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Title: *Sleep problems and solution seeking for children with cerebral palsy and their parents*

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Abstract

Aims: Sleep problems are common in school age children with cerebral palsy (CP). Despite the significant impact of sleep disturbance and deprivation, there is a paucity of research in the area. The aims of this study were to 1) investigate the frequency of sleep problems in children with CP and their parents 2) understand what happens when parents ask for help with sleep problems from their healthcare professionals.

Method: A prospective cohort study using an online survey sent to parents/primary caregivers of children with CP aged 6-12 years recruited through the Victorian Cerebral Palsy Register. The sleep assessment tools: The Children's Sleep Habits Questionnaire, the Pittsburgh Sleep Quality Index and the Gross Motor Function Classification System Parent Rating Tool were administered along with custom designed questions that were informed by a preceding qualitative scoping study.

Results: Complete data sets were received from 126 parents/caregivers. Almost half (46%) the parents reported their child had sleep problems. Of the 64 parents who reported seeking help for a child's sleep problem only 21 indicated that their attempt was successful. If a child had poor sleep, the parent was more likely to have a sleep problem than parents who did not report poor child sleeping.

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Conclusion and Implications: Sleep problems are common in children with CP and their parents. Parents do not always seek help and those that do may not find an effective solution. Future research should explore how sleep problems can be effectively prioritised for children with CP and their parents/caregivers.

Brief Points:

What is already known on this topic:

- International studies indicate that sleep problems are common in children with CP, yet accurate Australian data are lacking on both sleep problems and help seeking for sleep problems.
- The comorbidities associated with CP such as pain, discomfort, gastrointestinal issues and the need for repositioning can all impact on sleep.
- There is a paucity of descriptive data that could inform the implementation of effective management practices.

What this paper adds:

- Frequency and type of sleep problems in children with CP in Australia concur with similar international studies.
- Results provide additional evidence that sleep problems in a child with CP are concurrent with poor parental sleep quality.
- Parents seeking help for sleep issues often fail to find effective solutions from their healthcare team, demonstrating a real need for evidence-based approaches to make sleep a health priority and to improve access to sleep solutions for this cohort.

Keywords: Sleep, cerebral palsy, parent sleep, child

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1.1 Introduction

Cerebral palsy (CP) is a static neurological disorder caused by damage or malformation to the foetal or developing brain affecting movement and posture.(1) CP is associated with many comorbidities, including but not limited to: vision impairment, intellectual disability, respiratory conditions, gastroenterological conditions, epilepsy, and sleep disturbance.(2) Current research suggests that between 30%(3) and 60%(4) of children with CP have sleep problems. International research suggests that sleep issues impact on children with CP; a negative association has been found between sleep problems and physical and emotional health(4) and behaviour.(5) Similar data are not available about sleep in Australian children with CP. However, a report from the Victorian Equal Opportunity and Human Rights Commission states that sleep deprivation and night-time vigilance are associated with parents surrendering care of their children with disabilities.(6) It is clear that the impact of sleep problems on health and well-being in children with CP is significant (7), consequently there is a need for good quality evidence to inform the development of effective sleep solutions.

The majority of published research in CP and sleep for school-aged children consists of studies with small sample sizes that use prospective parent report sleep assessment tools such as the Child Sleep Habits Questionnaire (CSHQ) and the Sleep Disturbance Scale for Children.(3, 5, 8-10) These studies demonstrate that sleep problems are prevalent in children with CP. Few studies(3, 9-14) have explored the impact of the child's sleep problems on the parent. Again, there is a paucity of Australian based studies that specifically focus on CP, with only two published studies, a pilot(15) that led to the current study and a larger study,(16) which was a retrospective audit of children who attended a sleep service.

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There is little published evidence about whether parents seek help for sleep problems in children with CP, and if the interventions suggested are effective. Prior to determining how to address sleep problems, it is essential to understand if and when parents ask for help, and the subsequent outcome of their request. This study aimed to describe the sleep problems of children with CP and their parents and how families seek solutions in Victoria, Australia.

Specifically, this study aimed to explore:

i. The frequency and type of sleep problems in Victorian children with CP aged 6-12 years and whether parents concurrently had sleep difficulties.

ii. Whether, and from whom, parents/primary caregivers seek help for their children's sleep problems.

A secondary aim was to understand if the parents who sought help found the advice or treatment offered effective.

1.2 Materials and Methods

1.2.1 Study design

This was a population-based cohort study using an online survey that included validated sleep questionnaires and custom designed questions. The survey was managed and administered through the data management program REDCap(17) hosted by the Murdoch Children's Research Institute.

1.2.2 Study population

Parents/caregivers (hereafter referred to as parents) of children aged 6 to 12 years with CP, whose names were recorded on the Victorian Cerebral Palsy Register (VCPR), were invited to participate. The age group was chosen to exclude

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developmentally appropriate sleep problems that may occur during the pre-school years and to align with the age limits of the validated CSHQ.(17)

1.2.3 Recruitment process

Potential participants who met the study inclusion criteria were identified via the VCPR. The VCPR, established in 1987, collects data on people with CP born or living in the Australian state of Victoria since 1970. It is used to recruit people with CP and their families or carers who have provided consent to be contacted about research projects. Potential participants were sent the REDCap(17) survey link and a brief information letter via email or post over an eight-week period from December 2017 to February 2018. Email invitations were sent four times including three reminders after the initial invitation. The hard copy letter was sent once with one reminder.

1.2.4 Measures

The survey consisted of three validated questionnaires and the custom designed questions. The choice of validated sleep assessment tool and the content and structure of the custom designed questions were informed by a preceding scoping study of qualitative semi-structured interviews of nine parents of children aged 6-12 years with CP with sleep problems, as reported by their parents.(19) Parents reported that they preferred 'tick box' surveys (i.e. not free text answers) that could be completed quickly, and questionnaires that allowed for an answer of 'not applicable' for surveys not inclusive of children of all abilities. Based on these preferences, the CSHQ and the Pittsburgh Sleep Quality Index (PSQI) were chosen as the validated sleep tools for this study. All validated tools were transcribed, unaltered into the REDCap(17) program, and the custom designed questions were then added.

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1.2.4.1 Children's Sleep Habits Questionnaire

The CSHQ abbreviated version was used to measure the children's sleep. Individual scores can range from 31 to 97(18) with a score higher than 41 indicating problematic sleep. Use of this tool requires parents to report on their child's sleep in the preceding four weeks. It is a concise and accessible survey, designed and validated for typically developing children and has been used in the CP population.(8)

1.2.4.2 Pittsburgh Sleep Quality Index

The Pittsburgh Sleep Quality Index , a validated sleep screening tool,(20) was used to assess parent reported sleep quality over the month prior to completing the survey. The tool consists of 18 questions that are summed into seven components that give a global score where scores of five or more indicate a sleep problem.

1.2.4.3 Gross Motor Function Classification System Parent Rating Tool

The Gross Motor Function Classification System (GMFCS) is an internationally recognised classification system that describes the gross motor function of children with CP.(21) The GMFCS classifies gross motor function into five levels: people at levels I-II are ambulant; those in level III use mobility aids such as walking frames; people in levels IV and V primarily use wheelchairs. The GMFCS Family Report Questionnaire is a reliable and validated questionnaire(21) developed as a self-report tool for parents of children with CP. It contains five descriptors of the GMFCS levels, and parents are asked to select which descriptor best matches their children. This ensures an accurate report of the child's GMFCS level for data collection.

The custom designed questions included demographics of the parent and child, socioeconomic status, a single item asking parents about whether their child has

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sleep problems, methods used to seek sleep solutions and data about the child's healthcare team.

1.2.5 Ethics

Ethics approval was obtained from the Royal Children's Hospital, Melbourne Human Research and Ethics Committee (HREC 35166 and HREC 37300).

1.3 Data Analysis

Descriptive statistics were produced for the outcome measures, proportions for the categorical data (PQSI and parent report of child's sleep) and mean (sd) for the continuous outcome (CHSQ). Comparisons of the categorical variables were tested with chi-square tests while the continuous variables were compared using a t-test. Free text survey answers were grouped and labelled according to similarity which was decided by consensus between all authors.

1.4 Results

Figure 1 describes the participant recruitment process. The demographics of the parents and children are detailed in Tables 1 and 2 respectively. The study sample was mostly Australian born biological mothers aged between 26 and 50 years, tertiary educated with reported above-average education and household incomes for the Australian population as compared to Australian Census data.(23)

1.4.1 GMFCS and sleep

Given the children in the sample had varying severities of CP as measured by the GMFCS, the data were initially examined to investigate the proportion of sleep problems in children and their parents in each of the GMFCS classification groups. As shown in Table 3 the proportion of parents with poor sleep (PQSI) was similar across the three GMFCS classifications of their children (59% to 67%, chi square

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=0.510 p=0.775). There was also no evidence that the CSHQ scores varied between the GMFCS classifications (F=0.6, p=0.551), or parent reported presence of sleep problems differed by GMFCS classifications (46% to 59%, chi square =0.768 p=0.681).

1.4.2 Parent and child sleep

From the custom designed item *Do you think your child has a sleep problem?*, 59 (46.8%) parents reported that they thought their child had a sleep problem at the time of the survey, with an additional 30 (23.8%) indicating that their child had previously had sleep problems. Children whose parents reported that they had a sleep problem had a higher mean CSHQ score (mean=52), compared to children whose parents did not indicate their child had a sleep problem(mean=39; mean difference:12.1 (95%CI:9.2-15.0) (p<0.005)).

Overall 82 (65%) of parents were classified as having poor sleep by the PQSI, with 48 (81.4%) of the 59 parents who stated that their child had a sleep problem reporting poor sleep quality themselves, compared with 34 (50.8%) of 67 parents who reported their child did not have a sleep problem (chi-square=12.93, p<0.005). See Figure 2.

Of those parents who reported sleep problems in their child 59 provided brief free text answers for the type of sleep problems, most commonly experienced. These included night-time waking (n=31), sleep latency (issues getting to sleep) (n=20) and a sleep association (n=12). Notably 26 of the 59 responses reported more than one sleep problem.

1.4.3 Seeking sleep solutions

Parents were asked which healthcare professionals their child had seen in the previous twelve months. Physiotherapists (84%), general practitioners (GP)

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(78.5%), paediatricians (72%), occupational therapists (69%), speech therapists (44%), rehabilitation specialists (32%) and psychologists (15%) were the most commonly seen. For the parents who had ever asked for help with sleep problems at some point in their child's lifetime (n=64) help was sought from a paediatrician (72%) and/or a GP (19%), psychologist (15%), maternal and child health nurse (12%), physiotherapist (12%), and/or an occupational therapist (12%). Figure 3 shows the outcomes of those parents who asked for professional help with their child's sleep. Only 21 of 64 parents reported that their request for help led to advice/treatment that was effective.

1.4.4 Reasons for not seeking help with sleep

Eighteen parents who reported that their child had a sleep problem completed the free text question *Why haven't you asked for help with sleep?* Answers were grouped for similarity with the most common reasons being that they planned to manage it themselves (32%), did not see it as a problem (21%) (this is despite reporting in the survey that it was or is a current problem), or did not see the sleep problem as having a solution (21%).

1.5 Discussion

This study examined sleep problems and solution seeking in a cohort of children with CP and found that sleep is problematic for almost half of the cohort. Furthermore, a child with CP who sleeps poorly is likely to have a parent who sleeps poorly, as reflected in the parents' PSQI scores. Sleep problems were present for children across all GMFCS levels, aligning with international studies(3, 12) which report similar rates of frequency of sleep problems and associated sleep problems for parents of those children. Parent reports of their child having a sleep problem correlated with the child having a high score on the CSHQ.

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These results provide important new information about how not all parents ask for sleep solutions and of those that do ask for help, not all have a positive outcome of child sleep improvement. It might be hypothesised that parents tolerate chronic sleep problems because of their inability to find effective solutions from their healthcare professionals. It is essential to consider the long-term impact of living with sleep disturbance and deprivation. An argument could be made that a sleep screening tool specific for children with CP may be a useful clinical tool to determine if a child has a sleep problem. However, the results of this study indicate that parents are aware when sleep is a problem, therefore simply asking the parents about sleep may be the most efficient screening method. The issue may not be with identification of the sleep problem by the parent but that solutions for sleep problems are not easily accessible, for reasons that are not well understood. A recent Canadian study(24) described a significant knowledge gap for healthcare professionals concerning sleep and highlighted their lack of sleep education; some clinicians reported that they have less than three hours of training about sleep. Similar data are not available for Australia, however a recent Australian Government inquiry recommended that sleep knowledge of both doctors and nurses needed to be assessed and addressed.(25) Data from this study demonstrate these children have multidisciplinary care teams, with a paediatrician being the healthcare professional from whom help with sleep is most commonly sought. However, as the majority of these children have other healthcare professionals in their team, sleep education may also be sourced from them. It was not within the scope of this study to fully explore the use of the healthcare team and the process of seeking sleep help. The initial findings of this study indicate more research is needed in this area.

Notably, 21% of parents who had identified that their child had sleep problems reported they had not sought sleep help as they thought it was not a problem. Reasons for this discrepancy could not be determined by the current study;

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subsequent qualitative phases of this project will explore the family behaviours regarding seeking assistance.

There are a number of limitations that may impact on the generalisability of this study. The CSHQ has limitations in regard to sensitivity when compared to gold standard objective sleep assessment polysomnography (26), however this tool was chosen based on parent preferences with the aim to optimise participation rates. The participants were more educated and were of higher socioeconomic status than the general population of Victoria and this may have resulted in some sample bias. However, there is no data on sleep in Victorian children with CP and their parents. It might be argued that the results in this study substantially underestimate the extent of sleep problems as those with more significant resources and education are more likely to have success in advocating for themselves accessing private services. Indeed, Magee (27) in their study of sleep in Australian children aged 4-8 years found that lower maternal education and household income were factors that influenced the likelihood of incomplete data.

The recruitment rate of this study was 20% of the potential cohort. Considerable effort was made to maximise the recruitment but despite these efforts, participation was still low. This may demonstrate the difficulty in engaging this population due to time pressures, the additional care needs of a child with a disability and the fatigue associated with problematic sleep.

1.6 Conclusion and Recommendations

Sleep is a problem for many children with CP and this study highlights how sleep problems need more attention in the Australian setting. Many parents are aware of a sleep problem for their child but are not seeking help. For those parents seeking sleep solutions, they do not always find effective treatment. Future research needs to address three key areas: 1) exploration of barriers and

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facilitators for parents seeking sleep solutions for their child, 2) sleep as a health priority for healthcare professionals who care for children with CP, and 3) how best to assist both parents and healthcare professionals to make sleep a health priority.

It is further proposed that a mixed methods research approach might be best suited to investigate these issues that are both clinically and sociologically complex. Sleep is an important determinant of health that is frequently overlooked within the clinical setting. Improved sleep is likely to improve overall health and quality of life and there is considerable work to be done to address the evidence gaps for children with CP and their parents.

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1. Bax M, Goldstein M, Rosenbaum P, Leviton A, Paneth N, Dan B, et al. Proposed definition and classification of cerebral palsy, April 2005. Dev Med Child Neurol. 2005;47(8):571-6.

2. Novak I. Evidence-based diagnosis, health care, and rehabilitation for children with cerebral palsy. J Child Neurol. 2014;29(8):1141-56.

3. 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.

4. Zuculo GM, Knap CCF, Pinato L. Correlation between sleep and quality of life in cerebral palsy. Codas. 2014;26(6):447-56.

5. 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.

6. Victorian Equal Opportunity and Human Rights Commission. Desperate measures: the relinquishment of children with disability into state care in Victoria [Internet]. Carlton (VIC): Victorian Equal Opportunity and Human Rights Commission; 2012 [cited 2014 December 15]. Available from: http://www.humanrightscommission.vic.gov.au/index.php/our-resources-and-publications/reports/item/153-desperate-measures-the-relinquishment-of-children-with-disability-into-state-care-in-victoria-may-2012.

7. Verschuren O, McPhee P, Rosenbaum P, Gorter JW. The formula for health and well-being in individuals with cerebral palsy: physical activity, sleep, and nutrition. Developmental Medicine and Child Neurology

   . 2016;58(9):989,90.

8. Newman CJ, O'Regan M, Hensey O. Sleep disorders in children with cerebral palsy. Dev Med Child Neurol. 2006;48(7):564-8.

9. 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.

10. 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.

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11. Tietze A-L, Blankenburg M, Hechler T, Michel E, Koh M, Schlüter B, et al. Sleep disturbances in children with multiple disabilities. Sleep Med Rev. 2012;16(2):117-27.

12. Mörelius E, Hemmingsson H. Parents of children with physical disabilities: perceived health in parents related to the child's sleep problems and need for attention at night. Child Care Health Dev. 2014;40(3):412-18.

13. Ikeda T, Nagai T, Kato-Nishimura K, Mohri I, Taniike M. Sleep problems in physically disabled children and burden on caregivers. Brain Dev. 2012;34(3):223-29.

14. Hemmingsson H, Stenhammar AM, Paulsson K. Sleep problems and the need for parental night-time attention in children with physical disabilities. Child Care Health Dev. 2009;35(1):89-95.

15. Petersen S, Harvey A, Reddihough D, Newall F. Children with cerebral palsy: why are they awake at night? A pilot study. J Spec Pediatr Nurs. 2015;20(2):98-104.

16. McCabe SM, Blackmore AM, Abbiss CR, Langdon K, Elliott C. Sleep concerns in children and young people with cerebral palsy in their home setting. J Paediatr Child Health. 2015;51(12):1188-94.

17. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)—A metadata-driven methodology and workflow process for providing translational research informatics support. Journal of Biomedical Informatics. 2009;42(2):377-81.

18. Owens JA, Spirito A, McGuinn M. The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. Sleep. 2000;23(8):1043-51.

19. Petersen S, Harvey A, Reddihough D, Lima S, Newall F. The experience and impact of sleep problems for parents of children with cerebral palsy: a qualitative study. Australasian Academy of Cerebral Palsy and Developmental Medicine: Developmental Medicine and Child Neurology; 2018.

20. Buysse DJ, Reynolds CF, Monk TH, Berman SR, Kupfer DJ. The Pittsburgh Sleep Quality Index: a new instrument for psychiatric practice and research. Psychiatry Res. 1989;28(2):193-213..

21. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised gross motor function classification system. Dev Med Child Neurol. 2008;50(10):744-50.

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22. Dietrich A, Abercrombie K, Fanning J, Bartlett D. GMFCS Family and Self Report questionnaire. Hamilton (CAN): CanChild Centre for Childhood Disability; 2014.

23. Australian Bureau of Statistics. 2016 Census quickstats [Internet]. Canberra (ACT): Commonwealth of Australia; 2016 [cited 2018 December 17]. Available from: https://quickstats.censusdata.abs.gov.au/census_services/getproduct/census/2016/quickstat/2?opendocument.

24. Gruber R, Constantin E, Frappier JY, Brouillette RT, Wise MS. Training, knowledge, attitudes and practices of Canadian health care providers regarding sleep and sleep disorders in children. Paediatr Child Health. 2017;22(6):322-7.

25. House of Representatives Standing Committee on Health, Aged Care and Sport. Bedtime reading: inquiry into sleep health awareness in Australia. Canberra (ACT): Commonwealth of Australia; 2019.

26. Markovich A, Gendron M, Corkum P. Validating the Children's Sleep Habits Questionnaire against polysomnography and actigraphy in school-aged children. Frontiers in Psychiatry. 2015;5:188.

27. Magee CA, Lee JK, Vella SA. Bidirectional Relationships Between Sleep Duration and Screen Time in Early Childhood. JAMA PEDIATRICS. 2014;168(5):465-70.

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