluate sleep disturbances in childhood and adolescence. *J Sleep Res.* 1996;5(4):251–61.
- Selina H, Priambodo WS, Sakundarno M. Gangguan tidur pada anak palsy serebral. *Med Hosp.* 2012;1(2):108–12.
- Atmawidjaja RW, Wong SW, Yang WW, Ong LC. Sleep disturbances in Malaysian children with cerebral palsy. *Dev Med Child Neurol.* 2014;56(7):681–5.
- Lélis A, Cardoso M, Hall WA. Sleep disorders in children with cerebral palsy: an integrative review. *Sleep Med Rev.* 2016;30:63–71.
- Ropper AH, Samuels MA, Klein JP, Prasad S, editors. *Adams and Victor's principles of neurology.* 11th Edition. New York: McGraw Hill; 2019.
- Gamayani U. *Neurology in daily practice.* 1st Edition. Bandung: Bagian Ilmu Penyakit Saraf Fakultas Kedokteran Universitas Padjadjaran Rumah Sakit Hasan Sadikin Bandung; 2010.
- Adiga D, Gupta A, Khanna M, Taly AB, Thennarasu K. Sleep disorders in children with cerebral palsy and its correlation with sleep disturbance in primary caregivers and other associated factors. *Ann Indian Acad Neurol.* 2014;17(4):473–6.
- Grigg-Damberger MM. Sleep and sleep disorders in epilepsy. In: Sheldon SH, Ferber R, Kryger MH, Gozal D, editors. *Principles and practice of pediatric sleep medicine.* New York: Elsevier; 2014. p. 353–61.
- Löwing K, Gyllensvärd M, Tedroff K.

- Exploring sleep problems in young children with cerebral palsy - a population-based study. *Eur J Paediatr Neurol.* 2020;28:186–92.
22. Hulst RY, Gorter JW, Voorman JM, Kolk E, Van Der Vossen S, Visser-Meily JMA, et al. Sleep problems in children with cerebral palsy and their parents. *Dev Med Child Neurol.* 2021;63(11):1344–50.
  23. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy. *J Child Neurol.* 2011;26(10):1303–10.
  24. Lagunju IA, Adedokun BO. A comparison of quadriplegic and hemiplegic cerebral palsy. *J Pediatr Neurol.* 2008;6(1):25–30.