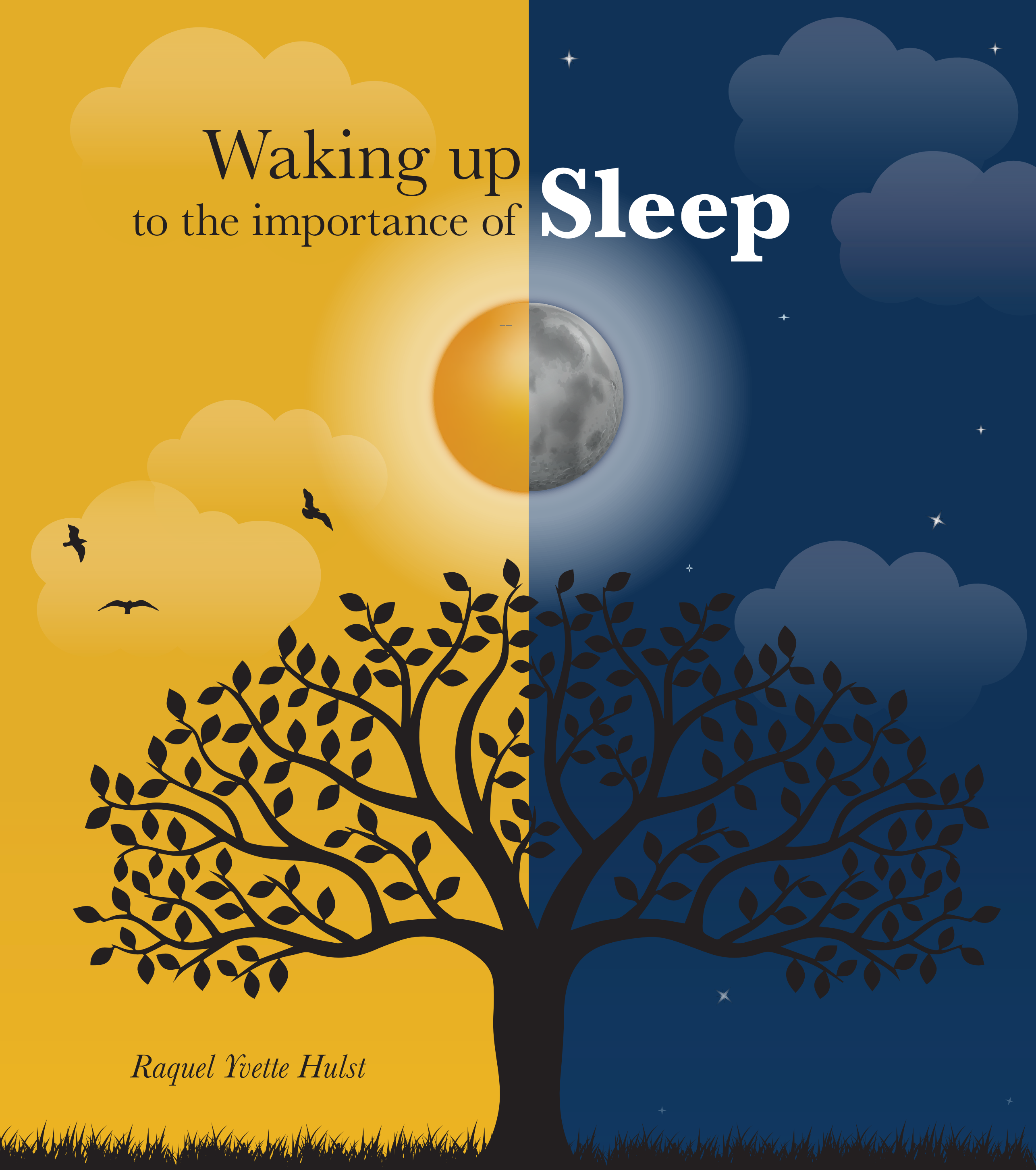


Waking up to the importance of Sleep

Raquel Yvette Hulst

UMC Utrecht Brain Center

Integrating sleep into care for children with cerebral palsy through a 24-hour activity approach

Waking up to the importance of sleep

Integrating sleep into care for children with cerebral palsy through a 24-hour activity approach

Raquel Yvette Hulst

Cover	Jochem Helleman Wendy Schoneveld | wenz iD
Layout	Renate Siebes | Proefschrift.nu
Printed by	Proefschriftmaken.nl | De Bilt
ISBN	978-90-393-7448-1
DOI	https://doi.org/10.33540/1133

Waking up to the importance of sleep

Integrating sleep into care for children with cerebral palsy through a 24-hour activity approach

Wakker worden voor het belang van slaap

Het integreren van slaap in de zorg voor kinderen met cerebrale parese
door middel van een 24-uurs activiteiten benadering

(met een samenvatting in het Nederlands)

Proefschrift

ter verkrijging van de graad van doctor aan de
Universiteit Utrecht op gezag van de rector magnificus,
prof. dr. H.R.B.M. Kummeling, ingevolge het besluit van het
college voor promoties in het openbaar te verdedigen
op maandag 4 april 2022 des middags te 2.15 uur

door

Raquel Yvette Hulst

geboren op 17 januari 1990
te Utrecht

Promotor:	Prof. dr. J.M.A. Visser-Meily
Copromotoren:	Dr. O.W. Verschuren Dr. J.M. Voorman

The studies reported in this thesis were financially supported by Innovatiefonds Zorgverzekeraars (project number 3521), JKF Kinderfonds and Kinderrevalidatiefonds Adriaanstichting (project number 20190017).

Financial support for the printing and distribution of this thesis has been generously provided by Stichting De Hoogstraat Onderzoeksfonds Utrecht, Nutricia Nederland B.V., Emfit Ltd., ChipSoft, ActiGraph and ProCare, Indoor Cliff Diving, CP Nederland, and my mom & dad.

Contents

PART I Understanding the perspectives of parents regarding the care for sleep of their child with cerebral palsy

PART II Evaluating the care for sleep in pediatric rehabilitation

PART III Integrating sleep into care for children with cerebral palsy using a 24-hour activity approach

PART IV Measuring the 24-hour activities in children with cerebral palsy

Chapter 1

General introduction

CEREBRAL PALSY

Cerebral palsy (CP) is one of the most frequent causes of childhood disability, with a global prevalence of 2.5 per 1000 live births.[1,2] The umbrella term CP refers to a group of permanent neurodevelopmental disorders characterized by impairments in motor function, which are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain.[3]

Consistent with the heterogeneous nature of CP, symptoms vary widely between individuals and may change as development progresses. Children can have spasticity, poor coordination, painful muscles, contractures, fatigue, and difficulties maintaining posture and balance, causing the majority of children with CP to experience some degree of mobility impairment, depending on the severity of the disability. Based on the day-to-day functional ability of children with CP, the Gross Motor Function Classification System (GMFCS) distinguishes between five levels of severity ranging from walking independently (level I) to being dependent for all types of mobility (level V).[4]

In addition to the motor impairments, CP is often accompanied by co-occurring conditions (comorbidities) which can be attributed to the same injury to the brain that caused CP (e.g. epilepsy, cognitive impairment, speech problems), while other comorbid disorders may be regarded as secondary complications (e.g. pain, eating difficulties, chronic fatigue, and sleep problems).[5,6] Altogether, these comorbidities can be as debilitating as the physical disability itself.[5,7]

Compared to children with typical development, children with CP have reduced physical health, and a lower health-related quality of life.[8,9] In addition, having CP increases the risk for chronic health problems in later life, including diabetes, stroke, and heart and lung diseases.[10] Clearly, achieving good health across the lifespan is challenging for individuals with CP. This raises critical questions about preventable health complications and strategies to improve overall health and well-being in this vulnerable population.

Even though the brain injury associated with CP is irreversible, there are many aspects of health and development that can be optimised. Modifiable lifestyle behaviours, for example, are shaped during childhood and known to track into adulthood, thereby providing opportunities with long-term implications for health maintenance. Therefore, promoting healthy lifestyle behaviours and other health-enhancing activities, such as regular physical activity, healthy nutrition, and good sleep practices, has the potential to provide children with CP a strong foundation with long-term health benefits throughout their lifespan.[11]

REHABILITATIVE CARE FOR CHILDREN WITH CP

Children with CP represent the largest diagnostic group treated in pediatric rehabilitation.[2,12] In the Netherlands, children with neurodevelopmental disorders, including CP, usually receive developmental or rehabilitative services in an outpatient setting of a rehabilitation center, or integrated in a school for special education. Rehabilitation is a complex process that requires a multidisciplinary approach.[13] Hence, the care for children with CP is delivered by a team of healthcare professionals from various disciplines that are often institutionalized under one roof, and coordinated by a rehabilitation physician.[14] Furthermore, the family is a key participant in pediatric rehabilitation under the concept 'family-centered care'.[15]

As there is no cure for CP, available interventions are aimed at limiting the damage secondary to the brain injury, and providing children the highest degree of independence so as to increase their participation in society.[16] This way, pediatric rehabilitation programs ultimately aim to improve the quality of life of children and their families.[13] Treatment options for children with CP include physical, occupational, and speech therapies, drug treatments for spasticity, and orthopedic and neurosurgical interventions.[17] Most children require combinations of these therapies.

Over the past decade, therapeutic goals of physical interventions for children with CP have primarily focused on facilitating motor development and skills, assistance with gait/mobility deficits, maintaining muscle length, prevention of secondary complications such as deformities, and improving the ability to walk or perform other functional activities in daily life.[16,18] In more recent years, health-promotion strategies and goals for rehabilitation programs have shifted to more activity-based interventions for improved fitness, functional mobility, and habitual physical activity.[19] This has led, for example, to the establishment of CP-specific physical activity and exercise recommendations,[10] which aim to promote healthy, active lifestyle behaviours (i.e. increasing physical activity, and reducing sedentary behaviour) during the day. However, focusing on daytime activities alone may not be enough to optimize health and development in children with CP.

SLEEP AND THE 24-HOUR ACTIVITIES

Healthy sleep is essential for children's physical and mental health and plays a crucial role in brain development.[20,21] Impaired sleep during childhood is associated with a broad spectrum of negative consequences, including poor growth, deficits in cognitive functioning, challenges with emotional regulation, adverse school performance, and a reduced quality of life.[20,22]

Children with CP are considered to be a population at risk for the occurrence of sleep problems.[23] Increasing evidence suggests that these children may be more vulnerable to the impact of poor sleep than their typically developing peers, with far-reaching implications for child health, functioning, and well-being of the family.[24–26]

Given the central role of sleep in recovery and repair processes,[27,28] it would be logical to assume that sleep deserves to receive proper attention in the rehabilitative care for children with neurodevelopmental disorders and neonatal brain injury, like CP. However, despite its potential to maintain (or disrupt) overall health and functioning, the night is often neglected and sleep problems are often overlooked in pediatric rehabilitation.[29] There is a dire need to bridge this -quite literal- yawning gap. This could be achieved by considering the day and night, and the activities within this 24-hour period, as a whole.

The way children spend their time within a 24-hour period, has important implications for (long-term) physical health.[30] From this whole-day activity perspective, a child can engage in various lifestyle behaviours, i.e. physical activity of different intensities, sedentary behaviour, and sleep. Together, these behaviours span the breadth of the so-called 24-hour activity continuum,[30] as illustrated in Figure 1.1. While the independent health benefits of high levels of physical activity, low levels of sedentary behaviour, and sufficient hours of good quality sleep have been well-established,[23,31,32] the collective importance of these behaviours is becoming increasingly recognized under the concept that "*the whole day matters*",[33] resulting in 24-hour activity guidelines for children.[34,35] The importance of achieving an optimal balance between the 24-hour activities is highlighted by evidence indicating that having one 'unhealthy' activity behaviour can moderate the health benefits of another.[30] Given that children with CP not only have inactive lifestyles (i.e. these children tend to

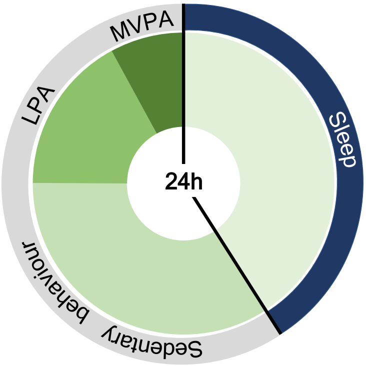


Figure 1.1 The 24-hour activity continuum captures the activity behaviours throughout a 24-hour period, ranging from zero/low intensity (sleep) to medium/high intensity (MVPA).
Abbreviations: LPA = light physical activity; MVPA = moderate-to-vigorous physical activity.

have low levels of physical activity and spend prolonged times sitting),[10,36] and are likely to experience impaired sleep,[23] this emphasizes the urge to acknowledge the entire 24-hour activities in the care for children with CP, with attention to day ánd night.

Adopting a 24-hour lens has the potential to advance the rehabilitative care for children with CP by providing a broader window of opportunity and a more holistic approach towards optimized health and well-being. Improving the quality of sleep in children with CP may not only improve their clinical outcomes, it could greatly ameliorate the quality of life of the child and the entire family.[37,38] Given that the major long-term goal of rehabilitation is to ensure children with CP and their families the best possible quality of life,[13,39] it is time to wake up to the importance of sleep in the rehabilitative care for this population.

AIMS AND OUTLINE OF THIS THESIS

In order to increase awareness of, knowledge about, and ultimately change attitudes and practices regarding care for sleep, research into the field of sleep in children with CP, and the impact of sleep problems on the family, is needed. It is hoped that an increased understanding of (and for) sleep problems in children with CP will pave the way for the development and implementation of new therapeutic approaches and pathways that consider and integrate care for sleep into pediatric rehabilitation practices.

Hence, the overall objective of this thesis is to provide a wake-up call on the importance of sleep in the (rehabilitative) care for children with CP. Therefore, this thesis is divided into four parts, each with separate aims:

PART I To understand the parental perspectives on the care for sleep of their child with CP (**Chapter 2**).

PART II To evaluate the care for sleep in pediatric rehabilitation (**Chapter 3**).

PART III To develop a clinical practice guide on how to integrate sleep in the care for children with CP using a 24-hour activity approach (**Chapter 4**), and to report the results of a newly developed practical tool for detecting 24-hour activity problems in children with CP, with a focus on sleep problems (**Chapter 5**).

PART IV To measure the 24-hour activities in ambulatory children with CP using actigraphy (**Chapter 6**), and to compare subjective and device-based measures of sleep so as to formulate recommendations for the use of these methods in research and clinical practice (**Chapter 7**).

REFERENCES

1. Oskoui M, Coutinho F, Dykeman J, et al. An update on the prevalence of cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol. 2013;55:509–19.
2. Odding E, Roebroeck ME, Stam HJ. The epidemiology of cerebral palsy: Incidence, impairments and risk factors. Disabil Rehabil. 2006;28:183–91 (2006).
3. Rosenbaum P, Paneth N, Leviton A, et al. A report: the definition and classification of cerebral palsy April 2006. Dev Med Child Neurol. Suppl. 2007;109:8–14.
4. Palisano R, Rosenbaum P, Walter S, et al. Development and reliability of a system to classify gross motor function in children with cerebral palsy. Dev Med Child Neurol. 1997;39:214–23.
5. Hollung SJ, Bakken IJ, Vik T, et al. Comorbidities in cerebral palsy: a patient registry study. Dev Med Child Neurol. 2020;62:97–103.
6. Gabis LV, Tsubary NM, Leon O, et al. Assessment of abilities and comorbidities in children with cerebral palsy. J Child Neurol. 2015;30:1640–5.
7. Novak I. Evidence-Based Diagnosis, Health Care, and Rehabilitation for Children With Cerebral Palsy. J Child Neurol. 2014;29:1141–56.
8. Varni JW, Burwinkle TM, Sherman SA, et al. Health-related quality of life of children and adolescents with cerebral palsy: hearing the voices of the children. Dev Med Child Neurol. 2005;47:592–7.
9. Beckung E, White-Koning M, Marcelli M, et al. Health status of children with cerebral palsy living in Europe: a multi-centre study. Child Care Health Dev. 2008;34:806–14.
10. Verschuren O, Peterson MD, Balemans ACJ, et al. Exercise and physical activity recommendations for people with cerebral palsy. Dev Med Child Neurol. 58, 798–808 (2016).
11. Verschuren O, McPhee P, Rosenbaum P, et al. The formula for health and well-being in individuals with cerebral palsy: physical activity, sleep, and nutrition. Dev Med Child Neurol. 2016;58:989–90.
12. Hadders-Algra M, Pangalila RF, Becher JG, et al. Kinderrevalidatie. Uitgeverij Koninklijke Van Gorcum; 2021.
13. Trabacca A, Vespino T, Di Liddo A, et al. Multidisciplinary rehabilitation for patients with cerebral palsy: improving long-term care. J Multidiscip Healthc. 2016;9: 455.
14. Gulati S, Sondhi V. Cerebral Palsy: An Overview. Indian J Pediatr 2017;85:1006–16.
15. Dirks T, Hadders-Algra M. The role of the family in intervention of infants at high risk of cerebral palsy: a systematic analysis. Dev Med Child Neurol. 2011;53:62–7.
16. Novak I, McIntyre S, Morgan C, et al. A systematic review of interventions for children with cerebral palsy: State of the evidence. Dev Med Child Neurol. 2013;55:885–910.
17. Jan MMS. Cerebral Palsy: Comprehensive Review and Update. Ann Saudi Med. 2006;26:123–32.
18. Patel DR, Neelakantan M, Pandher K, Merrick J. Cerebral palsy in children: a clinical overview. Transl Pediatr. 2020;9:S125.
19. Verschuren O, Darrah J, Novak I, et al. Health-Enhancing Physical Activity in Children With Cerebral Palsy: More of the Same Is Not Enough. Phys Ther. 2014; 94:297–305.
20. Chaput J, Gray CE, Poitras VJ, et al. Systematic review of the relationships between sleep duration and Health Indicators in School-Aged Children and Youth. Appl Physiol Nutr Metab. 2016;41:S266–82.
21. Zielinski MR, McKenna JT, McCarley RW. Functions and Mechanisms of Sleep. AIMS Neurosci. 2016;3:67.
22. Astill RG, Van der Heijden KB, Van IJzendoorn MH, et al. Sleep, cognition, and behavioral problems in school-age children: A century of research meta-analyzed. Psychol Bull. 2012;138:1109–38.
23. Simard-Tremblay E, Constantin E, Gruber R, et al. Sleep in children with cerebral palsy: A review. J Child Neurol. 2011;26:1303–10.
24. Lélis ALPA, Cardoso MVLM, Hall WA. Sleep disorders in children with cerebral palsy: An integrative review. Sleep Med Rev. 2015;30:63–71.
25. Lang CP, Boucaut A, Guppy M, et al. Children with cerebral palsy: A cross-sectional study of their sleep and their caregiver's sleep quality, psychological health and well-being. Child Care Health Dev. 2021;47:859–68.
26. Wayte S, McCaughey E, Holley S, et al. Sleep problems in children with cerebral palsy and their relationship with maternal sleep and depression. Acta Paediatr. 2012;101:618–23.

27. Venter RE. Role of Sleep in Performance and Recovery of Athletes: a Review Article. South African J Res Sport Phys Educ Recreat. 2012;34:167–84.
28. Duss SB, Seiler A, Schmidt MH, et al. The role of sleep in recovery following ischemic stroke: A review of human and animal data. Neurobiol Sleep Circadian Rhythm. 2017;2:94–105.
29. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: An underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
30. Chaput JP, Carson V, Gray C, et al. Importance of All Movement Behaviors in a 24 Hour Period for Overall Health. Int J Environ Res Public Health. 2014;11:12575–81.
31. Janssen I, LeBlanc AG. Systematic review of the health benefits of physical activity and fitness in school-aged children and youth. International Journal of Behavioral Nutrition and Physical Activity. 2010;7:40.
32. Cappuccio FP, Taggart FM, Kandala NB, et al. Meta-analysis of short sleep duration and obesity in children and adults. Sleep. 2008;31:619–26.
33. Rollo S, Antsygina O, Tremblay MS. The whole day matters: Understanding 24-hour movement guideline adherence and relationships with health indicators across the lifespan. J Sport Heal Sci. 2020;9:493–510.
34. Tremblay MS, Carson V, Chaput JP, et al. Canadian 24-hour movement guidelines for children and youth: An integration of physical activity, sedentary behaviour, and sleep. Appl Physiol Nutr Metab. 2016;41:S311–27.
35. Okely AD, Ghersi D, Hesketh KF, et al. A collaborative approach to adopting/adapting guidelines - The Australian 24-Hour Movement Guidelines for the early years (Birth to 5 years): An integration of physical activity, sedentary behavior, and sleep. BMC Public Health. 2017;17:869.
36. Obeid J, Balemans ACJ, Noorduyn SG, et al. Objectively measured sedentary time in youth with cerebral palsy compared with age-, sex-, and season-matched youth who are developing typically: An explorative study. Phys Ther. 2014;94:1163–7.
37. McDonald A, Joseph D. Paediatric neurodisability and sleep disorders: clinical pathways and management strategies. BMJ Paediatr Open. 2019;3:e000290.
38. Owens J. Classification and Epidemiology of Childhood Sleep Disorders. Prim Care - Clin Off Pract. 2008;35:533–46.
39. Schneider JW, Gurucharri LM, Gutierrez AL, et al. Health-related quality of life and functional outcome measures for children with cerebral palsy. Dev Med Child Neurol. 2001;43:601–8.

Part I

Understanding the perspectives of parents regarding the care for sleep of their child with cerebral palsy

Chapter 2

Parental perspectives on care for sleep in children with cerebral palsy: A wake-up call

Raquel Y Hulst, Jeanine M Voorman, Sigrid Pillen, Marjolijn Ketelaar,
Johanna MA Visser-Meily, Olaf Verschuren

Disabil Rehabil. 2022;44(3):458–67. doi: 10.1080/09638288.2020.1770873.

ABSTRACT

Purpose: Sleep problems are common in children with cerebral palsy (CP) and have a large impact on child health and family functioning. This qualitative study aimed to explore parental perspectives regarding the care for sleep of their young child (age 1–8 years) with CP.

Materials and methods: Individual, semi-structured interviews were conducted with eighteen parents of a child with CP (GMFCS levels I–V). Inductive thematic analysis of the data was performed within each of the three preidentified domains: 1) Current situation; 2) Concerns; 3) Needs.

Results: In total, sixteen themes were identified across the three domains. Parents expressed a range of sleep-related issues within the families' current situation, with significant concerns and needs regarding the care for sleep of their child. Themes mapped across the home environment included child and family aspects, whereas those identified within the healthcare setting included clinical practices and attitudes of healthcare professionals, as well as the broader organisation of care for sleep.

Conclusions: Parents face numerous challenges caring for their child's sleep and the burden placed on families by sleep problems is underappreciated. In order to break the vicious circle of sleep problems and their disastrous consequences on families' wellbeing, we need to wake up to parent-identified issues and shortcomings in healthcare. Care for sleep should be integrated into pediatric rehabilitation through routine inquiries, using a family-centred and multidisciplinary approach.

INTRODUCTION

Cerebral palsy (CP) is a common cause of childhood disability worldwide.[1] Between 23% and 46% of the children with CP are reported to experience sleep problems.[2–6] These sleep problems range from difficulties in initiating and maintaining sleep, to sleep-wake transition disorders, excessive daytime sleepiness and sleep-related breathing disorders.[2,3,6] Although the underlying causes of the sleep problems are not always clear, they may be related to (a combination of) comorbid physical and medical conditions that often accompany CP, such as spasticity, epilepsy, reflux, pain, a decreased ability to change body position at night, visual impairments, sensory sensitivity and behavioural problems.[7–13]

Sleep deficiency in childhood adversely affects health and development.[14] Lower sleep quality is related to deficits in cognitive functioning and significantly impacts on school performance of both typically developing children and children with CP.[2,15,16] In children with CP, sleep problems have been shown to increase the risk of impaired psychological health,[17] with insomnia and excessive daytime sleepiness being associated with a lower quality of life.[18]

In addition to the negative implications for the child, sleep problems in children with CP can place a heavy burden on the family.[2,5,6] Parents can become sleep deprived themselves as a result of their child requiring night-time attention.[5,19,20] Increased caregiving demands at night have been associated with elevated parental stress and psychological exhaustion.[20–23] As one study has shown, 40% of mothers of a child with CP experienced poor sleep quality, which in turn was associated with maternal depression.[13] In the same study, 74% of parents reported impairments in daytime functioning because of their child's sleep disorder.[13]

Clearly, sleep problems in children with CP and its consequences must be considered as a broader issue, concerning the entire family system.[6,7,24] Understanding the caregiving experiences of parents and the challenges they face, are necessary to better support these families. Therefore, the objective of this qualitative study is to explore the care for sleep of children with CP from a parental perspective.

METHODS

Study design and procedure

A qualitative, exploratory study was conducted using an inductive thematic analysis approach.[25,26] The research team consisted of researchers and healthcare professionals with varied backgrounds (i.e. pediatric rehabilitation, biomedical sciences, medicine and somnology), enabling us to take different interpretations of the data into account. The Medical Ethics Research Committee of the University Medical Centre Utrecht, the Netherlands, granted the study exempt from the Dutch Medical Research Involving Human Subjects Act (file number 19-066), and therefore no ethical approval was required.

Participants

Parents of children with CP were recruited via healthcare settings (rehabilitation centres and rehabilitation department of a children's hospital) and parent organisations (CP Nederland and OuderInzicht) in the Netherlands, using a convenience sampling method. The inclusion criteria were parents of a child with CP classified as Gross Motor Function Classification System (GMFCS) levels I–V, and under the age of 16. As sleep varies greatly across childhood, we focused on early childhood after infancy, spanning the period between the age of 1 and 8 years. To increase the sample size, we also included parents of children older than 8 years, who were asked to reflect on the period of their child's life between the age of 1 and 8 years. Exclusion criteria were not being able to understand or converse in Dutch. Parents were informed about the study by means of an information brochure, and were subsequently asked for permission by their treating physician (assistant) or therapist to be contacted by the researcher. Participation in the study was voluntary and all participants signed an informed consent form.

Data collection

Individual semi-structured interviews were conducted by the first author (RYH), and used for data collection. A topic guide (Appendix) was developed based on existing literature and expert knowledge of the research team, and was structured to cover three domains that were set a priori: 1) Current situation; 2) Concerns; 3) Needs. Open-ended questions were used to gain insight into parent's experiences regarding the care for their child's sleep within these three domains, aiming to identify sleep-related issues in the families' current situation, concerns parents may have regarding their child's sleep, and how these may affect their family, as well as parental needs with regard to improving the care for their child's sleep. Although the interviews were guided by pre-set topics, parents were allowed and encouraged to address topics and issues which they felt deserved receiving acknowledgement. Therefore, the term 'care for sleep' could be freely interpreted by parents as any care related to the child's sleep (problems), whether it be care provided by caregivers, parents, or nurses providing night care at home, or care delivered in pediatric clinical practice. Similarly, the phrasing 'sleep problem' was not tied to any particular sleep disorder, but rather served as a term to describe sleep as problematic when parents perceived and reported it as such. Parents were given the choice to have the interview conducted by telephone or face-to-face at a location of their preference. The sample size was not determined before the start of the study, but followed the qualitative approach of data saturation: this was achieved when no new themes emerged during three consecutive interviews. Interviews were audio-recorded with permission. To improve validity of the data and enhance credibility, a summary of the initial pair of interviews was shared with the corresponding participants for member-checking.

Data processing

Audio-taped interviews were transcribed verbatim. Final transcripts were cross-referenced against the audio tapes by an independent student-researcher to ensure accuracy. Personal details were removed from the transcripts and a unique numeric code was assigned to each interview transcript to ensure anonymity.

Data analysis

Data were coded independently by two researchers (RYH & OV) using MAXQDA (VERBI Software, version 2018.2). Inductive thematic analysis[26] of the data was undertaken within each of the three preidentified domains. First, the transcribed texts were repeatedly read to become familiar with the narratives and to gain an initial understanding of the content. Next, the texts were broken down into fragments of meaningful units, which were labelled with codes. Codes were compared and discussed between researchers until consensus regarding content and interpretation was reached. Subsequently, interpretive coding took place, during which the codes with comparable meaning were clustered into categories, and subsequently grouped into subthemes. Finally, subthemes were further collapsed into overarching main themes within each of the three domains (Current situation, Concerns, Needs). Preliminary results were discussed with experts in the field of qualitative research and refined accordingly. To ensure credibility of the findings, the entire research team was involved throughout the process of data analysis. Organisation of categories and construction of (sub)themes within domains were decided upon through discussions until agreement was reached about the interpretation of the data. Quotations from the interviews are enclosed in the results section to illustrate our findings and verify interpretations.

RESULTS

Participants

In total, eighteen parents (one parent per family) were interviewed. Interviews were between 33–74 minutes in duration. Four parents of children above the age of 8 were asked to reflect on the period of their child between the age of 1 and 8 years. Characteristics of participating parents and their child are summarised in Table 2.1.

Findings

The findings of this qualitative study revealed that, regardless of the presence and extent of possible sleep problems, all parents expressed a range of concerns and needs with regard to the care for sleep of their child. Within each of the three domains set a priori (Current situation, Concerns, Needs) several themes and subthemes (Tables 2.2–2.4) were identified; across domains there seemed to be a clear separation between themes described within the families' home environment versus those established within the healthcare setting. Figure 2.1 shows a representation of how identified themes map across domains and environmental settings. Themes are discussed below for each of the three domains, with illustrative quotes.

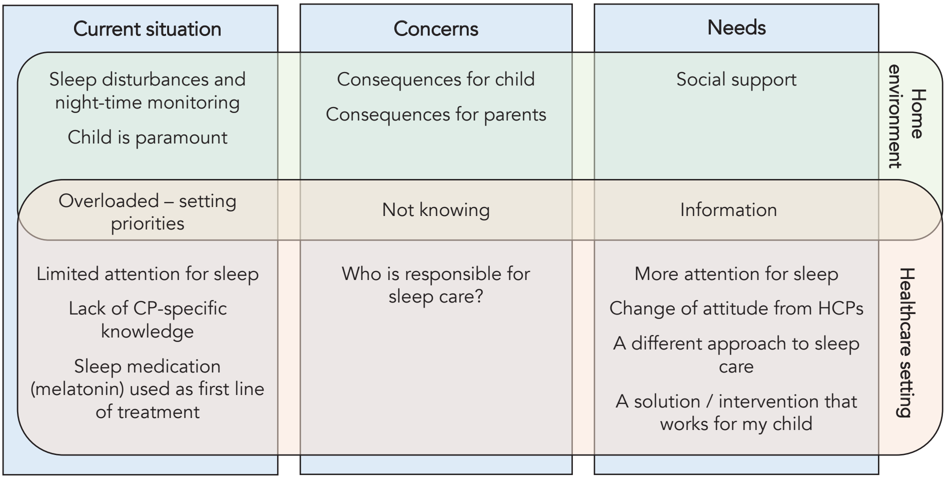


Figure 2.1 Representation of the identified themes mapped across the three domains (*Current situation*; *Concerns*; *Needs*) and environmental settings (*Home*; *Healthcare*).

Table 2.1 Characteristics of parents and their children

Characteristics (*N*=18)	*n* (%)
PARENT	
Role	
Father	2 (11)
Mother	16 (89)
Age	
20–30 years	1 (6)
31–40 years	12 (67)
41–50 years	4 (22)
51–60 years	1 (6)
Education	
Secondary vocational education	2 (11)
Higher professional education	7 (39)
University education	8 (44)
Not reported	1 (6)
CHILD	
Sex	
Boy	10 (56)
Girl	8 (44)
Age (years)	
Range	2–15
Average (SD)	6.2 (2.9)
Median	5.5
GMFCS level	
I	3 (17)
II	4 (22)
III	2 (11)
IV	4 (22)
V	5 (28)
Type of CP	
Spastic	13 (72)
Dyskinetic	2 (11)
Mixed spastic & dyskinetic	2 (11)
Unknown	1 (6)
Affected side of body	
Unilateral	4 (22)
Bilateral	13 (72)
Not reported	1 (6)
Siblings	
Yes	11 (61)
No	7 (39)

Current situation

This domain encompasses descriptions of the current situation or context in which sleep problems manifest at home, and how sleep is currently addressed by healthcare professionals from a parental viewpoint. Six themes were identified (Figure 2.1 and Table 2.2).

Table 2.2 Current situation

Current situation
Themes
Sleep disturbances and night-time monitoring
Child is paramount
Overloaded – setting priorities
Limited attention for sleep in healthcare
Lack of CP-specific knowledge
Sleep medication (melatonin) is used as first line of treatment

Sleep disturbances and night-time monitoring

Parents gave detailed descriptions of the sleep disturbances their child experiences at night, which could range from waking up a few times per night to hardly getting any sleep. The child's sleep problems were often accompanied or believed to be caused by co-morbid problems, such as breathing difficulties, epilepsy, reflux, or pain. These problems frequently required parents to be constantly alert and vigilant during the night:

> *"We are trained to hear the sound of her suffocating; it's the sound that alerts you. And when she cries, you hear it of course. But there are moments that we think we don't even notice. So we use camera and sound monitoring."* (Parent of a 6-year-old child)

As a consequence of night-time monitoring, many parents have to get out of bed multiple times per night. Some parents described this to be 'normal':

> *"In the beginning there were nights that I went out of bed, say 10 to 15 times. That was just quite normal."* (Parent of a 5-year-old child)

Child is paramount

For some parents the child's sleep comes first before everything else, including their own sleep. Parents try to do whatever it takes to make sure that their child sleeps well, either to prevent the negative effects that poor sleep has on the family, or because they feel there is no other option. In contrast, other parents reported to 'accept' the sleep problems of their child; instead of trying to do whatever it takes to fix the issue, they rather focus on coping with the current situation in the best possible way. For some, this simply meant ensuring that at least they themselves get enough sleep at night in order to be able to function the next day:

> *"Our criterion is not based on her being awake or not anymore. Our criterion is based on our ability to sleep. So, yes, she might be awake for hours, that is possible. It could be better perhaps, but we have tried many things. We still have the same sleep problems and the only thing we've done is to try and make sure we suffer as little as possible in our daily functioning. Ourselves."* (Parent of a 6-year-old child)

Overloaded – setting priorities

Having a child with CP requires parents to provide and manage multidimensional health and care needs simultaneously. Sleep is only one of the many domains that requires attention. Not having enough time to manage everything demands parents to make choices and set priorities.

Limited attention for sleep in healthcare

In describing the current healthcare of their child, the majority of parents stated that there is limited, if any, attention for sleep. Some parents reported never to have been asked about the topic sleep by their healthcare professional:

> *"No one ever really asks about it [sleep]."* (Parent of a 2-year-old child)

Other experiences described hospital visits, where awareness around the importance of sleep and the (long term) consequences of impaired or disturbed sleep for the (sick) child was lacking:

> *"We were in the hospital for a while. And they had to inject her – whether it was her time to sleep or not. So she would be woken by a complete stranger. It gave her a bit of a mini trauma which would go on for weeks, and she'd be afraid to go to sleep."* (Parent of a 6-year-old child)

Lack of CP-specific knowledge

A recurrent statement from parents was the healthcare professionals' lack of knowledge about CP and a constant comparison of their child (with CP) to typically developing children. Receiving 'generic' help or advice that is not tailored to their child with CP, brought along frustration and a feeling of being not taken seriously. This was the case in hospitals, but also in specialised sleep clinics:

> *"Then you get to an official sleep clinic, but they haven't got a clue about children like XY. It's all based on normal people. This is such a specific issue. So then they'd say: 'Oh well…' and they'd start to mess about with melatonin, but it has to do with so many different factors."* (Parent of an 11-year-old child, in retrospect)

Sleep medication (melatonin) is used as first line of treatment

As illustrated in the abovementioned quote, several parents reported that in current healthcare, sleep treatment is focussed on the use of pharmacological interventions. In their point of view, melatonin and sleep medication seem to be used as first line of treatment, despite the reported negative effects on the child's wellbeing:

> *"What I think is a shame, is that the first solution is always prescribing medication. XY had sleep medication at one time and, ah, it was a disaster. She would wake terribly - we really thought it made her ill."* (Parent of a 15-year-old child, in retrospect)

Concerns

This domain describes the four main concerns that parents have and the challenges they face in caring for their child's sleep (Figure 2.1). Themes and sub-themes are presented in Table 2.3.

Table 2.3 Concerns

Concerns	
Themes	*Sub-themes*
Consequences for the child	Health and wellbeing Vicious circle of poor sleep Future
Consequences for parents	Impact on family functioning Reaching limits
The unknown	Origin and causes of sleep problems
Responsibility	Who is responsible for sleep care?

Consequences for the child

The main concerns expressed by parents were related to the consequences of sleep problems for the child, as well as for the parents. The consequences for the child highlighted three areas of concern. Firstly, parents expressed health concerns for their child, like getting sick more easily as a result of poor sleep. Others worried about their child's wellbeing during the night. For example, statements were made about children suffering from pain caused by spasticity in their limbs, lying uncomfortably (e.g. from wearing night orthoses) or unknown reasons that keep them awake at night. The following quote from a mother illustrates the extent of her concerns:

> *"We really thought: if you have to live like this, it should be over, because you're in such pain. And this happens in the middle of the night, she would wake up screaming*

> *and crying, and after an hour you still can't get her to calm down and we would think 'If you have to live like this, then, rather not at all…"* (Parent of a 6-year-old child)

Secondly, parents mentioned concerns about poor sleep acting as a negative spiral, thereby impacting on everything else in their child's life and development:

> *"It's like a house of cards, if you lack sleep; it has an effect on all other areas."* (Parent of an 11-year-old child, in retrospect)

These statements indicate that parents consider sleep as a fundamental building block, necessary for their child to function properly.

Thirdly, concerns were expressed with regard to the child's future. Parents worried about their child's ability to sleep in the future in general, sleepovers, structure and routines. For various reasons, some children had developed a preferred sleeping position (usually on one specific side) which imposed future-related worries for this mother:

> *"I have been thinking about sleep as such – I'm worried about these severe spasms. He always wants to sleep in a specific position, on his side. And he can really tense up his legs, or he can be lying there totally relaxed. And then I sometimes think, when he gets older and gets more physical ailments, then he will not be able to sleep in a comfortable way anymore. I would worry how he would be able to sleep."* (Parent of a 7-year-old child)

Consequences for parents

In addition to consequences for the child, parents described concerns in terms of consequences for themselves. Comprehensive reports of the impact of sleep disturbances on many areas of family functioning were given by parents. The need for parental monitoring at night required them to be constantly alert for sounds, to check video/audio recordings, or to physically get out of bed multiple times per night. On top of that, the resulting deprived parental sleep led to adjustments in households, like rotating night 'duties' or deploying nurses that provide night care. All these consequences impacted greatly on families.

> *"We swap days and nights shifts and we recently had a week without any help at night. Well, we felt like dead! Now that we have help during the night, it is OK. But if I have one night without sleep, I think: 'How did I do it?'. It's just impossible, not real, and all you do is surviving, but you really can't do it."* (Parent of a 6-year-old child)

Another factor repeatedly reported to negatively affect daytime family functioning was their child's excessive sleepiness or fatigue during the day, which according to parents was often a consequence of them not getting enough sleep at night:

> *"It [fatigue of the child] is a very limiting factor in your life. You really have to plan things. We can do one or two things a day, then it's finished."* (Parent of a 7-year-old child)

Parents are struggling with their current situation and the consequences for the entire family, with some describing to have turned into 'survival mode' in order to cope with the circumstances. As a result, concerns were expressed about having to keep up with this daunting and exhaustive situation that parents are in. This brought along uncertainties about their future and raised questions like: When will we reach our limits of not being able to proceed any longer? – as this mother described:

> *"Perhaps it will change, in terms of sleep. But our main worry is: what will happen then? We are not thinking about respite care at all yet. But at some point, we may reach our limits, and then things will have to change."* (Parent of a 7-year-old child)

The unknown

In many cases, parents did not know the exact cause of their child's sleep problem. This concern is enhanced by the fact that some children with CP cannot speak, making them unable to explain what is bothering them:

> *"It's always so funny with a non-speaking child (sarcastically). It could be epilepsy, it could be constipation, it could be spasm or cramps, but it could also be a hairpin on her pillow that pricks in her ear, but she can't tell us. And then she lies there, she's strapped in one position and she can't even turn herself around."* (Parent of an 11-year-old child, in retrospect)

Others described not knowing whether the origin of the sleep problems is related to their child's developmental age, is inherent to their child's natural characteristics/behaviour, or whether it directly results from their child's brain damage:

> *"It's so difficult. What is normal toddler behaviour? And what is XY? And what comes out of his CP?"* (Parent of a 5-year-old child)

Responsibility

Parents repeatedly voiced concerns about the lack of clarity of responsibilities of healthcare professionals when it comes to care for sleep. From their point of view, nobody takes clear responsibility for the topic, as sleep does not 'fit' under one discipline, but clearly is an integral part of their child's health in general. Their uncertainty is accompanied by the fact that generally there are many professionals involved in their child's healthcare, leaving parents unsure who to approach:

> *"I wouldn't know. We talk to ten different parties. I have a physical therapist, a pediatrician, a school, a social worker, I have (…) Who do we ask? I wouldn't even know who to ask…"* (Parent of a 6-year-old child)

Needs

Six themes were identified as parental needs (Figure 2.1). Themes and sub-themes are presented in Table 2.4.

Table 2.4 Needs

Needs	
Themes	*Sub-themes*
Social support	Understanding Acknowledgement
Information	Insight into causes and consequences of sleep problems Tips, advice and guidance
More attention for sleep in healthcare	Routine inquiries Taking initiative and responsibility
Change of attitude from healthcare professionals	No judging A coaching and supportive attitude
A different approach to sleep care	Family-centred care Multidisciplinary collaboration Holistic approach – the child as a whole
A solution / intervention that works for my child	Tailor-made Finding the way

Social support

Parents mentioned not feeling understood by the people around them:

> *"Our direct environment, they just do not understand. Because you cannot understand when you're not in the middle of it."* (Parent of a 7-year-old child)

Since their friends and neighbours 'do not have a child like theirs', the adverse consequences of the child's sleep problems, like deprived parental sleep, fatigue or forgetfulness during the day, are too often not appreciated by the social environment. As a result, this mother constantly needs to remind others of her impaired sleep and exhaustion:

> *"Then at some point you start to think: 'I am so bone tired!' And then I'll explain again: 'Hey guys, I'm so... I don't sleep.' And then they understand. But you have to keep at it, and it just goes on. It's very invisible. It's always something you need to indicate: 'Gosh, I notice that I'm exhausted, I simply forget things, I haven't registered.'"* (Parent of an 11-year-old child, in retrospect)

Parents therefore expressed the need for social support, i.e. receiving understanding and acknowledgement from their environment.

Information

As many parents expressed their concern of not knowing the cause of their child's sleep problem, there is a logical need for them to gain insight into the cause. In addition, understanding the consequences of sleep problems was marked important for parents to prepare themselves for what is coming. They expressed a need for objective, reliable information about sleep that is clustered in one place, and specifically aimed at children with CP.

Furthermore, there was a strong need for receiving advice about possibilities to improve sleep. This ranged from specific information about sleeping devices or orthotics, to general advice about healthy sleep practices, or tips from other parents of a child with CP. Some parents seemed afraid to miss out on potential important information that may benefit them and their child, if they are not informed about such matters:

> *"I believe that there are advices and tools out there that could be helpful, but I'm not aware of them. So it would be good if others can inform you or point them out to you."* (Parent of a 10-year-old child, in retrospect)

More attention for sleep in healthcare

A unanimous necessity that parents expressed was the need for sleep to receive more attention in healthcare. Parents indicated that they had never really been asked about sleep by their child's rehabilitation physician, and that they would appreciate the topic being discussed routinely during check-ups.

> *"I believe that especially for children like XY it's important to sleep well, so (…) it should be addressed."* (Parent of a 2-year-old child)

Whilst participating in this research, a mother realised that she wished that healthcare professionals had asked her about her child's sleep before:

> *"Talking about it, I realise that although I had a lot of questions about it [sleep], I couldn't formulate them myself without people asking me first. It's only when other people ask about it, that you think: 'Oh, that's strange indeed'. And now that we're talking about it, I would have liked to hear more, and be questioned about it."* (Parent of a 4-year-old child)

According to parents, they often have to take the lead in bringing up issues concerning sleep. The majority indicated that they would like their healthcare professionals to take the initiative during consultations, and inquire with parents earlier.

> *"Very much at our own initiatives as parents. I would have liked the doctor to take initiatives and tell us or ask us about it. And start early, not when a parent is already deep in trouble, but before."* (Parent of a 15-year-old child, in retrospect)

Change of attitude from healthcare professionals

A different attitude from healthcare professionals was perceived as an important need by parents. Discussing the sleep problems that parents experience at home can be a sensitive topic that requires a coaching, understanding attitude from healthcare professionals, without being judgemental:

> *"Talking to parents, coaching, without judging. That's the most important thing. Parents often feel pushed into a corner, or being judged that they don't do enough. And we are vulnerable. I was less open. But when I realised I wasn't being judged I became more open."* (Parent of a 15-year-old child, in retrospect)

A different approach to sleep care

A significant theme identified as parental need, is for the care for sleep to receive a different approach. Parents described current healthcare to be very child-centred, i.e. focused on the child alone. The majority expressed a clear need for their child's healthcare to become more family-centred. This implies that parents would like healthcare professionals to consider their sleep and wellbeing in the context of their child's sleep issues:

> *"I think we need to be asked more questions, and also aimed at the parents, like: 'How is your sleep? How does that suffer from your child's sleep?' That doesn't happen enough yet. That this is also taken into consideration. This is just as important as*

> *looking at the child. Not enough is done to see how parents are coping."* (Parent of a 7-year-old child)

Parents reported healthcare professionals to be too focussed on their own discipline alone. Only by taking into account a wider scope of view, by combining aspects of different disciplines, the child can be considered as a whole. This parent advocated a holistic approach could benefit finding a solution to her child's sleep problem:

> *"I think we need to be more multi-disciplinary towards dealing with sleep problems. Because reflux might play a part… or other medical issues… sleeping position, cramps during the night. We do not look at the complete picture yet."* (Parent of a 15-year-old child, in retrospect)

In line with this holistic approach, parents view care for sleep to be the responsibility of every professional involved in their child's care. As multiple disciplines are typically involved in the child's healthcare, parents expressed the need for them to combine their knowledge and expertise as a way to approach sleep care:

> *"I always ask for a different attitude. We need to work together, look at the problem together. So the 'sleep-doctor', that's what I'll call him, and the rehabilitation services should work with us to solve the puzzle. Let's solve this problem, with joint knowledge."* (Parent of an 11-year-old child, in retrospect)

Clearly, parents would like to turn the care for sleep into a team effort.

A solution / intervention that works for my child

Ultimately, parents want to find a solution or intervention that works for their child:

> *"I would appreciate it if someone comes to a solution."* (Parent of a 6-year-old child)

Many parents reported to have undertaken actions to prevent or treat their child's sleep problems, of which some with successful outcomes. Anecdotes shared by the parents made it clear that there are dozens of different types of interventions that could optimise sleep, but due to the heterogeneity of the sleep problems in children with CP, only certain interventions seem effective for certain children. According to parents, sleep interventions should be tailored to the child's complaints, parental needs and home situation. Additionally, parents indicated lack of a clear overview of the possibilities and routes to follow when it comes to finding appropriate sleep interventions, and believe that clinicians should guide them in finding the solution they seek:

> *"They should know which route to follow to get help and where."* (Parent of a 6-year-old child)

DISCUSSION

Achieving a good night's sleep for both the child and the parents is important for health and wellbeing. In children with CP the prevalence of sleep problems is high and the resultant burden placed on families is underappreciated. Our qualitative study addressed parental perspectives on the care for sleep of children with CP, and identified various concerns and needs that warrant acknowledgement. Perceived shortcomings in current healthcare lead to under recognition of the challenges faced by parents in caring for their child's sleep. This paper serves as a wake-up call echoing the parents' voices to raise awareness about the importance of sleep in pediatric healthcare.

Similar to concerns of parents of children with physical disabilities,[23] parents in our study reported being concerned about their child's safety and wellbeing at night, which often resulted in the need to monitor the child's sleep at night. Additionally, long-term concerns about the negative consequences of poor sleep on the child's general health along with worries about the child's future sleep habits were frequently expressed. Secondary to the child, parents were concerned about adverse consequences for their own wellbeing. Increased caregiving demands at night were reported to lead to deprived parental sleep, extreme fatigue, and significantly impaired family functioning during daytime. In fact, coping with a child with sleep problems has been described as one of the main causes of parental stress in these families.[23,27] Our findings are in line with prior research on parents of children with physical disabilities who suffered from disrupted sleep, poor health and psychological exhaustion when their child had sleep problems.[5,20,23] It may be clear that the harmful effects of sleep problems on family functioning should be considered of great concern, especially given the previously reported lower quality of life and decreased mental health in parents of children with CP.[13,22,28,29] This study complements previous findings by showing that sleep problems can further operate in a downward spiral, with exhausted parents who experience day-to-day hassles that can pile up and cause a stress overload, while others quietly contemplate respite care in view of reaching their limits. In order to break the vicious circle of sleep problems, it is crucial for sleep care to be acknowledged within pediatric healthcare in a family-centred manner.

Despite the high prevalence of sleep problems in children with CP, they are still under-recognised in neurorehabilitation.[24] According to parents in our study, clinicians caring for their child rarely inquire about sleep and lack knowledge to detect sleep problems. This is supported by studies that have shown significant gaps in both knowledge and clinical practices regarding pediatric sleep disorders among physicians, pediatricians and child neuropsychiatrists,[30–32] yet little is described in the context of pediatric rehabilitation. From the experiences shared by parents, several other reasons can be deduced why sleep problems go by undetected, and serve as important lessons for clinical practice. Firstly, parents indicated they were overloaded by care demands for their child. As a result, sleep may not be at the forefront of their minds and fail to receive priority on their extensive list of topics that need to be discussed with their doctor. Therefore, it is essential for parents that clinicians take the initiative and responsibility by regularly asking them about sleep. Secondly, parents indicated that caring for their child's sleep is paramount and they feel that providing care at night is something that is expected from them as parents. As a consequence, they may not recognise their child's sleep as problematic. This is supported by previous research showing discrepancies between parent-reported sleep problems using objective criteria versus their subjective perceptions, i.e. parents do not always perceive their child as having a sleep problem even when it is present.[33] When clinicians ask parents about sleep, they should take the extra step to dig a little deeper, in order to get a true understanding of what their nights look like. Objective, measurable questions like '*Does your child wake up more than three times per night?*' or '*Does is take longer than 30 minutes for your child to fall asleep?*' would serve as a good starting point to identify sleep difficulties.[34] Thirdly, parents expressed a need to be heard and understood in a supportive manner. Those who felt judged by their healthcare professionals described feelings of insecurity for not taking good care of their child at night, and may therefore be less inclined to raise concerns.[35] Therefore, professionals should acknowledge the sensitivity that surrounds the care for sleep of these children, taking on a coaching attitude towards parents without being judgmental. Considering these perspectives in clinical practice may prove beneficial in meeting parents' collectively expressed need for sleep to receive more awareness within pediatric healthcare.

Recently, International Classification of Functioning, Disability and Health (ICF) Core Sets for children and youth with CP were developed during a consensus meeting with experts.[36] Among these Core Sets, sleep was reported as one of the 25 functions considered most relevant for describing the child across all age groups between 0–18 years. The identified Core Sets serve to guide professionals in identifying areas of functioning that need to be addressed in this population, and offers support for the parental need to incorporate sleep into standard clinical assessment. In doing so, care for sleep should consider all different aspects captured within the ICF model, that may influence sleep, including body functions (e.g. spasticity, pain, airway obstructions, reflux), participation (e.g. fatigue or daytime sleepiness during school), as well as environmental (e.g. bedroom, parents) and personal factors (e.g. behaviour, sleep hygiene practices). Applying this framework when a child with CP and their family visit during clinical follow-up may guide professionals to avoid overlooking important aspects relevant for child-family functioning, including sleep.

A team of (non)medical specialists of various disciplines is often involved in the child's rehabilitation care, but parents emphasised not knowing who exactly is responsible for care for sleep. According to parents, sleep is intertwined in all aspects of their child's functioning, and should therefore be approached holistically with the combination of knowledge and expertise of all disciplines. Although CP is best managed in a multidisciplinary setting,[37] currently there is no standardised approach to sleep management within pediatric rehabilitation. Given the fact that the rehabilitation physician serves as an important gatekeeper in these settings, we propose that they are encouraged to adopt it as their responsibility to detect and monitor sleep problems, necessary to initiate multidisciplinary interventions.

In addition to needs described within the healthcare setting, an important parent-identified need raised within their home environment was the availability of social support. The influence of social support provided by immediate family, friends, and neighbours has been reported to indirectly affect the psychological health of parents of children with CP,[21] and should therefore not be forgotten. The sleep problems were often described by parents to be invisible to others around them. Hence, it can be challenging for their social environment to truly understand a situation that they are unaware of, cannot imagine, or simply do not recognise to be so tough. In fact, sleep disturbances in mothers with CP have been described to be comparable to those experienced by mothers caring for new-borns.[13] Although peer caregivers of healthy children may experience similar problems during childhood, their child will likely outgrow the sleep issues and should realise that parents of a child with CP may carry this burden for a lifetime. Parents should feel encouraged to openly discuss these issues with their social network, who in turn are expected to offer support and acknowledgement for being tired, irritable or forgetful during the day as a result of being sleep deprived or (psychologically) exhausted.

Our findings on how parental concerns and needs map across different environmental settings, could provide a framework for targeting home-based versus healthcare-based intervention strategies. Home-based approaches should take into consideration specific concerns, and focus on what can be done to meet parents' needs, in order to engage families in (implementing) intervention processes. For example, even establishing a good sleep hygiene, which is considered a simple but effective first-line treatment for sleep problems in children with neurodevelopmental disorders,[38] can be challenging in a complex child and family situation. Actively involving and educating parents to ensure that healthy sleep practices are implemented and maintained at home is therefore crucial.[39] More efforts should be made to adopt a family-centred approach in relation to the care for child sleep. Intervention strategies related to the clinical setting may in turn require educating healthcare professionals to improve their sleep knowledge, as well as adaptations on an organisational level in order for sleep to become a standard item for review during routine health assessments. Moreover, effective sleep assessment should be broad enough to screen for all kinds of sleep issues, yet focused enough to target specific individual parental concerns and needs.[40] This approach should result in healthcare-based interventions that are tailored to the unique and diverse family situations.

Strengths and limitations

It should be noted that the term 'sleep problems' in this article is solely based on parent reports (i.e. whether they perceive their child's sleep as problematic or not) rather than on clinically diagnosed sleep problems. Though one could argue whether perceiving or having a problem is similar, the aim of this paper was to expose perspectives on care for sleep from a parental viewpoint.

Although we did not specifically recruit parents of children who experienced sleep problems in their child, those parents who participated may have been more concerned with sleep issues. Yet, not all parents included in this study mentioned sleep problems, and some of them still described concerns and needs related to their child's care for sleep.

Four parents were reflecting on a past period when their child was aged 1–8 years rather than being interviewed about their current experiences. A potential recall bias of these parents could have impacted the data due to e.g. faulty memory. On the other hand, the contributions of parents of older children could have enriched the data by introducing different perspectives on the care for sleep of their child during the same age period, but viewed in retrospect.

Parents of non-ambulatory children with CP have been reported to express more needs than parents of ambulatory children.[41] It may well be that parents of the most severely impaired children experienced a greater burden of care demands at night, with a greater impact on family functioning and wellbeing. Concerns about sleep have been found to vary across ages in children and young people with CP, but are present at every GMFCS level.[42] Despite the use of a convenience sampling method, we managed to reach great variation in age and GMFCS levels between children. This qualitative study was not designed to investigate correlations, but rather to explore the issues that families of a child with CP face with regard to care for sleep, across all levels of motor disability.

Similar to previous literature conducted in this field,[13,21,39,40] our parent sample was dominated by (highly-educated) mothers. Since the responsibilities for caring for a child with a disability are attributed differently, i.e. based on gender,[43] mothers likely experience sleep problems differently. Future research should elucidate perspectives of fathers, as well as those of siblings and the children with CP themselves.

Conclusions

This qualitative study allowed us to hear the voices of parents by addressing their perspectives on the care for sleep of their child with CP. Parents face numerous challenges caring for their child's sleep which are opposed by perceived shortcomings in healthcare. In order to prevent poor health outcomes in families of children with CP, sleep needs to be acknowledged within the pediatric setting. Clinicians should routinely inquire about sleep during clinical follow-ups, in order to identify sleep problems early on. A proactive, non-judgmental, and family-centred approach is needed from healthcare professionals.

Acknowledgements

The authors would like to express their gratitude to the parents who shared their experiences with us, and parent organisations CP Nederland and OuderInzicht for their collaboration. We thank Barbara Uelkes and Nicole IJff for transcribing the interviews, Ian & Hilde Hall for their support in translating quotations, and Floryt van Wesel, expert in qualitative research, for her assistance in structuring categories and construction of themes during the analysis.

Declaration of interest

The authors report no conflicts of interest.

REFERENCES

1. Oskoui M, Coutinho F, Dykeman J, Jetté N, Pringsheim T. An update on the prevalence of cerebral palsy: a systematic review and meta-analysis. Dev Med Child Neurol. 2013;55(6):509–19.
2. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in Children With Cerebral Palsy: A Review. J Child Neurol. 2011;26(10):1303–10.
3. Newman CJ, O'Regan M, Hensey O. Sleep disorders in children with cerebral palsy. Dev Med Child Neurol. 2006;48(7):564–8.

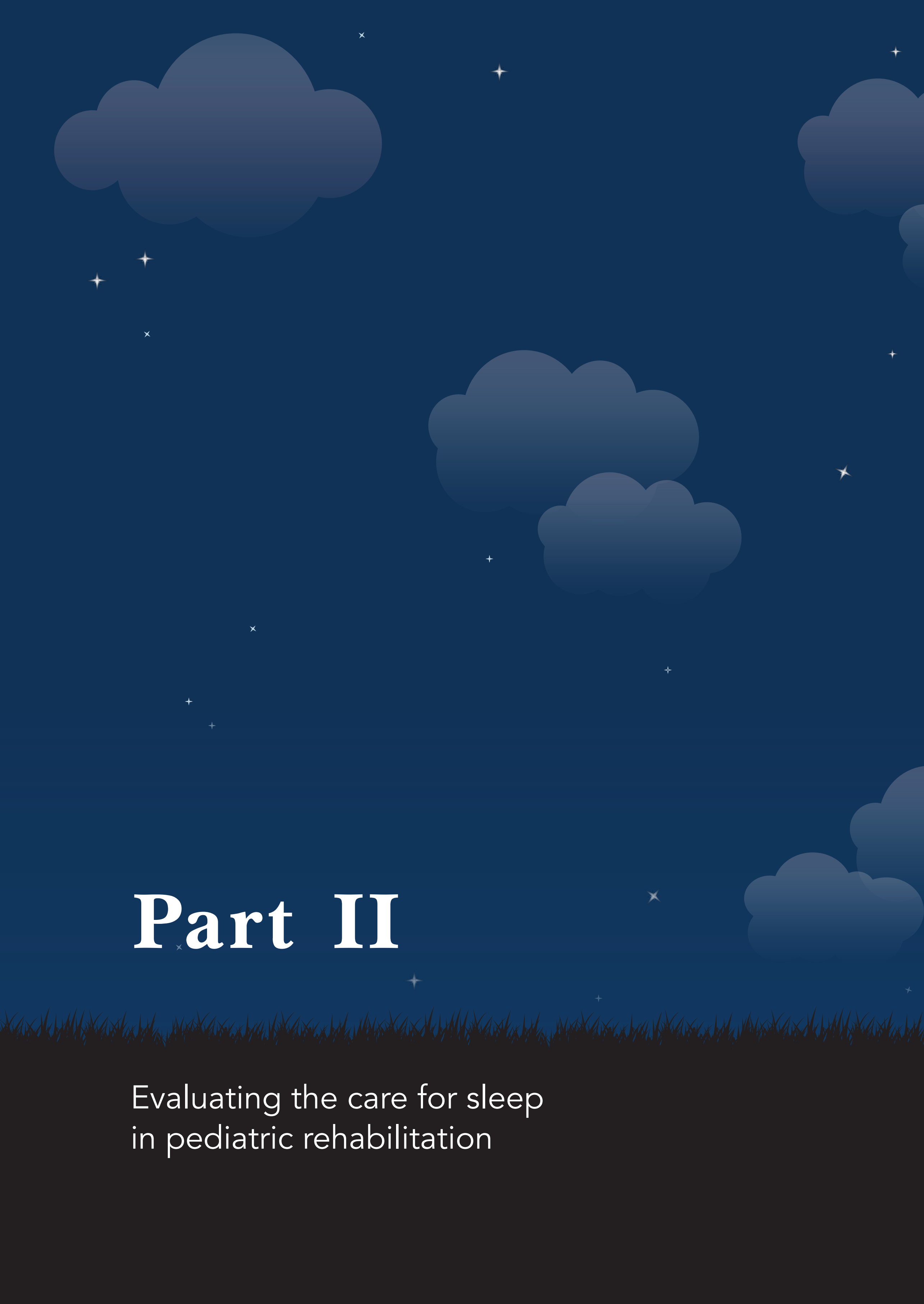
4. Horwood L, Mok E, Li P, Oskoui M, Shevell M, Constantin E. Prevalence of sleep problems and sleep-related characteristics in preschool- and school-aged children with cerebral palsy. Sleep Med. 2018;50:1–6.
5. Hemmingsson H, Stenhammar AM, Paulsson K. Sleep problems and the need for parental night-time attention in children with physical disabilities. Child Care Health Dev. 2009;35(1):89–95.
6. Dutt R, Roduta-Roberts M, Brown C. Sleep and Children with Cerebral Palsy: A Review of Current Evidence and Environmental Non-Pharmacological Interventions. Children. 2015;2(1):78–88.
7. 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.
8. Kotagal S, Gibbons VP, Stith JA. Sleep abnormalities in patients with severe cerebral palsy. Dev Med Child Neurol. 2008;36(4):304–11.
9. Garcia J, Wical B, Wical W, Schaffer L, Wical T, Wendorf H, et al. Obstructive sleep apnea in children with cerebral palsy and epilepsy. Dev Med Child Neurol. 2016;58(10).
10. Lélis ALPA, Cardoso MVLM, Hall WA. Sleep disorders in children with cerebral palsy: An integrative review. Sleep Med Rev. 2015;30:63–71.
11. 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):213–218.
12. Horwood L, Li P, Mok E, Oskoui M, Shevell M, Constantin E. Behavioral difficulties, sleep problems, and nighttime pain in children with cerebral palsy. Res Dev Disabil. 2019;95:103500.
13. 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.
14. Chaput J, Gray CE, Poitras VJ, Carson V, Gruber R, Olds T, et al. Systematic review of the relationships between sleep duration and Health Indicators in School-Aged Children and Youth. Appl Physiol Nutr Metab. 2016;41:266–282.
15. Meijer, Habekothé, Van Den Wittenboer. Time in bed, quality of sleep and school functioning of children. J Sleep Res. 2000;9(2):145–53.
16. Astill RG, Van der Heijden KB, Van IJzendoorn MH, Van Someren EJW. Sleep, cognition, and behavioral problems in school-age children: A century of research meta-analyzed. Psychol Bull. 2012;138(6):1109–38.
17. Horwood L, Li P, Mok E, Oskoui M, Shevell M, Constantin E. Health-related quality of life in Canadian children with cerebral palsy: what role does sleep play? Sleep Med. 2019;54:213–22.
18. Sandella DE, O'Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. Sleep Med. 2011;12(3):252–256.
19. Jan MMS. Cerebral palsy: comprehensive review and update. Ann Saudi Med. 2006;26(2):123–32.
20. 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):412–8.
21. Raina P, O'Donnell M, Rosenbaum P, Brehaut J, Walter SD, Russell D, et al. The health and well-being of caregivers of children with cerebral palsy. Pediatrics. 2005;115(6):e626-36.
22. Barlow JH, Cullen-Powell LA, Cheshire A. Psychological well-being among mothers of children with cerebral palsy. Early Child Dev Care. 2006;176(3–4):421–8.
23. Wright M, Tancredi A, Yundt B, Larin H. Sleep Issues in Children with Physical Disabilities and Their Families. Phys Occup Ther Pediatr. 2006;26(3):55–72.
24. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: An underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
25. Boeije H. Analysis in qualitative research. Sage publications; 2009.
26. Virginia Braun &, Victoria Clarke. Using thematic analysis in psychology. Qual Res Psychol. 2006;3(2006):77–101.
27. Quine L. Sleep problems in primary school children: comparison between mainstream and special school children. Child Care Health Dev. 2001 May 1;27(3):201–21.
28. Guillamón N, Nieto R, Pousada M, Redolar D, Muñoz E, Hernández E, et al. Quality of life and mental health among parents of children with cerebral palsy: The influence of self-efficacy and coping strategies. J Clin Nurs. 2013;22(11–12):1579–90.

29. Okurowska –Zawada B., Kułak W., Wojtkowski J., Sienkiewicz D. P-PG. Quality of life of parents of children with cerebral palsy. Prog Heal Sci. 2011;1(1):116–123.
30. Owens JA. The practice of pediatric sleep medicine: results of a community survey. Pediatrics. 2001;108(3):e51-e51.
31. 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 Heal. 2017;22(6):322–7.
32. Bruni O, Violani C, Luchetti A, Miano S, Verrillo E, Di Brina C, et al. The sleep knowledge of pediatricians and child neuropsychiatrists. Sleep Hypn. 2004;6(3):130–8.
33. Robinson AM, Richdale AL. Sleep problems in children with an intellectual disability: parental perceptions of sleep problems, and views of treatment effectiveness. Child Care Health Dev. 2004;30(2):139–50.
34. Moore M. Behavioral sleep problems in children and adolescents. J Clin Psychol Med Settings. 2012;19(1):77–83.
35. Norman C, Moffatt S, Rankin J. Young parents' views and experiences of interactions with health professionals. J Fam Plan Reprod Heal Care. 2016;42(3):179–86.
36. Schiariti V, Selb M, Cieza A, O'Donnell M. International Classification of Functioning, Disability and Health Core Sets for children and youth with cerebral palsy: a consensus meeting. Dev Med Child Neurol. 2015;57(2):149–58.
37. Panteliadis CP, editor. Cerebral Palsy: A Multidisciplinary Approach. Springer; 2018.
38. 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.
39. Bourke-Taylor H, Pallant JF, Law M, Howie L. Relationships between sleep disruptions, health and care responsibilities among mothers of school-aged children with disabilities. J Paediatr Child Health. 2013;49(9):775–82.
40. Unsal-Delialioglu S, Kaya K, Ozel S, Gorgulu G. Depression in mothers of children with cerebral palsy and related factors in Turkey: a controlled study. Int J Rehabil Res. 2009 Sep;32(3):199–204.
41. Palisano RJ, Almarsi N, Chiarello LA, Orlin MN, Bagley A, Maggs J. Family needs of parents of children and youth with cerebral palsy. Child Care Health Dev. 2010;36(1):85–92.
42. 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.
43. Traustadottir R. Mothers Who Care. J Fam Issues. 1991;12(2):211–28.

APPENDIX

Table 2.A Interview topic guide

Topic	Example questions
Current situation regarding child’s sleep (care)	• Description of average 24h day/night at home. • Possible challenges you/your family encounter(s) during the night, or subsequent day, related to your child’s sleep? - How do you/your family deal with them? - How does this affect you/your family? • Description of current care for sleep in child’s healthcare. - Do you discuss sleep with your healthcare professional? - Who takes initiatives? - Satisfied?
Perceptions of child’s sleep and care for sleep	• Do you believe your child sleeps well? Are you satisfied with his/her sleeping? - Explain, why, what not? • What do you find important aspects of your child’s sleep? - Why? Do you monitor those aspects? How? • Do you worry or have concerns about (elements of) your child’s sleep, or in the challenges you face caring for your child’s sleep? - In what way? Examples. • Does your child’s sleep affect you/your family? - In what way? Examples. - Sleep problems? What kinds of issues do you encounter, how do you deal with them? - Consequences for child, family functioning/wellbeing • What would help you/your family in providing better sleep care for your child? - Home/social environment - Healthcare setting • What expectations do you have, or what would you need from your clinician or healthcare center regarding care for sleep of your child? - Are those needs met? - What is missing, what would help you? - How can care for sleep be optimised?



Part II

Evaluating the care for sleep in pediatric rehabilitation

Chapter 3

Sleep health practices and sleep knowledge among healthcare professionals in Dutch pediatric rehabilitation

Raquel Y Hulst, Sigrid Pillen, Jeanine M Voorman, Neele Rave,
Johanna MA Visser-Meily, Olaf Verschuren

Child Care Health Dev. 2020;46(6):703–10. doi: 10.1111/cch.12799.

ABSTRACT

Background: Sleep disorders are highly prevalent in children with neurodisabilities, yet they seem under-recognised in pediatric rehabilitation settings. The aim of this study was to assess among two groups of healthcare professionals (HCPs) working in pediatric rehabilitation: 1) sleep health practices, and 2) knowledge about sleep physiology, sleep disorders, and sleep hygiene.

Methods: We performed a cross-sectional sleep survey among medical and non-medical HCPs, and the general population. Participants (30 rehabilitation physicians (RPs), 54 allied health professionals (AHPs) and 63 controls) received an anonymous 30-item survey consisting of three domains: 1) general information; 2) application of sleep health practices; 3) sleep knowledge.

Results: RPs address sleep issues more frequently in clinical practice than AHPs. Sleep interventions mostly consist of giving advice about healthy sleep practices, and are given by the majority of HCPs. While RPs demonstrated the highest scores on all knowledge domains, total sleep knowledge scores did not exceed 50% correct across groups, with AHPs and controls showing equal scores. Sleep hygiene rules closest to bedtime and related to the sleep environment were best known, whereas those related to daytime practices were rarely mentioned across all groups. A small minority of HCPs (RPs 20%; AHPs 15%) believed to possess sufficient sleep knowledge to address sleep in clinical practice. No association was found between self-perceived knowledge and sleep knowledge scores among HCPs.

Conclusions: Sleep should become a standard item for review during routine health assessments in pediatric rehabilitation settings. HCPs' limited exposure to sleep education may result in feelings of incompetence and inadequate sleep knowledge levels, affecting their sleep health practices. Appropriate sleep training programs should be implemented to empower HCPs with knowledge, skills, and confidence, needed to recognise and treat sleep disorders in children with neurodisabilities, as well as to be able to guide parents.

INTRODUCTION

Sleep disorders are common in children with neurodevelopmental disabilities (NDDs), with a reported prevalence as high as 86%.[1,2] In addition to affecting the children's physical and cognitive health and development,[3] sleep disorders may greatly impact on the wellbeing of both the children and their families.[4–6] Therefore, improving the quality of sleep in children with NDDs not only has the potential benefit of improving their clinical outcomes,[7] it can greatly ameliorate the quality of life of the entire family.[8]

In recent years, there has been growing acknowledgement of the importance of sleep and the need for recognition of sleep problems by physicians.[9] Yet, there continues to be only limited education in sleep medicine across medical school curricula.[8,10] The minimal training received is accompanied by shortcomings in confidence and sleep knowledge, all of which may contribute to sleep issues not being inquired about routinely when children are seen.[11,12]

For children receiving developmental or rehabilitative services, like those with NDDs, pediatric rehabilitation provides an ideal place to address their sleep health as part of the routine assessments. The multidisciplinary team of healthcare professionals (HCPs) that is typically involved in the rehabilitative care of these children, thereby serves a joint role in both detecting and managing sleep problems. Hence it is crucial that (non-)medical HCPs working in pediatric rehabilitation settings are not only aware of the importance of sleep, but also possess current knowledge of basic sleep physiology, can recognise symptoms of common pediatric sleep disorders, and are familiar with good sleep hygiene practices.

However, despite the high prevalence of sleep disorders in children with NDDs, according to parents sleep has received only limited attention in pediatric rehabilitation.[13] Indeed, sleep problems are not always appropriately addressed in these populations,[2,14] leaving sleep an underemphasised aspect of health in neurorehabilitation.[15] This raises the question whether HCPs have sufficient knowledge and competence to address sleep issues in clinical practice. Therefore, this survey study aimed to assess the: 1) sleep health practices, and 2) sleep knowledge (sleep physiology, sleep disorders, and sleep hygiene) in two groups of HCPs (i.e. medical and non-medical professionals) working within pediatric rehabilitation settings. To effectively guide parents, HCPs are required to have more sleep knowledge than the general population, and therefore a control group was added to allow comparisons.

METHODS

Study design

A cross-sectional quantitative survey study was conducted. The study was deemed exempt from review under the Dutch Medical Research Involving Human Subjects Act by the Medical Ethics Research Committee of the University Medical Centre Utrecht, the Netherlands (file number 19-066).

Respondents

HCPs from three pediatric rehabilitation settings (rehabilitation centre, school for special education, rehabilitation department of a children's hospital) in the Netherlands participated in this study. In Dutch rehabilitation, a medically schooled physician serves a gatekeeping role in detecting child-related problems during clinical encounters, and consequently coordinates the child's rehabilitative care. When, in this case, a problem with sleep of the child is detected, the physician gives first-line treatment advice or can decide to set up referral to a non-medical professional or sleep clinic for subsequent sleep assessments and/or interventions. Depending on the nature of the sleep problem, a non-medical professional may further assess the child's sleep, bed routine and/or behaviour, and perform therapies like behavioural interventions and implementing healthy sleep practices. In this way, the roles of HCPs involved in sleep care in Dutch rehabilitation settings are distinct. The pen-and-paper surveys were distributed during live meetings among the following two groups of pediatric HCPs:

1. rehabilitation physicians (RPs). This group included pediatric rehabilitation physicians, physician assistants, pediatricians, and doctors in specialist training to become RP.
2. allied health professionals (AHPs). This group included physical therapists, occupational therapists, developmental behavioural therapists, speech and language therapists, social workers, and psychologists.

An additional control group was drawn from the general population, comprising individuals without a background or current profession in healthcare, to allow comparisons of sleep knowledge levels. The control group consisted mostly of parents of (young) children, who were recruited via the social networks of colleagues and acquaintances, and filled out the pen-and-paper surveys during face-to-face encounters.

Data collection

A 30-item structured questionnaire was designed based on relevant literature and consultations with experts (researchers and clinicians) working in the field of pediatric rehabilitation and sleep medicine (see online Supporting Information). Pilot testing was conducted to ensure that the questionnaire was easily understood and could be completed within a short time window (15–20 minutes). The questionnaire comprised three sections:

1. general information including age, sex, profession, educational level, and hours of sleep education received;

2. application of sleep health practices in daily clinical practice. HCPs were asked to indicate how often they address the topic sleep during clinical encounters, choosing between *never/seldom* (less than once per month), *sometimes* (1–3 times per month) or *often* (once per week or more often). HCPs who reported to address sleep *sometimes* or *often* in clinical practice, were asked to indicate the type(s) of sleep interventions or therapies they apply.

3. sleep knowledge within the domains of a) basic sleep physiology (i.e. recommended sleep durations, sleep architecture), b) symptoms and characteristics of common pediatric sleep disorders, and c) sleep hygiene rules (i.e. healthy sleep practices). Additionally, the participants' self-perceived knowledge sufficiency was assessed.

Apart from the general information, responses were measured using multiple choice questions, and included a "*don't know*" answer option. An open-ended question was used for collecting information about knowledge of sleep hygiene rules; respondents were asked to name three sleep hygiene rules other than the example given regarding limiting screen time two hours before bedtime. A cover letter which explained the aim of the study, and emphasised the need for honest responses (i.e. to answer with "*don't know*" instead of guessing if one does not know) was attached to the survey.

Data analysis

Surveys that returned largely incomplete (>25%) were excluded from analysis. Data were analysed using IBM SPSS Statistics 26. Sleep knowledge scores (i.e. number of correctly answered questions) were calculated for each group and converted into percentages; these are presented as mean ± standard deviation scores (%) for all knowledge questions in total, and per domain sleep physiology and sleep disorders. After testing for normality, means were compared using Kruskal-Wallis test followed by post-hoc Mann-Whitney tests. Answers to the open-ended question regarding sleep hygiene rules were categorised according to those presented by the National Sleep Foundation,[16] and relative frequencies were calculated per group. Categorical variables (i.e. sleep education, sleep health practices, and knowledge about sleep hygiene rules) are displayed as percentage frequency distributions. To determine the relationship between categorical data, relative frequencies were compared using Fisher's exact tests. The critical value for significance was set at .05, and a correction for multiple comparisons was applied during post-hoc analyses.

RESULTS

Respondents' general information

In total, 84 HCPs completed the survey. Based on their profession, HCPs were divided between the RP group (*n*=30) and the AHP group (*n*=54). An additional control group (*n*=63) completed the survey questions with exception of the section regarding sleep health practices. The majority of all respondents were female, and between the age of 31–50 years. Across all groups, over 75% indicated to have received less than 5 hours of sleep education throughout their entire school curricula (RPs 75.9%; AHPs 90.6%; controls 88.5%), and this amount was independent of group (*p*>.05, Fisher's exact test). Group characteristics are summarised in Table 3.1.

Table 3.1 Group characteristics

	Rehabilitation physicians [a] *n*=30 % (*n*)	Allied health professionals [b] *n*=54 % (*n*)	Control group *n*=63 % (*n*)
Sex			
Female	76.7 (23)	92.6 (50)	73.0 (46)
Age			
20–30 years	33.3 (10)	13.0 (7)	31.8 (20)
31–40 years	40.0 (12)	33.3 (18)	23.8 (15)
41–50 years	16.7 (5)	24.1 (13)	27.0 (17)
51–60 years	10.0 (3)	24.1 (13)	14.3 (9)
61–70 years	0 (0)	5.6 (3)	3.2 (2)
Level of education			
Secondary vocational	0 (0)	0 (0)	15.9 (10)
Higher professional	3.3 (1)	77.8 (42)	20.6 (13)
University or higher	97.7 (29)	22.2 (12)	63.5 (40)

[a] This group consisted of pediatric rehabilitation physicians (*n*=9), doctors in specialist training to become RP (*n*=19), physician assistant (*n*=1), pediatrician (*n*=1).

[b] This group consisted of physical therapists (*n*=17), occupational therapists (*n*=15), developmental behavioural therapists (*n*=8), speech and language therapists (*n*=7), social workers (*n*=4), psychologists (*n*=3).

Sleep health practices

RPs (*often* 56.7%, *n*=17; *sometimes* 40%, *n*=12; *never/seldom* 3.3%, *n*=1) reported to more frequently address sleep issues than AHPs (*often* 11.8%, *n*=6; *sometimes* 51%, *n*=26; *never/seldom* 37.2%, *n*=19), a difference found to be statistically significant (*p*<.001*, Fisher's exact test).

Sleep interventions

Those who reported to *sometimes* or *often* address sleep, indicated the types of sleep interventions they apply (Figure 3.1). The majority of both HCPs groups (RPs 97%, *n*=29; AHPs 79%, *n*=38) reported to give advice about sleep hygiene rules. RPs more often mentioned giving advice compared to AHPs (*p*<.05*, Fisher's exact test). Half of RPs (50%, *n*=15) indicated to prescribe medication (including melatonin) compared to 6% (*n*=3) of AHPs (*p*<.001*, Fisher's exact test). One-third of RPs (33%, *n*=10) indicated to refer to a sleep clinic, compared to 6% (*n*=3) of AHPs (*p*<.01*, Fisher's exact test). Less than a quarter of HCPs (RPs 23%, *n*=7; AHPs 13%, *n*=6) reported to perform behavioural therapy to treat sleep problems (*p*>.05, Fisher's exact test). None of the HCPs reported to perform light therapy.

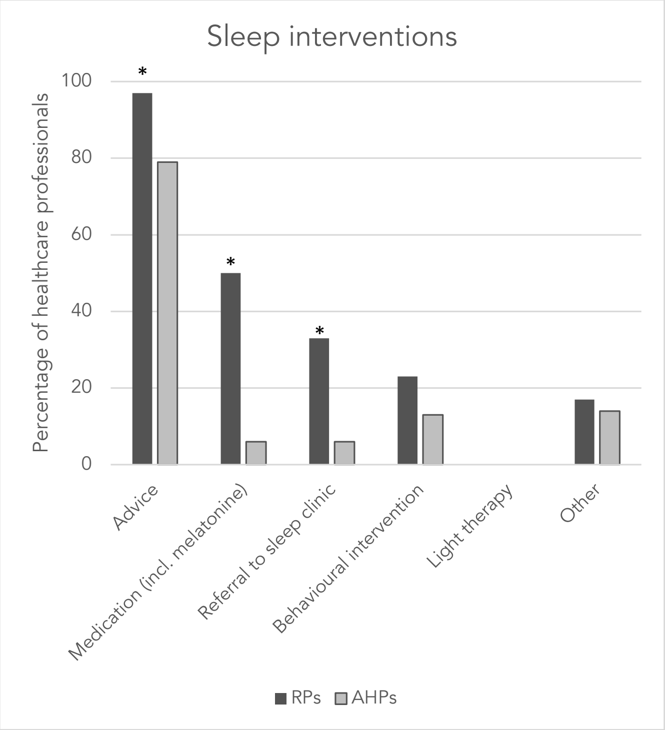


Figure 3.1 Types of sleep interventions applied by RPs (*n*=30) and AHPs (*n*=54).
* Denotes *p*<.05 (Fisher's exact test); RPs: rehabilitation physicians; AHPs: allied health professionals.

Sleep knowledge

Sleep physiology & sleep disorders

Table 3.2 shows the sleep knowledge scores (correct answer rates) across groups. Mean total sleep knowledge scores were found to be statistically different across groups (H(2) = 24.322, $p<.001$*), with RPs demonstrating significantly higher scores than AHPs ($p<.001$*) and controls ($p<.001$*), whereas no difference was found between AHPs and controls ($p>.05$).

To allow subgroup analyses between different domains of sleep knowledge, total sleep knowledge scores were divided between questions covering the domains sleep physiology and sleep disorders. All three groups demonstrated lower scores on questions about symptoms and characteristics of sleep disorders, compared to questions related to basic sleep physiology (Table 3.2). Subgroup analysis revealed different scores across groups on both domains, with RPs scoring significantly higher than AHPs (sleep physiology $p<.001$*; sleep disorders $p<.001$*) and controls (sleep physiology $p=.002$*; sleep disorders $p<.001$*). Controls showed higher scores than AHPs within the domain sleep physiology, but this difference failed to reach statistical significance after correcting for alpha ($p>.05$). Within the domain sleep disorders, AHPs demonstrated significantly higher scores than controls ($p<.05$*).

Table 3.2 Sleep knowledge and self-perceived sleep knowledge scores

	Rehabilitation physicians *n*=30 mean±SD	**Allied health professionals** *n*=54 mean±SD	**Control group** *n*=63 mean±SD
Sleep knowledge scores (%)			
Total sleep knowledge	48.9±12.4*	35.4±10.9	34.5±13.3
Domain sleep physiology	53.3±12.6*	37.4±15.9	43.0±16.3
Domain sleep disorders	41.3±17.1*	26.6±15.9#	19.4±17.8
Self-perceived sleep knowledge scores (%)			
Total sleep knowledge score in response to the question: *"Do you believe you have sufficient sleep knowledge to address sleep issues in clinical practice?"*			
Yes	54.76±9.87 (*n*=6)	34.52±7.94 (*n*=8)	N/A
No	50.40±14.7 (*n*=12)	33.75±10.14 (*n*=23)	N/A
Don't know	44.44±10.02 (*n*=12)	35.15±10.70 (*n*=21)	N/A

* Statistically significant ($p<.05$) compared to allied health professionals and control group.
# Statistically significant ($p<.05$) compared to control group.

Sleep hygiene

The frequency of sleep hygiene rules mentioned by RPs, AHPs, and controls is shown in Figure 3.2. RPs were able to more frequently recall the majority of sleep hygiene rules. Significant differences were observed between groups for *establishing a relaxing bedtime routine* ($p<.05$*, Fisher's exact test) and *establishing a regular sleep schedule* ($p<.05$*, Fisher's exact test), with these rules being mentioned more often by RPs and AHPs compared to controls, respectively. The sleep hygiene rules closest to bedtime and related to the sleep environment were best known across groups, whereas those related to daytime practices (i.e. *promoting physical exercise during the day*, *exposure to natural light*, and *limiting daytime naps*) were rarely mentioned across all groups.

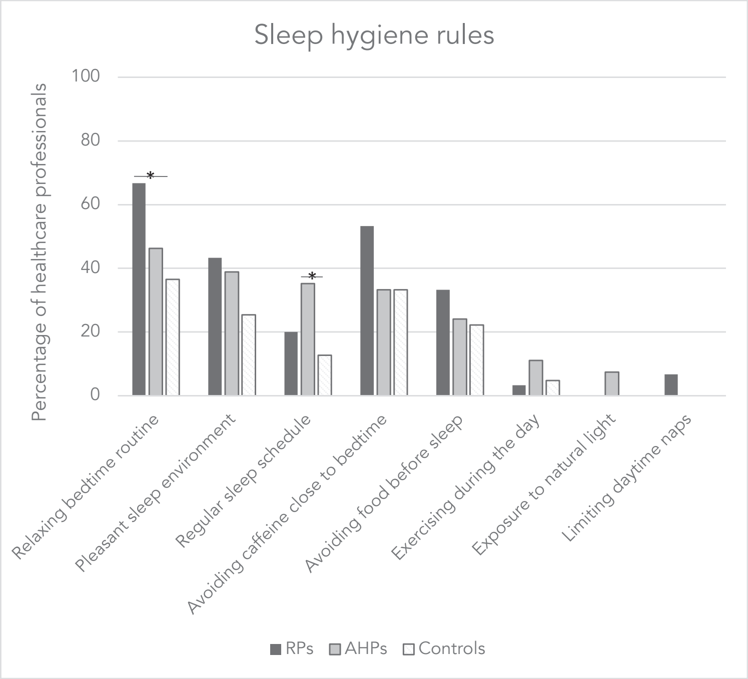


Figure 3.2 Frequency of sleep hygiene rules mentioned by RPs (n=30), AHPs (n=54) and controls (n=63), in addition to the example rule regarding limiting screen time two hours before bedtime. * Denotes $p<.05$ (Fisher's exact test); RPs: rehabilitation physicians; AHPs: allied health professionals.

Self-perceived sleep knowledge

The minority of all HCPs (RPs 20%, n=6; AHPs 14.8%, n=8) reported to believe that they possess sufficient sleep knowledge to address sleep problems in daily clinical practice. In contrast, most HCPS reported either to not have sufficient knowledge (RPs 40%, n=12; AHPs 42.6%, n=23) or that they "*don't know*" (RPs 40%, n=12; AHPs 38.9%, n=21) (Table 3.2).

No difference was found in total sleep knowledge scores between HCPs who believed they had sufficient knowledge about sleep versus those who believed they did not have sufficient sleep knowledge (RPs: t(16) = .652, *p*>.05; AHPs: t(29) = .196, *p*>.05).

DISCUSSION

This study assessed the sleep health practices and knowledge about sleep physiology, sleep disorders, and sleep hygiene among two groups of HCPs in Dutch pediatric rehabilitation. The frequency of sleep being addressed during clinical encounters varies greatly between medical and non-medical HCPs, and more efforts should be made for sleep assessments to become a standard item for surveillance during routine healthcare practices. Although RPs showed higher sleep knowledge scores than AHPs, neither group exceeded 50% correct scores, suggesting limited sleep knowledge, particularly in the area of sleep disorders. We also noticed limited familiarity with healthy sleep behaviours that can be practiced during the day. Our findings emphasise the need to educate and empower HCPs with sound knowledge, skills and confidence required to address sleep problems in children with NDDs, and to support their parents.

There are several strengths and limitations to this study that should be considered. The questionnaire used was developed in co-creation with researchers and clinicians in the field of pediatric rehabilitation and sleep medicine, based on currently available literature in these fields, and trialled before use, yet it should be noted that it has not been validated. HCPs may have felt obliged to respond favourably to questions concerning their sleep health practices. Also, the relatively small sample size may limit the generalisability of our findings. Unlike other survey studies on sleep knowledge levels, we did include a control group to allow comparisons of HCP scores to the general population.

In line with our results, survey studies of practicing physicians and pediatricians have consistently found poor knowledge about sleep, and significant gaps in clinical practices regarding pediatric sleep disorders.[17–21] For example, Bruni et al. found low scores in all areas of sleep knowledge and particularly in sleep disorders among pediatricians and child neuropsychiatrists.[19] Papp and colleagues reported an average knowledge of 34% among primary care physicians, with only 10% rating themselves as *good*.[21] Similarly, we found the self-perceived knowledge among pediatric rehabilitation professionals to be low, i.e. only one in five RPs, and one in seven AHPs rated their own sleep knowledge to be sufficient. Without proper training and experience, HCPs may lack confidence or feel incompetent to address sleep problems properly, resulting in sleep problems left unaddressed and untreated.[10] As more HCPs acquire a greater awareness of sleep, more consistent processes for screening and assessment can be developed across pediatric rehabilitation settings.

Given that parental knowledge about children's sleep is typically poor,[22] they should be provided with appropriate information and advices to ensure that healthy sleep practices are implemented and maintained at home.[23,24] Nearly all HCPs reported to give such advices on a frequent basis, yet their knowledge deficits are indicated by equally low total sleep knowledge scores as our control sample, which consisted mostly of parents. A recent study on sleep problems and solution seeking for children with cerebral palsy and their parents, reported that out of the 63 parents that asked for professional help with their child's sleep, only 21 reported that their request for help led to effective treatment or advice from their HCP.[25] In addition, we found HCPs' familiarity with sleep hygiene rules to be confined to those closest to bedtime and related to the sleep environment, whereas they appeared unfamiliar to daytime practices (like exposure to daylight, adequate exercise, and limiting daytime naps). This knowledge gap is worrisome since sleep hygiene is considered the first line of treatment for sleep problems in children with NDDs.[26]

A review of the lifestyle practices that contribute to good quality sleep, can be valuable in providing a starting point to improve sleep, and more broadly in adopting healthy and protective lifestyles. In fact, promoting the entire triad of healthy behaviours, which in addition to physical activity and nutrition, also includes sleep itself, has recently been described as '*the formula for health and wellbeing*' in vulnerable patient populations with neurodevelopmental[27] and neuropsychiatric disorders.[28] Clearly, and in alignment with the global medical trend towards preventive healthcare,[29] the need to protect, promote and maintain healthy sleep as part of a healthy lifestyle, especially in children with NDDs, is evident. But if we want doctors to preach a healthy lifestyle to their patients (and families), expect them to detect and prevent sleep problems early on, and require them to effectively guide parents, obviously they need to be adequately equipped with proper training before they enter the clinic.

Unfortunately, there is only very limited coverage of sleep education in medical schools, which has previously been identified in 409 medical schools across twelve countries,[9] and appears to persist. Consistent with their findings from nearly a decade ago, we found that the majority of Dutch physicians received less than 5 hours of sleep education, similar to non-medical professionals and controls. This alarmingly low number may in turn explain their limited sleep knowledge and feelings of incompetence to address sleep in clinical practice. Indeed, limited exposure to sleep education can predict medical trainees' confidence and knowledge levels, thereby forecasting their future clinical practices regarding sleep health.[30]

This advocates the continued need for sleep medicine education to be fully incorporated into medical school curricula.[31]

Taking into account the confines of an already packed medical curriculum, additional educational efforts like postgraduate training, clinical opportunities, and other sleep education tools for current HCPs are warranted. It has been shown that sleep knowledge can be successfully increased through provision of sleep education, both when delivered face-to-face and through online webinars.[32] Our results showed that the knowledge scores of HCPs who believed to possess sufficient sleep knowledge, did not differ from those who believed they lacked sleep knowledge, demonstrating the importance to undertake such sleep training programs regardless of self-perceived knowledge. The goal of educational sleep trainings should not be to turn HCPs into sleep experts, but rather to provide them with the knowledge required to recognise symptoms of major pediatric sleep disorders by asking the right questions, to give the right general advice regarding good sleep hygiene, and to enable them to know when to refer to a (sleep) specialist for further assessment or to initiate sleep treatment strategies.[33]

Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of this article.

Key messages

- Sleep problems are highly prevalent among children with neurodevelopmental disabilities (NDDs), yet they are under-reported, under-recognised, under-diagnosed, and often untreated.
- Limited exposure to sleep education likely contributes to low levels of sleep knowledge, and confidence to guide patients, among healthcare professionals.
- There is a need to educate and empower healthcare professionals with knowledge and skills that are required to address sleep problems in children with NDDs and their families.
- Sleep should become a standard item for review during routine health assessments in pediatric rehabilitation settings.

REFERENCES

1. Robinson-Shelton A, Malow BA. Sleep Disturbances in Neurodevelopmental Disorders. Current Psychiatry Reports. 2016;18(6):1–8.
2. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy: A review. J Child Neurol. 2011;26(10):1303–10.
3. Turnbull K, Reid GJ, Morton JB. Behavioral Sleep Problems and their Potential Impact on Developing Executive Function in Children. Sleep. 2013;36(7):1077–84.
4. 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.
5. Sandella DE, O'Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. Sleep Med. 2011;12(3): 252–256.
6. 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):412–8.
7. Owens J. Classification and Epidemiology of Childhood Sleep Disorders. Prim Care - Clin Off Pract. 2008;35(3):533–46.
8. McDonald A, Joseph D. Paediatric neurodisability and sleep disorders: clinical pathways and management strategies. BMJ Paediatr Open. 2019;3(1).
9. Perry GS, Patil SP, Presley-Cantrell LR. Raising Awareness of Sleep as a Healthy Behavior. Prev Chronic Dis. 2013;10(8):E133.
10. Mindell JA, Bartle A, Wahab NA, Ahn Y, Ramamurthy MB, Huong HTD, et al. Sleep education in medical school curriculum: A glimpse across countries. Sleep Med. 2011;12(9):928–31.
11. Honaker SM, Meltzer LJ. Sleep in pediatric primary care: A review of the literature. Sleep Med Rev. 2016;25:31–9.
12. Chervin RD, Archbold KH, Panahi P, Pituch KJ. Sleep problems seldom addressed at two general pediatric clinics. Pediatrics. 2001;107(6):1375–80.
13. Hulst RY, Voorman JM, Pillen S, Ketelaar M, Visser-Meily JMA, Verschuren O. Parental perspectives on care for sleep in children with cerebral palsy: a wake-up call. Disabil Rehabil. 2020:1–10.
14. Didden R, Korzilius H, van Aperlo B, Overloop C van, de Vries M. Sleep problems and daytime problem behaviours in children with intellectual disability. J Intellect Disabil Res. 2002;46(7):537–47.
15. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: An underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
16. National Sleep Foundation. What is sleep hygiene? 2019. Retrieved from https://www.sleepfoundation.org/articles/sleep-hygiene
17. Owens JA. The practice of pediatric sleep medicine: results of a community survey. Pediatrics. 2001;108(3):E51.
18. 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 Heal. 2017;22(6):322–7.
19. Bruni O, Violani C, Luchetti A, Miano S, Verrillo E, Di Brina C, et al. The sleep knowledge of pediatricians and child neuropsychiatrists. Sleep Hypn. 2004;6(3):130–8.
20. BaHammam AS. Knowledge and attitude of primary health care physicians towards sleep disorders. Saudi Med J. 2000;21(12):1164–7.
21. Papp KK, Penrod CE, Strohl KP. Knowledge and attitudes of primary care physicians toward sleep and sleep disorders. Sleep Breath. 2002;6(3):103–9.
22. McDowall PS, Galland BC, Campbell AJ, Elder DE. Parent knowledge of children's sleep: A systematic review. Sleep Med Rev. 2017;31:39–47.
23. Mindell JA, Owens JA. Sleep problems in pediatric practice: Clinical issues for the pediatric nurse practitioner. J Pediatr Heal Care. 2003;17(6):324–31.
24. Blackmer AB, Feinstein JA. Management of Sleep Disorders in Children With Neurodevelopmental Disorders: A Review. Pharmacother J Hum Pharmacol Drug Ther. 2016;36(1):84–98.

25. Petersen S, Francis KL, Newall F, Reddihough DS, Lima S, Harvey A. Sleep problems and solution seeking for children with cerebral palsy and their parents. J Paediatr Child Health. 2020; 56(7):1108–13.
26. Jan JE, Owens JA, Weiss MD, Johnson KP, Wasdell MB, Freeman RD, et al. Sleep hygiene for children with neurodevelopmental disabilities. Pediatrics. 2008;122:1343–50.
27. 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. Dev Med Child Neurol. 2016;58:989–90.
28. Briguglio M, Vitale JA, Galentino R, Banfi G, Dina CZ, Bona A, et al. Healthy eating, physical activity, and sleep hygiene (HEPAS) as the winning triad for sustaining physical and mental health in patients at risk for or with neuropsychiatric disorders: Considerations for clinical practice. Neuropsychiatr Dis Treat. 2020;16:55–70.
29. Egger, G., Binns, A., Rossner, S., Sagner M. Lifestyle Medicine: Lifestyle, the environment and preventive medicine in health and disease. 3rd ed. Academic Press; 2017.
30. Salas RE, Gamaldo A, Collop NA, Gulyani S, Hsu M, David PM, et al. A step out of the dark: Improving the sleep medicine knowledge of trainees. Sleep Med. 2013;14(1):105–8.
31. Salas RME, Strowd RE, Ali I, Soni M, Schneider L, Safdieh J, et al. Incorporating sleep medicine content into medical school through neuroscience core curricula. Neurology. 2018;91(13):597–610.
32. Osborne JM, Blunden S. Evaluating Accessible Sleep Health Information in Rural and Urban Contexts: Delivery Face-to-Face or Online? Clin Med Insights Pediatr. 2018;12:117955651881516.
33. Luginbuehl M, Kohler WC. Screening and Evaluation of Sleep Disorders in Children and Adolescents. Child Adolesc Psychiatr Clin N Am. 2009;18:825–38.

Part III

Integrating sleep into care
for children with cerebral palsy
using a 24-hour activity approach

Chapter 4

24-hour activity for children with cerebral palsy: A clinical practice guide

Olaf Verschuren, Raquel Y Hulst, Jeanine Voorman, Sigrid Pillen,
Nicole Luitwieler, Jeroen Dudink, Jan Willem Gorter

Dev Med Child Neurol. 2021;63(1):54–9. doi: 10.1111/dmcn.14654.

ABSTRACT

The association between physical activity and health has been clearly established, and the promotion of physical activity should be viewed as a cost-effective approach that is universally prescribed as a first-line treatment for nearly every chronic disease. Health care providers involved in the care for individuals with cerebral palsy (CP) are encouraged to take an active role in promoting their health and well-being. Balancing activity behaviours across the whole day, with improved physical activity, reduced sedentary time, and healthy sleep behaviours, can set up infants, preschool, and school-aged children with CP for a healthy trajectory across their lifetime. However, most clinicians do not apply a systematic surveillance, assessment, and management approach to detect problems with physical activity or sleep in children with CP. Consequently, many children with CP miss out on an important first line of treatment. This article presents an evidence-informed clinical practice guide with practical pointers to help practitioners in detecting 24-hour activity problems as a critical step towards adoption of healthy lifestyle behaviours for children with CP that provide long-term health benefits.

INTRODUCTION

During childhood the body's physical and cognitive development are rapid, and the attitudes and habits formed at an early age can set the stage for the child's health in later life. Therefore, promoting an active lifestyle early on is key for optimal development and includes engaging in physical activity as well as minimizing sedentary behaviour. Owing to several factors, children with cerebral palsy (CP) usually have low levels of physical activity and spend prolonged times sitting.[1] Given the high prevalence of inactive lifestyles and the concomitant risk of chronic health conditions in adults with CP, it is no surprise that recently published guidelines for children with CP are informed by the health implications of physical activity and sedentary behaviour.[1]

However, these recommendations might not be enough to optimize health and development. For typically developing children, there has been a recent shift towards healthy activity throughout the whole day. Referred to as the 24-hour movement guidelines, the recommendations acknowledge that the entire activity continuum should be considered, which, in addition to physical activity and sedentary behaviour, also includes sleep.[2] A child's 24-hour day can be spent in several activity domains. This means that a day could be divided into time spent sleeping, being sedentary, or being physically active at different intensities, while there is never a time when a child is not engaging in one of these activities. In other words, if time spent in one activity is increased, there is less time available in the day for the remaining activity domains. For example, 1 hour of sleep may be lost to compensate for an extra hour of sedentary behaviour, or, alternatively, for an extra hour of physical activity. In both examples the hour of sleep has been exchanged for another activity domain by exactly the same duration, but the two scenarios are likely to be quite different in their health implications. What may sound as a small shift in behaviour could have a significant long-term impact on a child's health trajectory; hence, the importance in considering the whole-day activity pattern for children.

Emerging literature demonstrates a positive association between meeting the recommended hours of sleep and positive (brain) health outcomes in typically developing children, including global cognition.[3] Healthy sleep seems especially important for more vulnerable children, yet the prevalence of sleep problems in children with CP is high.[4] Following 'the whole day matters' philosophy, we need to adopt a proactive approach to ensure adequate hours of uninterrupted sleep in children with CP. The relation between physical activity and sleep is probably bidirectional where both acute and regular physical activity can improve sleep,[5,6] and sleep duration and quality may likewise influence physical activity behaviour.[7] Getting adequate sleep helps typically developing children to feel energized in the morning and participate fully in their school day and after-school activities. It allows them to be creative, play sports, socialize with friends, and be active during the day - which in turn helps children to sleep better. Exercise-based interventions promote sleep efficiency and duration in the general population regardless of the mode and intensity of the activity,[8] yet the effects of such interventions in children with CP may be less predictable. Although sleep-time recommendations will probably be beneficial for all children with CP, they seem especially promising for those who are classified in Gross Motor Function Classification System (GMFCS) levels IV and V, who have limited capabilities and opportunities to be active in the moderate to vigorous activity spectrum of the movement continuum. Optimizing their sleep quality and quantity might be an effective intervention for lifetime health promotion, in particular when combined with the physical activity recommendations.

In summary, a balanced interplay between optimal yet feasible proportions of activity behaviours throughout the day (i.e. limited sitting, increased physical activity, and enough sleep) is recommended for a healthy life for all children; hence, the importance of adopting the 24-hour activity guideline approach for children with CP. Key to promoting physical activity in the clinical setting is the use of tools in which every child's 24-hour habits are routinely assessed and recorded in their medical record. Those not meeting the recommendations, and their families, should be supported to optimize their 24-hour activity levels and sleep pattern.

THE ROLE OF PEDIATRIC (RE)HABILITATION

The time is now to open a window of opportunity for 24-hour activity assessment to be integrated into routine health care practice for children with CP. Pediatric physiatrists, (developmental) pediatricians, and other clinicians involved in the care of children with CP are particularly well-positioned to address physical inactivity and sleep problems, because they often have an established relationship and routine with children with CP and their parents or primary caregivers. By incorporating physical activity across the 24-hour activity continuum as a recognized vital sign (similar to height and weight), it can be documented and tracked routinely over time, facilitating meaningful and personalized counselling initiatives.

Surveillance of sleep and physical activity problems

Although inquiring about sleep is widely advised in pediatric health care,[9] sleep problems are still under-reported and under-recognized in children with CP.[10] A recent qualitative study among parents of children with CP found that health care professionals rarely ask them about sleep issues during clinical encounters, and parents expressed a strong wish for sleep to receive more attention in pediatric rehabilitation settings.[11] In addition, it is known that the physical inactivity and sedentary behaviour levels of children with CP are high and that these may require further attention to preserve and enhance their health and well-being/development.[1]

Therefore, implementation of an assessment tool within a routine follow-up or monitoring clinic is a good first option for clinicians interested in 24-hour activity. We advocate a brief checklist with useful questions for discussing physical activity and sleep in children with CP. This checklist needs to be practical and feasible to implement in clinical practice. For a recent innovation project, we have developed a 24-hour activity checklist, specifically for children with CP. This tool includes questions carefully designed to determine whether a more thorough evaluation for a particular problem is warranted. Parents can complete the checklist at home before their appointment with the health care provider.

DEVELOPMENT OF THE 24-HOUR ACTIVITY CHECKLIST

For surveillance, clinicians need to be able to ask the right questions to recognize the signs of pediatric physical activity and sleep problems. Questions must be more specific than quickly asking parents whether their child has trouble being physically active or sleeping. For example, more than half of parents who have a child with a sleep disorder will deny such problems when asked directly using a general question only,[12] whereas others may be unaware of what is normal or problematic sleep.[9]

For the questions related to sleep, a detailed description of the steps required for the development of pediatric sleep questionnaires was outlined by Spruyt and Gozal.[13] The initial steps are important in providing evidence of content validity, namely the extent to which the questionnaire measures the intended construct and is appropriate for its intended use. Our 24-hour activity checklist assesses the occurrence of different types of physical activity and sleep problems that are not necessarily correlated (i.e. a formative model); therefore, analysis about structural validity and internal consistency is not relevant and subsequently not reported for this checklist.[14]

Exploratory interviews

Before the development of the checklist, 18 parents of children with CP (GMFCS levels I–V, aged 2–15y) were interviewed to explore their current situation, concerns, and needs about the care for sleep,[11] and physical activity of their child, to gain an initial understanding of the areas within the 24-hour activity spectrum that warrant attention from a parental perspective.

Item generation and content review

For children with CP, Capio et al.[15] have systematically reviewed the literature on physical activity assessment measures, including questionnaires. Unfortunately, the current physical activity measurement tools are not practical for implementation in routine care. Specifically, for children with CP, the feasibility of existing questionnaires is limited owing to their length and the time required for the parents to complete them, as well as for the clinician to evaluate them.[16] Moreover, existing instruments have not included sedentary behaviour (as part of physical activity) nor considered sleep to be part of the (24-hour) physical activity spectrum. Using questions from previously validated questionnaires developed for other pediatric populations, such as the Family Nutrition and Physical Activity screening tool,[17] and physical activity recommendations for children with CP,[1] we have applied a structured approach with interviews involving both health professionals and parents of children with CP to adapt items and construct the new checklist. Existing pediatric sleep questionnaires were identified through literature review and reviewed by our project steering group with various backgrounds (i.e. pediatric rehabilitation, biomedical sciences, [pediatric] physical therapy, experts in the field of exercise physiology and sleep medicine, neonatology, and parenting). The steering group consisted of three researchers (aged 30–54y), seven clinicians (aged 38–60y), and three parents (aged 34–48y) to identify a range of questions, which related to symptoms relevant to identifying sleep problems in children with CP. We searched for questionnaires that could be used for children with CP across all ability levels. The following questionnaires were identified: the BEARS sleep screening tool,[18,19] the Sleep Disturbance Scale for Children,[20] the Paediatric Sleep Questionnaire,[21] and the Children's Sleep Habits Questionnaire.[22] These questionnaires have all been previously used in a variety of children with CP, and some have been validated in typically developing pediatric populations against objective measures, such as polysomnography.[23] The health care professionals and parents liked the brief, simple nature of the BEARS sleep screening tool. The questions in the sleep section of the checklist were therefore based on items of the BEARS and supplemented with questions that could be more relevant for children with CP, such as pain/discomfort during the night. After examining existing questionnaires within the project steering group, relevant items were selected along with a response format (based on the Sleep Disturbance Scale for Children) that would be appropriate for all questions. A 'don't know' option was added to help avoid non-response. The addition of a '*don't know*' option can introduce some challenges to the interpretation and scoring of responses. However, since our aim was to develop a checklist (and not an outcome instrument), this was not considered a problem.

Focus group

A focus group with three parents of children with CP (in GMFCS level I [aged 3y], level III [aged 8y], and level V [aged 12y]) was organized to discuss the developed checklist and, if necessary, modify the phrasing of the instructions and questions. Parents indicated that the checklist should also acknowledge parental sleep, which ultimately resulted in a checklist with 10 questions related to the sleep of the child and three questions related to the sleep of parents (each with an open-ended question with room for remarks, questions, or concerns that parents might have had). In addition, parents valued the importance of incorporating questions for the children who were able to understand items such as how they thought they slept, and whether they thought being physically active was fun; these child-oriented items were therefore added to the pilot checklist.

CLINICAL EXPERIENCES AND IMPLICATIONS

In our innovation project, we have observed some clinical implications of the 24-hour activity checklist, its use by clinicians, and its contributions to the care of children and their families.

After 12 months of pilot testing in three health care settings with quarterly evaluation meetings, we received mostly positive written feedback from participating parents (*n*=79; a response rate of 80% in a consecutive sample of patients) about usability, content, and clarity of the checklist. Their children represented a cross-section of those with CP regarding their GMFCS levels (I–V) and ages (0–12y). In addition, verbal feedback was collected from pediatric physiatrists and developmental pediatricians in various settings: a children's hospital (*n*=1), a school for special education (*n*=2), and a rehabilitation centre (*n*=6) in the Netherlands, about the use and applicability of the checklist in the clinical setting. The feedback and experiences of parents and clinicians can be summarized as follows. (1) The checklist, which was filled out by parents at home before their visit to the health care setting, was clear and easy to answer for parents. However, for some children it was not possible to answer the child-oriented questions (e.g. the children were too young or unable to understand/answer). The children who were capable of filling out the child-specific questions were very enthusiastic that they were asked about these issues. (2) On average, it took between 5 and 10 minutes to fill out the checklist, which was considered acceptable by all parents and children. (3) The pediatric physiatrists indicated that the questions yielded sufficient information related to physical activity and sleep to make an informed decision. (4) Reviewing and interpreting the checklist took clinicians no more than 2 minutes. (5) The checklist led to increased awareness and discussions with parents about 24-hour activity behaviours. The fact that the parents filled out the checklist at home before the consultation resulted in a more prepared doctor's visit; parents thought about possible consequences of not having enough activity throughout the day and the relation with their complaints. This facilitated the conversation between parents and health care professionals. In some cases, the appointments took longer than before implementation of the checklist. However, this was not considered a problem because the issues that were important for parents were now addressed. This process greatly increased the likelihood of identifying problems related to 24-hour activities among children with CP, with referrals to specialized sleep clinics. (6) The findings were encouraging and made us decide to continue to keep the checklist in its current form, with additional instructions related to the questions for the child. See Table 4.1 for the final checklist.

Given that counselling on sleep can be time-consuming and clinicians often feel incompetent to address sleep problems properly once encountered, many pediatric clinicians may inadvertently overlook sleep concerns.[24] Moreover, many parents do not routinely share information about their child's sleep,[10] urging the need for incorporating questions about sleep into routine health assessment for children of all ages. Health care professionals are encouraged to proactively ask patients (and their caregivers) questions about their 24-hour activity levels, and to provide specific counselling to assist with accessibility strategies for physical activity as well as suggestions for activity/exercise prescription and better sleep. Routine surveillance once a year using the checklist developed specifically for children with CP is a first step in the 24-hour activity approach for this group of children. We have noticed that counselling serves as an approach to increase awareness of the 24-hour continuum. For counselling and management, it is important that the clinician first informs parents of children with CP who have problems, or who are at risk of them, about the importance of sleep and physical activity, and educates them about the consequences that can occur if sleep or physical activity problems are not addressed. Follow-up assessment needs to be performed when appropriate.

At present the 24-hour activity checklist does not come with a validated algorithm or clinical care pathway. That said, clinicians are encouraged to take a common-sense approach to determining the likelihood of a child (or parents) having one or more problems with sleep or physical activity. In other words, every question that has been answered negatively could be interpreted as a 'red flag' requiring follow-up. For example, an endorsement of snoring on most days of the week (often or always) is suggestive and might be an indication to look for signs of obstructive sleep apnoea. In this situation, one single question will lead to the suspicion of a serious problem that should result in additional assessment. In case more than one question has been answered negatively, the clinician's confidence that the child has a sleep problem requiring further investigation will probably increase. In our innovation project we used the following rule of thumb: once the pediatric physiatrist determined that there was a probability of one sleep or physical activity problem, the child was then referred for a comprehensive sleep or physical activity evaluation, respectively.

It is important to realize that the evaluation of sleep or physical activity problems requires a more thorough history of the child's 24-hour routine with a focus on bedtime habits, night-time behaviour, naps, and daytime physical activity behaviour. This information can be obtained through (some form of) daily logs. Because of their easy accessibility and usability, such diaries may prove suitable for clinicians who are interested in physical activity and sleep duration, sleep schedule, and sleep-related behaviours and interactions. However, when the focus is on sleep quality or sleep architecture, more sophisticated measures such as 24-hour actigraphy for sleep and physical activity[25] serve as a more reliable choice for those interested in objective physical activity and sleep assessment for extended periods in the child's natural environment.

To make 24-hour activity recommendations simpler both for clinicians to use and for parents (and children) to understand, we created an infographic (Figure 4.1). This infographic was created by an expert committee, including researchers, experts in the field of physical activity and sleep medicine, physicians, and parents. To improve inclusivity of the infographic, an additional round of feedback from parents of children with more severe CP, namely those in GMFCS levels IV and V, was undertaken. The primary goal of this infographic is to facilitate effective 24-hour activity counselling and education for parents and professionals in pediatric health care. A recently published study explored how parents of children and young people with disabilities perceive the Canadian 24-hour Movement Guidelines for Children and Youth.[26] The results of this study indicate that although mothers of children and young people with disabilities have positive perceptions of the concept of the guidelines, the guideline recommendations and the brand messaging strategy are not inclusive or compatible with the abilities, needs, and previous experiences of children and young people with disabilities.[26] It is important to realize that these guidelines are not based on any evidence specific to these children and young people. The guidelines that we present in this infographic are based on CP-specific physical activity recommendations,[1] use the generally accepted sleep recommendations,[2] and take into account the suggestions to increase inclusivity by Handler et al.[26] We call on everyone with responsibility for the care of young children with CP to join us in educating and empowering parents and clinicians on

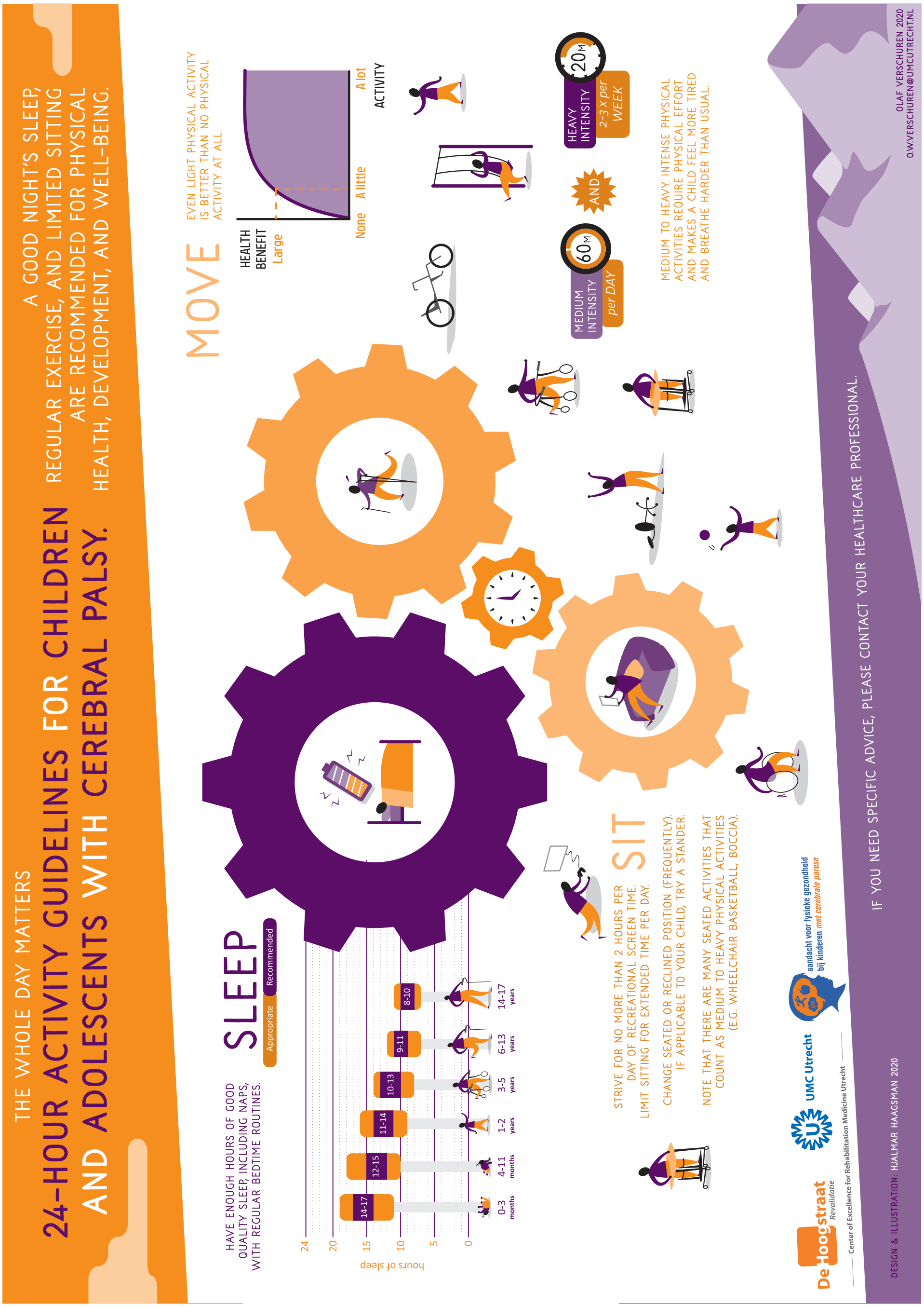


Figure 4.1 The 24-hour activity guidelines infographic.

the importance of the 24-hour activity approach for this group of children. This infographic should be used at every available opportunity to help all children with CP achieve the recommendations, and develop well and healthily.

In a recent study[27] it has been shown that transitioning from a seated to a standing position contributes to the accumulation of light activity and reduces sedentary behaviour. This activity might be a feasible option for children with CP who are classified in GMFCS levels IV and V. However, for most clinicians this type of counselling for patients who need to increase physical activity is not routine practice, even though breaking up sedentary behaviour is generally accepted as an intervention. For more activities that can be used to increase the physical activity levels of children with CP, we highlight the Ability Toolkit, developed by Handler et al.[26] This Toolkit (available at https://cdpp.ca/resources-and-publications/ability-toolkit) is a resource for children and young people with a disability, and is meant to supplement the 24-hour movement guidelines for typically developing children, similar to the guidelines for children with CP. The Ability Toolkit provides information relevant to adapting the guidelines to the unique movement abilities of children or teenagers with any type of disability. Some information may be especially useful for parents and guardians of children and teenagers with CP.

Unfortunately, there are limited data available on therapeutic approaches related to sleep in children with CP.[29] However, a good starting point would be to promote good sleep hygiene.[30] Sleep hygiene is defined as a set of sleep-related behaviours that expose children to cues and activities that prepare them for and promote appropriately timed and effective sleep.[31] These sleep promotion practices are grouped into four categories:[30] (1) environmental (e.g. bedroom temperature and blackout curtains); (2) scheduling (e.g. regular bed- and wake-times); (3) sleep practices (e.g. a relaxing bedtime routine, limiting screen time before bed); and (4) physiological (e.g. regular exercise and light exposure during the day).

Since successful management of chronic sleep disruption may decrease family stress and improve child functioning and development, it is timely for health care professionals who work with children and young people with CP and other developmental disabilities to change practice by using the 24-hour activity checklist as a springboard for meaningful conversations. Despite the emphasis in the literature on physical activity over the past 10 years, recommendations for physical activity are only beginning to be applied by health care practitioners.[32] We hope this 24-hour activity checklist will accelerate change of practice.

Table 4.1 The 24-hour activity checklist

Sleep

Sleep satisfaction	never	seldom	sometimes	often	always	don't know
1. Are you satisfied with the sleep of your child?						
Initiating or maintaining sleep	**never**	**seldom**	**sometimes**	**often**	**always**	**don't know**
2. Does it take more than 30 minutes before your child falls asleep?						
3. Does your child wake up more than 3 times a night, OR is your child awake for more than 20 minutes during the night?						
4. Do you think your child wakes up too early?						
Snoring and pain/discomfort in bed	**never**	**seldom**	**sometimes**	**often**	**always**	**don't know**
5. How often does your child snore at night?						
6. Do you think your child experiences pain or discomfort in bed?						
Nightmares	**never**	**seldom**	**sometimes**	**often**	**always**	**don't know**
7. How often does your child experience nightmares?						
Fatigue	**never**	**seldom**	**sometimes**	**often**	**always**	**don't know**
8. Does your child seem overtired or sleepy during the day?						
Sleep medication	**no**	**yes**				
9. Does your child use sleep medication/tablets (e.g. melatonin)?		My child uses: ………………………………… (*name medication*) (*dosage*) ………………… mg (*number*) ………… times a week				
Open question regarding the sleep of your child						
10. Do you have questions, remarks or concerns related to the *sleep of your child*?						
Questions related to your own sleep	**never**	**seldom**	**sometimes**	**often**	**always**	**don't know**
1. Are you satisfied with your own sleep?						
2. Do you think you have lack of sleep?						
3. Do you have questions, remarks or concerns related to *your own sleep*?						

Physical activity

Physical activity: walking	yes	no				
1. Is your child able to walk (with or without an assistive device)?						
Physical activity: movement	<30 minutes a day		30-60 minutes a day		>60 minutes per day	
2. How many minutes does your child do something physically active when he/she has free time? *You can think of one of the following activities: Active play, walking, playing outdoors, running, cycling, swimming, dancing, horse-riding, playing sport (e.g. boccia, wheelchair basketball), (toddlers)gymnastics, playing on the floor, crawling, propelling a wheelchair.*						
Fun in physical activity	yes	no	don't know			
3. Does your child like to be physically active? Does he/she experience fun in being active?						
Stimulating physical activity	yes		sometimes, but not always		no	
4. Do you know how you can help your child to be physically active? *Examples you can think about:* - *<u>playing together</u>; playing or horsing around on the floor together, playing at the playground together, play sports together, walk the dog, do groceries etc.* - *<u>moving independently</u>; crawling, walking (with or without assistive device), riding a wheelchair, cycling, being mobile using a walker/handbike etc.* - *<u>physical challenges</u>; For the children that are **able to walk** you can think about: walking stairs independently, walking long(er) distances, playing outdoors, etc. For the children that are **not able to walk** you can think about: getting in and out of the wheelchair, activities on the floor/couch, sitting unsupported (under supervision) on the couch, playing on the floor, etc.*						
			Would you like some help/advice in this area? ☐ Yes, please ☐ No, thank you			
Screen time (sedentary behaviour)	<1 hour a day		1–2 hours a day		>2 hours a day	
5. How many minutes a day does your child have 'screen time' in his/her free time? (e.g. TV, computer, game system, or any mobile device with visual screens)						
Pain/fatigue	never	seldom	sometimes	often	always	don't know
6. Do you think your child experiences pain or fatigue while being physically active?						
Open question regarding the physical activity of your child						
7. Do you have questions, remarks or concerns related to the *physical activity of your child*?						

Table 4.1 *Continued*

Questions for children

When possible, ask your child to answer the following questions (together with your help):

Sleep	
1. How do you sleep at night?	 ☐ ☐ ☐ ☐ ☐
Physical activity	
2. Do you like to move?	 ☐ ☐ ☐ ☐ ☐
Comments	
3. Any additional comment(s) about your own sleep / physical activity?	

Instructions: Please answer the questions (reflecting on the past month) at home prior to your appointment with the healthcare professional. It approximately takes 10 minutes to complete the checklist. There are three questions that can be answered by your child if he/she is capable and willing to answer the questions (with your help), but this is not mandatory.
never=this never happens; seldom=this rarely happens, less than once a week; sometimes=less than half of the week; often=more than half of the week; always=almost every day or night

Moving forward with the 24-hour activity checklist

In the past, clinical practice guidelines have been viewed as static documents. However, the science that informs clinical decision making continues to evolve. In this case, where the science related to sleep and physical activity in children with CP is rapidly evolving, guideline creation should best be viewed as a continuous improvement process with new studies reviewed and graded as they become available. It is important to realize that the checklist is based on the currently available literature and developed in co-creation with parents and researchers. Although the checklist has been well received it does need further validation. Nevertheless, we hope that health care professionals will start using and incorporate the 24-hour activity checklist into routine health assessment, experiment with and learn from its use, and provide us with feedback for continued improvement.

CONCLUSION

In the care for children with CP there is little attention to healthy development through a 24-hour approach. So, despite the emerging literature on the importance of a 24-hour activity approach, there is a knowledge-to-action gap in this area for children with CP and their families. Tools such as the proposed 24-hour activity checklist, have the potential to contribute to increasing awareness and changing practice.

Having an easy-to-use tool to identify problems within the 24-hour activity continuum is only one step in the process of identifying and resolving pediatric sleep and physical activity problems in children with CP and their families. Once the results indicate a high probability of a problem, clinicians need to be prepared for follow-up with counselling and a management plan. Often a multi- or interdisciplinary approach is essential, including medical specialists (sleep expertise, exercise medicine, child neurology, pain specialists) as well as service providers (including physical, occupational, and recreational therapists) and psychosocial expertise (developmental behavioural therapists, child life, early childhood educators, psychologists, social workers). It is hoped that a systematic integration of 24-hour physical activity and sleep assessment into clinical settings can effectively identify a large population of at-risk patients and act as a facilitator for comprehensive management as part of a broader-based strategy to increase adoption of healthy lifestyle behaviours for children with CP.

ACKNOWLEDGEMENTS

We are grateful for the contribution of the parents (OuderInzicht), parent organization (CP-Nederland), researchers, and clinicians of De Hoogstraat Rehabilitation. JWG holds the Scotiabank Chair in Child Health Research. The authors have stated that they no interests that might be perceived as posing a conflict or bias.

What this paper adds

- The 24-hour activity checklist detects problems in children with cerebral palsy (CP).
- A CP-specific infographic facilitates effective 24-hour activity counselling and education.

REFERENCES

1. Verschuren O, Peterson MD, Balemans AC, Hurvitz EA. Exercise and physical activity recommendations for people with cerebral palsy. Dev Med Child Neurol. 2016;58:798–808.
2. Tremblay MS, Carson V, Chaput JP, et al. Canadian 24-hour movement guidelines for children and youth: an integration of physical activity, sedentary behaviour, and sleep. Appl Physiol Nutr Metab. 2016;41:S311–27.
3. Walsh JJ, Barnes JD, Cameron JD, et al. Associations between 24 hour movement behaviours and global cognition in us children: a cross-sectional observational study. Lancet Child Adolesc Health. 2018;2:783–91.
4. 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;5:555–71.
5. Kredlow MA, Capozzoli MC, Hearon BA, Calkins AW, Otto MW. The effects of physical activity on sleep: a meta-analytic review. J Behav Med. 2015;38:427–49.
6. Driver HS, Taylor SR. Exercise and sleep. Sleep Med Rev. 2000;4:387–402.
7. Chennaoui M, Arnal PJ, Sauvet F, Leger D. Sleep and exercise: a reciprocal issue? Sleep Med Rev. 2015;20:59–72.
8. Dolezal BA, Neufeld EV, Boland DM, Martin JL, Cooper CB. Interrelationship between sleep and exercise: a systematic review. Adv Prev Med. 2017;2017:1364387.
9. Honaker SM, Meltzer LJ. Sleep in pediatric primary care: a review of the literature. Sleep Med Rev. 2016;25:31–9.
10. 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 Feb 26, online ahead of print.
11. Hulst RY, Voorman JM, Pillen S, Ketelaar M, Visser-Meily JMA, Verschuren O. Parental perspectives on care for sleep in children with cerebral palsy: a wake-up call. Disabil Rehabil. 2020 Jun, online ahead of print.
12. Luginbuehl M, Kohler WC. Screening and evaluation of sleep disorders in children and adolescents. Child Adolesc Psychiatr Clin N Am. 2009;18:825–38.
13. Spruyt K, Gozal D. Development of pediatric sleep questionnaires as diagnostic or epidemiological tools: a brief review of dos and don’ts. Sleep Med Rev. 2011;15:7–17.

14. Prinsen CAC, Mokkink LB, Bouter LM, et al. Cosmin guideline for systematic reviews of patient-reported outcome measures. Qual Life Res. 2018;27:1147–57.
15. Capio CM, Sit CH, Abernethy B, Rotor ER. Physical activity measurement instruments for children with cerebral palsy: a systematic review. Dev Med Child Neurol. 2010;52:908–16.
16. Lavelle G, Noorkoiv M, Theis N, et al. Validity of the international physical activity questionnaire short form (IPAQ-SF) as a measure of physical activity (PA) in young people with cerebral palsy: a cross-sectional study. Physiotherapy. 2019;107:209–15.
17. Ihmels MA, Welk GJ, Eisenmann JC, Nusser SM. Development and preliminary validation of a family nutrition and physical activity (FNPA) screening tool. Int J Behav Nutr Phys Act. 2009;6:14.
18. Owens JA, Dalzell V. Use of the 'BEARS' sleep screening tool in a pediatric residents' continuity clinic: a pilot study. Sleep Med. 2005;6:63–9.
19. Bastida-Pozuelo MF, Sanchez-Ortuno MM. Preliminary analysis of the concurrent validity of the spanish translation of the BEARS sleep screening tool for children. J Psychiatr Ment Health Nurs. 2016;23:513–20.
20. Bruni O, Ottaviano S, Guidetti V, 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;5:251–61.
21. Chervin RD, Hedger K, Dillon JE, Pituch KJ. Pediatric sleep questionnaire (PSQ): validity and reliability of scales for sleep-disordered breathing, snoring, sleepiness, and behavioral problems. Sleep Med. 2000;1:21–32.
22. Goodlin-Jones BL, Sitnick SL, Tang K, Liu J, Anders TF. The children's sleep habits questionnaire in toddlers and preschool children. J Dev Behav Pediatr. 2008;29:82–8.
23. Spruyt K, Gozal D. Pediatric sleep questionnaires as diagnostic or epidemiological tools: a review of currently available instruments. Sleep Med Rev. 2011;15:19–32.
24. Gruber R, Carrey N, Weiss SK, et al. Position statement on pediatric sleep for psychiatrists. J Can Acad Child Adolesc Psychiatry. 2014;23:174–95.
25. Sadeh A. III. Sleep assessment methods. Monogr Soc Res Child Dev. 2015;80:33–48.
26. Handler L, Tennant EM, Faulkner G, Latimer-Cheung AE. Perceptions of inclusivity: the Canadian 24-hour movement guidelines for children and youth. Adapt Phys Activ Q. 2019;36:1–18.
27. Verschuren O, Peterson MD, Leferink S, Darrah J. Muscle activation and energy-requirements for varying postures in children and adolescents with cerebral palsy. J Pediatr. 2014;165:1011–6.
28. Canadian Disability Participation Project, Canadian Society for Exercise Physiology, ParticipACTION, Healthy Active Living and Obesity Research (HALO) CHEO RI. The Ability Toolkit: a resource for parents of children and youth with a disability. Ottawa, ON: Canadian Society for Exercise Physiology, 2018.
29. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: an underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
30. Jan JE, Owens JA, Weiss MD, et al. Sleep hygiene for children with neurodevelopmental disabilities. Pediatrics. 2008;122:1343–50.
31. Meltzer LJ, Mindell JA. Nonpharmacologic treatments for pediatric sleeplessness. Pediatr Clin North Am. 2004;51:135–51.
32. Carson V, Leblanc CM, Moreau E, Tremblay MS. Paediatricians' awareness of, agreement with and use of the new Canadian physical activity and sedentary behaviour guidelines for children and youth zero to 17 years of age. Paediatr Child Health. 2013;18:538–42.

Chapter 5

Sleep problems in children with cerebral palsy and their parents

Raquel Y Hulst, Jan Willem Gorter, Jeanine M Voorman, Eveline Kolk,
Sanne van der Vossen, Johanna MA Visser-Meily, Marjolijn Ketelaar,
Sigrid Pillen, Olaf Verschuren

Dev Med Child Neurol. 2021;63(11):1344–50. doi: 10.1111/dmcn.14920.

ABSTRACT

Aim: To describe: (1) the frequency and types of sleep problems; (2) parent-rated satisfaction with their child's and their own sleep; and (3) child factors related to the occurrence of sleep problems in children with cerebral palsy (CP) and their parents. The secondary objective was to compare the sleep outcomes of children with CP with those from typically developing children and their parents.

Method: The Sleep section of the 24-hour activity checklist was used to assess the sleep of children with CP and their parents and the sleep of typically developing children and their parents.

Results: The sleep outcomes of 90 children with CP (median age 5y, range 0–11y, 53 boys, 37 girls, 84.4% ambulatory) and 157 typically developing peers (median age 5y, range 0–12y; 79 boys, 78 girls) and their parents were collected. Children with CP were more likely to have a sleep problem than typically developing children. Non-ambulatory children with CP were more severely affected by sleep problems than ambulatory children. The parents of non-ambulatory children were less satisfied about their child's and their own sleep. Waking up during the night, pain/discomfort in bed, and daytime fatigue were more common in children with CP and more prevalent in children who were non-ambulatory.

Interpretation: These findings highlight the need to integrate sleep assessment into routine pediatric health care practice.

INTRODUCTION

A growing body of evidence supports the importance of sleep for optimal child health and development.[1] Cerebral palsy (CP) represents one of the most common physical childhood disabilities worldwide.[2] Children with CP are a population at risk for the occurrence of sleep problems, with parent-reported prevalence rates varying between 23% and 46%[3–5] compared to 20% to 30% in typically developing children.[6] Commonly reported sleep problems in children with CP include difficulty falling asleep, frequent night-time waking, sleep-related breathing disorders, early-morning waking, and excessive daytime fatigue.[7–11]

Despite widespread recommendations for systematically enquiring about sleep and sleep problems in pediatric health care,[12,13] sleep health is frequently overlooked in rehabilitation settings.[14] According to the parents of children with CP, clinicians rarely ask about sleep during routine health assessments, with even less attention paid to parental sleep and the impact on the family.[15] This is worrisome because in addition to the negative implications for the child, sleep problems in children with CP are a major source of family stress.[15,16] A recent review showed that parents of children with neurodevelopmental disabilities, including CP, report poorer sleep quality than parents of typically developing children.[17] To improve the sleep of both children and parents, and thereby optimize their health and well-being, we first have to recognize sleep problems.

Originating from a need for a brief, practical tool that can be easily applied in routine care, we recently developed a 24-hour activity checklist as part of a more general clinical practice guide for children with CP.[18] In addition to providing an insight into the physical activities of children with CP, the checklist was designed to guide clinicians in determining whether a more thorough assessment or referral is warranted when sleep problems are detected. Now, 1 year after the implementation of the 24-hour activity checklist, we have the opportunity to explore the extent to which sleep problems can be detected in children with CP and their parents.

The primary purpose of this study was to describe in children with CP and their parents: (1) the frequency and types of parent-reported sleep problems; (2) parent-rated satisfaction with their child's sleep and their own sleep; and (3) child factors related to the occurrence of sleep problems. Since children with CP have both sleep problems that are similar to typically developing children and specific sleep problems related to CP, it is important to place findings in the context of children without CP. Therefore, the secondary purpose of this study was to compare the sleep outcomes of children with CP and their parents with comparative data collected from typically developing children and their parents.

METHOD

A multicentre, cross-sectional study was conducted involving children with CP and their parents. In addition, a convenience sample of typically developing children and their parents was recruited from the general population as a comparison group. The study was approved by the medical ethics research committee of De Hoogstraat Rehabilitation Center, Utrecht, the Netherlands.

Participants

The study population consisted of children with CP (aged 0–11y, 53 males, 37 females, 84.4% ambulatory) and their parents receiving care from three pediatric rehabilitation settings (a rehabilitation centre, a special education school, and the rehabilitation department of a children's hospital) in the Utrecht province of the Netherlands. Inclusion criteria were children diagnosed with CP across all Gross Motor Function Classification System (GMFCS) levels and between the ages of 0 to 12 years. All families who were scheduled for a follow-up appointment with their rehabilitation physician received a paper-and-pencil 24-hour activity checklist at home as part of routine care. Parents were instructed to complete the questionnaire before their appointment and hand in the checklist during the upcoming consultation with their rehabilitation physician, who would in turn discuss the responses with the parents and collect the checklists. The parents of children with CP who agreed for their questionnaire to be used for research signed an informed consent for participation in the study. All checklists collected between April 2019 and August 2020 that were returned complete with signed consent and met the inclusion criteria were used for data analysis.

The comparison group consisted of typically developing children (79 males, 78 females) aged between 0 and 12 years without any known physical, developmental, intellectual, or behavioural disability (based on parental report). The parents of typically developing children were recruited via the (social) networks of colleagues and acquaintances. They were invited to participate in the study by e-mail and their responses were collected via a Web-based version of the same questionnaire. To ensure anonymity, no socio-demographic information, other than sex and age of the child, was collected.

The 24-hour activity checklist

Parents completed the 24-hour activity checklist, a brief questionnaire developed to assess the physical activity and sleep patterns of the child and parental sleep.[18] For the purpose of the present study, responses to the Sleep section of the 24-hour activity checklist (see Table

4.1, Sleep section) were used. In this section, parents were asked to rate the frequency of child and parent sleep problems within the past month on a 5-point Likert-type scale (Table 5.1). Since the checklist assesses the occurrence of different types of sleep problems that are not necessarily correlated (i.e. a formative model), analyses regarding structural validity and internal consistency were not relevant[19] and subsequently not reported.

Table 5.1 Items, questions, and response format of the Sleep section of the 24-hour activity checklist

Item	Question	Response format[a]
Child sleep		
C1	Are you satisfied with the sleep of your child?	5-point scale from never to always
C2	Does it take more than 30 minutes before your child falls asleep?	5-point scale from never to always
C3	Does your child wake up more than three times a night OR is your child awake for more than 20 minutes during the night?	5-point scale from never to always
C4	Do you think your child wakes up too early?	5-point scale from never to always
C5	How often does your child snore at night?	5-point scale from never to always
C6	Do you think your child experiences pain or discomfort in bed?	5-point scale from never to always
C7	How often does your child experience nightmares?	5-point scale from never to always
C8	Does your child seem overtired or sleepy during the day?	5-point scale from never to always
C9	Does your child use sleep medication/ tablets (e.g. melatonin)?	Yes/no. If yes: type, dose, and frequency of drug
Parent sleep		
P1	Are you satisfied with your own sleep?	5-point scale from never to always
P2	Do you think you have lack of sleep?	5-point scale from never to always

[a] The 5-point scale options are as follows: 1=never (less than once per month), 2=seldom (1–2 times per month), 3=sometimes (1–3 times per week/less than half of the week), 4=often (4–6 times per week/more than half of the week), 5=always (daily/nightly). All items, except for C9, included a 'don't know' option.

Statistical analysis

Data were analysed per study group (children with CP and typically developing children) and within CP subgroups. CP subgroup analyses were performed by sex, age group (preschool age, 0–3y vs school-age, 4–12y) and walking ability (ambulatory, GMFCS levels I–III vs non-ambulatory, GMFCS levels IV and V) to determine child factors related to the occurrence of sleep problems in children with CP.

Descriptive statistics were performed on the responses given to each of items C2 to C8 (Table 5.1) separately. To determine whether these sleep problems were more often reported by children with CP compared to typically developing children and whether there were differences within subgroups of CP, the difference in distribution of responses (i.e. the proportion of sleep problems reported to occur never, seldom, sometimes, often, or always) for each item was analysed using a non-parametric Mann–Whitney *U* test.

To calculate the total number of sleep problems per child, the responses to items C2 to C8 were dichotomized for the presence of a sleep problem (yes/no) and summed, yielding a possible score ranging from 0 to 7 sleep problems; difficulties occurring often or always (i.e. more than half of the week) were considered a sleep problem, whereas items answered with never, seldom, or sometimes (i.e. less than half of the week) or as 'don't know' were scored as no problem. Based on these dichotomized values, the frequencies of individual sleep problems and the distribution of the total number of sleep problems per child across study groups and within CP subgroups were calculated. To analyse whether children with CP were more likely to have a sleep problem than typically developing children and whether non-ambulatory children were more likely to have a sleep problem than ambulatory children with CP, Fisher's exact tests were performed on the proportion of children having at least one sleep problem based on dichotomized values between groups. To determine whether children with CP suffered from more sleep problems than typically developing children and whether there were differences within subgroups of CP, the difference in distribution of the total number of sleep problems (i.e. the proportion of children having zero, one, two, three, or more than three sleep problems based on dichotomized values) were assessed using Mann–Whitney *U* tests.

Since the use of sleep medication can be considered both a problem or solution to a sleep problem, item C9 (Table 5.1) was not included in the dichotomized 'total number of sleep problems' per child; the proportion of medication use across groups was analysed separately using Fisher's exact test.

The two items on sleep satisfaction (i.e. C1, P1; Table 5.1) measured different aspects of sleep, that is, subjective perceptions, in contrast to all other items of the questionnaire, which were more objective in nature (i.e. frequencies of factual events, such as snoring); therefore, their outcomes are reported separately and were analysed using Mann–Whitney *U* tests.

Data were analysed using SPSS v26 (IBM Corp., Armonk, NY, USA). Significance level was set at $p<.05$.

RESULTS

In total, 110 responses were collected from children with CP and their parents; one checklist was returned largely (>75%) incomplete and 19 checklists were returned by the same family for a second follow-up evaluation and were therefore excluded from analysis. For the typically developing comparison group, a total of 168 responses were collected from typically developing children and their parents, of which three responses were excluded because of the presence of a disability, six responses had unknown ages, and two children were older than 12 years and were therefore excluded from the analysis. The final sample consisted of 90 children with CP and 157 typically developing children (group characteristics are presented in Table 5.2). Study groups did not differ with respect to sex or age (both *p*>.05). The majority of children with CP (84.4%) were ambulatory (i.e. classified in GMFCS levels I–III), with a similar distribution across age groups (preschool age 88.9%, school-age 82.5%; Fisher's exact test, *p*>.05).

Table 5.2 Group characteristics of children with CP and their typically developing peers

	Children with CP *n*=90	Typically developing children *n*=157	*p*
Sex, *n* (%)			
Male	53 (58.9)	79 (50.3)	.233[a]
Female	37 (41.1)	78 (49.7)	
Age, y			
Range	0–11	0–12	.974[b]
Median	5	5	
Age group, *n* (%)			
0–3y	27 (30)	57 (36.3)	.332[a]
4–12y	63 (70)	100 (63.7)	
GMFCS level, *n* (%)			
I	52 (57.8)		
II	19 (21.1)		
III	5 (5.6)		
IV	7 (7.8)		
V	7 (7.8)		

CP: cerebral palsy; GMFCS: Gross Motor Function Classification System. [a] Fisher's exact test; [b] Mann–Whitney *U* test.

Frequency and types of parent-reported sleep problems

Child sleep

Total number of sleep problems

Children with CP were more likely to have a sleep problem than typically developing children (Fisher's exact test, $p<.001$); 72.2% of children with CP were reported to have at least one sleep problem occurring often or always compared to 46.4% in typically developing children. The child-by-child distribution of the total number of sleep problems in children with CP was significantly different from those observed in typically developing children, with a considerably higher total number of sleep problems reported by parents of children with CP (U=4779.5, z=−4.53, $p<.001$); in children with CP, zero sleep problems were present in 27.8%, one sleep problem was present in 38.9%, two sleep problems were present in 17.8%, three sleep problems were present in 12.2%, and more than three sleep problems were present in 3.3% compared to 53.5%, 31.8%, 12.7%, 1.9%, and 0% in typically developing children respectively.

Types of sleep problems

Figure 5.1 shows the frequencies of different parent-reported sleep problems in children with CP compared to typically developing children. Daytime fatigue (33.3%), difficulty falling sleep (30%), and early-morning waking (26.7%) were the most frequently reported sleep problems occurring often or always in children with CP. The most frequently reported sleep problems in typically developing children were difficulty falling asleep (22.3%) and early-morning waking (19.7%). Significant differences were observed between children with CP and typically developing children in the distribution of responses to the items regarding daytime fatigue (U=4249, z=−5.42, $p<.001$), waking up during the night (U=4800, z=−4.31, $p<.001$), and pain or discomfort in bed (U=4412, z=−5.49, $p<.001$), with the parents of children with CP consistently reporting a higher frequency in the occurrence of these sleep problems. Use of medication for sleep purposes was reported in 6.7% (n=6; n=4 melatonin; n=2 baclofen) of children with CP, a proportion that did not differ from the 3.2% (n=5, all melatonin) observed in typically developing children (Fisher's exact test, $p>.05$).

Sleep satisfaction

The parents of typically developing children were more satisfied with their child's sleep than the parents of children with CP (U=5728, z=−2.8, $p<.01$; Figure 5.2). The parents of children with CP reported to be never or seldom satisfied about their child's sleep in 8.9% of cases compared to a significantly lower proportion (1.3%) of parents of typically developing children (Fisher's exact test, $p<.01$).

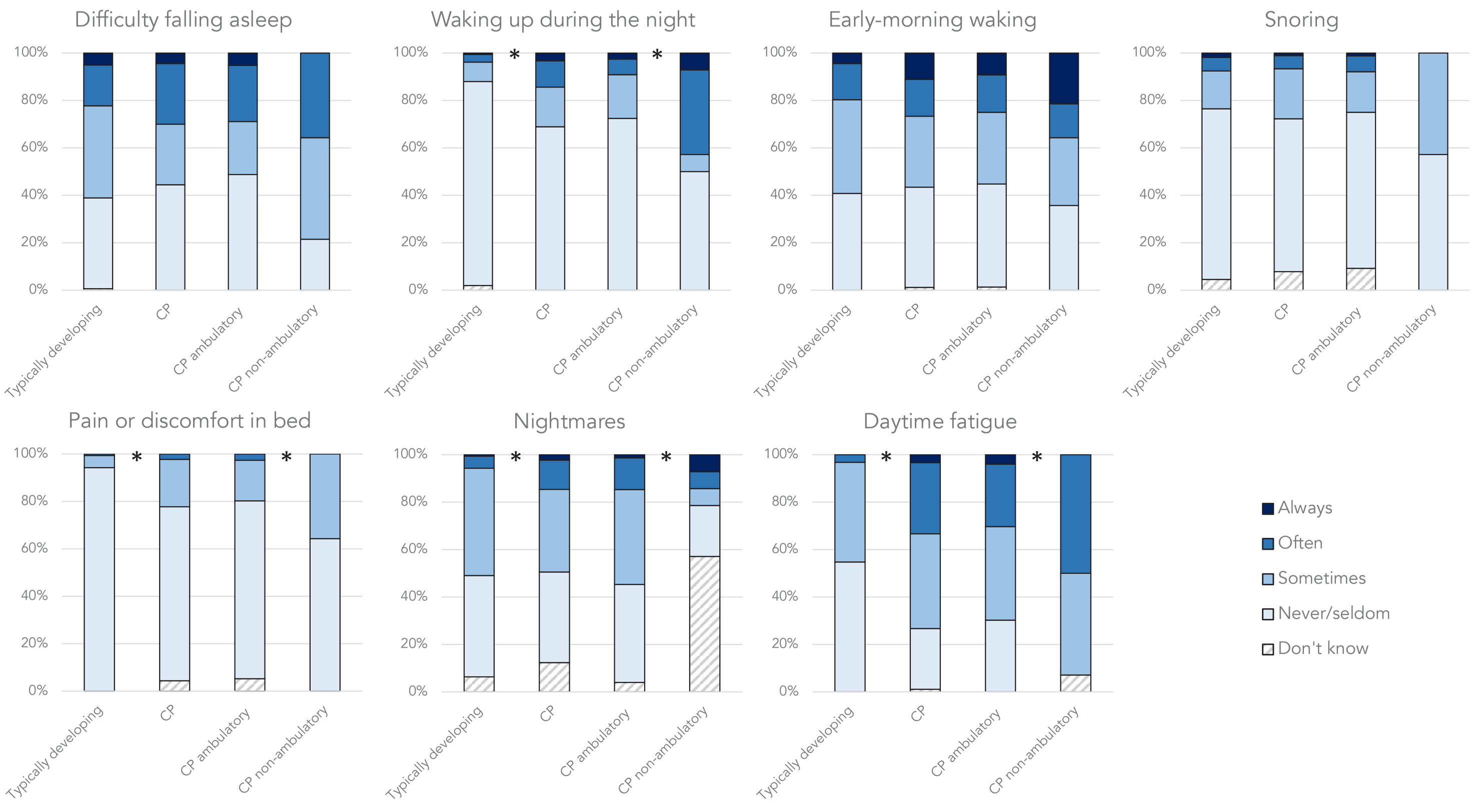


Figure 5.1 Parent-reported sleep problems in children with cerebral palsy (CP) (total, ambulatory, and non-ambulatory) compared to typically developing children. The single asterisk indicates a significant difference (Mann–Whitney *U* test, $p<.05$) between the CP and typically developing study groups or between ambulatory and non-ambulatory children with CP.

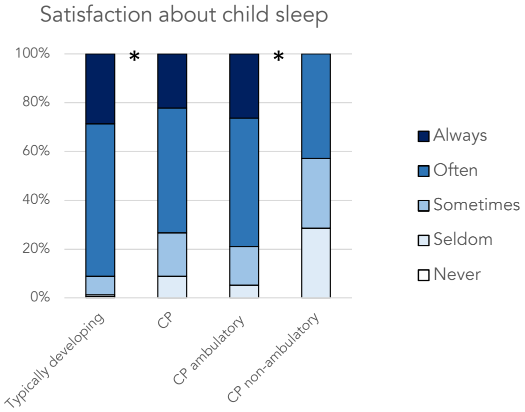


Figure 5.2 Parent-reported satisfaction about child sleep in children with cerebral palsy (CP) (total, ambulatory, and non-ambulatory) compared to typically developing children. The single asterisk indicates a significant difference (Mann–Whitney *U* test, *p*<.05) between the CP and typically developing study groups or between ambulatory and non-ambulatory children with CP.

Parent sleep

No differences were observed in the distribution of responses on the two parental sleep items of sleep satisfaction (P1) and sleep deprivation (P2) between the parents of children with CP and the parents of typically developing children (both *p*>.05). Of the parents of children with CP, 13.6% reported to be never or seldom satisfied about their own sleep compared to 8.9% recorded in the parents of typically developing children (Figure 5.3). When parents were asked whether they felt sleep-deprived, one-third (34.1%) of parents of children with CP reported feeling sleep-deprived often or always compared to a quarter (24.8%) of parents of typically developing children.

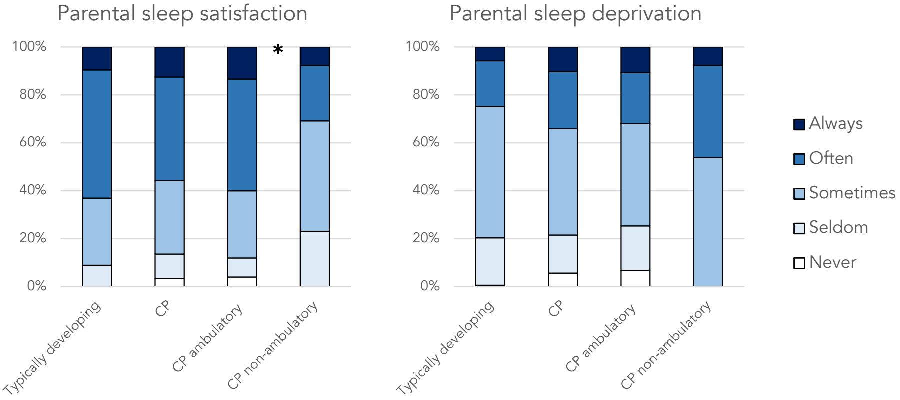


Figure 5.3 Self-rated sleep satisfaction and sleep deprivation in the parents of children with cerebral palsy (CP) (total, ambulatory, and non-ambulatory) compared to the parents of typically developing children. The single asterisk indicates a significant difference (Mann–Whitney *U* test, *p*<.05) between ambulatory and non-ambulatory children with CP.

Child factors and sleep problems in children with CP

Sex

No differences were found between males and females regarding child sleep problems (individual items and total number of sleep problems), use of sleep medication, sleep satisfaction, or parental sleep (all $p>.05$).

Age

The parents of older children reported that their child would wake less frequently during the night than the parents of younger children ($U=671.5$, $z=-1.71$, $p<.05$) and the parents of older children were more often satisfied with their child's sleep compared to those of younger children ($U=625.5$, $z=-2.08$, $p<.02$). No other differences were found with regard to age (all $p>.05$).

Walking ability

Although no association was found between walking ability and the presence of a sleep problem (i.e. having at least one sleep problem occurring often or always; Fisher's exact test, $p>.05$), the child-by-child distribution of the total number of sleep problems was significantly higher for non-ambulatory children ($U=355.50$, $z=-2,06$, $p<.05$). Specifically, waking up during the night ($U=286$, $z=-2.89$, $p<.01$), pain or discomfort in bed ($U=348.5$, $z=-1.96$, $p<.05$), and daytime fatigue ($U=319$, $z=-2.14$, $p<.05$) were significantly more frequently reported by the parents of non-ambulatory children compared to ambulatory children (Figure 5.1), while the parents of non-ambulatory children more often reported to 'not know' whether their child had nightmares. No difference was found between the proportion of children using sleep medication across walking ability (Fisher's exact test, $p>.05$).

Satisfaction about child sleep was significantly differently distributed across walking ability; the parents of ambulatory children were more often satisfied about their child's sleep ($U=264$, $z=-3.24$, $p<.001$, Figure 5.2).

With regard to parental sleep, the parents of ambulatory children were more often satisfied about their own sleep ($U=347.5$, $z=-1.75$, $p<.05$) than the parents of non-ambulatory children (Figure 5.3). Moreover, the parents of non-ambulatory children seemed to feel sleep-deprived more often than the parents of ambulatory children, although this difference was not significant ($U=364$, $z=-1.54$, $p=.06$).

DISCUSSION

We found that sleep problems are commonly reported by the parents of children with CP and that these children are more likely to have a sleep problem compared to their typically developing peers. In addition, non-ambulatory children with CP were more severely affected by sleep problems than ambulatory children and their parents were less satisfied about their child's and their own sleep. Also, a large group of parents reported feeling sleep-deprived.

Children with CP have a spectrum of sleep problems similar to typically developing children, with both difficulty falling sleep and early-morning waking being signs of (behavioural) insomnia, the most prevalent sleep disorder across childhood. This is supported by findings from our study, which found that the most frequently reported sleep problems in typically developing children were difficulty falling sleep (22.3%) and early-morning waking (19.7%), which were reported in comparably high frequencies by the parents of children with CP (30% and 26.7% respectively). In addition to these similarities, we found that children with CP were more likely to have other sleep disruptions, especially night-time waking and pain or discomfort in bed, compared to typically developing children. Also, daily functioning was more impaired; one-third (33.3%) of children with CP were reported to suffer from fatigue during the day often or always compared to 3.2% of typically developing children.

Although recognizing and addressing sleep problems will likely be beneficial for all children with CP, this may be especially worthwhile for those classified in higher GMFCS levels. In our study, non-ambulatory children showed a higher total number of sleep problems, which is in line with previous studies reporting higher frequencies of abnormal total sleep scores with increasing GMFCS level.[20,21] First, waking up during the night for the total group of children with CP (14.4%) is lower than in other studies reporting night-time waking in children with CP, ranging from 23.2% to 36%.[22,23] However, for children with CP who are non-ambulatory, we found that 42.9% (compared to 9.2% of ambulatory children) of parents reported frequent night-time waking. Second, pain or discomfort in bed was more frequently reported in children who were more severely affected. Physical discomfort and pain are increased by muscle spasms and joint contractures; together with an impaired ability to change sleep positions, these are likely to adversely affect the child's sleep quality.[24] Third, children with CP who are non-ambulatory were reported to suffer from daytime fatigue or sleepiness often or always in 50% of cases compared to 30.3% of ambulatory children. These rates are comparable to previous studies reporting on the prevalence of daytime sleepiness in children with CP, which ranges from 12.8% to 63.5%.[3]

In line with previous research on sleep outcomes in the parents of children with neurodevelopmental disabilities,[7,17] we found that poor sleep quality was reported by a large group of parents and especially by those with non-ambulatory children. However, it should be noted that the parental sleep items did not specifically enquire about the cause of poor parent sleep quality; therefore, there could have been other disruptors at play unrelated to or in addition to their child's sleep. Children with physical disabilities, including CP, are more likely to require night-time parental attention,[25] which may contribute to greater sleep disturbance for caregivers.[16] Mothers experiencing the highest number of sleep disruptions have children with the highest care needs.[26] In addition to affecting parental sleep, child sleep problems and night-time care requirements have repeatedly been associated with increased psychological exhaustion and decreased well-being among parents.[7,16,27–29] Clearly, sleep problems in children with CP and the impact that they have on the family should be addressed, and this includes paying attention to parental sleep.

Children with CP showed higher frequencies of sleep problems compared to typically developing children but these differences were not as great as expected. This might be explained by the fact that frequencies, not severity, were reported; therefore, more severe problems might be at play that are reported with the same frequency. Also, the reference of parents with children with CP may be different from that of parents with typically developing children, giving rise to parents accepting abnormal sleep patterns because they think it is to be expected when dealing with CP.[15]

This study has several strengths and limitations. First, all sleep outcome measures were based on parental reports; although this is likely to be the primary source of information in the pediatric clinical setting, reporter bias may have occurred. However, the results reflect the extent to which parents perceive their child's sleep as problematic, which has also been shown to affect family well-being.[15] Second, the checklist is not a validated screening instrument but was designed as a practical tool to open and facilitate the conversation about sleep with parents in the health care setting.[18] In contrast to more extensive sleep screening tools like the Sleep Disturbance Scale for Children, the 24-hour activity checklist includes questions about parental sleep. This not only gives an insight into the impact of sleep problems beyond the child, it also stimulates a family-centred care for sleep, an important wish of parents of children with CP.[15] Although the number of children classified in the higher GMFCS levels IV and V in our study is small (n=14; 15.6%), the distribution of ambulatory and non-ambulatory children with CP is comparable to cohort data from the Netherlands.[30] Children with CP who have additional comorbidities such as epilepsy, visual, and cognitive impairments have more sleep problems than children without comorbidities, although the effects of comorbidities on the prevalence of sleep problems are not consistent.[3] We did not collect such additional information from the children's medical files; therefore, comorbidities could potentially have contributed, especially in the more complex non-ambulatory group, to excess night-time waking and daytime fatigue. Also, no further background information on the socio-demographic aspects of our samples were collected and this may influence the generalizability of the findings. Finally, a strength of this study is the use of reference data, which allowed us to place the findings on sleep problems in children with CP in the context of those reported by the parents of typically developing children.

In conclusion, the findings of this study demonstrate that: (1) sleep problems are more common in children with CP than in their typically developing peers; (2) non-ambulatory children with CP are more severely affected by sleep problems than ambulatory children both in frequency and number of sleep problems; and (3) a large group of parents (of both children with CP and typically developing children) is sleep-deprived. These results call for more attention to be placed on the importance of sleep in pediatric health care, especially in more vulnerable populations like children with CP and their parents, to improve their sleep health and well-being.

ACKNOWLEDGEMENTS

The authors express their gratitude to the children and parents who participated in this study and the health care professionals and colleagues who helped to collect data from patients and from the general population. We thank Ron Hulst for entering the data, Eline Scholten for statistical advice, Sebas van Baarsen for providing feedback on graphic colour design, and CP Nederland and OuderInzicht for their contribution to the development of the 24-hour activity checklist.

The authors have stated they had no interests that might be perceived as posing a conflict or bias.

What this paper adds

- Children with cerebral palsy (CP) are more likely to have a sleep problem than typically developing peers.
- Non-ambulatory children with CP are more severely affected by sleep problems.
- One-third of parents of children with CP report feeling sleep-deprived often or always compared to a quarter of parents of typically developing children.

REFERENCES

1. Chaput J-P, Gray CE, Poitras VJ, et al. Systematic review of the relationships between sleep duration and health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016; 41:S266–82.
2. Odding E, Roebroeck ME, Stam HJ. The epidemiology of cerebral palsy: incidence, impairments and risk factors. Disabil Rehabil. 2006;28:183–91.
3. 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;5:555–71.
4. Dutt R, Roduta-Roberts M, Brown C. Sleep and children with cerebral palsy: a review of current evidence and environmental non-pharmacological interventions. Children (Basel). 2015;2:78–88.
5. 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: 1108–13.
6. Owens J. Classification and epidemiology of childhood sleep disorders. Prim Care. 2008;35: 533–46.
7. 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;17:473–6.
8. Horwood L, Li P, Mok E, Oskoui M, Shevell M, Constantin E. Health-related quality of life in Canadian children with cerebral palsy: what role does sleep play? Sleep Med. 2019;54:213–22.
9. Newman CJ, O’Regan M, Hensey O. Sleep disorders in children with cerebral palsy. Dev Med Child Neurol. 2006;48:564–8.
10. Sandella DE, O’Brien LM, Shank LK, Warschausky SA. Sleep and quality of life in children with cerebral palsy. Sleep Med. 2011;12:252–6.
11. Romeo DM, Brogna C, Quintiliani M, et al. Sleep disorders in children with cerebral palsy: neurodevelopmental and behavioral correlates. Sleep Med. 2014;15:213–8.
12. Honaker SM, Meltzer LJ. Sleep in pediatric primary care: a review of the literature. Sleep Med Rev. 2016;25:31–9.
13. Hulst RY, Pillen S, Voorman JM, Rave N, Visser-Meily JMA, Verschuren O. Sleep health practices and sleep knowledge among healthcare professionals in Dutch paediatric rehabilitation. Child Care Health Dev. 2020;46:703–10.
14. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: an underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
15. Hulst RY, Voorman JM, Pillen S, Ketelaar M, Visser-Meily JMA, Verschuren O. Parental perspectives on care for sleep in children with cerebral palsy: a wake-up call. Disabil Rehabil. 2020. DOI: https://doi.org/10.1080/09638288.2020.1770873 (Online ahead of print).
16. 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;40:412–8.
17. Micsinszki SK, Ballantyne M, Cleverley K, Green P, Stremler R. Sleep outcomes for parents of children with neurodevelopmental disabilities: a systematic review. J Fam Nurs. 2018;24:217–49.
18. Verschuren O, Hulst RY, Voorman J, et al. 24-hour activity for children with cerebral palsy: a clinical practice guide. Dev Med Child Neurol. 2021;63:54–9.
19. Prinsen CAC, Mokkink LB, Bouter LM, et al. COSMIN guideline for systematic reviews of patient-reported outcome measures. Qual Life Res. 2018;27:1147–57.
20. Munyumu K, Idro R, Abbo C, et al. Prevalence and factors associated with sleep disorders among children with cerebral palsy in Uganda: a cross-sectional study. BMC Pediatr. 2018;18:26.
21. Romeo DM, Brogna C, Musto E, et al. Sleep disturbances in preschool age children with cerebral palsy: a questionnaire study. Sleep Med. 2014;15:1089–93.
22. Zuculo GM, Knap CC, Pinato L. Correlation between sleep and quality of life in cerebral palsy. Codas. 2015;26:447–56.

23. Elsayed RM, Hasanein BM, Sayyah HE, El-Auoty MM, Tharwat N, Belal TM. Sleep assessment of children with cerebral palsy: using validated sleep questionnaire. Ann Indian Acad Neurol. 2013;16:62–5.
24. Lélis AL, Cardoso MVLM, Hall WA. Sleep disorders in children with cerebral palsy: an integrative review. Sleep Med Rev. 2016;30:63–71.
25. 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;35:89–95.
26. Bourke-Taylor H, Pallant JF, Law M, Howie L. Relationships between sleep disruptions, health and care responsibilities among mothers of school-aged children with disabilities. J Paediatr Child Health. 2013;49:775–82.
27. Wright M, Tancredi A, Yundt B, Larin HM. Sleep issues in children with physical disabilities and their families. Phys Occup Ther Pediatr. 2006;26:55–72.
28. Barlow JH, Cullen-Powell LA, Cheshire A. Psychological well-being among mothers of children with cerebral palsy. Early Child Dev Care. 2006;176:421–8.
29. 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;101:618–23.
30. Smits DW, Gorter JW, Riddell CA, et al. Mobility and self-care trajectories for individuals with cerebral palsy (aged 1–21 years): a joint longitudinal analysis of cohort data from the Netherlands and Canada. Lancet Child Adolesc Health. 2019;3:548–57.

Part IV

Measuring the 24-hour activities in children with cerebral palsy

Chapter 6

Accelerometer-measured physical activity, sedentary behaviour, and sleep in children with cerebral palsy and their adherence to the 24-hour activity guidelines

Raquel Y Hulst, Jan Willem Gorter, Joyce Obeid, Jeanine M Voorman,
Ilse M van Rijssen, Anke Gerritsen, Johanna MA Visser-Meily, Sigrid Pillen,
Olaf Verschuren

Under review.

ABSTRACT

Aim: To: (1) measure and describe the 24-hour activities (i.e. physical activity (PA), sedentary behaviour (SB), and sleep); and (2) examine adherence to the 24-hour activity guidelines among children with cerebral palsy (CP) using actigraphy.

Method: Children's 24-hour activities were recorded over 7 days using hip- and wrist-worn ActiGraph wGT3X-BT accelerometers.

Results: In total, 362 days and 340 nights from 54 children with CP (Gross Motor Function Classification System (GMFCS) levels I–III; 44% girls; median age [range] 6.5 [3–12] years) were included in the analyses. Average daily wear time was 746.2±48.9 min, of which children spent on average 33.8% in light PA (251.6±58.7 min/day), 5.2% in moderate-to-vigorous PA (38.5±20.1 min/day), and the remaining 61.1% being sedentary (456.1±80.4 min/day). PA decreased while SB increased with increasing GMFCS level. In total, 13% of all children met the PA recommendations, and 35% met the age-appropriate sleep duration recommendation. The proportion of children meeting the combined 24-hour guidelines for PA and sleep was low (5.9%), especially in those classified at GMFCS level III (0%).

Interpretation: The observed low 24-hour guideline adherence rates underscore the importance of considering the entire continuum of movement behaviours in the care of children with CP, in efforts to promote healthy lifestyle behaviours and prevent negative health outcomes.

INTRODUCTION

Physical activity (PA), sedentary behaviour (SB), and sleep, collectively known as movement behaviours, are modifiable lifestyle factors affecting child health and wellbeing. In the last decade, accumulating evidence has highlighted the independent health benefits of high PA levels (especially moderate-to-vigorous physical activity (MVPA)), low SB, and sufficient hours of good quality sleep among children.[1] More recently, the collective importance of these behaviours has been recognized under the concept that "*the whole day matters*", which has led to the development of integrated 24-hour movement guidelines for children.[2] Good health outcomes may be attained by optimizing the balance between PA, SB and sleep for the individual child. Although the health benefits of meeting the 24-hour movement guidelines are well-established for typically developing (TD) children,[2,3] little is known about the 24-hour activities of children with physical disabilities.

Cerebral palsy (CP) is a common cause of physical disability in childhood. Children with CP demonstrate reduced PA levels and engage in significantly more sedentary time compared to their healthy peers.[4,5] In addition, children with CP are more likely to have sleep problems than TD children, such as difficulties falling asleep, frequent night awakenings, pain or discomfort in bed, and waking up too early.[6] Along with reduced sleep quality, a large proportion (41%) of children with CP aged 4–18 years were reported to have a sleep duration of less than 8 hours per night,[7] suggesting that these children may not be getting enough sleep. Given that sleep is an important determinant for the health-related quality of life in children with CP,[8] together with the high prevalence of inactive lifestyles, and the concomitant risk of chronic health conditions in adults with CP,[9] it is likely that this population stands to benefit from meeting the 24-hour movement guidelines throughout childhood and adolescence. It is therefore no surprise that the 24-hour activity guideline approach has been adopted for children with CP in a recently published clinical practice guide,[10] to promote healthy lifestyle behaviours and prevent negative health outcomes in this vulnerable population.

Studies reporting on 24-hour movement behaviours and guideline adherence in pediatric populations with heterogeneous conditions, have predominantly relied on subjective measures like surveys or questionnaires.[11–13] In children with CP, the few studies that have measured PA and SB objectively, either did not include sleep as part of the 24-hour activities,[5,14] or assessed sleep using parental reports,[15] which can be unreliable and can introduce different sources of reporting bias.[16] For example, subjective reports have been shown to consistently overestimate sleep duration.[17] In contrast, actigraphy provides a device-based measure of activity frequency, duration, and intensity, and is increasingly used to assess PA and SB in children.[18] Actigraphy is also recognized as a valid measure to quantify periods of sleep and wakefulness.[16] Nevertheless, no studies have objectively examined the combined 24-hour activities and guideline adherence in children with CP using solely device-based measures. Therefore, the aims of the present study were to: (1) measure and describe the 24-hour activities (i.e. PA, SB, and sleep); and (2) examine adherence to the 24-hour activity guidelines for PA and sleep among a sample of children with CP using actigraphy. Additionally, group comparisons were explored across the levels of functional ability (Gross Motor Function Classification System (GMFCS) levels I–III) and by age groups (preschool and school-age).

METHOD

A multicenter, cross-sectional, observational study was conducted. The study was approved by the Medical Ethics Research Committee of the University Medical Centre Utrecht (file number 19-630), the Netherlands.

Participants

Children with CP were recruited from the outpatient clinics of five rehabilitation settings (three pediatric rehabilitation centers, a school for special education, and the rehabilitation department of a children's hospital) in the Netherlands. Inclusion criteria were children: (1) diagnosed with CP; (2) classified at GMFCS levels I–III (i.e., able to walk independently with or without an assistive device); and (3) between the ages of 3–12 years. This age range was chosen to allow us to describe adherence to the international, age-appropriate sleep duration recommendations for preschool (3–5y) and school-aged (6–12y) children.[19] Parents of children who met the inclusion criteria were informed of the study via their healthcare professional. All parents and children aged 12 years provided written consent/assent prior to participation in the study.

Consenting families were briefed by telephone and received a box with study materials (accelerometers, activity logbooks, and a sleep diary) along with instructions. Measurements took place in the child's home environment, while the researcher was available to address any questions or issues via telephone or e-mail. Upon completion of the study week, parents returned the study materials.

24-hour activity measurements

Device

Habitual PA, SB, and sleep were measured using the ActiGraph wGT3X-BT triaxial accelerometer (ActiGraph Corporation, Pensacola, FL, USA). At the beginning of each data collection session, the device was initialized to collect data at a 30 Hz resolution using the Actilife software version 6.13.4 (ActiGraph Corporation, Pensacola, FL, USA). Although more challenging than in TD children (e.g. due to hypersensitivity), actigraphy has been shown to be feasible and acceptable in children with CP.[20,21]

Data collection

Participants were instructed to wear the accelerometer for 24 h/day over seven consecutive days. A seven-day period was selected to ensure that the measured activities, accounting for missing days/nights due to technical failures or unforeseen circumstances, was representative of habitual PA and sleep measures.[22] Typical accelerometer placement site for PA measurements is the hip as it allows detection of the body's acceleration and deacceleration during locomotion, while wrist placement is generally used for sleep assessment to optimize the recording of small movements that occur at the distal extremities when the individual is in supine position.[16] Therefore, in this study, participants were instructed to wear the accelerometer over the right hip, at the mid-axillary line, using an elastic belt during waking hours, and to exchange accelerometer placement upon bedtime to the non-dominant wrist using a watch strap; after getting out of bed the next morning, accelerometer placement was changed back to the hip, etc. Parents and teachers/educators were asked to record the times that the accelerometer was worn and removed in an activity log, and to record naps and overnight bed times in a sleep diary. Accelerometers were to be removed during water-based activities, and all times and reasons for non-wear were recorded in the activity log. Upon completion of the study week, accelerometers and diaries were obtained from the participants and data were extracted and processed for further analyses. Data were collected between January and December 2020 during non-holiday periods. Due to the COVID-19 pandemic, data collection was paused during the school lockdown period from mid-March to mid-April 2020.

Data validation and processing

Raw accelerometer data were downloaded and converted into 15-sec epochs for hip-worn PA and SB data, while sleep data were downloaded into 60-sec epochs. Data were visually inspected for any technical malfunctions or spurious counts. All analyses were completed in Actilife software (ActiGraph Corporation, Pensacola, FL, USA).

Physical activity & sedentary behavior

Activity data from the hip-worn devices were cleaned using a semi-automated protocol, whereby any five minutes of zero counts were flagged and verified. Only non-wear periods identified in the participant logbook were excluded. The average vertical axis (VA) counts and combined triaxial average vector magnitude (VM) counts were extracted. Activity counts were normalized to actual wear time to account for differences in device wear time among participants, and reported as counts per minute (cpm) of wear time. Next, activity by intensity was calculated, and included SB, light physical activity (LPA) and moderate-to-vigorous activity (MVPA), as defined by the Evenson cut-points (using the VA counts).[23] These cut-points have been validated in ambulatory children with CP,[20] and have since then been used in several studies among children, youth, and adolescents with CP.[5,14,21] Activities by intensity are presented as percentages of total wear time. Only data from participants that met the minimum wear time of ≥6 hours per day on ≥4 days with at least 1 weekend day (Saturday or Sunday) were included in the analyses. The estimated reliability coefficient for 4 days of wear in ambulatory children with CP is .73 (95% confidence interval .58–.83), and an average of 6 days of wear would achieve reliability of .8.[24]

Sleep

Data from the wrist-worn devices were manually and visually inspected against the sleep diary records containing the child's bedtimes (in bed, lights off, out of bed). The difference between lights off and out of bed times (or between in bed and out of bed times, in those cases when lights were turned off after sleep onset), as noted in the sleep diary, were used to calculate total time in bed (TIB) and subsequently used to define the start and end of the sleep analysis period. Note that TIB includes time out of bed during night-time awakenings, a manifestation of sleep discontinuity. Within the identified TIB, the Sadeh algorithm,[25] the most commonly used algorithm for sleep-wake determination for children,[16] was used to calculate sleep onset latency (SOL: the time between lights off and the first epoch scored as sleep), total sleep time (TST: the sum of all periods scored as sleep), and wakefulness after sleep onset (WASO: the sum of all periods scored as awake after sleep onset); these outcomes are reported in hours. Sleep efficiency (SE) was calculated as the ratio of TST to TIB (in %). A night was excluded when the difference between sleep times identified by the algorithm deviated more than 10% from the bed times entered in the sleep diary. Participants with valid sleep measures for ≥4 nights with at least one weekend night (i.e. Friday to Saturday, or Saturday to Sunday) were included in the analyses.

Criteria for 24-hour activity guideline adherence

To describe guideline adherence, the 24-hour activity guidelines for children with CP[10] were applied, from which the following operational definitions were derived and applied in our study:

Physical activity

Children were classified as meeting the PA recommendations if their average daily MVPA was at least 60 minutes per day.

Sedentary behaviour

With regard to SB, the guidelines recommend: (1) "no more than 2 hours per day of recreational screen time"; and (2) "limit sitting for extended periods".[10] Because no measures for screen time were included in this study to assess the first recommendation, and there is no specification on the length of the sedentary period in the second recommendation, we could not quantify participant adherence to the SB component of the guidelines. Therefore, the SB component is not included in the 24-hour guideline adherence but rather reported descriptively as proportion of wear time (%) and total time (min/d) spent in SB, as outlined in *Daytime activities*.

Sleep

Children were classified as meeting the sleep duration recommendations if their average daily total sleep time (defined as TST) was between 10–13 hours per night for preschool children (aged 3–5 years), and 9–11 hours per night for school-aged children (aged 6–12 years).

Statistics

Data were analyzed using IBM SPSS Statistics version 26 (SPSS Inc, Chicago, Illinois). Descriptive statistics were calculated and variables were visually inspected for normality based on their boxplot and quantile-quantile-plots. Day and night variables were averaged per child, normalized to wear days and daily wear times, and analyzed for all children with CP and across subgroups for GMFCS level (I–III) and by age group (preschool and school-age). Between-group differences were assessed using ANCOVA, with GMFCS level and age group as fixed factors, and sex, age, or GFMCS level entered as covariates (depending on the group comparisons). Tukey's post-hoc tests were performed between subgroups (GMFCS level) with Bonferroni corrections for multiple pairwise comparisons. To describe 24-hour guideline adherence, the proportion of children meeting the individual and combined recommendations for PA and sleep were calculated, and group differences were assessed using Fisher's Exact test. The level of significance was set at $p<.05$.

RESULTS

Study group characteristics

In total, 60 children with CP participated in this study. Six children were excluded because no actigraphy data was collected due to hypersensitivity (*n*=4), unwillingness of the child to wear an accelerometer on their body (*n*=1), or unknown reasons (*n*=1). Therefore, the final study population consisted of 54 children (median age 6.5y), of which 30 (55.6%) were male (median age 7y), and 24 (44.4%) were female (median age 6y); 38.9% were categorized into the preschool-age group (3–5y) and 61.1% into the school-aged (6–12y) group. The majority (88.9%) of the children presented with a spastic CP, and just over half of the sample (55.6%) were classified at GMFCS level I. Detailed characteristics of the total study sample, and by GMFCS level, are presented in Table 6.1. No statistically significant age differences were observed across sex and GMFCS level (both $p>.05$). Similarly, the two age groups did not differ with respect to sex or GMFCS level, and the proportion of boys/girls was not different across GMFCS levels (all $p>.05$).

Table 6.1 Characteristics of the total study sample, and by GMFCS level

	All children *n*=54		GMFCS I *n*=30		GMFCS II *n*=15		GMFCS III *n*=9	
Sex, *n* (%)								
Girl	24	(44.4)	11	(36.7)	9	(60)	4	(44.4)
Boy	30	(55.6)	19	(63.3)	6	(40)	5	(55.6)
Age, y								
Range	3–12		3–12		5–11		3–8	
Median	6.5		6		7		4	
Age group, *n* (%)								
Preschool (3–5y)	21	(38.9)	14	(46.7)	2	(13.3)	5	(55.6)
School-aged (6–12y)	33	(61.1)	16	(53.3)	13	(86.7)	4	(44.4)
CP subtype, *n* (%)								
Spastic	48	(88.9)	26	(86.7)	14	(93.3)	8	(88.9)
Dyskinetic	2	(3.7)	2	(6.7)	-	-	-	-
Mixed	3	(5.6)	1	(3.3)	1	(6.7)	1	(11.1)
Unclassified/unknown	1	(1.9)	1	(3.3)	-	-	-	-
Affected side, *n* (%)								
Unilateral	34	(63)	23	(76.7)	10	(66.7)	1 (11.1)	
Bilateral	20	(37)	7	(23.3)	5	(33.3)	8 (88.9)	

Daytime activities: physical activity and sedentary behaviour

With regard to daytime activities (i.e. SB, LPA, and MVPA), out of the total 367 days measured, *n*=5 (1.4%) days (deriving from four children) were excluded from PA analysis due to non-wear (sickness, *n*=1, hip-monitor not worn tightly around the waist, *n*=1, not meeting the minimum daily wear time of ≥6 hours, *n*=3). The final sample with valid PA data comprised 54 children, from which a total of 362 days were included in the daytime analyses. On average, children had 6.7±0.7 valid wear days (72% had 7 days of wear time; 26% wore the device for 4-6 days, and 2% wore the device between 8–9 days) with a mean daily wear time of 746.2±48.9 min/d (12.4±0.8 hours/d) (Table 6.2).

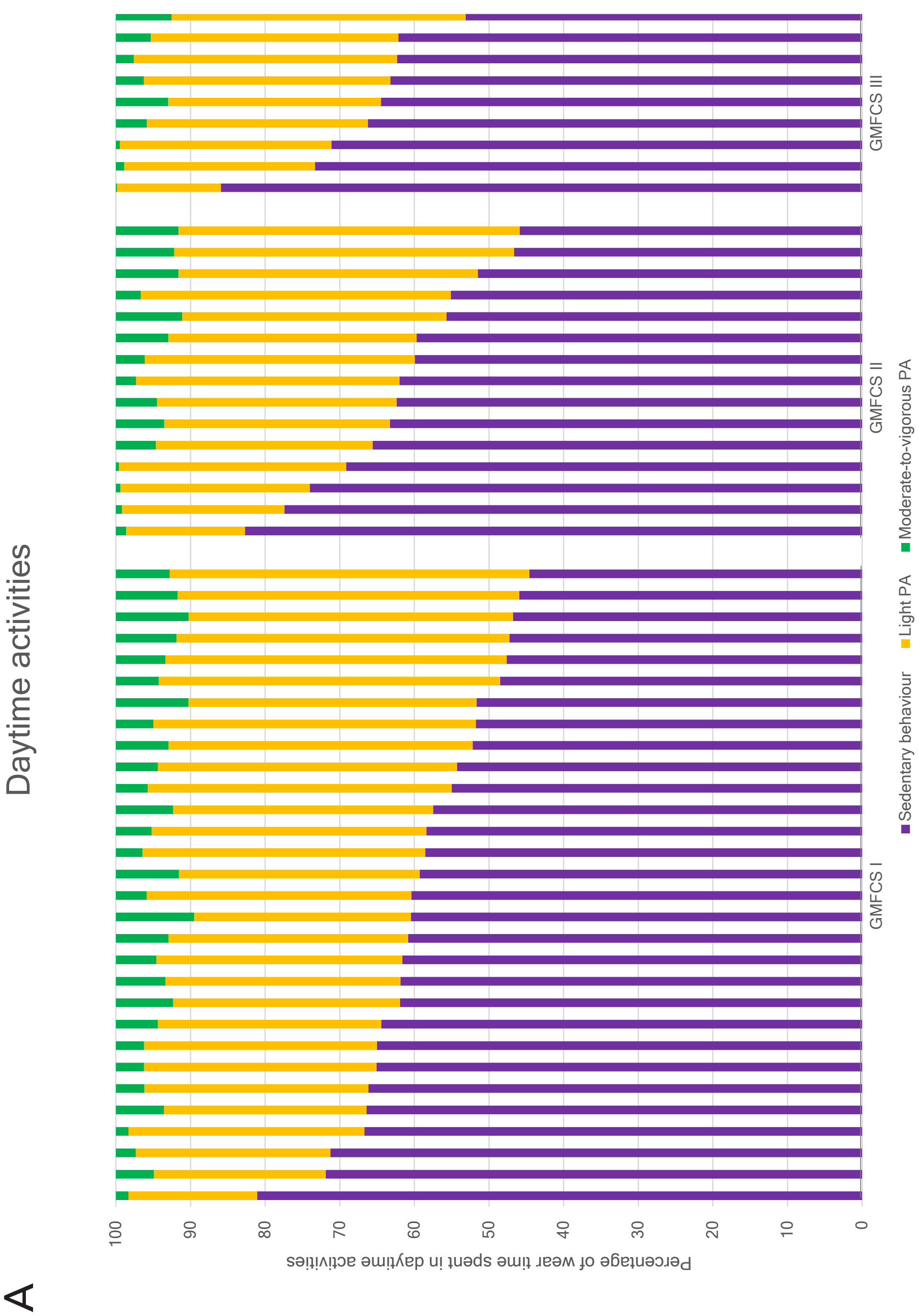
Table 6.2 Accelerometer-based data for PA, SB, and sleep, for all children and by GMFCS level

	All children		GMFCS I		GMFCS II		GMFCS III	
Physical activity data[a], mean ±SD	*n*=54		*n*=30		*n*=15		*n*=9	
Daily wear time (min/day)	746.2	±48.9	750.4	±50.2	756.6	±42.3	715.1	±47.4
SB (% of wear time)	61.1	±9.6	58.8	±8.7	62.1	±10.6	66.9	±9.2
LPA (% of wear time)	33.8	±7.8	35.3	±7.5	33.2	±8.3	29.7	±7.2
MVPA (% of wear time)	5.2	±2.7	5.9	±2.3	4.7	±3.1	3.5	±2.7
Total MVPA (min/day)	38.5	±20.1	44.3	±17.3	35.4	±22.8	24.2	±17.7
Sleep data[b], mean ±SD	*n*=51		*n*=30		*n*=12		*n*=9	
TIB, h	11.0	±0.8	10.9	±0.8	10.9	±0.6	11.4	±0.8
TST, h	9.0	±0.8	8.9	±0.8	9.1	±0.9	9.4	±0.9
SOL, h	0.3	±0.2	0.3	±0.2	0.3	±0.2	0.3	±0.3
WASO, h	1.6	±0.6	1.6	±0.6	1.5	±0.7	1.7	±0.8
SE (%)	82.4	±6.0	82.0	±5.6	83.4	±6.9	82.8	±6.6

[a] Derived from the hip-worn device; [b] derived from the wrist-worn device.
Abbreviations: PA = physical activity; SB = sedentary behaviour; LPA = light physical activity; MVPA = moderate-to-vigorous physical activity; TIB = time in bed; TST = total sleep time; SOL = sleep onset latency; WASO = wakefulness after sleep onset; SE = sleep efficiency.

To gain a better understanding of how children with CP accumulate their daytime activities, the average daily time spent in either SB, LPA, or MVPA was calculated and normalized to actual wear time. Figure 6.1 shows the proportion (%) of time spent in each of the different PA intensities for all individual children clustered by GMFCS level, sorted from highest to lowest percentage of SB (Figure 6.1A). Subgroup data are shown across GMFCS level (Figure 6.1B), and age group (Figure 6.1C).

On average, children spent 33.8% of their total wear time engaging in LPA (mean±SD, 251.6±58.7 min/day), 5.2% (38.5±20.1 min/day) in MVPA, and the remaining 61.1% of their time (456.1±80.4 min/day) being sedentary.


A
Daytime activities
Percentage of wear time spent in daytime activities
100
90
80
70
60
50
40
30
20
10
0
GMFCS I
GMFCS II
GMFCS III
■ Sedentary behaviour
■ Light PA
■ Moderate-to-vigorous PA

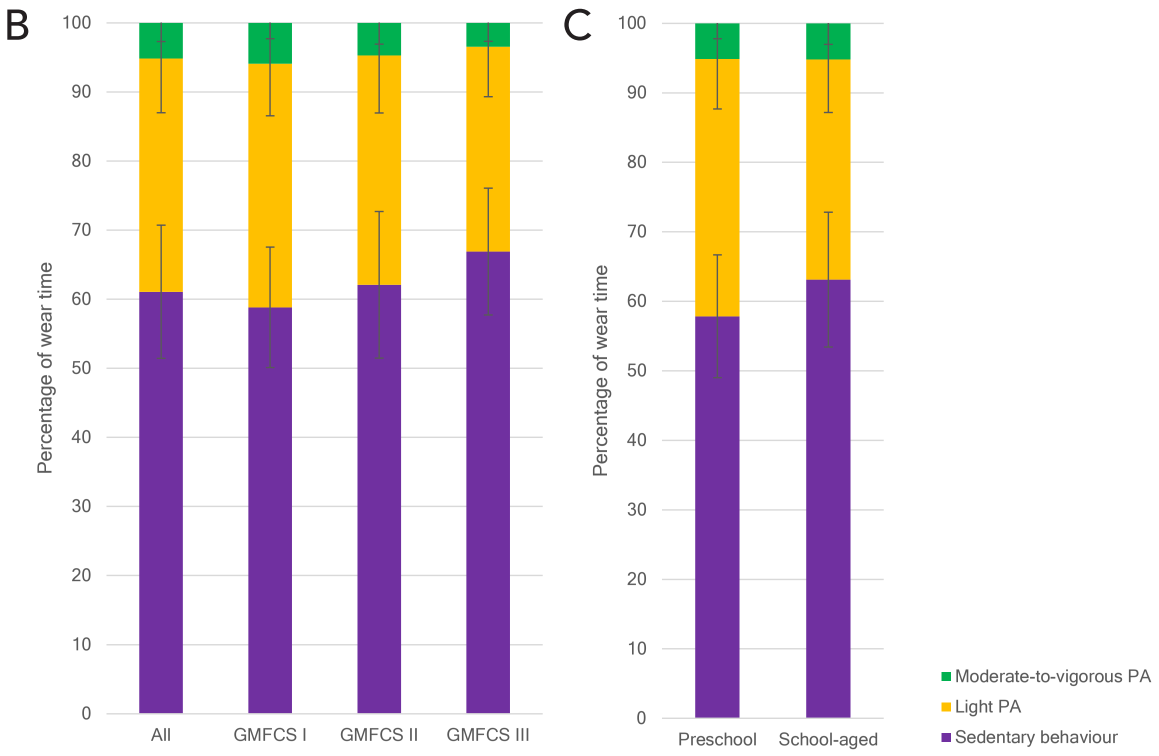


Figure 6.1 Normalized wear time spent in SB, LPA, and MVPA for **(A)** all individual participants clustered by GMFCS level and sorted from highest to lowest SB, and subgroup data across **(B)** GMFCS level and **(C)** age group. Abbreviations: SB = sedentary behaviour; LPA = light physical activity; MVPA = moderate-to-vigorous physical activity.

GMFCS level

Average daily wear time was equal across the different GMFCS levels (Table 6.2). After controlling for sex and age, a significant effect of GMFCS level was found on the percentage of wear time spent in SB (F(2, 49)=6.1, *p*<.01), LPA (F(2, 49)=5.8, *p*<.01) and MVPA (F(2, 49)=3.4, *p*<.05). Despite the significant effect for GMFCS level, post-hoc tests revealed no significant differences between individual GMFCS levels after correcting for multiple pairwise comparisons. Sedentary time tended to increase with increasing GMFCS level from I (58.8%; 441.9±74.9 min/day) to III (66.9%; 478.0±69.0 min/day), while the opposite pattern was found for time spent in MVPA, which tended to decrease from level I (5.9%; 44.3±17.2 min/day) to level III (3.5%; 24.2±17.8 min/day) (Table 6.1). The covariate age was significantly related to the percentage of wear time spent in SB, (F(1,49)=18.6, *p*<.001) and LPA (F(1,49)=25.6, *p*<.001), but not on MVPA (*p*>.05).

Age group

Average daily wear time was significantly higher in the school-aged group (762.9±46.8 min/day) compared to the preschool-aged group (720±40.9 min/day; t(52)=-3.4, *p*=.001). Sedentary time in preschool-aged children (415.5±60.3 min/day) was roughly one hour less than that of school-aged children (482.0±81.6 min/day), while time spent in MVPA was similar (preschool: 37.0±16.0 min/day; school-aged: 39.4±16.0 min/day). After controlling for GMFCS level and sex, a significant effect of age group was found on the percentage of wear time spent in LPA (F(1, 50)=5.4, *p*<.05), with preschool aged children accumulating significantly more time in LPA (37.0%; 267.6±58.5 min/day) compared to school-aged children (31.2%; 241.5±57.4 min/day). The covariate GMFCS level was significantly related to the percentage of time spent in SB, (F(1,50)=4.7, *p*<.05) and MVPA (F(1,50)=6.1, *p*<.05).

The Appendix shows the count-based PA parameters for the vertical axis (VA) and the combined triaxial vector magnitude (VM), both normalized to wear time, for all children and by GMFCS level (Figure 6.A, A), and by age group (Figure 6.A, B).

Nighttime activities: sleep and wakefulness

From the total of 54 children, three participants did not meet the minimum number of valid monitoring nights (≥4 of which 1 weekend night) and were excluded from the sleep analysis. Of the remaining children, a total of 418 nights were recorded, of which 78 nights (deriving from 24 children) were excluded due to absence of actigraphy data (*n*=52), sickness (*n*=4), missing sleep diary data to confirm bed times (*n*=6), >10% difference between actigraphy data and bed times stated in the sleep diary (*n*=7), accelerometer fell off during the night (*n*=1), accelerometer was worn around the ankle (*n*=1), or unknown reasons (*n*=7). Therefore, the final sample with valid sleep data comprised 51 children, from which a total of 340 nights were included in the sleep analysis. On average, children had 6.7±0.8 monitored nights (61% had 7 nights of monitoring; 33% wore the device for 5–6 nights, and 6% wore the device for 8–9 nights). The total time children spent in bed (TIB, in hours) are displayed in Figure 6.2, represented by the total length of the bars. The sleep algorithm further divided TIB into time spent trying to fall asleep (SOL), time asleep (TST), or time being awake (WASO), represented by the different colours. Data are presented for all individual children with valid sleep data (*n*=51) clustered by age group in Figure 6.2A, as well as for subgroups across GMFCS level (Figure 6.2B), and age group (Figure 6.2C).

On average, children spent 11.0±0.2 hours in bed (TIB), of which 9.0±0.9 hours asleep (TST), 1.6±0.6 hours awake (WASO), with a sleep latency (SOL) of 0.3±0.2 hours (Table 6.2). The majority (78.4%) of the children had an average SOL of <30 minutes. The sleep efficiency (SE) of the total study sample was 82.4±6.0 %, with *n*=21 (41.2%) children having an average SE above 85%.

GMFCS level

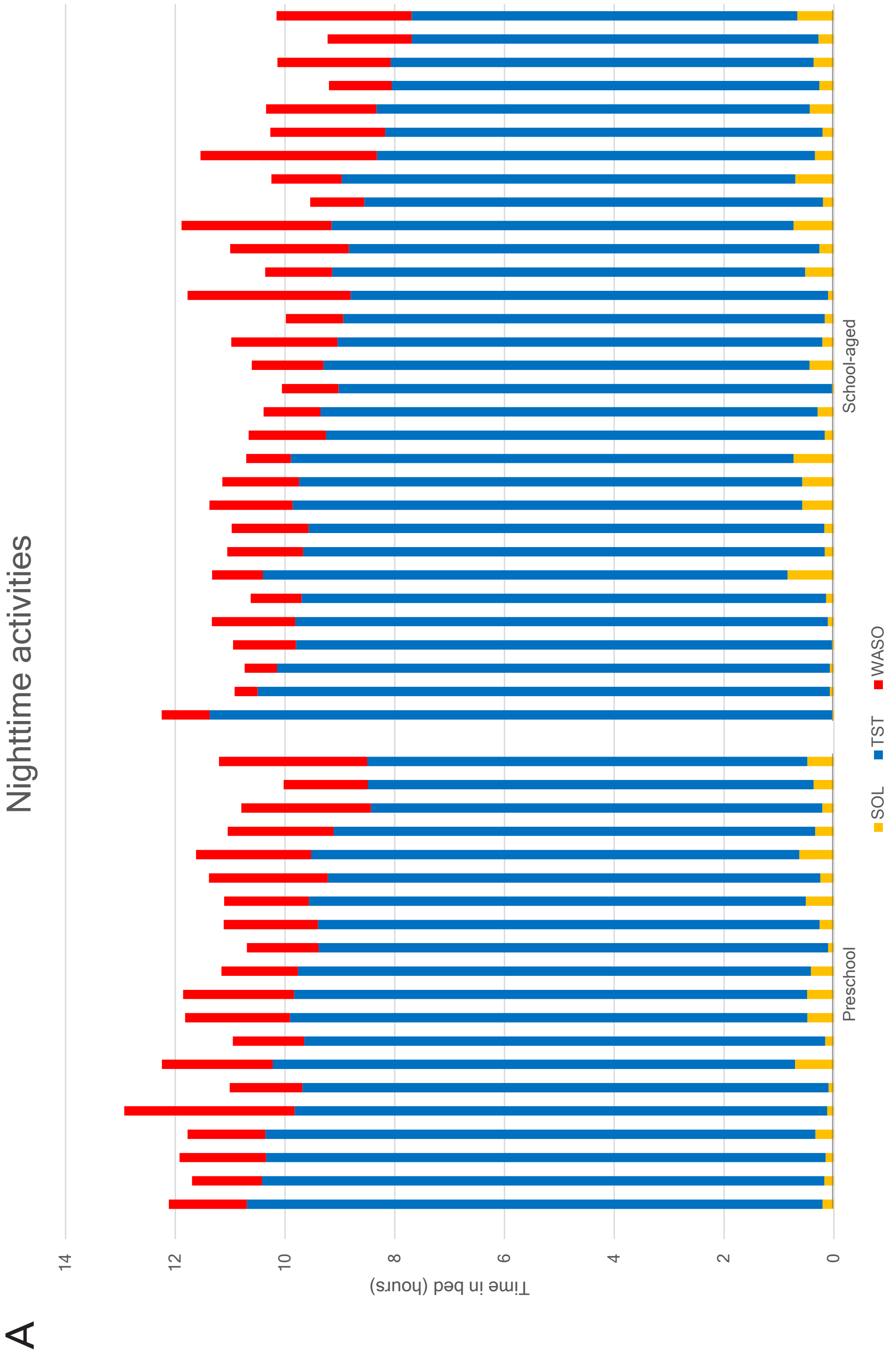
No significant differences were observed between any of the sleep parameters for GMFCS level, after adjusting for age and sex (Table 6.2 and Figure 6.2B).

Age group

After controlling for GMFCS level and sex, a significant effect of age group was found on TIB (F(1, 47)=13.7, *p*=.001), with preschool-aged children spending more hours in bed (mean 11.4±0.6 hours) on average than school-aged children (mean 10.7±0.7 hours) (Figure 6.2C). Despite these differences, no differences were found between age groups on average TST and SOL (both: *p*>.05). Specifically, preschool-aged children spent on average 9.3±0.7 hours asleep compared to 8.9±0.9 hours for school-aged children. Preschool-aged children trended towards spending a greater amount of time (108±30.5 min) awake (i.e. WASO) than school-aged children (89.6±41.5 min), but this difference failed to reach significance (F(1, 47)=3.6, *p*=.06).

Adherence to the 24-hour activity guidelines

Figure 6.3 shows the proportion of children meeting individual components (i.e. the PA recommendations and age-appropriate sleep recommendations) as well as the combined 24-hour activity guidelines for children with CP,[10] organized by GMFCS level (Figure 6.3A) and age group (Figure 6.3B).


A
Nighttime activities
Time in bed (hours)
0
2
4
6
8
10
12
14
Preschool
School-aged
SOL
TST
WASO

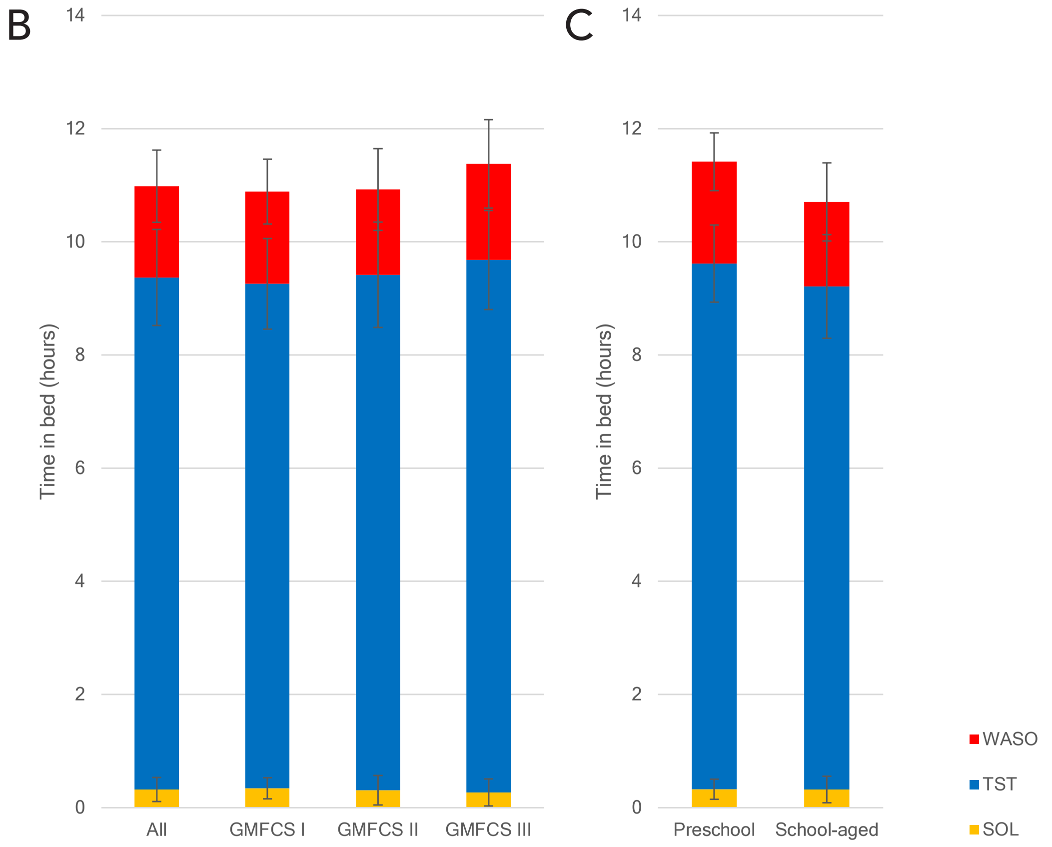


Figure 6.2 Total time in bed (hours) spent in SOL, TST, and WASO for (A) all individual participants sorted by age group, and subgroup data across (B) GMFCS level and (C) age group. Abbreviations: SOL = sleep onset latency; TST = total sleep time; WASO = wakefulness after sleep onset.

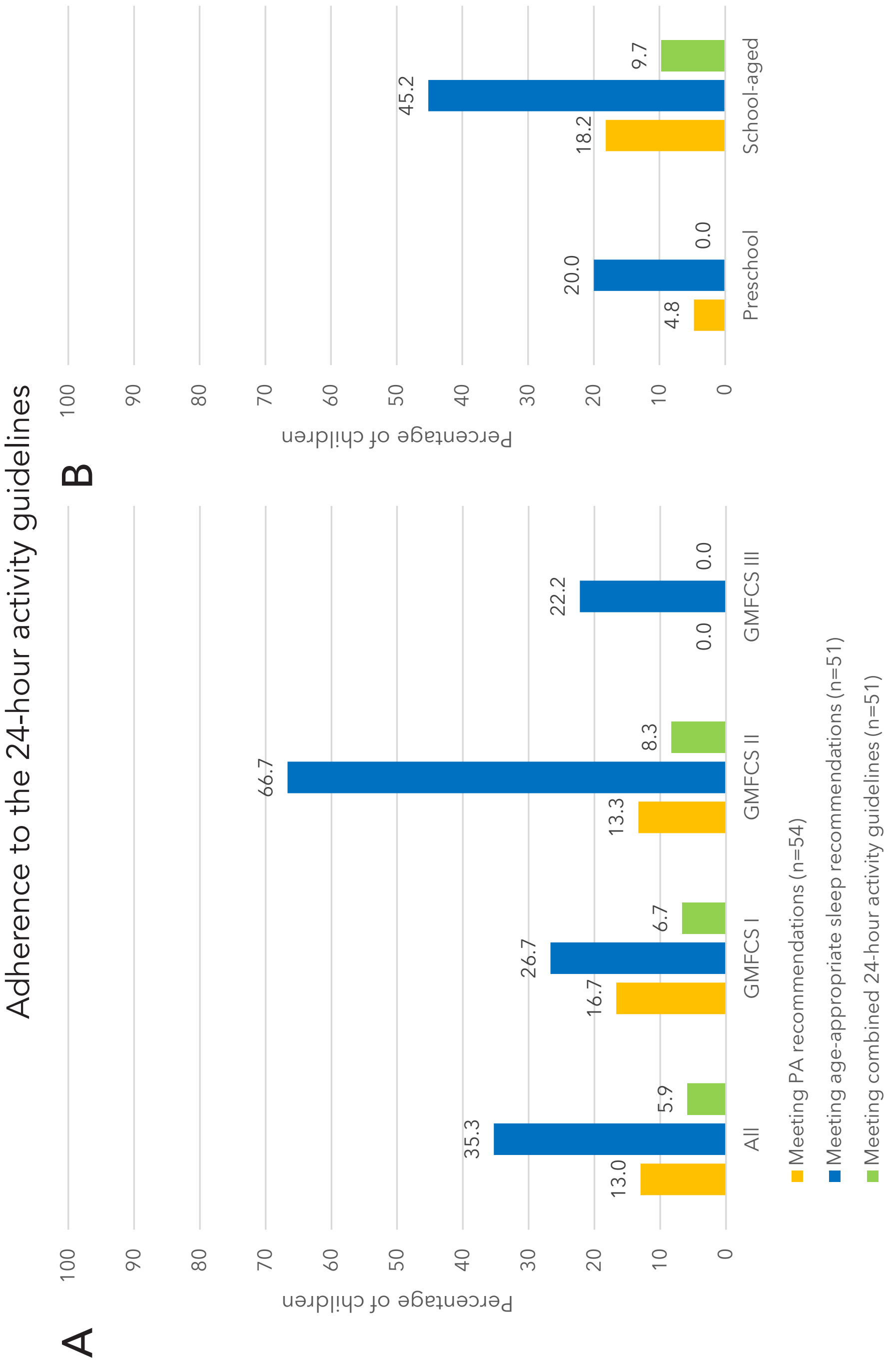


Figure 6.3 Adherence to individual components of, and to the combined 24-hour activity guidelines for children with CP for **(A)** all children and by GMFCS level and **(B)** by age group.

Of all children with CP, looking at the individual components of the 24-hour activity guidelines, a small proportion (13%) met the PA recommendations, and one-third (35.3%) met the age-appropriate sleep recommendations. In all cases, not meeting the sleep recommendations was due to shortened sleep duration (i.e., too little sleep rather than too much sleep). More than half (56.9%) of all children with CP did not meet the combined recommendations for both PA and sleep, while a mere *n*=3 (5.9%) children met both the PA and sleep recommendations (Figure 6.3A).

GMFCS level

Significant group differences were observed between GMFCS levels regarding the proportion of children who did not meet either the PA or age-appropriate sleep recommendations (p<.05; Fisher's Exact), and the proportion of children meeting the sleep recommendations alone (p<.05; Fisher's Exact, Figure 6.3A). Children classified in GMFCS level II showed the highest adherence rates for meeting the age-appropriate sleep recommendations. GMFCS level III showed the highest proportion of children who did not comply with the PA or sleep recommendations (77.8%), and the lowest number of children meeting the PA recommendations alone (0%), meeting the sleep recommendations alone (22.2%), and meeting the combined 24-hour activity guidelines (0%).

Age group

With regard to age groups, a higher proportion of preschool aged children (75%) failed to meet both the PA recommendations as well as the sleep recommendations, compared to 45.2% of school-aged children (p<.05; Fisher's Exact). Of the younger group, a mere 4.8% met the PA recommendations, one in five (20%) met the age-appropriate sleep duration recommendations, and none of them complied with the combined 24-hour activity guidelines in their entirety (Figure 6.3B). Of the school-aged children, 18.2% met the PA recommendations, 45.2% met the recommended hours of sleep, and 9.7% complied with the combined 24-hour activity guidelines.

DISCUSSION

The current study describes the 24-hour activity behaviours (i.e. PA, SB, and sleep) of ambulatory children with CP (GMFCS level I–III) aged 3–12 years, and their adherence to the 24-hour activity guidelines for PA and sleep, using device-based measures. We found that the proportion of children with CP meeting the 24-hour activity guidelines is low, with an overall adherence rate of 5.9%. The vast majority (87%) of ambulatory children with CP are not engaging in sufficient MVPA to meet the PA recommendations, and two-thirds (64.7%) do not meet the age-appropriate recommended hours of sleep. Based on these findings, it is clear that awareness, knowledge, and/or adoption of the 24-hour activity guidelines in pediatric rehabilitation is needed to optimize health outcomes and well-being in individuals with CP over the life course.

Our findings support a body of literature demonstrating high levels of physical inactivity among children with CP, with decreasing amounts of PA with increasing GMFCS level.[14,21] The 24-hour activity guidelines recommend 60 minutes of MVPA per day for children with CP,[10] which was achieved by 13% of the children in our study. These findings are consistent with a recent study reporting a 19% PA guideline compliance rate among a mixed sample of children with one or more (neuro)developmental disabilities.[26] Notably, results from the Dutch Report Card+ showed that the percentages of children meeting the PA guidelines were similar for children with (26%) and without disabilities (29%),[27] suggesting that the MVPA recommendations are often not met across pediatric populations, including TD children. Similar to what has been observed in TD children,[28] we found PA to be the behaviour least complied with across all subgroups. This is especially notable given that PA, and specifically MVPA, is most consistently associated with desirable health indicators in school-aged children and youth.[2,29] For young persons with CP, the consequences of low levels of PA may be even more devastating. For example, higher functioning children with CP are at increased risk to lose ambulatory skills in adulthood, which has been attributed to both chronic inactivity and secondary complications common among adults with CP such as pain, fatigue, loss of strength, balance and reduced physical fitness.[30] Our findings underscore the continued need for more efforts towards stimulating PA in children with CP, for example by creating opportunities for them to acquire more active lifestyles and by increasing awareness among parents of the importance of exercise as a health-promoting behaviour with long-term implications for health maintenance.

Time during a 24-hour period can only be spent engaging in one activity behaviour at a time (i.e., if time spent in one activity is increased, there is less time available in the day for the remaining activity domains).[10] Therefore, in addition to focusing on increasing PA, part of achieving a more active lifestyle includes reducing sedentary time. Children in our study spent on average 61% of their time engaging in SB. This proportion is lower than the previously reported 79% SB in ambulatory children with CP aged 8–17 years,[5] which could be explained by our much younger sample of children. This suggestion is supported by our finding showing that the preschool-aged children engaged in significantly more LPA (at the expense of SB) compared to school-aged children. In this context, it is important to realize that too much sedentary time is distinct from too little PA. Reducing and/or breaking up sedentary time (i.e. replacing SB with LPA) may be easier for some to achieve than increasing higher intensity PA levels, and can have substantial (additional) health benefits. Given that both PA and SB are modifiable behaviours known to track from early childhood into adulthood,[31] they represent viable targets of intervention for children with CP. For those with high levels of inactivity, aiming to improve behaviour in one domain can have a positive effect on the activity composition of the entire 24-hour period.

Our study further adds to the literature by adopting a 24-hour lens, specifically integrating measures of sleep, which has not been considered in earlier studies in children with CP. We found that only one-third of the children in our study met the age-appropriate sleep duration recommendations. This is much lower than reported in a previous study,[32] in which over 81% of the preschool-aged and 92% of the school-aged children with CP adhered to the sleep recommendations. However, it should be noted that this study used subjective tools (parent-reported questionnaires) to measure sleep duration. While self/parent-reported sleep outcomes can provide important insights into sleep perception, these measures can overestimate sleep duration, making it difficult to accurately compare findings across studies.[17] It is also important to note that actigraphy measures movement (i.e., accelerations) and not sleep, per se. Thus, activity during the night scored as wakefulness, may also reflect body movements in bed (e.g. spasms, restlessness or other types of involuntary/unconscious movements) while the child is sleeping, which may be more pronounced in children with CP. It is recommended to use sleep diaries as an important adjunct to accelerometer-based sleep measures.

As the 24-hour activity guidelines for children with CP were adapted from those originally developed for TD children, the question remains to what extent these guidelines are applicable for children with physical or other disabilities. Nevertheless, there is evidence for a dose-response pattern between the number of recommendations achieved within the 24-hour guidelines and the odds of better health outcomes for children with and without disabilities.[3,13] None of the 24-hour activity behaviours are independent of each other, and there are indications of a mutually beneficial relationship between the different behaviors. For example, in children with autism spectrum disorder, improved sleep outcomes (i.e. fewer wake minutes and higher sleep efficiency) were associated with more minutes of MVPA the following day,[33] while increased PA was favorably associated with restoring sleep in a non-clinical sample of adolescents.[34] Future research is needed to explore the complex bidirectional relationship between the 24-hour activities in children with CP, and the health outcomes associated with meeting the individual and combined 24-hour activity guidelines. Nonetheless, increasing guideline adherence is likely to be clinically beneficial in a population that is at risk of various secondary health complications. Given the physical challenges that children with CP and their parents face daily, tailored interventions aimed at improving one (or more) of the 24-hour activity behaviours in children with CP should be considered as a viable, initial target towards long-term health benefits.

Strengths and limitations

A strength of this study is that the 24-hour activities in children with CP were measured using device-based measures for both PA and sleep over a period of one week. Although actigraphy is more challenging in children with CP compared to TD children, our relatively high adherence rates (only 5 out of 60 children did not tolerate wearing the device) are consistent with previous research, and suggest that actigraphy is feasible and acceptable in ambulatory children with CP.[20,21]

Our findings must be interpreted in light of some limitations. We did not assess screen time as a marker of sedentary behaviour, and as such, could not assess adherence to the SB recommendations. Children with neurodevelopmental disorders may have more difficulties disengaging from screen devices, and their average screen use duration has been reported to be 4 hours per day.[35] Also, children with CP not only engage in more sedentary time compared to their TD peers, their SB is also characterized by less frequent breaks.[5] This suggests that the overall 24-hour guideline adherence rate may in fact be even lower than the 5.9% reported in the present study, if the SB component were to be taken into account as well.

A second limitation of this study is the lack of children with more severe functional limitations (GMFCS levels IV and V). As non-ambulatory children with CP are known to be more inactive,[4] and more severely affected by sleep problems than ambulatory children with CP,[6] this group of children is expected to have even lower guideline adherence rates. Also, information about comorbidities was not obtained from the children’s medical files, and these may have potentially influenced our outcomes (e.g. excess night-time wakefulness), especially in the higher GMFCS level III.

For the younger children who still needed daytime naps, parents were asked to record nap times in the activity logbook and take off the device, so as not to confuse sleep for sedentary time during PA analysis. As nap times were based on subjective reports, and could not be confirmed using the accelerometer, it should be noted that naps were not accounted for in the TST of the children. This could have caused an underestimation of the TST, and therefore, could have affected the proportion of children meeting the sleep duration recommendations. However, naps were only reported in a relatively small number (9%, all preschool-aged) of children.

Finally, with regard to PA, as participants were instructed to remove the accelerometer during water-activities, this could have led to an underestimation of MVPA in children who participated in swimming (as part of therapy or leisure activity). According to the logbooks, hip-worn devices were taken off for swimming on a total of 19 days from a total of 15 children across all GMFCS levels (I, *n*=9; II, *n*=4; III, *n*=2), resulting in a total 1450-min (average 76 min per swimming episode) that were not accounted for in the PA analysis.

Conclusion

In conclusion, the results of this study contribute to our understanding of the 24-hour activities among children with CP, and draw attention to the importance of considering the entire continuum of movement behaviours (i.e. PA, SB, and sleep) into the (rehabilitative) care for this population. The very low guideline adherence rates underscore the need for tailored intervention strategies aimed at improving the activity behaviours throughout a 24-hour period, including increased PA, limited sitting, and sufficient hours of good quality sleep. Promoting healthy lifestyle behaviours in (early) childhood has the potential to provide children a strong foundation for lifelong physical and mental health, and this may be especially critical for more vulnerable populations like children with CP.

Acknowledgements

This study was kindly supported by a grant from JKF Kinderfonds and Kinderrevalidatiefonds Adriaanstichting, foundations aimed at improving the quality of life of children and adolescents with physical disabilities in The Netherlands.

The authors are grateful to each of the children and their parents for participation in this study. We thank the healthcare professionals from the rehabilitation settings for their kind assistance in participant recruitment, in particular Sanne van der Vossen (De Hoogstraat Revalidatie), Rick van de Ven (Tolbrug/Atlent, Jeroen Bosch Ziekenhuis) and Mirjam Jeurissen (Rijndam Revalidatie). Also, we would like to acknowledge Wibe van Grunderbeek for his assistance with writing macros.

What this paper adds

- Physical activity decreases while sedentary behaviour increases with increasing GMFCS level in children with CP.
- Only 13% of children with CP meets the daily physical activity recommendation.
- Only 35% of children with CP meets the age-appropriate sleep duration recommendation.
- The proportion of children meeting the CP-specific 24-hour activity guidelines is low.

REFERENCES

1. Poitras VJ, Gray CE, Borghese MM, et al. Systematic review of the relationships between objectively measured physical activity and health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016;41(6 Suppl 3):S197–239.
2. Rollo S, Antsygina O, Tremblay MS. The whole day matters: Understanding 24-hour movement guideline adherence and relationships with health indicators across the lifespan. J Sport Heal Sci. 2020;9(6):493–510.
3. Janssen I, Roberts KC, Thompson W. Is adherence to the Canadian 24-Hour movement behaviour guidelines for children and youth associated with improved indicators of physical, mental, and social health? Appl Physiol Nutr Metab. 2017;42(7):725–31.
4. Verschuren O, Peterson MD, Balemans ACJ, Hurvitz EA. Exercise and physical activity recommendations for people with cerebral palsy. Dev Med Child Neurol. 2016;58(8):798–808.
5. Obeid J, Balemans ACJ, Noorduyn SG, Gorter JW, Timmons BW. Objectively measured sedentary time in youth with cerebral palsy compared with age-, sex-, and season-matched youth who are developing typically: An explorative study. Phys Ther. 2014;94(8):1163–7.
6. Hulst RY, Gorter JW, Voorman JM, Kolk E, Vossen S Van Der, Visser-Meily JMA, et al. Sleep problems in children with cerebral palsy and their parents. Dev Med Child Neurol. 2021;63(11):1344–50.
7. 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.
8. Horwood L, Li P, Mok E, Oskoui M, Shevell M, Constantin E. Health-related quality of life in Canadian children with cerebral palsy: what role does sleep play? Sleep Med . 2019;54:213–22.
9. Peterson MD, Ryan JM, Hurvitz EA, Mahmoudi E. Chronic conditions in adults with cerebral palsy. JAMA - J Am Med Assoc. 2015;314(21):2303–5.
10. Verschuren O, Hulst RY, Voorman J, Pillen S, Luitwieler N, Dudink J, et al. 24-hour activity for children with cerebral palsy: a clinical practice guide. Dev Med Child Neurol. 2021;63(1):54–9.
11. Healy S, Foley J, Haegele JA. Physical Activity, Screen Time, and Sleep Duration Among Youth With Chronic Health Conditions in the United States. Am J Heal Promot. 2020;34(5):505–11.
12. Arbour-Nicitopoulos KP, Bassett-Gunter RL, Leo J, Sharma R, Olds T, Latimer-Cheung AE, et al. A cross-sectional examination of the 24-hour movement behaviours in Canadian youth with physical and sensory disabilities. Disabil Health J. 2021;14(1):100980.
13. Brown DMY, McPhee PG, Kwan MY, Timmons BW. Implications of Disability Severity on 24-Hour Movement Guideline Adherence Among Children With Neurodevelopmental Disorders in the United States. J Phys Act Heal. 2021;18(11):1325–31.
14. Mitchell LE, Ziviani J, Boyd RN. Habitual physical activity of independently ambulant children and adolescents with cerebral palsy: Are they doing enough? Phys Ther. 2015;95(2):202–11.

15. Smit DJM, Zwinkels M, Takken T, Hulst RY, de Groot JF, Lankhorst K, et al. Sleep quantity and its relation with physical activity in children with cerebral palsy; insights using actigraphy. J Paediatr Child Health. 2020;56(10):1618–22.
16. Quante M, Kaplan ER, Rueschman M, Cailler M, Buxton OM, Redline S. Practical considerations in using accelerometers to assess physical activity, sedentary behavior, and sleep. Sleep Heal. 2015;1(4):275–84.
17. Lauderdale DS, Knutson KL, Yan LL, Liu K, Rathouz PJ. Self-reported and measured sleep duration: How similar are they? Epidemiology. 2008;19(6):838–45.
18. Reilly JJ, Penpraze V, Hislop J, Davies G, Grant S, Paton JY. Objective measurement of physical activity and sedentary behaviour: Review with new data. Arch Dis Child. 2008;93(7):614–9.
19. Hirshkowitz M, Whiton K, Albert SM, Alessi C, Bruni O, DonCarlos L, et al. National sleep foundation's sleep time duration recommendations: Methodology and results summary. Sleep Heal. 2015;1(1):40–3.
20. Clanchy KM, Tweedy SM, Boyd RN, Trost SG. Validity of accelerometry in ambulatory children and adolescents with cerebral palsy. Eur J Appl Physiol. 2011;111(12):2951–9.
21. Gorter JW, Noorduyn SG, Obeid J, Timmons BW. Accelerometry: A Feasible Method to Quantify Physical Activity in Ambulatory and Nonambulatory Adolescents with Cerebral Palsy. Int J Pediatr. 2012;2012:1–6.
22. Trost SG, Pate RR, Freedson PS, Sallis JF, Taylor WC. Using objective physical activity measures with youth: How many days of monitoring are needed? Med Sci Sports Exerc. 2000;32(2):426–31.
23. Evenson KR, Catellier DJ, Gill K, Ondrak KS, McMurray RG. Calibration of two objective measures of physical activity for children. J Sports Sci. 2008;26(14):1557–65.
24. Mitchell LE, Ziviani J, Boyd RN. Variability in measuring physical activity in children with cerebral palsy. Med Sci Sports Exerc. 2015;47(1):194–200.
25. Sadeh A, Sharkey KM, Carskadon MA. Activity-based sleep-wake identification: An empirical test of methodological issues. Sleep. 1994;17(3):201–7.
26. Case L, Ross S, Yun J. Physical activity guideline compliance among a national sample of children with various developmental disabilities. Disabil Health J. 2020;13(2):100881.
27. Burghard M, de Jong NB, Vlieger S, Takken T. 2017 Dutch Report Card+: Results from the first physical activity report card plus for Dutch youth with a chronic disease or disability. Front Pediatr. 2018;6:122.
28. De Craemer M, McGregor D, Androutsos O, Manios Y, Cardon G. Compliance with 24-h movement behaviour guidelines among belgian pre-school children: The toybox-study. Int J Environ Res Public Health. 2018;15(10):1–10.
29. Saunders TJ, Gray CE, Poitras VJ, Chaput JP, Janssen I, Katzmarzyk PT, et al. Combinations of physical activity, sedentary behaviour and sleep: Relationships with health indicators in school-aged children and youth. Appl Physiol Nutr Metab. 2016;41(6):S283–93.
30. TURK MA. Health, mortality, and wellness issues in adults with cerebral palsy. Dev Med Child Neurol. 2009;51(SUPPL. 4):24–9.
31. Telama R, Yang X, Leskinen E, Kankaanpää A, Hirvensalo M, Tammelin T, et al. Tracking of physical activity from early childhood through youth into adulthood. Med Sci Sports Exerc. 2014; 46(5):955–62.
32. Horwood L, Mok E, Li P, Oskoui M, Shevell M, Constantin E. Prevalence of sleep problems and sleep-related characteristics in preschool- and school-aged children with cerebral palsy. Sleep Med. 2018;50:1–6.
33. Benson S, Bender AM, Wickenheiser H, Naylor A, Clarke M, Samuels CH, et al. Differences in sleep patterns, sleepiness, and physical activity levels between young adults with autism spectrum disorder and typically developing controls. Dev Neurorehabil. 2019;22(3):164–73.
34. Lang C, Brand S, Feldmeth AK, Holsboer-Trachsler E, Pühse U, Gerber M. Increased self-reported and objectively assessed physical activity predict sleep quality among adolescents. Physiol Behav. 2013;120:46–53.
35. Lin J, Magiati I, Chiong SHR, Singhal S, Riard N, Ng IHX, et al. The Relationship among Screen Use, Sleep, and Emotional/Behavioral Difficulties in Preschool Children with Neurodevelopmental Disorders. J Dev Behav Pediatr. 2019;40(7):519–29.

APPENDIX

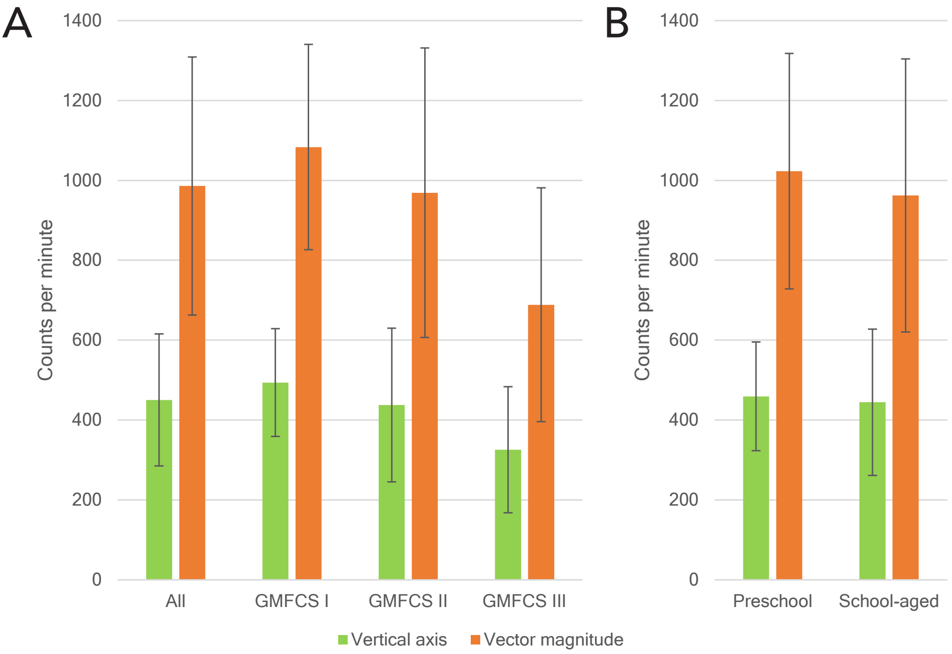


Figure 6.A Count-based physical activity parameters.
Normalized count-based PA parameters for the vertical axis (VA) and combined triaxial vector magnitude (VM) for **(A)** all individual participants and across GMFCS level; and **(B)** by age group. On average, children accumulated 450.2 (±165.2) counts per minute (cpm) for VA, and a total average of 985.8 (±322.9) cpm for VM. After accounting for sex and age, a significant effect of GMFCS level was found on both VA cpm (F(2, 49)=5.2, *p*<.01) and on VM cpm (F(2, 49)=8.7, *p*=.001). Children classified at GMFCS level I displayed the highest VA (493.8±135.1 cpm) and VM (1083.4±257.0 cpm), whereas those classified at GMFCS level III showed the lowest values (VA cpm: 325.4±157.9; VM cpm 688.3±292.7). The covariate age was significantly related to the average VM cpm (F(1, 49)=6.4, *p*<.05), but not to VA cpm (*p*>.05). No other group differences were observed for the count-based PA data.

Chapter 7

Device-based and subjective measurements of sleep in children with cerebral palsy: A comparison of sleep diary, actigraphy, and bed sensor data

Ilse M van Rijssen, Raquel Y Hulst, Jan Willem Gorter, Anke Gerritsen,
Johanna MA Visser-Meily, Jeroen Dudink, Jeanine M Voorman, Sigrid Pillen,
Olaf Verschuren

Under review.

ABSTRACT

Objective: To investigate how subjective assessments and device-based measurements of sleep relate to each other in children with cerebral palsy (CP).

Methods: Sleep of children with CP classified at Gross Motor Function Classification System (GMFCS) levels I-III, was measured during 7 consecutive nights using one subjective (i.e. sleep diary), and two device-based (i.e. actigraphy and bed sensor) instruments. The agreement between the instruments was assessed by intraclass correlation coefficients (ICC) and Bland-Altman plots.

Results: A total of 227 nights from 38 children with CP (53% boys; median age [range] 6 [2–12] years), were included in the analyses. We found poor agreement for all parameters between i) actigraphy and sleep diary, and ii) between actigraphy and bed sensor, except for Total Bed Time (ICC>0.86 and ICC>0.84, respectively). Furthermore, iii) the agreement between sleep diary and bed sensor was satisfactory for Total Bed Time (ICC>0.83), Total Sleep Time (ICC>0.70), and Wakefulness After Sleep Onset during weekend nights (ICC=0.55). Other sleep parameters showed poor agreement between sleep diary and bed sensor.

Conclusions: Researchers and clinicians need to be aware of the discrepancies between instruments for sleep monitoring in children with CP. We recommend combining both subjective and device-based measures to provide information on the perception as well as an unbiased estimate of sleep. Further research needs to be conducted on the use of a bed sensor for sleep monitoring in children with CP.

INTRODUCTION

Increasing evidence shows that the prevalence of sleep problems in children with cerebral palsy (CP) exceeds that in typically developing (TD) children.[1–4] Sleep problems in children with CP include sleep-wake transition disorders, parasomnias, disorders of initiation and maintenance of sleep, among others.[3,5] Poor quality or insufficient sleep can negatively impact (health-related) quality of life in children with CP,[6,7] and greatly affect parental well-being or family functioning.[8–10]

Accurately assessing sleep in children with CP within a routine follow-up or monitoring clinic is an important step for the identification and management of sleep problems. A variety of subjective and device-based tools have been used in children with CP and are available to clinicians. Subjective tools, such as sleep diaries or questionnaires, offer insight into the parent- and self-reported perception of sleep.[11,12] Subjective methods can be used in combination with device-based methods, such as polysomnography (PSG) or accelerometry.[13] Although PSG is considered the gold standard for characterizing sleep rhythms, it is a relatively costly procedure performed in an unfamiliar environment such as a sleep clinic and is often used for only one night. In contrast, an accelerometer, a watch-like device containing a highly sensitive sensor, can continuously measure body movements and estimate sleep-wake patterns using activity-based algorithms.[14] This method is less intrusive and less expensive than PSG. It can also measure sleep over extended periods in the home environment. Nevertheless, the accelerometer is a physically worn device and has limited battery life, which requires frequent recharging. Furthermore, the analysis of accelerometer data is time consuming, hindering its implementation in clinical practice.

Over the past years, several new methods for sleep monitoring have been introduced. One of these developments is the bed sensor; a mattress for unobtrusive and contactless analysis of sleep.[15–17] A bed sensor is not dependent on batteries and can be used as long as there is electricity in the home. The technology behind the bed sensor is based on ballistocardiography, which combines measurements of heart rate, respiratory rate, and movement activity to assess sleep quantity and quality.[18] This technique has been implemented and validated in diagnosing sleep-disordered breathing,[19,20] and has been deemed a promising device for continuous sleep monitoring in adults.[21]

Different methods for assessing sleep quantity and quality in children are prone to inconsistencies in measuring sleep parameters.[22] Studies in TD children.[23,24] as well as in children with insomnia,[25] anxiety disorder,[26] and epilepsy,[27] consistently describe a discrepancy between subjective and device-based measurements of sleep. In children with

CP, neuromuscular impairments such as hypertonicity, are likely to cause heterogeneity in their sleeping postures.[28] This might affect sleep monitoring when using movement-based methods and can therefore influence the agreement between instruments. However, it is unclear how subjective and device-based instruments relate to each other when used in children with CP. Therefore, the objective of our study is to examine the agreement between sleep diary, actigraphy and bed sensor data in children with CP. Based on our findings, we aim to inform researchers and practitioners about the differences between subjective and device-based measurements of sleep and to formulate recommendations for the use of these methods in research and clinical practice.

METHOD

A cross-sectional, observational study design was used. The study protocol was approved by the Medical Ethics Research Committee of the University Medical Centre Utrecht (file number 19-630), the Netherlands.

Participants

Children with CP were recruited via five pediatric healthcare settings (three rehabilitation centres, a school for special education, and the rehabilitation department of a children's hospital) in the Netherlands. Children were not recruited based on existing sleep problems. All participants were children with CP classified as Gross Motor Function Classification System (GMFCS) levels I-III and between the age of 2 and 12 years. Children were excluded when parents had insufficient knowledge of the Dutch language, such that measurements were not possible or required an excessive amount of time.

Parents of children with CP were informed about the study employing an information brochure and were subsequently asked for permission by their treating physician to be contacted by the researcher. All parents and children aged 12 years provided written consent/assent prior to participation in the study. Parents were asked to answer general questions on the experience of sleep problems and the use of sleep medication.

Data collection

Sleep was measured for one week (5 school days, 2 weekend days) in the child's home environment during a representative (non-holiday) period. Sleep was measured using three instruments: (i) a sleep diary, (ii) an accelerometer, and (iii) a bed sensor. Data were collected between January 2020 and January 2021. Due to the COVID-19 pandemic, data collection was paused during the school lockdown period from mid-March to mid-April 2020.

Instruments

Subjective sleep assessment

Sleep diary

In the sleep diary, each night was represented by a continuous timeline divided into boxes, with each box corresponding to 15 minutes. Parents were told to indicate awake in bed phases with shaded boxes and sleep phases with coloured boxes. They were also instructed to write down the presumed number of nocturnal awakenings and any other particularities such as an intercurrent illness of the child.

Parameters derived from the sleep diary (see Table 7.1) included: (1) Total Bed Time (TBT), defined by the time of lights out until getting out of bed; (2) Total Sleep Time (TST), defined by all night time sleep (excluding daytime naps); (3) Sleep Onset Latency (SOL), the delay between lights out and sleep onset; (4) Wakefulness After Sleep Onset (WASO), the total time awake between sleep onset and getting out of bed; (5) Awakenings, the indicated number of nocturnal awakenings; and (6) Sleep Efficiency (SE), the ratio of TST to TBT (in %). See Figure 7.1 for a schematic diagram of data extracted for the time in bed.

Table 7.1 Sleep parameters for each instrument

	Subjective	Device-based	
	Sleep diary	Actigraphy	Bed sensor
Total bed time	X	X*	X
Total sleep time	X	X	X
Sleep onset latency	X	X	X
Wakefulness after sleep onset	X	X	X
Awakenings	X	X	X
Sleep efficiency	X	X	X
Bed exits	X	-	X
Sleep architecture (REM/non-REM)	-	-	X
Tossing & turning	-	-	X
Sleep perception parents	X	-	-
Sleep perception child	X	-	-

* Derived from times noted in the sleep diary.

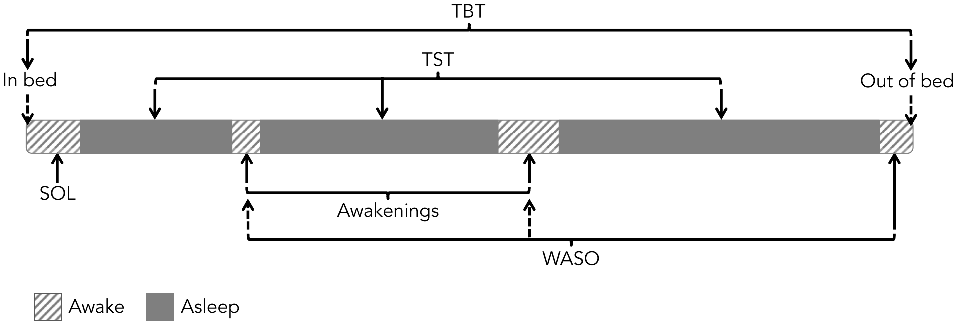


Figure 7.1 Representation of a night containing all sleep parameters. Abbreviations: TBT = Total Bed Time; TST = Total Sleep Time; SOL = Sleep Onset Latency; WASO = Wakefulness After Sleep Onset.

Device-based sleep assessment

Actigraphy

The Actigraph GT3X accelerometer (ActiGraph Corporation, Pensacola, FL, USA) was initialized at a 30Hz sampling rate and 15 seconds epoch length using the ActiLife software (version 6, ActiGraph Corporation, Pensacola, FL, USA). Parents were instructed to attach the Actigraph to their child's non-dominant wrist in the evening before bedtime and to remove it in the morning after waking up. The sleep diary was used to mark the start ('lights out' time) and end ('out of bed' time) of the sleep period. Data were extracted and converted into 60 seconds epoch lengths. Nights were excluded when technical malfunctions caused a deviation of more than 10% between sleep times identified by the algorithm and bedtimes entered in the sleep diary.

The following sleep parameters were extracted: (1) TBT, total time spent in bed was calculated using the times of the nearest activity around 'lights out' and 'out of bed' times from the sleep diary, so that no clear non-wear periods were invalidly scored as sleep; (2) TST, the estimated time scored as sleep within the TBT according to the Sadeh algorithm[14]; (3) SOL, amount of time elapsed between bedtime ('lights out') and the first epoch of scored sleep; (4) WASO, number of minutes scored as wakefulness during TST including lying awake in the morning; (5) Awakenings, number of awakenings during TBT; and (6) SE, which was calculated as the ratio of TST to TBT.

Bed sensor

A portable bed sensor, the *Emfit*® QS sleep tracker (Emfit Ltd, Vaajakoski, Finland; 542mm x 70mm x 1.4mm) has recently been validated in healthy adult participants using an electrocardiography-based reference device. The Emfit QS showed good validity and reliability in measuring heart rate during the sleep period.[29] The portable bed sensor was placed underneath the bed mattress at the thoracic area of the child.. The bed sensor was installed by parents and automatically registered sleep. Sleep data were automatically uploaded to the secured Emfit server (accessible at https://qs.emfit.com) after each sleep period.

The Emfit bed sensor epoch length was 30 seconds. The bed sensor automatically provided data on (1) TBT, the total time spent in bed; (2) TST, total time spent asleep; (3) SOL, amount of time elapsing from bedtime to the first moment of sleep; (4) Awakenings, number of awakenings during the sleep period. WASO was calculated using the raw time-stamps for awake and sleep periods, and included the number of wakeful minutes during TST and in the morning before getting out of bed. SE was again calculated as the ratio of TST to TBT. Nights were excluded based on a >10% deviation from TST according to the sleep diary, due to technical malfunctions (such as wireless internet disconnection) or a change in the child's sleeping location.

Data analysis

Only nights with a complete set of three instruments per child were included. Analyses were performed on sleep measurement data of children with at least four nights including at least one weekend night (i.e. Friday on Saturday, or Saturday on Sunday).

Analyses between instruments were performed between means across multiple nights per child. Normality was visually examined using QQ plots. For comparison between instruments, repeated measures ANOVAs were conducted on each sleep parameter with "Instrument" as within subjects' factor. If a significant main effect of "Instrument" was found, Post Hoc tests were conducted for pairwise comparison using a Bonferroni correction. Intraclass correlations (ICC) estimates and their 95% confidence intervals (CIs) were calculated to assess agreement between the three instruments using the guidelines by Koo & Li.[30] Bland Altman plots were used as an extra control method to visually examine the agreement between two measurements at a time.[31] The ideal Bland-Altman plot would demonstrate narrow limits of agreement, around a mean bias of zero.

As in previous research differences were found in the comparison between instruments during school- and weekend nights,[23] all analyses for the current research were run for all nights, and separately for school- and weekend nights. Statistical analyses were performed with R version: 3.6.3.[32] The level of statistical significance was set at $p<.05$.

RESULTS

A total of 60 children were measured. Fourteen children were excluded due to one missing instrument: two children did not fill out the sleep diary, three children had missing

Actigraph data, and nine children had less than four nights registered by the bed sensor. Three children were excluded based on two missing instruments: two children had missing Actigraph data and less than four nights of the bed sensor, and one child self-reported the sleep diary and had missing Actigraph data. Additionally, four children had no weekend night registered by the bed sensor, and one child did not have a weekend night registered by both Actigraph and bed sensor.

A total of 38 children had a complete set of a sleep diary, actigraphy and bed sensor data and were included in the analyses. The final dataset included 227 nights, with a mean number of six nights (range: 4–9) per child. Other group characteristics are summarized in Table 7.2.

Table 7.2 Group characteristics

Characteristic	*N*=38
Sex, *n* (%)	
Boy	20 (53)
Age, y	
Median (range)	6 (2–12)
GMFCS level, *n* (%)	
I	24 (63)
II	9 (24)
III	5 (13)
CP type, *n* (%)	
Spastic	35 (92)
Dyskinetic	1 (3)
Mixed	1 (3)
Unclassified/unknown	1 (3)
Affected side, *n* (%)	
Unilateral	25 (66)
Bilateral	13 (34)
Reported sleep problems, *n* (%)	10 (26)
Reported use of melatonin, *n* (%)	4 (11)

Comparison of sleep parameters assessed by the different instruments

Mean (SD) of TBT, TST, SOL, WASO, awakenings and SE measured by the three different instruments are presented in Table 7.3. Repeated measures ANOVA performed on TBT and SOL showed no significant main effect of “Instrument” considering all nights, school nights and weekend nights.

When considering TST, a significant main effect of Instrument ($F_{2,108}$=8.72, p<.001) was observed for all nights. Pairwise comparisons revealed significant differences between all

Table 7.3 Mean, SD and range for sleep parameters according to sleep diary, actigraphy or bed sensor during all nights, school nights and weekend nights

	Sleep diary Mean (SD) [range]	Actigraphy Mean (SD) [range]	Bed sensor Mean (SD) [range]
All nights			
TBT (hours)	11.22 (0.80) [8.69–12.63]	10.98 (0.72) [8.86–12.12]	11.12 (0.67) [9.01–12.86]
TST (hours)	10.46 (0.73) [8.69–11.65]	9.01 (0.85) [6.89–10.52]	10.09 (0.66) [8.24–11.76]
SOL (min)	25.40 (19.92) [0–90.00]	19.34 (14.11) [2.17–57.00]	31.98 (6.36) [21.88–46.70]
WASO (min)	19.85 (24.13) [0–97.50]	99.23 (44.48) [29.60–214.67]	38.97 (19.29) [17.65–104.11]
Awakenings (#)	0.60 (0.73) [0–2.50]	23.50 (6.29) [12.00–43.43]	1.03 (0.88) [0.17–4.83]
SE (%)	93.43 (4.29) [83.08–100.00]	82.19 (7.25) [64.52–94.75]	90.60 (2.15) [85.89–93.78]
School nights			
TBT (hours)	11.15 (0.73) [9.13–12.50]	11.94 (0.65) [9.43–11.91]	11.10 (0.15) [9.78–12.63]
TST (hours)	10.42 (0.78) [8.65–11.69]	8.99 (0.83) [7.01–10.40]	10.08 (0.66) [8.58–11.70]
SOL (min)	27.38 (22.92) [0–96.00]	20.00 (14.76) [1.25–58.33]	32.71 (6.21) [20.83–47.88]
WASO (min)	16.34 (19.35) [0–75.00]	97.00 (42.04) [32.67–213.75]	37.99 (16.95) [15.33–90.21]
Awakenings (#)	0.54 (0.66) [0–2.4]	23.76 (6.17) [12.25–40.20]	0.98 (0.77) [0–3.75]
SE (%)	93.54 (4.48) [81.56–100.00]	82.31 (7.04) [64.67–94.15]	90.55 (2.15) [85.34–93.71]
Weekend nights			
TBT (hours)	11.39 (1.17) [8.13–13.25]	11.11 (1.03) [7.88–13.13]	11.23 (1.10) [8.25–13.43]
TST (hours)	10.59 (0.94) [7.63–12.25]	9.10 (1.10) [6.42–10.83]	10.17 (0.97) [7.53–12.00]
SOL (min)	19.93 (16.57) [0–60.00]	17.49 (16.00) [0–67.00]	30.12 (11.38) [12.00–52.17]
WASO (min)	27.96 (36.99) [0–142.50]	103.03 (59.78) [25.00–248.00]	38.92 (30.21) [3.73–154.71]
Awakenings (#)	0.58 (0.93) [0–4.00]	23.44 (8.54) [10.50–51.50]	1.11 (1.30) [0–7.00]
SE (%)	93.29 (5.02) [76.42–100.00]	82.17 (8.76) [61.60–95.64]	90.98 (3.25) [81.87–95.53]

Abbreviations: TBT, Total Bed Time; TST, Total Sleep Time; SOL, Sleep Onset Latency; WASO, Wakefulness After Sleep Onset; SE, Sleep Efficiency.

three instruments. Parents reported significantly longer TST compared to actigraphy or bed sensor, and the bed sensor measured longer TST compared to actigraphy (all *ps*<.001). This discrepancy was also found when separately assessing school- and weekend nights ($F_{2,108}$=9.56, *p*<.001; $F_{2,108}$=4.53, *p*<.05, respectively).

Regarding the number of awakenings and WASO, a significant main effect of "Instrument" was observed for all nights ($F_{2,106}$=112; $F_{2,108}$=15.39). The differences between instruments remain when measuring school nights ($F_{2,105}$=118.8; $F_{2,108}$=21.02), and weekend nights ($F_{2,105}$=65.02; $F_{2,108}$=7.45) separately (all *ps*<.001). The Actigraph significantly reported more awakenings and WASO, compared to both sleep diary and bed sensor (all *ps*<.001). During all nights and school nights, parents reported lower numbers of awakenings and WASO compared to the bed sensor (all *ps*<.05). However, during the weekend, there were no significant differences between the number of awakenings and WASO reported by parents or bed sensor (*p*=.09; *p*=.1, respectively).

For SE, a significant main effect of "Instrument" was found for all nights and school nights ($F_{2,108}$=10.09; $F_{2,108}$=11.43; *p*<.001), as well as during weekend nights ($F_{2,108}$=6.92, *p*<.01). The Actigraph showed a lower SE compared to sleep diary and bed sensor (all *ps*<.001). Furthermore, the bed sensor indicated a lower SE in comparison with the sleep diary (all *ps*<.01).

Agreement between the instruments

TBT showed good agreement between all three instruments irrespective of days of measuring (ICC values between 0.83–0.88, see Table 7.4). The Bland-Altman plots also revealed satisfactory levels of agreement between the instruments for TBT (see Figure 7.2). The bias remained constant between all nights, school nights and weekend nights. Almost all data fell between the limits of agreement. During weekend nights the limits of agreement were generally wider.

TST demonstrated moderate to good agreement between the sleep diary and bed sensor during all nights, and when school nights and weekend nights were assessed separately (ICC values between 0.70–0.77, see Table 7.4). However, between sleep diary and actigraphy or bed sensor and actigraphy, the ICC values fell between 0.16 and 0.36, describing a poor agreement between these instruments.

Similar results were found for SOL, awakenings and SE, with poor agreements between the instruments (all ICC values < 0.43). For WASO, a moderate agreement was found between sleep diary and bed sensor, but only during weekend nights (ICC = 0.55). Other agreement values for WASO were poor (ICC<0.39).

Table 7.4 ICC (CI) between sleep parameters assessed by sleep diary, actigraphy and bed sensor during all nights, school nights and weekend nights

		All nights		School nights		Weekend nights	
		Sleep diary	Actigraphy	Sleep diary	Actigraphy	Sleep diary	Actigraphy
TBT	Sleep diary	-	0.87 (0.60–0.95)	-	0.86 (0.61–0.94)	-	0.86 (0.69–0.94)
	Bed sensor	0.86 (0.74–0.92)	0.88 (0.76–0.94)	0.84 (0.71–0.91)	0.84 (0.67–0.92)	0.83 (0.70–0.91)	0.86 (0.75–0.93)
TST	Sleep diary	-	0.16 (-0.09–0.46)	-	0.17 (-0.09–0.47)	-	0.23 (-0.10–0.55)
	Bed sensor	0.72 (0.21–.89)	0.21 (-0.10–0.51)	0.70 (0.32–0.86)	0.20 (-0.09–0.49)	0.77 (0.38–0.90)	0.36 (-0.07–0.66)
SOL	Sleep diary	-	0.40 (0.12–0.63)	-	0.46 (0.17–0.67)	-	0.22 (-0.10–0.50)
	Bed sensor	0.21 (-0.09–0.48)	0.22 (-0.08–0.50)	0.13 (-0.18–0.42)	0.12 (-0.10–0.36)	0.27 (-0.02–0.53)	0.38 (-0.04–0.67)
WASO	Sleep diary	-	0.05 (-0.06–0.22)	-	0.02 (-0.05–0.13)	-	0.13 (-0.09–0.37)
	Bed sensor	0.39 (-0.02–0.67)	0.13 (-0.09–0.39)	0.21 (-0.08–0.50)	0.09 (-0.08–0.31)	0.55 (0.29–0.74)	0.19 (-0.09–0.47)
Awakenings	Sleep diary	-	0.00 (-0.02–0.03)	-	0.00 (-0.02–0.03)	-	0.00 (-0.03–0.06)
	Bed sensor	0.15 (-0.14–0.43)	0.00 (-0.02–0.03)	0.19 (-0.09–0.47)	0.00 (-0.02–0.03)	0.15 (-0.15–0.44)	-0.01 (-0.04–0.05)
SE	Sleep diary	-	0.05 (-0.07–0.21)	-	0.03 (-0.08–0.18)	-	0.06 (-0.09–0.26)
	Bed sensor	0.26 (-0.04–0.53)	0.08 (-0.09–0.28)	0.18 (-0.08–0.44)	0.05 (-0.09–0.23)	0.43 (0.12–0.66)	0.13 (-0.09–0.37)

Abbreviations: ICC, intraclass correlation; CI, confidence interval; TBT, Total Bed Time; TST, Total Sleep Time; SOL, Sleep Onset Latency; WASO, Wakefulness After Sleep Onset; SE, Sleep Efficiency.

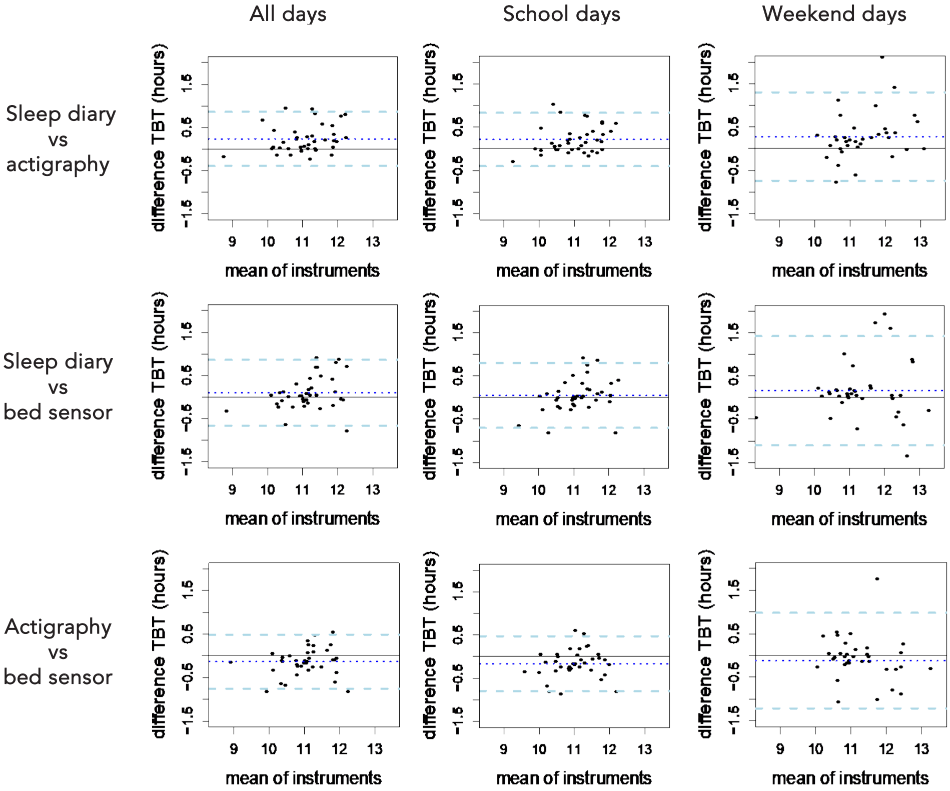


Figure 7.2 Bland-Altman plots to visually assess the bias and limits of agreement of TBT between sleep diary and actigraphy, sleep diary and bed sensor, and actigraphy and bed sensor for all nights, school nights and weekend nights. Y-axis represents the differences between the two measurements; x-axis represents the average TBT (in hours) of the two instruments. The blue horizontal lines represent the bias and the 95% limits of agreement.

DISCUSSION

This study builds upon vast literature on the comparison between subjective and device-based assessments of sleep. The originality of this study stems from the target group of children with CP, for which the agreement between subjective and device-based instruments has not yet been investigated. Overall, the results show statistically significant differences in sleep parameters derived from subjective and device-based measures. Exceptions were TBT, which showed good agreement between the three instruments, and TST, which showed good agreement between sleep diary and bed sensor. Our findings underscore substantial discrepancies between instruments of which researchers and clinicians should be aware when used to measure sleep in children with CP.

Our findings show similar patterns as studies in other pediatric populations comparing subjective measurements of sleep and actigraphy. Estimates of TBT often show satisfactory agreement, which was also shown in the present study using Bland-Altman plots. This agreement is unsurprising since actigraphic TBT data is based on bedtimes derived from the sleep diary. In contrast, agreement for TST is often poor.[24,27,33,34] In the present study, parents' estimates of TST were 87 minutes longer compared to actigraphy, which is similar to a recent study in children with epilepsy (82 minutes),[27] as well as in TD children (between 36 and 155 minutes).[23,24,35] This discrepancy can be explained by several factors, including the susceptibility of parental reports of sleep to reporting biases.[36] Several concerns have arisen regarding the use of accelerometry among pediatric populations.[37] Compared to PSG and videosomnography, various studies have shown an overestimation of awakenings and WASO by actigraphy.[38–40] It is important to note that actigraphy measures body movements and not sleep in itself. Therefore, activity during the night (e.g. spams, restlessness or other involuntary movements), which may be more pronounced in children with CP, might be scored as wakefulness. This is also represented by the high number of awakenings and high levels of WASO measured in the present study. These results advocate for the exploration of other methods for sleep analysis in children with CP.

This is the first study to use a bed sensor for the assessment of sleep in children with CP. In contrast to actigraphy, the bed sensor showed satisfactory agreement with sleep diary for TST. The deviation regarding awakenings and WASO was less between bed sensor and sleep diary compared to actigraphy and sleep diary (0.43 vs. 22.90 for the number of awakenings, and 19.12 vs. 79.38 minutes for WASO, during all nights, respectively), which is also represented by the good agreement for WASO during weekend nights. Nevertheless, poor agreement was found for other sleep parameters between sleep diary and bed sensor. Poor agreement (ICC<0.22 for all nights, except TBT) was also found between both device-based instruments. The agreement between actigraphy and bed sensor for the number of awakenings was negative .01. This negative value was due to the large variety within the number of awakenings measured by actigraphy. A negative ICC represents poor agreement, in which the accordance between awakenings measured by actigraphy and bed sensor are truly random.[41] As suggested by Lockley et al.,[42] even though all instruments strive to measure parameters of sleep, they have different manners of assessment. The sleep diary is based on the observations of parents, while device-based instruments are based on (motor) activity. The bed sensor was able to capture complete data (>4 nights) for 44 out of 60 children (73%). This finding supports the feasibility of a bed sensor, and underscores the need to continue investigating its application for sleep monitoring in children with CP.

Each method of sleep assessment comes with advantages and disadvantages. A sleep diary is based on subjective insights of sleep, and therefore gives valuable information on the parental perception. Moreover, a sleep diary provides information on behavioral consequences of sleep and manifestations of, for example, nighttime fears. However, parents might not always be aware of every aspect of sleep and might for example not notice a child awakening during the night. Although actigraphy is a device-based method, it is not a truly objective measure as it still requires a sleep logbook for the entry of bedtimes and the moment someone first attempts to fall asleep ('lights out'). We advise against the application of actigraphy as a diagnostic tool for children in CP as it might overestimate the amount of awakenings due to restlessness during the night. A promising device-based instrument for sleep monitoring is the bed sensor. This method is far less time consuming to interpret compared to the Actigraph and is therefore favourable for clinicians. However, the bed sensor will not register sleep when a child moves its sleeping location during the night. Its validity and reliability in monitoring sleep in children with CP needs to be further investigated. Preferably, a combination of subjective and device-based measures can best be used as complementary sources to inform researchers and clinicians on many different dimensions of sleep.[13]

Strengths & limitations

A strength of this study is the comparison of one subjective and two device-based methods of sleep assessment in a group of children with a movement or posture disorder. Moreover, we used a novel instrument for sleep measurement: ballistocardiography. All measurements were performed at home, with minimally invasive methods. This produced negligible distortion in sleep patterns of the children.

The findings of this study need to be interpreted considering some limitations. Our study population consisted of ambulatory children with CP, without including children with GMFCS levels IV and V. Non-ambulatory children with CP are known to be more severely affected by sleep problems.[43] Furthermore, the severe functional limitations of non-ambulatory children causes less frequent activity during the night,[28] and might influence device-based measurements of sleep. This suggests that the overall discrepancy between subjective and device-based measurements of sleep may be even larger in non-ambulatory children with CP.

A second limitation of this study is that the means of sleep parameters calculated for weekend nights were based on one or two measurements. This might be insufficient to provide reliable estimations of sleep.[44] However, differences between averages of school- and weekend nights were minor.

Finally, this study did not include PSG as a gold standard for sleep measurement. We advocate for more research into the agreement of sleep instruments in children with CP, including PSG as a comparison against both subjective and device-based instruments.

CONCLUSION

Our findings are particularly useful for practitioners who are interested in assessing sleep parameters in situations where traditional PSG is challenging to implement. These situations include documentation of sleep patterns in the home setting, over prolonged periods. Our findings show discrepancies between subjective and device-based methods for assessing sleep in children with CP. As the use of actigraphy is time consuming and some concerns have arisen regarding its measurement of wakefulness after sleep onset, we advise against the use of actigraphy as a single diagnostic tool. However, actigraphy may still be preferred as a device-based measure in subjects who do not sleep in the same bed consistently. Overall, the bed sensor is a promising method to provide an objective report of sleep, but needs to be further investigated for its feasibility in children with CP. When assessing sleep, we recommend to take into account the characteristics of the child and to utilize both subjective and device-based instruments in order to get a complete impression of sleep in children with CP.

Acknowledgements

This study was kindly supported by a grant from JKF Kinderfonds, a foundation aimed at improving the quality of life of children and adolescents with physical disabilities in The Netherlands.

The authors are grateful to the children and their parents for participation in this study. We thank the healthcare professionals from the rehabilitation settings for their kind assistance in participant recruitment, in particular Sanne van der Vossen (De Hoogstraat Revalidatie), Rick van de Ven (Tolbrug/Atlent, Jeroen Bosch Ziekenhuis) and Mirjam Jeurissen (Rijndam Revalidatie). Last but not least, we would like to thank Joyce Obeid for her expert support during the actigraphy data analysis.

REFERENCES

1. Horwood L, Mok E, Li P, Oskoui M, Shevell M, Constantin E. Prevalence of sleep problems and sleep-related characteristics in preschool- and school-aged children with cerebral palsy. Sleep Med. 2018;50:1–6.
2. Dutt R, Roduta-Roberts M, Brown C. Sleep and Children with Cerebral Palsy: A Review of Current Evidence and Environmental Non-Pharmacological Interventions. Children. 2015;2(1):78–88.
3. Newman CJ, O'Regan M, Hensey O. Sleep Disorders in Children With Cerebral Palsy. Dev Med Child Neurol. 2006;48(7):564–68.
4. Angriman M, Caravale B, Novelli L, Ferri R, Bruni O. Sleep in children with neurodevelopmental disabilities. Neuropediatrics. 2015;46(3):199–210.
5. Lélis ALPA, Cardoso MVLM, Hall WA. Sleep disorders in children with cerebral palsy: An integrative review. Sleep Med Rev. 2016;30:63–71.
6. Horwood L, Li P, Mok E, Oskoui M, Shevell M, Constantin E. Health-related quality of life in Canadian children with cerebral palsy: what role does sleep play? Sleep Med. 2019;54:213–22.
7. 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.
8. Hulst RY, Voorman JM, Pillen S, Ketelaar M, Visser-Meily JMA, Verschuren O. Parental perspectives on care for sleep in children with cerebral palsy: a wake-up call. Disabil Rehabil. 2020;0(0):1–10.
9. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy: A review. J Child Neurol. 2011;26(10):1303–10.
10. 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 Int J Paediatr. 2012;101(6):618–23.
11. Steur LMH, Grootenhuis MA, Terwee CB, et al. Psychometric properties and norm scores of the sleep self report in Dutch children. Health Qual Life Outcomes. 2019;17(1):1–9.
12. Lewandowski AS, Toliver-Sokol M, Palermo TM. Evidence-based review of subjective pediatric sleep measures. J Pediatr Psychol. 2011;36(7):780–93.
13. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: An underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
14. Sadeh A, Sharkey KM, Carskadon MA. Activity-based sleep-wake identification: An empirical test of methodological issues. Sleep. 1994;17(3):201–7.
15. Walsh L, Moloney E, McLoone S. Identification of nocturnal movements during sleep using the non-contact under mattress bed sensor. Proc Annu Int Conf IEEE Eng Med Biol Soc EMBS. Published online 2011:1660–3.
16. Guerrero-Mora G, Elvia P, Bianchi AM, et al. Sleep-wake detection based on respiratory signal acquired through a Pressure Bed Sensor. Proc Annu Int Conf IEEE Eng Med Biol Soc EMBS. Published online 2012:3452–5.
17. Mendez MO, Matteucci M, Cerutti S, Bianchi AM, Kortelainen JM. Automatic detection of sleep macrostructure based on bed sensors. 31st Annu Int Conf IEEE Eng Med Biol Soc. Published online 2009.
18. Roebuck A, Monasterio V, Gederi E, et al. A review of signals used in sleep analysis. Physiol Meas. 2014;35(1):R1–57.
19. Rauhala E, Virkkala J, Himanen SL. Periodic limb movement screening as an additional feature of Emfit sensor in sleep-disordered breathing studies. J Neurosci Methods. 2009;178(1):157–61.
20. Tenhunen M, Elomaa E, Sistonen H, Rauhala E, Himanen SL. Emfit movement sensor in evaluating nocturnal breathing. Respir Physiol Neurobiol. 2013;187(2):183–9.
21. Hendriks MMS, van Lotringen JH, van der Hulst MV, Keijsers NLW. Bed sensor technology for objective sleep monitoring within the clinical rehabilitation setting: Observational feasibility study. JMIR mHealth uHealth. 2021;9(2).
22. Sadeh A. The role and validity of actigraphy in sleep medicine: An update. Sleep Med Rev. 2011; 15(4):259–67.
23. Mazza S, Bastuji H, Rey AE. Objective and Subjective Assessments of Sleep in Children: Comparison of Actigraphy, Sleep Diary Completed by Children and Parents' Estimation. Front Psychiatry. 2020;11(June):1–11.

24. Werner H, Molinari L, Guyer C, Jenni OG. Agreement rates between actigraphy, diary, and questionnaire for children's sleep patterns. Arch Pediatr Adolesc Med. 2008;162(4):350–8.
25. Dayyat EA, Spruyt K, Molfese DL, Gozal D. Sleep estimates in children: Parental versus actigraphic assessments. Nat Sci Sleep. 2011;3:115–23.
26. Alfano CA, Patriquin MA, De Los Reyes A. Subjective - objective sleep comparisons and discrepancies among clinically-anxious and healthy children. J Abnorm Child Psychol. 2015;43(7): 1343–53.
27. Tsai SY, Lee WT, Lee CC, Jeng SF, Weng WC. Agreement Between Actigraphy and Diary-Recorded Measures of Sleep in Children With Epilepsy. J Nurs Scholarsh. 2018;50(2):143–50.
28. Sato H, Iwasaki T, Yokoyama M, Inoue T. Monitoring of body position and motion in children with severe cerebral palsy for 24 hours. Disabil Rehabil. 2014;36(14):1156–60.
29. Vesterinen V, Rinkinen N, Nummela A. A Contact-Free, Ballistocardiography-Based Monitoring System (Emfit QS) for Measuring Nocturnal Heart Rate and Heart Rate Variability: Validation Study. JMIR Biomed Eng. 2020;5(1):e16620.
30. Koo TK, Li MY. A Guideline of Selecting and Reporting Intraclass Correlation Coefficients for Reliability Research. J Chiropr Med. 2016;15(2):155–63.
31. Bland JM, Altman DG. Statistical methods for assessing agreement between two methods of clinical measurement. Lancet. 1986;1(8476):307–10.
32. R Core Team. R: A language and environment for statistical computing. Published online 2020. http://www.r-project.org/
33. Kushnir J, Sadeh A. Correspondence between reported and actigraphic sleep measures in preschool children: The role of a clinical context. J Clin Sleep Med. 2013;9(11):1147–51.
34. Sadeh A. Evaluating night wakings in sleep-disturbed infants: A methodological study of parental reports and actigraphy. Sleep. 1996;19(10):757–62.
35. Brazendale K, Beets MW, Weaver RG, et al. Comparing measures of free-living sleep in school-aged children. Sleep Med. 2019;60:197–201.
36. Iwasaki M, Iwata S, Iemura A, et al. Utility of subjective sleep assessment tools for healthy preschool children: A comparative study between sleep logs, questionnaires, and actigraphy. J Epidemiol. 2010;20(2):143–9.
37. Meltzer LJ, Montgomery-Downs HE, Insana SP, Walsh CM. Use of actigraphy for assessment in pediatric sleep research. Sleep Med Rev. 2012;16(5):463–75.
38. Sitnick SL, Goodlin-Jones BL, Anders TF. The use of actigraphy to study sleep disorders in preschoolers: Some concerns about detection of nighttime awakenings. Sleep. 2008;31(3):395–401.
39. Insana SP, Gozal DD, Montgomery-Downs HE. Invalidity of one actigraphy brand for identifying sleep and wake among infants. Sleep Med. 2010;11(2):191.
40. So K, Buckley P, Adamson TM, Horne RSC. Actigraphy correctly predicts sleep behavior in infants who are younger than six months, when compared with polysomnography. Pediatr Res. 2005; 58(4):761–5.
41. Taylor PJ. An Introduction to Intraclass Correlation that Resolves Some Common Confusions. Programs Sci Technol Values, Crit Creat Thinking, Public Policy. 2009;7656:1–9.
42. Lockley SW, Skene DJ, Arendt J. Comparison between subjective and actigraphic measurement of sleep and sleep rhythms. J Sleep Res. 1999;8(3):175–83.
43. Hulst RY, Gorter JW, Voorman JM, et al. Sleep problems in children with cerebral palsy and their parents. Dev Med Child Neurol. 2021;63(11):1344–50.
44. Short MA, Arora T, Gradisar M, Taheri S, Carskadon MA. How many sleep diary entries are needed to reliably estimate adolescent sleep? Sleep. 2017;40(3).

Chapter 8

Summary & General discussion

SUMMARY OF THE MAIN FINDINGS

This thesis aimed to provide a wake-up call on the importance of sleep in the (rehabilitative) care for children with cerebral palsy (CP). In order to increase awareness of, knowledge about, and ultimately change attitudes and practices regarding care for sleep, we need more research on sleep in children with CP and the impact of sleep problems on the family. It is hoped that a more thorough understanding of sleep problems in children with CP will pave the way for the development and implementation of new therapeutic approaches that integrate care for sleep into pediatric rehabilitation practices. The following section summarizes the main findings of the studies in this thesis, which include a mix of both quantitative and qualitative studies in children with CP, their parents, and the rehabilitation professionals caring for this population.

PART I Understanding the perspectives of parents regarding the care for sleep of their child with CP

Chapter 2 reports the findings of a qualitative study aimed at exploring and understanding parental perspectives on care for sleep for their child with CP. Individual, semi-structured interviews were conducted with eighteen parents of a young child (age 1–8 years) with CP across all levels of functional ability (Gross Motor Function Classification System (GMFCS) levels I–V). The interviews were recorded, transcribed verbatim, and analyzed using inductive thematic analysis. Detailed descriptions were provided about the families' current situation regarding their child's sleep problems and the resulting profound consequences to not only child well-being, but also to parental sleep and family functioning. Parents described the numerous challenges they face in caring for their child's sleep, which are hampered by perceived shortcomings in healthcare such as limited attention to sleep and lack of CP-specific knowledge among health professionals. Parents expressed concerns and needs in their home environment (child and family aspects, and the availability of social support) and the healthcare setting (clinical practices and attitudes of healthcare professionals, as well as the broader organisation with respect to care for sleep). In conclusion, this qualitative study demonstrated that the significant burden placed on families by sleep problems is underappreciated.

PART II Evaluating the care for sleep in pediatric rehabilitation

Chapter 3 evaluates the care for sleep by assessing the sleep health practices and sleep knowledge (covering the domains of sleep physiology, sleep disorders, and sleep hygiene) among healthcare professionals working in pediatric rehabilitation. A total of 30 rehabilitation physicians (RPs), 54 allied health professionals (AHPs), and 63 controls participated in this cross-sectional survey study. Results indicated that sleep was not consistently addressed by rehabilitation professionals, and only a minority (≤20%) believed that they possess sufficient sleep knowledge to confidently address sleep in clinical practice. Although RPs demonstrated the highest scores on all knowledge domains, no group exceeded 50% correct scores for total sleep knowledge, with AHPs and controls showing equal scores. We observed limited familiarity with healthy sleep behaviours that can be practiced during the day, such as exposure to natural light and daytime exercise. Healthcare professionals' limited exposure to sleep education may result in feelings of incompetence and inadequate sleep knowledge levels, which in turn may affect their sleep health practices.

PART III Integrating sleep into the care for children with CP using a 24-hour activity approach

Chapter 4 presents an evidence-informed clinical practice guide that encourages healthcare professionals to adopt a 24-hour approach to the care for children with CP and their parents. We describe the rationale behind and the development of *The 24-Hour Activity Checklist*, as well as clinical experiences and implications of this brief, practical tool that can be easily applied in routine care to detect problems related to physical activities and sleep. Furthermore, the clinical practice guide presents a CP-specific 24-hour activity guidelines infographic, which aims to facilitate effective 24-hour activity counselling and education for parents and professionals in pediatric healthcare. The tools provided in this chapter contribute to increasing awareness and changing practice, by offering support to clinicians in identifying and improving 24-hour activity problems and promoting healthy lifestyle behaviours in children with CP.

Chapter 5 reports the results of the Sleep section of *The 24-Hour Activity Checklist* one year after implementation into three pediatric rehabilitation settings. Checklists were collected from 90 children with CP (age range 0–11y; median age 5y; 41.1% girls; 84.4% ambulatory) and 157 typically developing children (age range 0–12y; median age 5y; 49.7% girls) and their parents. We found that sleep problems were commonly reported by the parents of children with CP, and that these children were more likely to have a sleep problem compared to their typically developing peers. In addition, non-ambulatory children with CP were more severely affected by sleep problems than ambulatory children both in frequency and number of sleep problems. Waking up during the night, pain/discomfort in bed, and daytime fatigue were more common in children with CP and more prevalent among children with a non-ambulatory motor status. Parents of non-ambulatory children were less satisfied about their child's sleep, as well as their own sleep. One-third of parents of children with CP reported feeling sleep-deprived often or always compared to a quarter of parents of typically developing children. The results of this study call for more attention to be placed on the importance of sleep in pediatric health care, especially in more vulnerable populations like children with CP and their parents.

PART IV Measuring the 24-hour activities in children with CP

In **Chapter 6**, we measured and described the 24-hour activities (i.e. physical activity (PA), sedentary behaviour, and sleep), and examined adherence to the 24-hour activity guidelines for PA and sleep among a sample of ambulatory children with CP (GMFCS levels I–III) using actigraphy. Children's 24-hour activities were recorded for seven consecutive days using hip- (for day) and wrist-worn (for night) accelerometers. In total, 362 days and 340 nights from 54 children with CP (age range 3-12y; median age 6.5y; 44.4% girls; GMFCS distribution: level I (*n*=30), level II (*n*=15), level III (*n*=9)) were included in the analyses. Average daily wear time was 746.2±48.9 min, of which children spent on average 33.8% in light PA (251.6±58.7 min/day), 5.2% in moderate-to-vigorous PA (38.5±20.1 min/day), and the remaining 61.1% being sedentary (456.1±80.4 min/day). Physical activity decreased while sedentary behaviour increased with increasing GMFCS level. In total, 13% of all children met the physical activity recommendations, and 35.3% met the age-appropriate sleep duration recommendation. The proportion of children meeting the combined 24-hour guidelines was low (5.9%), especially in those classified at GMFCS level III (0%). The results of this study contribute to our understanding of the 24-hour activities among children with CP, and draw attention to the importance of considering the entire continuum of movement behaviours into the (rehabilitative) care for this population.

Chapter 7 compared subjective and device-based measures of sleep in a sample of ambulatory children with CP (GMFCS levels I-III). Sleep outcomes were recorded for seven consecutive nights in the child's home using a sleep diary, actigraphy, and a bed sensor. Parameters derived from the instruments included: Total Bed Time (TBT), Total Sleep Time (TST), Sleep Onset Latency (SOL), number of awakenings, Wakefulness After Sleep Onset (WASO), and Sleep Efficiency (SE). The agreement between the measurements was assessed using Bland-Altman plots and intraclass correlation coefficients (ICCs) with separate analyses for all nights, school nights, and weekend nights. A total of 277 nights from 38 children with CP (age range 2–12y; median age 6y; 47% girls) were included in the analyses. Poor agreement was found for all parameters between i) actigraphy and sleep diary (except for

TBT; ICC>.86) and ii) actigraphy and bed sensor (except for TBT; ICC>.84). The agreement between sleep diary and bed sensor was satisfactory for TBT (ICC>.83), TST (ICC>.70), and WASO during weekend nights (ICC=.55). Other sleep parameters showed poor agreement between sleep diary and bed sensor. Researchers and clinicians ought to be aware of the discrepancies between instruments for sleep monitoring in children with CP. These findings are particularly useful for practitioners who are interested in documenting sleep patterns in the home setting over prolonged periods when traditional polysomnography may be challenging to implement. We recommend using a combination of device-based and subjective measures of sleep, taking into account the characteristics of the child, in order to get a complete impression of sleep in children with CP.

DISCUSSION OF THE MAIN FINDINGS

Time to wake up

Sleep is a universal need and a critical determinant of health and well-being across the lifespan.[1] It constitutes a period of physiological activity during which essential restorative processes occur, enabling recovery from the wear and tear of everyday life; it allows our body, brain, and mind to rest and restore, ensuring optimal physical and mental performance during subsequent periods of wakefulness. However, as a result of an increasingly modern 24/7 world, insufficient sleep duration and poor sleep quality have become widespread in our society.[2] The increased use of smart phones and electronic devices only serves to worsen this major public health problem.[3] Many view sleep as a luxury or a waste of time, but the perceived benefits of limiting the hours spent asleep do not outweigh the short- and long-term damage incurred by sleep loss. Impaired sleep not only results in reduced psychomotor performance thereby increasing the risk for (fatal) mistakes and vehicle crashes, it is also associated with a wide range of chronic health conditions including diabetes, cancer, obesity, depression, heart attack, dementia, and stroke.[4–6] Altogether, inadequate sleep poses a significant burden to society, affecting people across the lifespan.

In more vulnerable populations like children with neurodevelopmental disabilities (NDDs) or neonatal brain injury like cerebral palsy (CP), the impact of impaired sleep can be even more pronounced than in typically developing (TD) children,[7] as they can further compromise their health, development, and quality of life.[8–10] And yet, the importance of sleep and the deleterious consequences of sleep problems are underrecognized in pediatric rehabilitation.[11] Although obtaining healthy and sufficient sleep is an urgent matter for all, this thesis focused on pediatric rehabilitation and specifically the care for children with CP and their parents. Good sleep is as important as healthy nutrition or regular physical activity. Therefore in the 'package' for good health for this population, sleep should receive the same level of attention as waking factors.[12] Unfortunately, for children with CP, this has not been the case to date. Adopting a 24-hour lens offers a broader window of opportunity and a more holistic approach towards optimized health and well-being. This thesis aims to provide a wake-up call for pediatric rehabilitation to integrate sleep into care for children with CP by adopting a "whole-day-matters"[13] vision for healthy living (Textbox 8.1); we hope this serves as a call-to-action for the broader healthcare community to do the same in their respective specialties.

Textbox 8.1 Healthy Living

University Medical Centres (UMCs) across the globe increasingly consider health in the broader sense. The focus is no longer merely on curing sickness and restoring health, but has shifted its emphasis on maintaining and promoting people's (long-term) health. Preventive medicine and lifestyle medicine have become key elements of both research and healthcare, as exemplified by 'Healthy Living' as an important strategic focus of the UMC Utrecht. This vision builds on previous projects like 'Hospital in Motion' ('UMC Utrecht in Beweging'), which focused on improving inpatient movement behaviours by implementing physical activity into usual care.[14] Healthy living, however, does not stop at night. Indeed, future initiatives geared towards promoting healthy lives are encouraged to apply a 24-hour approach that acknowledges sleep as equally important in lifestyle medicine prescription.

Sleep and rehabilitation: a perfect match?

The potential for sleep to facilitate rehabilitation

There are a myriad of benefits of sleep, and while each deserves its own dissertation, two of these benefits are especially notable in the context of rehabilitation. First, sleep provides a period of recovery from preceding waking activities, thereby ensuring optimal daytime functioning.[15] From the perspective of rehabilitation programs, these 'waking activities' can be viewed as therapies, practicing new skills, body movements, or functional tasks and activities of daily living. Following these physical endeavours, sleep not only provides a period of 'passive' rest and relaxation, but also constitutes an 'active' state of physiological repair of the body and brain. Indeed, during sleep, brain waste products are removed and other essential restorative processes such as the release of growth hormones, synthesis of proteins, muscle growth, and tissue repair take place.[16] Furthermore, these restorative mechanisms also occur at the neuronal level, as demonstrated by the ability of sleep to promote neuroplasticity and thereby recovery after cerebral stroke.[17,18] In fact, treatment of sleep problems in stroke patients is associated with improved functional and motor outcomes of rehabilitation.[19] The above illustrates how the restorative functions of (improved) sleep could aid the physical rehabilitation training and (motor) recovery of children with brain injury, like those with CP.

Second, sleep plays a critical role in learning and memory. It has been repeatedly shown that newly acquired knowledge or a newly learned skill is retained more effectively after a good night's sleep.[20] Hence, after learning a new (motor) skill or action, the brain continues to learn and improve this skill in the absence of further practice, that is, during sleep.[21] This implies that improved motor function does not only depend on the actual training period, but also on the so-called 'offline' learning and memory consolidation (i.e. stabilization of what was learned during the training period) that comes with sleep.[22] Indeed, it is not only practice, but rather practice with sleep, that makes perfect.[23] Sleep-dependent learning has important implications in the context of pediatric rehabilitation services such as physical therapy, which aims to help children acquire, maintain, or improve motor skills.[24]

Thus, given the beneficial roles of sleep for physical and neural recovery, and learning of new skills, both of which are essential aspects within rehabilitative care, we should make use of this powerful 'tool'. Sleep has the ability to help children with NDDs optimize the full potential of their bodies, and that ability should be put to better use by pediatric rehabilitation. Therefore, therapists should not only be concerned with the daytime activities, training sessions, or therapies, but also acknowledge the subsequent offline learning and repair period captured by the night. Moreover, insufficient or impaired sleep caused by sleep problems should be prioritized to provide the optimal physical and mental functioning needed for effective rehabilitation.

Pediatric rehabilitation as an ideal place for multidisciplinary sleep care

The first two parts of this thesis (**Chapter 2 and 3**) aimed to understand parental perceptions regarding the care for sleep of their child with CP, and to evaluate the care for sleep in pediatric rehabilitation. Parents reported that rehabilitation professionals rarely ask about sleep, thereby overlooking potential sleep problems in children with CP and the associated burden they place on family functioning (**Chapter 2**). This confirms that the care for sleep is underappreciated in pediatric rehabilitation.[11] From the perspective of parents, healthcare professionals do not take responsibility for the topic of sleep as it does not 'fit' under their discipline, as opposed to viewing sleep as an integral part of child health and therefore being the responsibility of every professional involved in their child's care process (**Chapter 2**). In fact, a multidisciplinary approach is suggested to be ideal for the assessment and management of pediatric sleep problems reflecting the multi-dimensional evaluation and treatment required to manage sleep problems.[25] Yet despite involvement of a multidisciplinary team in the care of children with CP, sleep health and sleep problems are not routinely assessed in pediatric rehabilitation practices (**Chapter 3**). Healthcare professionals may lack the knowledge, skills, and confidence needed to address sleep and identify sleep problems (**Chapter 3**). Consequently, parents often feel left in the dark, do not know which route to follow or where to find appropriate sleep help (**Chapter 2**). Also, most hospitals lack the facilities and multidisciplinary know-how required to treat sleep problems. As a result, families of children with sleep problems often end up navigating through a series of referrals and appointments with different healthcare providers, and/or fail to receive the help they seek and need.[26,27]

Those families that do receive help for their child are often referred to specialized sleep clinics that offer specialized diagnoses, treatment and support to people with complex sleeping disorders. Somnologists (sleep medicine specialists), in collaboration with behavioural experts, provide this specialized sleep care using a multidisciplinary practice model as they are required to have expertise covering disciplines like neurology, pulmonary medicine, pediatrics, psychiatry, and psychology. However, despite their expertise along the full width of sleep medicine, somnologists are not typically familiar with the complex underlying (often comorbid) disorders in children with NDDs, like CP. Moreover, referral to sleep clinics pose additional barriers to parents, such as geographical location (e.g. sleep clinics are scarcely located in The Netherlands), long waiting lists, and the burden of extra hospital visits involving additional doctors/healthcare providers.[28]

It could therefore be argued that the treatment and management of sleep problems in children with NDDs, should not be centered in sleep clinics, but rather be incorporated in the healthcare setting that already provides the (multidisciplinary) rehabilitative services to these children and their families. In fact, there are several clear advantages to integrating sleep care into pediatric rehabilitation: 1) multidisciplinary infrastructure – the multidisciplinary teams offer a depth and breadth of knowledge and expertise under one roof, facilitating treatment planning, and communication whilst minimizing the fragmentation of care; 2) saving time and travel – centering sleep care and rehabilitative care in one place makes it possible to combine hospital visits or integrate sleep care into regular visits; 3) accessibility – pediatric rehabilitation offers regular access to children and families through routine assessments, which are necessary to monitor sleep (interventions); 4) therapeutic relationship - the sleep care would be provided by the same members of the team that provide the usual care, and thus are already familiar and/or have a personal relationship with the child and his/her caregivers; this therapeutic relationship is critical to treatment success;[29] 5) holistic approach - a team that is familiar to the child's complex medical/neurodevelopmental history, is likely to be better equipped to address sleep problem in the context of physiological (e.g. spasticity, pain), mental (e.g. anxiety, trauma), or behavioural (e.g. poor sleep hygiene, sensory processing issues) aetiologies which are common in children with CP; 6) family-centered care as a key element of pediatric rehabilitation – recognizing the family as a whole is essential given that sleep difficulties in children with CP significantly impact on their caregivers and families (**Chapter 2**). Clearly, pediatric rehabilitation represents an ideal setting for the assessment and management of sleep problems in children with CP and their families, provided that the rehabilitation team has the time, and is equipped with the specific knowledge and tools to evaluate and treat these problems.

Integrating sleep into existing care pathways

Over the past four years, we have aligned our efforts to raise awareness for sleep in pediatric rehabilitation, and we have successfully integrated sleep into our existing care pathways for children with CP. The following paragraphs will highlight some lessons learned that are considered important for effectively embedding sleep within pediatric rehabilitation, along with recommendations for clinical practice, future research, and education.

Identifying sleep problems

Rehabilitation physicians serve a gatekeeping role in detecting child-related problems, and as such, they need to be able to identify sleep problems when families present during clinical encounters. However, parents do not always raise concerns about their child's sleep, while others may be unaware of what is normal or problematic sleep.[30] Therefore, parents expressed the need for clinicians to take the initiative to inquire about sleep during consultations (**Chapter 2**). This means that clinicians have to be able to ask the right questions that are more specific than broad questions about trouble sleeping. Instead, more objective, measurable questions like "Does is take longer than 30 minutes for your child to fall asleep?" would serve as a good starting point to identify sleep difficulties.[31]

Existing pediatric sleep questionnaires to assess sleep disorders (e.g. Sleep Disturbance Scale for Children, Paediatric Sleep Questionnaire, Children's Sleep Habits Questionnaire) have been widely used in research with children with CP despite lack of validation in this population.[32] However, these questionnaires are less suitable for use in routine practice, as they typically take a long time to complete and have scoring systems that are difficult to apply. Hence, originating from a need for a brief, practical tool that can be easily applied in routine care, we developed a 24-hour activity checklist to identify problems related to physical activity and sleep of children with CP and their parents (**Chapter 4**). The checklist is not designed to comprehensively assess the full complexity of sleep, rather, the questions are meant to be practical for busy clinicians and designed to guide them in determining whether a more thorough assessment or referral is warranted when sleep problems are detected. After implementation of the 24-hour activity checklist into the existing care pathways of rehabilitation settings in Utrecht (The Netherlands), we showed that the checklist was able to identify a large population of children with CP, as well as a substantial group of parents, experiencing sleep problems on a regular basis (**Chapter 5**). This allowed families to be referred to our newly established 'sleep team' for further evaluation, and to receive sleep counselling and/or treatment as needed. Rehabilitation physicians are encouraged to use this checklist as a springboard for meaningful conversations with parents and their children, in an effort to routinely monitor sleep and detect possible sleep problems early on.

Recommendations for clinical practice

- Rehabilitation professionals are encouraged to take the responsibility for sleep care: proactively ask about sleep during family encounters and don't wait for parents to raise concerns.
- Integrate sleep into routine assessments/follow-ups, e.g. using the 24-hour activity checklist.
- Address and discuss sleep using a family-centered approach: i.e. also ask about parental sleep.
- Be aware that the sleep problems that families face can be a sensitive topic that requires a coaching, and understanding, non-judgemental attitude.

Recommendations for future research

- Further evaluate the validity, reliability, and usability of the 24-hour activity checklist among families of children with CP and other brain-based developmental disabilities.
- Assess 24-hour activity checklist outcomes in other pediatric populations (e.g. neuromuscular disorders, oncology) and age groups (e.g. adolescents, adults) to facilitate comparisons and better learn from each other.

Increasing knowledge and awareness about sleep

If we expect healthcare professionals in pediatric rehabilitation to be able to recognize, assess, and treat sleep problems in children with CP, then they need to possess current knowledge of basic sleep physiology and common pediatric sleep disorders, and be familiar with good sleep hygiene practices. As shown in **Chapter 3**, we found that healthcare professionals in Dutch pediatric rehabilitation received inadequate sleep education and, likely as a consequence, demonstrated limited sleep knowledge. Lack of sleep training has been shown to result in healthcare providers being unsure about not only what to ask, but also about what to do, when a sleep issue is identified.[33] Indeed, we reported that approximately 40% of pediatric professionals believed that they have insufficient sleep knowledge to address sleep issues in clinical practice, and another 40% reported feeling hesitant (**Chapter 3**).

A closer look at the curricula of medical schools across the globe reveals that the average amount of total time spent on sleep education is just under 2.5 hours.[34] Therefore, there is an urgent need to integrate sleep education as an essential component of all medical school curricula as a first step towards better equipping (future) doctors with knowledge and confidence about sleep medicine before transitioning to practice. In the same way that medical curricula are increasingly recognizing and developing educational modules on "exercise is medicine", so too should we recognize and incorporate "sleep as medicine" into training. There are various avenues to incorporate and increase sleep medicine exposure into medical school curricula, which include, but are not limited to, neuroscience and neurology courses, delivered in the form of lectures, active learning experiences, clinical opportunities, and online educational tools.[35]

Continuing to the clinic, additional post-graduate and on-the-job sleep training programs, as well as clinical opportunities are highly recommended to educate and empower (rehabilitation) professionals with the knowledge, skills and confidence, needed to appropriately address sleep problems in children, and to be able to guide parents. The goal of such educational sleep trainings should not be to turn all healthcare professionals into sleep experts, but rather to provide them with the knowledge required to recognize symptoms of major pediatric sleep disorders by asking the right questions, to give the right advice regarding healthy sleep hygiene practices, and to enable them to know when to refer to a (sleep) specialist for further assessment or to initiate sleep therapies.[36]

Furthermore, given that parental knowledge about child sleep is usually poor,[37] and treatment of inadequate sleep hygiene typically begins with the education of parents,[31] they should be provided with appropriate information and advice to ensure that healthy sleep practices are implemented and maintained at home.[38,39] Parents are considered the 'agents of change' for children, and therefore, increasing parental knowledge, awareness, and engagement, can facilitate therapeutic alliance and support parents to adopt behavioural changes needed for sleep treatment strategies to succeed. Informative tools such as the 24-hour activity guidelines infographic (**Chapter 4**) were designed to facilitate effective counselling and education for both parents and healthcare professionals. We call on everyone with responsibility for the care of children with NDDs to join us in educating and empowering parents and clinicians on the importance of sleep.

Recommendations for clinical practice

- Educate parents and take the opportunity to raise their awareness on the importance of sleep e.g. by using infographics such as Figure 8.2 and 8.3.
- Not only educate parents about their child's sleep, but involve their own sleep (needs, concerns) in relation to their child – i.e., inquire about sleep in a family-centered manner.

Recommendations for education

- Sleep medicine education should receive a more prominent role in medical school curricula and educational programs in order to better equip and prepare (future) doctors and allied health professionals with sleep knowledge, skills and confidence needed to address sleep in clinical practice.
- Additional educational efforts like professional on-the-job training programs, clinical opportunities and other sleep education tools for current healthcare professionals are warranted for continued learning and to update their sleep knowledge and skills.
- Sleep health should become a topic that is discussed already during high schools to raise awareness about the importance of sleep to older children/teenagers.
- Stepping further into childhood, introducing sleep health into early childhood programs could teach children why sleep is important, e.g. through stories about their need for sleep, a bedtime routine, and healthy sleep habits.[40]

Recommendations for future research

- Investigate whether a professional education program aimed at healthy living leads to an increase in sleep knowledge and more frequent discussion of sleep in clinical practice.
- Evaluate which forms of education and guidance work best for which target group (e.g. (young) children, teenagers, parents).

Management of sleep problems using a stepped care approach

To apply care for sleep in a more efficient and cost-effective way, a health service delivery model that is based on the principles of 'stepped care' offers an opportunity that would fit these requirements.[41] Stepped care is often conceptualised as a pyramid illustrating incremental levels (steps) of intervention complexity: high patient volume is managed at the base of the pyramid using low intensity treatments, while towards the top of the pyramid a progressively smaller amount of patients are centered who require greater expertise in assessment and treatment (Figure 8.1).

Based on this approach, a readily accessible form of treatment, such as provision of sleep (hygiene) education through information on websites (e.g. www.cpnederland.nl/fysieke-gezondheid/slaap/) and referring parents to reliable health resources/tools (e.g. Figure 8.2 and 8.3) are posited at the 'entry level' for stepped care for sleep problems. Despite being a minimal intervention, the entry level treatment should be evidence-based to provide significant health gain.[41] Sleep hygiene education is considered the first line treatment for sleep problems in children with NDDs.[42,43]

Climbing up the pyramid, counselling and treatment by a 'sleep team' within pediatric rehabilitation is suggested at the mid-level. As a first step, this sleep team could provide psycho-education to parents by explaining the basics of sleep physiology (e.g. the importance of sleep, what is (ab)normal sleep, sleep hygiene rules, bedtime routines) with personal advice and individualized recommendations that are more specific and tailored to the family situation. Next, the same sleep team could provide sleep therapies (e.g. behavioural interventions, light therapy) and counsel parents on how to implement healthy sleep practices and therapeutic strategies at home. For those children with complex underlying medical and/or sleep disorders (e.g. sleep apnea) requiring specialized sleep care, referral to a sleep clinic would be posited at the top end of the pyramid.

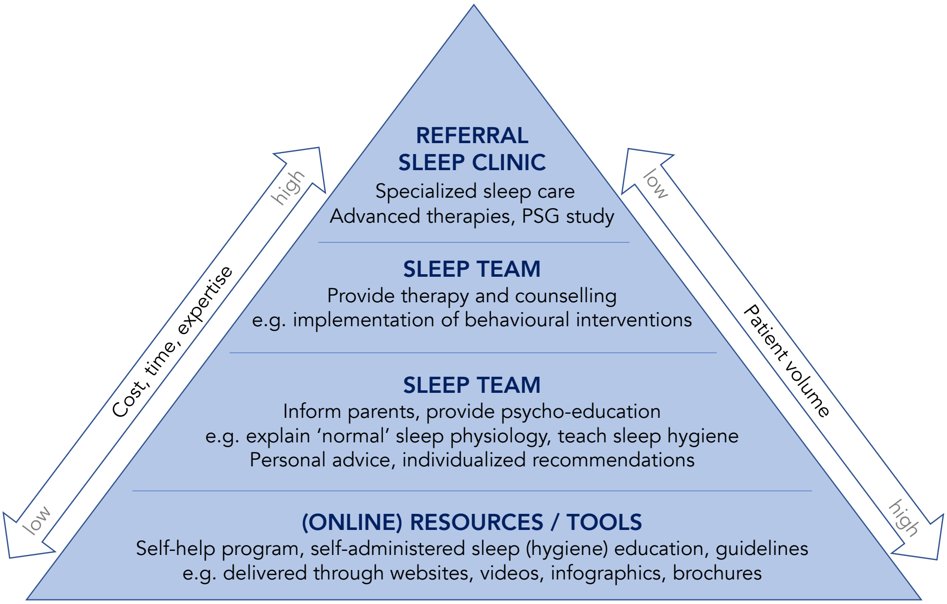


Figure 8.1 Suggested stepped care model for sleep care, in which the (rehabilitation) sleep team is positioned at the centre of the pyramid.

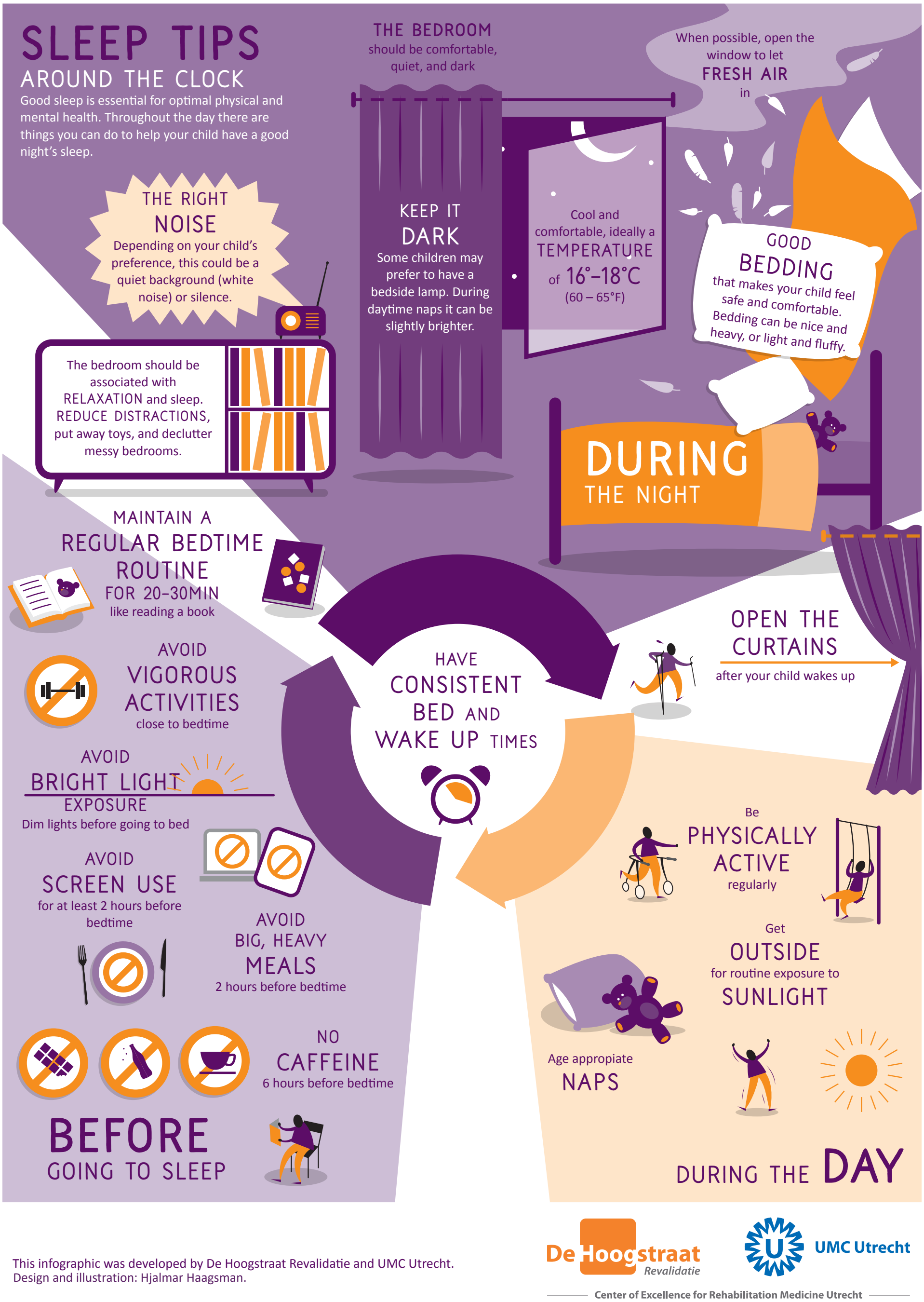
As allocation to a particular level represents increased resource requirements in terms of costs, time, and expertise, good triage of sleep problems is essential. In many cases, a combination of intrinsic and extrinsic factors may predispose, precipitate, or perpetuate a sleep problem.[44] Hence, different child-related characteristics (e.g. sensory profile, location of brain damage, comorbidities, walking status) as well as other extrinsic factors (e.g. parenting style, bed/room sharing, environmental stimuli, mental health of caregivers) are likely to have different contributions to sleep problems in terms of type and severity of problems. For example, children with CP with a non-ambulatory motor status (i.e. those classified at GMFCS levels IV–V) were more frequently reported to wake up during the night, have pain/discomfort in bed, and suffer from daytime fatigue, and these children also had more sleep problems compared to ambulatory children with CP (**Chapter 5**). Further research into child-related characteristics and environmental factors for sleep problems in children with CP are warranted to guide and facilitate healthcare professionals making informed decisions regarding good triage or next steps in sleep care, tailored to the unique child and family situation.

Recommendations for clinical practice

- Think of possibilities how to apply a stepped care approach to the care for sleep problems in your setting.
- Refer families to (online) resources/tools to inform them about sleep hygiene (e.g. Figure 8.2) or 24-hour activity guidelines (e.g. Figure 8.3)
- Consider setting up an internal 'sleep team' consisting of an occupational therapist, behavioural therapist/psychologist, and rehabilitation physician (and preferably a pediatrician/neurologist for additional consultation and/or examination) to follow-up on sleep problems as needed.
- If the main contributing factor to the sleep problem of the child appears behavioural, lifestyle, or environmental in nature, the sleep team can be deployed to provide psycho-education and counselling for parents, as well as targeted recommendations and/or sleep interventions.
- If there are indications of an underlying medical condition or sleep disorder (e.g. sleep apnea), consult with a neurologist/somnologist whether referral to a sleep clinic is warranted.
- Keep in mind that care for sleep should be individually tailored to the unique and diverse child and parental needs, and family situation; one size does not fit all.

Recommendations for future research

- Future research into child-related characteristics (e.g. location of brain damage, sensory profile, comorbidities) and extrinsic factors (e.g. parenting style, environmental stimuli, mental health of caregivers) are warranted to guide and facilitate clinicians making informed decisions regarding good triage and more targeted support.
- Investigate the effectiveness of the suggested stepped care approach for the management of sleep problems (Figure 8.1), and apply the findings to improve or complement the model e.g. with a treatment decision tree.



SLEEP TIPS
AROUND THE CLOCK

Good sleep is essential for optimal physical and mental health. Throughout the day there are things you can do to help your child have a good night's sleep.

THE BEDROOM should be comfortable, quiet, and dark

When possible, open the window to let FRESH AIR in

THE RIGHT NOISE
Depending on your child's preference, this could be a quiet background (white noise) or silence.

KEEP IT DARK
Some children may prefer to have a bedside lamp. During daytime naps it can be slightly brighter.

Cool and comfortable, ideally a TEMPERATURE of 16°–18°C (60 – 65°F)

GOOD BEDDING that makes your child feel safe and comfortable. Bedding can be nice and heavy, or light and fluffy.

The bedroom should be associated with RELAXATION and sleep. REDUCE DISTRACTIONS, put away toys, and declutter messy bedrooms.

DURING THE NIGHT

MAINTAIN A REGULAR BEDTIME ROUTINE FOR 20–30MIN like reading a book

AVOID VIGOROUS ACTIVITIES close to bedtime

AVOID BRIGHT LIGHT EXPOSURE
Dim lights before going to bed

AVOID SCREEN USE for at least 2 hours before bedtime

AVOID BIG, HEAVY MEALS 2 hours before bedtime

NO CAFFEINE 6 hours before bedtime

BEFORE GOING TO SLEEP

HAVE CONSISTENT BED AND WAKE UP TIMES

OPEN THE CURTAINS after your child wakes up

Be PHYSICALLY ACTIVE regularly

Get OUTSIDE for routine exposure to SUNLIGHT

Age appropiate NAPS

DURING THE DAY

This infographic was developed by De Hoogstraat Revalidatie and UMC Utrecht.
Design and illustration: Hjalmar Haagsman.

De Hoogstraat Revalidatie
UMC Utrecht
Center of Excellence for Rehabilitation Medicine Utrecht

Figure 8.2 Sleep tips around the clock infographic.

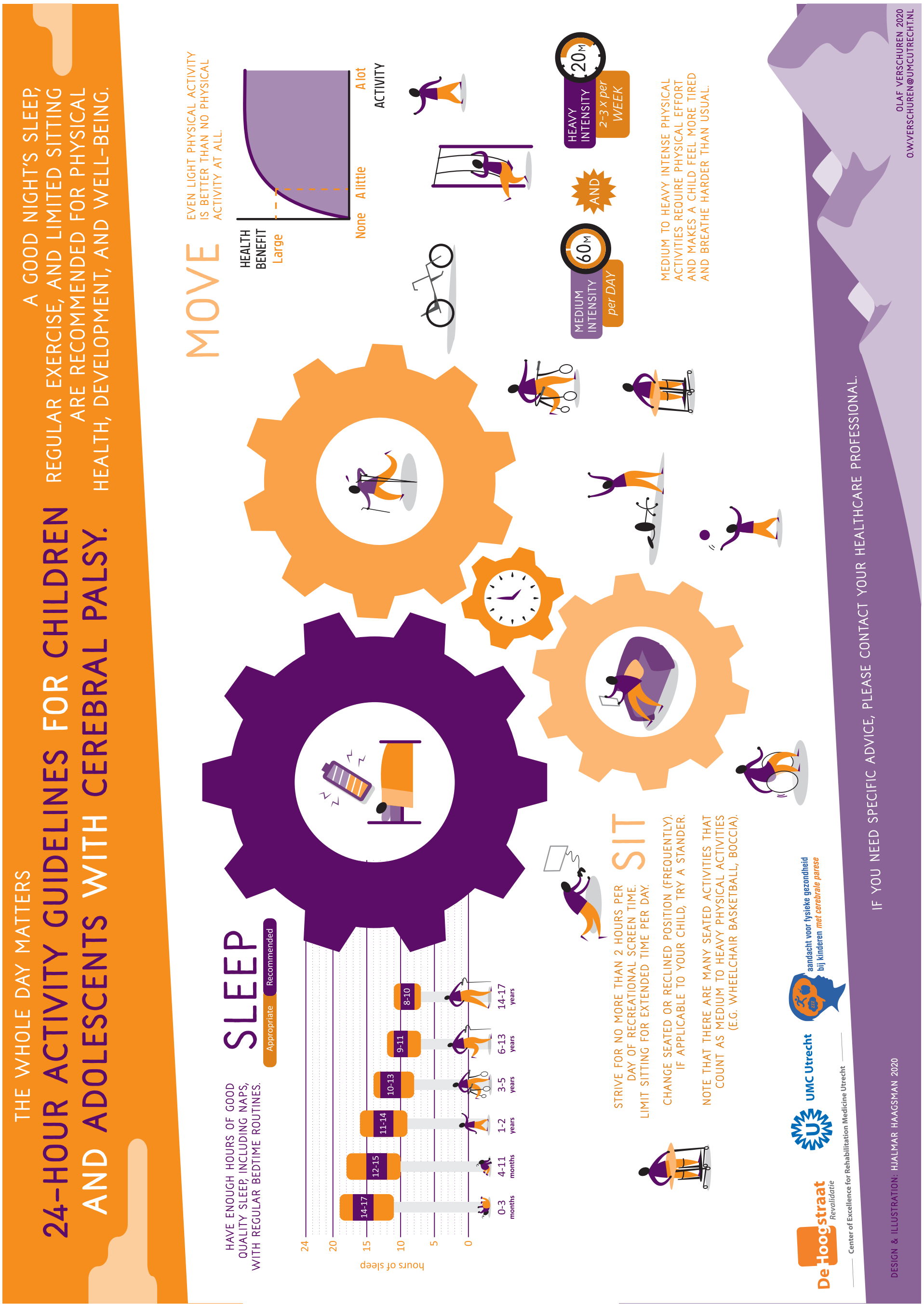


Figure 8.3 The 24-hour activity guidelines infographic.

e-Health

The COVID-19 pandemic closed the doors of outpatient clinics in rehabilitation centres in 2020, putting a halt on face-to-face consultations and therapies. To continue caring for patients, a forced transition from physical to digital care took place, with usual care delivered through online or blended care (mixing online with face-to-face therapy). Although the rationale for setting up online/blended care was previously often lacking,[45] and adoption of these new forms of therapy in rehabilitative routine care were limited,[46] we are now increasingly recognizing the added value of these digital approaches to care.

When we integrated sleep into the existing care pathways for children with CP, we noticed that many children with sleep problems that had been identified based on the 24-hour activity checklist (**Chapter 5**) now required a follow-up or intervention by our sleep team. This resulted in an unforeseen increased pressure on the treatment capacity of the healthcare system. On the receiving end, we also noted families often felt overloaded by their child's (health)care demands and fitting these into their daily schedules (**Chapter 2**). For them, accessing and receiving multidisciplinary rehabilitative care can be time-consuming (e.g. travel distances, long clinic days) and physically demanding (e.g. due to the child's physical disability). Scheduling additional appointments with a sleep team can cause a considerable extra burden of care. Online or electronic health (e-health) interventions delivered through the internet hold great potential for reducing such barriers, and increasing accessibility.

A recent study exploring the barriers and facilitators to access, use, and provision of treatment for sleep problems experienced by parents of children with NDDs and healthcare professionals who work with these children, highlighted the need to develop accessible interventions for sleep treatment.[47] Having an online sleep intervention would offer an efficient, time- and cost-effective strategy that can be delivered remotely and at a time and place that is convenient for the family. In fact, internet-based interventions designed to be delivered by parents for a broad range of clinical concerns have been shown effective for both TD children,[48] and children with NDDs.[49] For example, a recently developed parent-guided e-health sleep intervention was shown to be usable, useful, acceptable, and feasible among a sample of Canadian parents of children with NDDs, with parents rating the opportunity to participate by distance as a helpful and important aspect of the intervention.[50] To our knowledge, an online sleep intervention specifically adapted to children with NDDs including CP, is not available in The Netherlands. Sleep research groups are encouraged to join forces to improve sleep and quality of life of families of children with NDDs, share their knowledge, and collaborate to investigate the potential of (cross-culturally) translating existing e-health programs, followed by testing for feasibility and effectiveness.

A commonly reported disadvantage of e-health interventions is the lack of guidance through therapeutic relationship and limited communication.[51] However, combining the benefits of face-to-face with online care in a blended form of care may represent the best of both worlds. For example, complementing an online sleep intervention with live face-to-face or video-based sessions with a therapist may provide the child and their family with the necessary and ideal balance for monitoring their progress while ensuring real-time personal support and counselling.

Recommendations for clinical practice

- Capitalize on e-health and blended care (e.g. video consultations) to support sleep care in an effort to reduce the burden on families (e.g. travelling to appointments) and increase accessibility.

Recommendations for future research

- Cross-culturally translate existing online sleep interventions that have proven to be effective for parents of children with NDDs (e.g. the Canadian e-Health program 'Better Nights, Better Days for Children with NDDs') to be disseminated and implemented in The Netherlands and other countries. Also, investigate the application and need for modification of this behavioural intervention across all levels of GMFCS in children with CP. Additionally, explore whether adjustments to the current online format can be made to create a blended care program.
- Evaluate and compare the usability, feasibility, and effectiveness of delivering sleep interventions to parents of children with NDDs using an online versus blended format.
- Investigate how to effectively incorporate e-Health into the suggested stepped care model presented in Figure 8.1.

FINAL CONCLUSIONS

Sleep problems are common in children with CP, and can have profound impacts on child health and functioning, as well as on the well-being of the entire family. Given that the major long-term goal of pediatric rehabilitation is to ensure that children with CP and their families achieve the best possible quality of life, it is time that we wake up to the importance of sleep in the rehabilitative care for this population. Sleep should be acknowledged as a core component of lifestyle medicine prescription, and considered as one that is equally important to -or even as the foundation for- healthy nutrition and physical activity. Promoting healthy lifestyle behaviours around the clock has the potential to provide children with CP a strong foundation with long-term health benefits throughout their lifespan. To do this, sleep must receive a more prominent role in education, research, and clinical care. This thesis underscores the importance of considering the entire spectrum of activities into the (rehabilitative) care for children with CP, with attention to the day and night.

In recent years, we have noted the growing interest in and recognition of sleep (problems) in other medical specialties. Every sub-specialty approaches sleep from a different perspective, each with their own expertise, and with the clinical and personal characteristics of their patient population in mind. Yet despite these differences, we can all learn from and inspire each other to make positive changes for the benefit of better healthcare and treatment for patients. Therefore, we hope that the knowledge and experiences gained and disseminated through this thesis (and the parallel healthcare innovation project "Aandacht voor fysieke gezondheid bij kinderen met cerebrale parese") contribute to increased awareness about the importance of sleep in children with CP, other NDDs, and beyond. It would be a dream come true for sleep to become widely integrated as part of routine care across healthcare settings, and a common topic of conversation within clinical practices; this would represent an important step towards supporting children and families to have better nights, and as a result, live healthier, happier lives.

REFERENCES

1. Chaput J, Gray CE, Poitras VJ, Carson V, Gruber R, Olds T, et al. Systematic review of the relationships between sleep duration and Health Indicators in School-Aged Children and Youth. Appl Physiol Nutr Metab. 2016;41(6):266–82.
2. Basner M, Fomberstein KM, Razavi FM, Banks S, William JH, Rosa RR, et al. American Time Use Survey: Sleep Time and Its Relationship to Waking Activities. Sleep. 2007;30(9):1085–95.
3. Chattu VK, Manzar MD, Kumary S, Burman D, Spence DW, Pandi-Perumal SR. The Global Problem of Insufficient Sleep and Its Serious Public Health Implications. Healthcare. 2018;7(1):1.
4. Bixler E. Sleep and society: An epidemiological perspective. Sleep Med. 2009;10:S3–6.

5. Cappuccio FP, D'Elia L, Strazzullo P, Miller MA. Sleep Duration and All-Cause Mortality: A Systematic Review and Meta-Analysis of Prospective Studies. Sleep. 2010;33(5):585–92.
6. Wu H, Dunnett S, Ho YS, Chang RCC. The role of sleep deprivation and circadian rhythm disruption as risk factors of Alzheimer's disease. Front Neuroendocrinol. 2019;54:100764.
7. Sadeh A. Consequences of Sleep Loss or Sleep Disruption in Children. Vol. 2, Sleep Medicine Clinics. Elsevier; 2007. p. 513–20.
8. Simard-Tremblay E, Constantin E, Gruber R, Brouillette RT, Shevell M. Sleep in children with cerebral palsy: A review. J Child Neurol. 2011;26(10):1303–10.
9. Arboledas GP. The sleep in children with neurodevelopmental disorders. Medicina. 2019;79(1):44–50.
10. 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.
11. Verschuren O, Gorter JW, Pritchard-Wiart L. Sleep: An underemphasized aspect of health and development in neurorehabilitation. Early Hum Dev. 2017;113:120–8.
12. 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. Dev Med Child Neurol. 2016;58(9):989–90.
13. Rollo S, Antsygina O, Tremblay MS. The whole day matters: Understanding 24-hour movement guideline adherence and relationships with health indicators across the lifespan. J Sport Heal Sci. 2020;9(6):493–510.
14. Van Delft LMM, Bor P, Valkenet K, Slooter AJC, Veenhof C. The Effectiveness of Hospital in Motion, a Multidimensional Implementation Project to Improve Patients' Movement Behavior During Hospitalization. Phys Ther. 2020;100(12):2090–8.
15. Venter RE. Role of Sleep in Performance and Recovery of Athletes: a Review Article. South African J Res Sport Phys Educ Recreat. 2012;34(1):167–84.
16. Zielinski MR, McKenna JT, McCarley RW. Functions and Mechanisms of Sleep. AIMS Neurosci. 2016;3(1):67.
17. Siengsukon CF, Boyd LA. Sleep to learn after stroke: Implicit and explicit off-line motor learning. Neurosci Lett. 2009;451(1):1–5.
18. Duss SB, Seiler A, Schmidt MH, Pace M, Adamantidis A, Müri RM, et al. The role of sleep in recovery following ischemic stroke: A review of human and animal data. Neurobiol Sleep Circadian Rhythm. 2017;2:94–105.
19. Ryan CM, Bayley M, Green R, Murray BJ, Bradley TD. Influence of continuous positive airway pressure on outcomes of rehabilitation in stroke patients with obstructive sleep apnea. Stroke. 2011;42(4):1062–7.
20. Chittora R, Sharma D, Bhatnagar M. Rehabilitation or Recovery Sleep: A Future Potential Therapeutic Target for Sleep Deprivation. J Sleep Disord Ther. 2018;7(2).
21. Walker MP, Brakefield T, Morgan A, Hobson JA, Stickgold R. Practice with Sleep Makes Perfect: Sleep-Dependent Motor Skill Learning. Neuron. 2002;35(1):205–11.
22. Krakauer JW, Shadmehr R. Consolidation of motor memory. Trends Neurosci. 2006;29(1):58–64.
23. Walker MP, Stickgold R. It's practice, with sleep, that makes perfect: Implications of sleep-dependent learning and plasticity for skill performance. Clinics in Sports Medicine. 2005; 24(2):301–317.
24. Gudberg C, Johansen-Berg H. Sleep and motor learning: Implications for physical rehabilitation after stroke. Front Neurol. 2015;6(11):241.
25. Sevecke JR, Meadows TJ. It Takes a Village: Multidisciplinary Approach to Screening and Prevention of Pediatric Sleep Issues. Med Sci. 2018;6(3):77.
26. 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.
27. Petersen S, Reddihough DS, Lima S, Harvey A, Newall F. Barriers and Facilitators to Seeking Sleep Solutions for Children With Cerebral Palsy: A Qualitative Study. Front Psychiatry. 2021;17:1883.
28. Sutton JE, Huws JC, Burton CR. Experiences of sleep hygiene education as an intervention for sleep problems in children with developmental disabilities: Findings from an exploratory study. Br J Learn Disabil. 2019;47(3):165–73.

29. Martin DJ, Garske JP, Katherine Davis M. Relation of the therapeutic alliance with outcome and other variables: A meta-analytic review. J Consult Clin Psychol. 2000;68(3):438–50.
30. Honaker SM, Meltzer LJ. Sleep in pediatric primary care: A review of the literature. Sleep Med Rev. 2016 Feb 1;25:31–9.
31. Moore M. Behavioral sleep problems in children and adolescents. J Clin Psychol Med Settings. 2012;19(1):77–83.
32. Bautista M, Whittingham K, Edwards P, Boyd RN. Psychometric properties of parent and child reported sleep assessment tools in children with cerebral palsy: a systematic review. Dev Med Child Neurol. 2018;60(2):162–72.
33. Faruqui F, Khubchandani J, Price JH, Bolyard D, Reddy R. Sleep Disorders in Children: A National Assessment of Primary Care Pediatrician Practices and Perceptions. Pediatrics. 2011;128(3):539–46.
34. Mindell JA, Bartle A, Wahab NA, Ahn Y, Ramamurthy MB, Huong HTD, et al. Sleep education in medical school curriculum: A glimpse across countries. Sleep Med. 2011;12(9):928–31.
35. Salas RME, Strowd RE, Ali I, Soni M, Schneider L, Safdieh J, et al. Incorporating sleep medicine content into medical school through neuroscience core curricula. Neurology. 2018;91(13):597–610.
36. Luginbuehl M, Kohler WC. Screening and Evaluation of Sleep Disorders in Children and Adolescents. Child Adolesc Psychiatr Clin N Am. 2009;18:825–38.
37. McDowall PS, Galland BC, Campbell AJ, Elder DE. Parent knowledge of children's sleep: A systematic review. Sleep Medicine Reviews. 2017;31:39-47.
38. Blackmer AB, Feinstein JA. Management of Sleep Disorders in Children With Neurodevelopmental Disorders: A Review. Pharmacother J Hum Pharmacol Drug Ther. 2016;36(1):84–98.
39. Gottesman MM, Mindell JA, Owens JA. Sleep problems in pediatric practice: Clinical issues for the pediatric nurse practitioner. J Pediatr Heal Care. 2003;17(6):324–31.
40. Bonuck KA, Blank A, True-Felt B, Chervin R. Promoting sleep health among families of young children in head start: Protocol for a social-ecologicalapproach. Prev Chronic Dis. 2016;13(9).
41. Espie CA. "Stepped Care": A Health Technology Solution for Delivering Cognitive Behavioral Therapy as a First Line Insomnia Treatment. Sleep. 2009;32(12):1549.
42. Jan JE, Owens JA, Weiss MD, Johnson KP, Wasdell MB, Freeman RD, et al. Sleep Hygiene for Children With Neurodevelopmental Disabilities. Pediatrics. 2008;122(6):1343–50.
43. Angriman M, Caravale B, Novelli L, Ferri R, Bruni O. Sleep in Children with Neurodevelopmental Disabilities. Neuropediatrics. 2015;46(03):199–210.
44. Newton AT, Honaker SM, Reid GJ. Risk and protective factors and processes for behavioral sleep problems among preschool and early school-aged children: A systematic review. Sleep Med Rev. 2020;52:101303.
45. Wentzel J, Van der Vaart R, Bohlmeijer ET, Van Gemert-Pijnen JEWC. Mixing online and face-to-face therapy: How to benefit from blended care in mental health care. JMIR Ment Health. 2016;3(1):e9.
46. Hennemann S, Beutel ME, Zwerenz R. Ready for eHealth? Health Professionals' Acceptance and Adoption of eHealth Interventions in Inpatient Routine Care. 2017;22(3):274–84.
47. Tan-MacNeill KM, Smith IM, Jemcov A, Keeler L, Chorney J, Johnson S, et al. Barriers and facilitators to treating insomnia in children with autism spectrum disorder and other neurodevelopmental disorders: Parent and health care professional perspectives. Res Dev Disabil. 2020;107.
48. Hall CM, Bierman KL. Technology-assisted interventions for parents of young children: Emerging practices, current research, and future directions. Early Child Res Q. 2015;33:21–32.
49. Meadan H, Daczewitz ME. Internet-based intervention training for parents of young children with disabilities: a promising service-delivery model. Early Child Dev Care. 2015;185(1):155–69.
50. Tan-MacNeill KM, Smith IM, Weiss SK, Johnson SA, Chorney J, Constantin E, et al. An eHealth insomnia intervention for children with neurodevelopmental disorders: Results of a usability study. Res Dev Disabil. 2020;98:103573.
51. Craig, Holloway K, Riley G, Auret K. Online Mental Health Resources in Rural Australia: Clinician Perceptions of Acceptability. J Med Internet Res. 2013;15(9):e2772.

Addendum

Nederlandse samenvatting (Summary in Dutch)
Sleep tips
Parental involvement
Dankwoord (Acknowledgements)
Curriculum vitae
List of publications

NEDERLANDSE SAMENVATTING

Dit proefschrift gaat over slaap bij kinderen met cerebrale parese (CP). CP is een bewegingsstoornis die wordt veroorzaakt door een beschadiging van de hersenen tijdens de vroege ontwikkeling (d.w.z. voor het eerste levensjaar) van het kind. De lichamelijke beperkingen van kinderen met CP gaan vaak gepaard met bijkomende stoornissen of aandoeningen zoals epilepsie, cognitieve stoornissen, of voedingsproblemen, die samen bijdragen aan een verminderde fysieke gezondheid en een lagere kwaliteit van leven. Daarnaast verhoogt het hebben van CP het risico op chronische gezondheidsproblemen op latere leeftijd. Hierdoor ontstaan er kritische vragen over preventieve gezondheidscomplicaties en strategieën om de gezondheid en het welzijn van deze kwetsbare populatie te verbeteren.

In de afgelopen jaren zijn gezondheidsbevorderende strategieën met name gericht op het stimuleren van een actieve leefstijl, door het verhogen van fysieke activiteiten en het verminderen van sedentair gedrag. Ook binnen de kinderrevalidatiebehandeling voor kinderen met CP wordt veel aandacht besteed aan het verbeteren van motorisch functioneren en optimaliseren van activiteiten gedurende de dag. Toch zou er meer winst kunnen worden behaald door een holistische 24-uurs visie te hanteren waarbij er aandacht is voor het héle spectrum van activiteiten gedurende zowel de dag én de nacht.

Gezonde slaap is essentieel voor de fysieke en mentale gezondheid van kinderen en speelt een cruciale rol bij de ontwikkeling van de hersenen. Kinderen met CP hebben een verhoogd risico op het ontwikkelen van slaapproblemen, welke serieuze negatieve gevolgen kunnen hebben voor zowel de gezondheid en het functioneren van het kind als voor het welbevinden van het hele gezin. Zo kunnen slaapproblemen bijvoorbeeld bestaande lichamelijke beperkingen van het kind in stand houden of verergeren, de kwaliteit van leven van het kind verder verlagen, alsook bijdragen aan psychologische uitputting van de ouders. Toch wordt er maar weinig aandacht besteed aan slaap in de kinderrevalidatie en worden slaapproblemen bij kinderen met CP vaak over het hoofd gezien. Het is dan ook van belang dat dit letterlijke 'gapende gat' wordt gedicht.

Om de bewustwording en kennis over het belang van slaap bij kinderen met CP te vergroten, en om uiteindelijk de manier waarop er naar slaap wordt gekeken en gehandeld in de praktijk te veranderen, is onderzoek nodig op dit gebied. We hopen dat een beter begrip van (en voor) slaapproblemen bij kinderen met CP, en de impact van deze problemen op het functioneren van het hele gezin, een opstap biedt voor de ontwikkeling en implementatie van nieuwe behandelingen en trajecten die de zorg voor slaap integreren in de huidige kinderrevalidatiepraktijk. Het doel van dit proefschrift is dan ook om de lezer wakker te schudden over het belang van slaap binnen de (revalidatie)zorg voor kinderen met CP. Hiertoe is een mix van zowel kwantitatieve als kwalitatieve onderzoeken uitgevoerd onder kinderen met CP, hun ouders, alsook onder de zorgprofessionals die voor deze populatie zorgen. De volgende sectie geeft een overzicht van de bevindingen uit de onderzoeken die zijn opgenomen in dit proefschrift.

HOOFDBEVINDINGEN

DEEL I De perspectieven van ouders van kinderen met CP met betrekking tot de zorg voor slaap

Hoofdstuk 2 rapporteert de bevindingen van een kwalitatief onderzoek gericht op het begrijpen van de perspectieven van ouders met betrekking tot de zorg voor de slaap van hun kind met CP. Individuele, semi-gestructureerde interviews werden afgenomen bij achttien ouders van een jong kind (leeftijd 1–8 jaar) met CP op alle niveaus van functionele vaardigheden (Gross Motor Function Classification System (GMFCS) niveaus I–V). De interviews zijn opgenomen, getranscribeerd en geanalyseerd met behulp van inductieve thematische analyse. Er werden gedetailleerde beschrijvingen gegeven van de huidige situatie van de gezinnen met betrekking tot de slaapproblemen van hun kind en de daaruit voortvloeiende gevolgen. Deze bleken niet alleen van invloed te zijn op het welzijn van het kind, maar ook door te werken op de slaap van de ouders en het functioneren van het gezin als geheel. Ouders beschreven de talrijke uitdagingen waarmee ze worden geconfronteerd bij de zorg voor de slaap van hun kind, die worden belemmerd door vermeende tekortkomingen in de gezondheidszorg, zoals beperkte aandacht voor slaap en gebrek aan kennis bij zorgverleners. Ouders uitten hun zorgen en behoeften met betrekking tot hun thuisomgeving (kind- en gezinsaspecten, en de beschikbaarheid van sociale steun) en de zorgomgeving (klinische praktijken en attitudes van zorgverleners, evenals de bredere organisatie van slaapzorg). Concluderend toont deze kwalitatieve studie aan dat de impact van slaapproblemen op gezinnen, ondergewaardeerd is. Om de vicieuze cirkel van slaapproblemen en de desastreuze gevolgen voor het welzijn van gezinnen van kinderen met CP te doorbreken, moeten we ons bewust worden van de door ouders geïdentificeerde problemen en het belang van slaap erkennen binnen de kinderrevalidatie. Slaap moet routinematig worden aangekaart door clinici tijdens controlemomenten door middel van een gezinsgerichte en multidisciplinaire benadering. Hierbij wordt een proactieve, ondersteunende, begripvolle, niet-oordelende houding gevraagd van zorgprofessionals.

DEEL II Evaluatie van de zorg voor slaap in de kinderrevalidatie

Hoofdstuk 3 evalueert de zorg voor slaap door de klinische praktijken en kennis met betrekking tot slaap (binnen de domeinen slaapfysiologie, slaapstoornissen en slaaphygiëne) te beoordelen onder zorgprofessionals werkzaam binnen de kinderrevalidatie. In totaal namen 30 revalidatieartsen, 54 paramedici, alsook 63 controles deel aan dit cross-sectionele enquête-onderzoek. De resultaten lieten zien dat slaap niet consequent wordt aangekaart door revalidatieprofessionals. Slechts een minderheid (≤20%) was van mening dat ze over voldoende slaapkennis beschikten om in de klinische praktijk met vertrouwen om te gaan met het onderwerp slaap. Hoewel revalidatieartsen de hoogste scores lieten zien op alle domeinen van slaapkennis, overschreed geen van beide zorgverlenergroepen een score van 50% correct op totale slaapkennis, terwijl paramedici en controles gelijke scores lieten zien. Er werd beperkte kennis waargenomen over slaaphygiëneregels die betrekking hebben op de dag, zoals blootstelling aan natuurlijk licht en voldoende lichaamsbeweging. De zeer beperkte blootstelling aan slaapeducatie van zorgverleners kan leiden tot gevoelens van incompetentie en onvoldoende slaapkennis, welke hun handelen in de klinische praktijk kunnen beïnvloeden. Deze bevindingen benadrukken de noodzaak om zorgprofessionals op te leiden en te empoweren met gedegen kennis, vaardigheden en vertrouwen die nodig zijn om slaapproblemen bij kinderen met neurologische ontwikkelingsstoornissen zoals CP adequaat aan te kunnen pakken, alsook om ouders hierbij te begeleiden.

DEEL III Het integreren van slaap in de zorg voor kinderen met CP door middel van een 24-uurs activiteiten benadering

Hoofdstuk 4 presenteert een evidence-informed klinische praktijkgids die zorgprofessionals aanmoedigt om een 24-uurs benadering te hanteren in de zorg voor kinderen met CP en hun ouders. In deze praktijkgids wordt de rationale achter, de ontwikkeling van, evenals klinische ervaringen met en implicaties van *De 24-uurs Activiteiten Checklist* beschreven. Deze checklist is een beknopt, praktisch hulpmiddel dat gemakkelijk kan worden ingezet in de routinematige zorg om problemen rondom fysieke activiteiten en slaap te kunnen signaleren. Daarnaast presenteert de praktijkgids een infographic met daarin een 24-uurs activiteiten richtlijn voor kinderen met CP, welke als doel heeft om begeleiding van en voorlichting voor ouders en zorgprofessionals op dit gebied te faciliteren. De hulpmiddelen in dit hoofdstuk dragen bij aan het vergroten van de bewustwording en aan een verandering van de praktijk, door clinici te ondersteunen bij het signaleren en begeleiden van kinderen die een risico lopen op problemen met bewegen en slapen. Gehoopt wordt dat door het systematisch integreren van een 24-uurs activiteiten beoordeling in de klinische praktijk, een belangrijke stap wordt gezet in de richting van het bevorderen van een gezonde leefstijl van kinderen met CP.

Hoofdstuk 5 rapporteert de resultaten van de *Slaap* sectie van *De 24-uurs Activiteiten Checklist* één jaar na implementatie binnen drie kinderrevalidatie-instellingen in Utrecht. Checklists werden verzameld van 90 kinderen met CP (leeftijdsrange 0–11j; mediane leeftijd 5j; 41,1% meisjes; 84,4% ambulant) en 157 zich normaal ontwikkelende kinderen (leeftijdsrange 0–12j; mediane leeftijd 5j; 49,7% meisjes) en hun ouders. Uit de resultaten kwam naar voren dat slaapproblemen vaak worden gerapporteerd door de ouders van kinderen met CP, en dat deze kinderen een grotere kans hebben op het hebben van een slaapprobleem dan hun zich normaal ontwikkelende leeftijdsgenoten. Bovendien werd gevonden dat niet-ambulante kinderen met CP ernstiger worden getroffen door slaapproblemen dan ambulante kinderen, zowel in frequentie van optreden als in het aantal slaapproblemen. 's Nachts wakker worden, pijn/ongemak in bed en vermoeidheid overdag kwamen vaker voor bij kinderen met CP en vaker bij kinderen met een niet-ambulante motorische status. Ouders van niet-ambulante kinderen waren minder tevreden over de slaap van hun kind en hun eigen slaap. Een derde van de ouders van kinderen met CP gaf aan dat zij vaak of altijd last hebben van slaaptekort, vergeleken met een kwart van de ouders van zich normaal ontwikkelende kinderen. De resultaten van deze studie vragen om meer aandacht voor het belang van slaap in de kindergezondheidszorg, en met name in kwetsbare populaties zoals kinderen met CP en hun ouders.

DEEL IV Het meten van de 24-uurs activiteiten van kinderen met CP

Hoofdstuk 6 beschrijft de 24-uurs activiteiten (d.w.z. fysieke activiteit, sedentair gedrag en slaap) alsook de naleving van de 24-uurs activiteiten richtlijn, bij een steekproef van ambulante kinderen met CP (GMFCS niveaus I–III) zoals gemeten door middel van actigrafie. De 24-uurs activiteiten van kinderen werden gedurende zeven opeenvolgende dagen gedurende 24 uur per dag geregistreerd met behulp van activiteitenmonitors gedragen om de heup (overdag) en pols ('s nachts). In totaal zijn er 362 dagen en 340 nachten van 54 kinderen met CP (leeftijdsrange 3-12j; mediane leeftijd 6,5j; 44,4% meisjes; GMFCS-verdeling: niveau I (*n*=30), niveau II (*n*=15), niveau III (*n*=9)) meegenomen in de analyses. De gemiddelde dagelijkse draagtijd was 746,2±48,9 min, waarvan kinderen gemiddeld 33,8% in lichte fysieke activiteit (251,6±58,7 min/dag), 5,2% in matige-tot-intensieve fysieke activiteit (38,5±20,1 min/dag), en de resterende 61,1% sedentair (456,1±80,4 min/dag) doorbrachten. Fysieke activiteit nam af, terwijl sedentair gedrag toenam met toenemend GMFCS-niveau. In totaal voldeed 13% van alle kinderen aan de richtlijn voor fysieke activiteit en 35,3% voldeed aan de richtlijn voor leeftijdsafhankelijke slaapduur. Het aandeel kinderen dat voldeed aan de gecombineerde 24-uurs richtlijn was laag (5,9%), vooral bij de kinderen met GMFCS-niveau III (0%). De resultaten van deze studie dragen bij aan een beter begrip van de 24-uurs activiteiten van kinderen met CP, en vestigen de aandacht op het belang van het betrekken van het gehele activiteitencontinuüm in de (revalidatie)zorg voor deze populatie.

Hoofdstuk 7 vergelijkt de uitkomsten van subjectief gemeten slaap met uitkomsten van apparatuur voor het meten van slaap in een steekproef van ambulante kinderen met CP (GMFCS-niveaus I–III). De slaap van de kinderen werd geregistreerd over een periode van zeven opeenvolgende nachten in de thuissituatie door middel van een slaapdagboek, actigrafie en een bedsensor. Slaapuitkomsten van de meetinstrumenten omvatten: totale bedtijd (TBT), totale slaaptijd (TST), inslaaplatentie, aantal ontwakingen, waaktijd na in slaap vallen (WAK) en slaapefficiëntie. De overeenstemming tussen de meetinstrumenten werd beoordeeld met behulp van Bland-Altman-plots en intraclass correlatiecoëfficiënten (ICC's) met afzonderlijke analyses voor alle nachten, schoolnachten en weekendnachten. In totaal werden 277 nachten van 38 kinderen met CP (leeftijdsrange 2–12j; mediane leeftijd 6j; 47% meisjes) meegenomen in de analyses. Er werd een slechte overeenstemming gevonden voor alle parameters tussen i) actigrafie en slaapdagboek (met uitzondering van TBT; ICC>.86) en ii) actigrafie en bedsensor (met uitzondering van TBT; ICC>.84). Er was een redelijk tot goede overeenstemming tussen slaapdagboek en bedsensor voor TBT (ICC>.83), TST (ICC>.70) en WAK tijdens weekendnachten (ICC=.55). Andere slaapuitkomsten lieten een slechte overeenstemming zien tussen het slaapdagboek en de bedsensor. Onderzoekers en clinici worden geacht zich bewust te zijn van de discrepanties tussen instrumenten voor slaapmonitoring bij kinderen met CP. Deze bevindingen zijn met name nuttig voor clinici die geïnteresseerd zijn in het documenteren van slaappatronen gedurende langere periodes in de thuissituatie waarin traditionele polysomnografie een uitdaging is om te implementeren. Aangeraden wordt om een combinatie van apparatuur en subjectieve metingen van slaap te gebruiken, rekening houdend met de kenmerken van het kind, om een volledig beeld te krijgen van de slaap bij kinderen met CP.

SLEEP TIPS

From parents for parents:

- Doing things that provide little stimulation and/or relaxation, e.g. listening to quiet music together, reading a book, taking a bath or shower, massage. Try to avoid TV/iPad/screens. For more active children for whom these activities are too quiet, you can think of active relaxation to make them calm. This depends on your child.
- Use fixed bedtime rituals every evening.
- Make clear agreements with your child when he/she wakes up during the night: what is the desired behaviour you would like to see?
- Leave on a small light/bed lamp for children who are afraid of the dark or who are anxious to go to sleep. Or open the door and leave a light on in the hallway.
- Some children find the pressure of a weighted blanket to be very calming and comforting at night, but note that this does not work for all children.
- Take a good look at your child's sleeping position with the occupational therapist: is he/she comfortable, and well supported?
- Regular physical activity during the day can help your child sleep better.
- Consult an expert who can tell you more about the sleep cycle to better understand sleeping habits, so that you can more easily tackle sleep problems when they occur.
- Use pictograms to make the bedtime ritual visible to your child.
- Try to avoid drinking too much before going to bed. This can help reduce or avoid your child from waking up during the night to pee.
- Be aware that some children may be more sensitive to noise and may wake up to sounds (e.g. the wind, family members talking, creaking of the stairs). A special shutter on the window can help to reduce noise (and also light, is the room dark enough?)
- Bear in mind the temperature of the bedroom: not too hot/too cold.
- It is nice to be able to ask for advice from experts, but also listen to your gut feelings as a parent: you know your child best, and this knowledge is so important.
- Sometimes sleep medication can help your child sleep better. Discuss this with your doctor first.

From children for children:

- Cuddling with mom/dad before going to sleep.
- A small bedside lamp instead of those big lamps. And turning off the light on time!
- Listening to the sound of a waterfall.
- Two hours before I go to bed I run two laps around the courtyard.
- Don't worry when you're in bed.
- Count your inhalations and exhalations 7 times.
- Fake yawning, to make you feel tired.
- Warm milk with honey and anise, mmmh!

PARENTAL INVOLVEMENT

Parental involvement in a pediatric healthcare innovation project

Over the last three years, I have participated as a parent co-researcher from OuderInzicht in a combined healthcare innovation project and (PhD) research project led by Raquel Hulst. This project focused on sleep, nutrition, and physical activity among children (aged 0–8) with cerebral palsy (CP) and their families. I took part in the project group and Karen van Meeteren was involved as my buddy from OuderInzicht. Within this project, parents cooperated, on the basis of equal partnership, with researchers and healthcare professionals.

OuderInzicht is a non-profit organization that is committed to promoting parental involvement in research. Our focus is on projects around families of children with disabilities and high-intensity needs. Together with our partners we set up new projects and join ongoing projects using the Involvement Matrix.[1] This tool provides handles to project leaders on how to involve experts by experience (e.g. parents) in projects and research.

Throughout this cooperation, OuderInzicht took an active part in each phase of the combined project. Before the start, Karen and I sat down with the other members of the project group to make agreements about our roles in the different project phases. Looking back, we have contributed to the project in several ways:

Contributions to the healthcare innovation project:

A. Several parents contributed to the development of the 24-hour activity checklist (item generation, defining questions for parents and children) presented in Chapters 4 and 5;
B. Karen participated in a consensus meeting (to develop the 24-hour activity checklist, described in Chapter 4) with researchers and healthcare professionals;
C. In co-creation with the researchers, parents developed information sources on the website of CP Nederland (videos, infographics, sleep cards).[2]
D. Moreover, I gave a presentation about parental involvement within this project at an online symposium on behalf of the national roll-out of the project.

Contributions to the research project:

A. I supported the recruitment of respondents for the interview study about sleep (Chapter 2) and I also took part as a mother myself;
B. Together with other parents and researchers we wrote a Clinical Practice Guide which was published in the journal Developmental Medicine & Child Neurology

(Chapter 4). Moreover, together we thought about how to make the 24-hour activity guidelines infographic more inclusive for children classified at GMFCS level IV or V;

C. Furthermore, I was actively involved in the preparation of the pilot of the sleep study (reported in Chapters 6 and 7) and the recruitment of participants.

The cooperation between parents, researchers, and healthcare professionals within this project proved an added value and resulted in some important insights. First, the parental input led to products (checklist, infographic, and information sources for the website) that better met the needs and characteristics of end-users. Second, the additional investments made at the start of the sleep study such as collecting feedback from end-users resulted in benefits at the time of performance because many difficulties had already been overcome.

Despite all good intentions, there were also a number of obstacles during our collaboration. While clear agreements about the roles had been made in advance, sometimes confusion arose in practice. To maintain clarity about the division of roles between the researchers and myself as a parent-researcher it turned out to be important to plan dialogue sessions, making use of the Involvement Matrix, at the start of every new project phase. Moreover, the provided information on the progress of the project was also found to be an important matter of attention. It proved necessary to plan more structured consultations between Raquel and myself and to search for better ways to inform the other parents involved in this project, for example by using factsheets and newsletters. Fortunately, all these challenges could be quickly overcome due to our common interest and open communication.

In conclusion, I look back at very constructive and positive cooperation with Raquel Hulst and her fellow researchers. For us, this partnership has been a great opportunity to include the parents' perspectives in scientific research and to gain useful experiences for future projects.

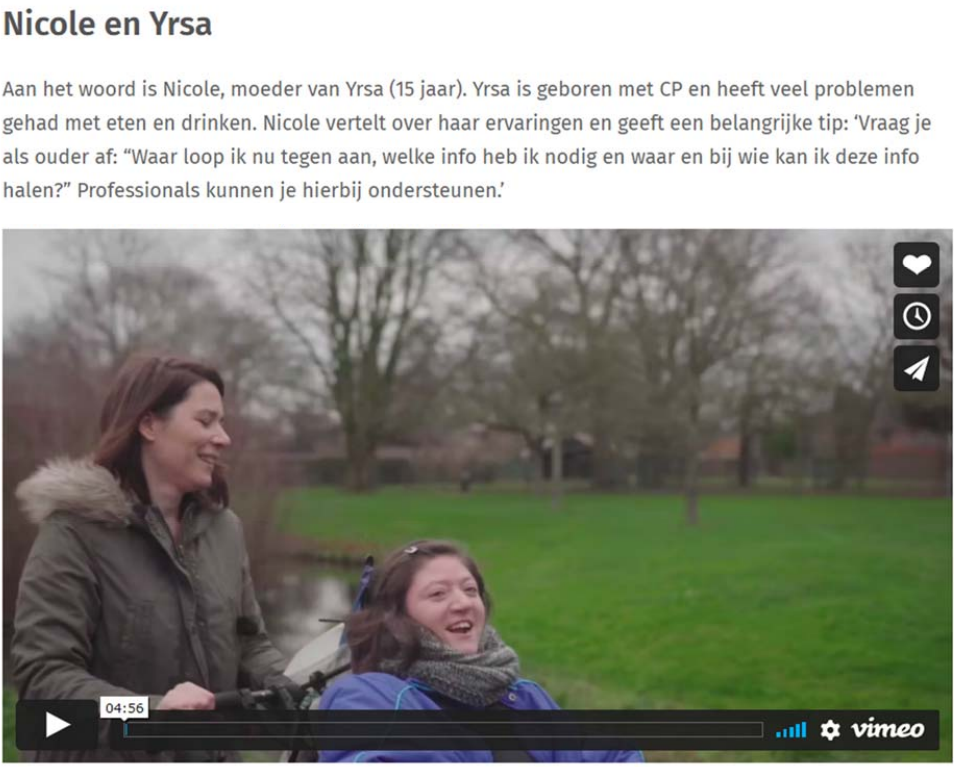
Nicole van den Dries-Luitwieler

OuderInzicht

---

[1] https://www.kcrutrecht.nl/involvement-matrix/

[2] https://cpnederland.nl/fysieke-gezondheid/



Nicole en Yrsa

Aan het woord is Nicole, moeder van Yrsa (15 jaar). Yrsa is geboren met CP en heeft veel problemen gehad met eten en drinken. Nicole vertelt over haar ervaringen en geeft een belangrijke tip: 'Vraag je als ouder af: "Waar loop ik nu tegen aan, welke info heb ik nodig en waar en bij wie kan ik deze info halen?" Professionals kunnen je hierbij ondersteunen.'

Screenshot of the video contribution from Nicole and her daughter Yrsa for the website of CP Nederland.[2]

DANKWOORD | ACKNOWLEDGEMENTS

Wauw, het is eindelijk zo ver! Nu ik dit Dankwoord mag schrijven besef ik dat mijn proefschrift en promotie een realiteit zijn. Hoewel velen van jullie kunnen beamen dat mijn eigen slaap dankzij dit promotietraject ironisch genoeg niet veel beter is geworden (naast talloze slapeloze nachten heb ik een ietwat uit de pas gelopen slaap-waakritme ontwikkeld…) en mijn 'liefde' voor het schrijven niet veel groter is geworden, kijk ik met een tevreden lach terug op vier onvergetelijke jaren. Ik had van tevoren niet durven dromen over hoeveel impact een PhD-traject zou hebben op mij en mijn professionele en persoonlijke ontwikkeling als mens, en ben ontzettend dankbaar dat ik dit avontuur heb mogen beleven met zulke fijne mensen om me heen. Ik ben apetrots op het prachtige werk dat we met elkaar hebben neergezet: *Teamwork makes the dream work!* Graag wil ik iedereen bedanken die direct of indirect betrokken is geweest bij mijn promotietraject, met wie ik mijn lief en leed mocht delen, en bovenal voor het bieden van een onuitputtelijke bron van support én gezelligheid de afgelopen jaren. Een aantal personen wil ik graag in het bijzonder bedanken.

Participanten

Allereerst wil ik graag mijn dank uitspreken aan alle families die bereid zijn geweest om mee te doen met de verschillende onderzoeken. **Ouders**, bedankt voor het delen van jullie persoonlijke verhaal, ervaringen en perspectieven: ik hoop dat ik jullie stemmen heb weten te laten klinken in Hoofdstuk 2 welke hopelijk fungeert als 'Wake-up call'. Ook dank aan alle ouders die trouw *De 24-uurs Activiteiten Checklist* invulden en beschikbaar stelden voor wetenschappelijk onderzoek, en alle vrienden en collega's met kinderen voor het creëren van een mooie referentiegroep. Tot slot, heel veel dank aan de **kinderen en families** die een week lang 24/7 een activiteitenmonitor hebben gedragen en hun slaap registreerden met dagboeken en slaapmatjes. (Huisdieren: dank dat jullie ongevraagd op de matjes zijn gaan liggen en daarmee soms de nodige verwarring hebben veroorzaakt 😉). Wat een schat aan waardevolle data hebben deze onderzoeken opgeleverd, zonder jullie inspanningen was dit nooit gelukt.

Fondsen

Graag wil ik het **Innovatiefonds Zorgverzekeraars** bedanken voor het financieren van het zorginnovatieproject "Aandacht voor fysieke gezondheid bij kinderen met cerebrale parese", en het **JKF Kinderfonds** en **Kinderrevalidatiefonds Adriaanstichting** voor financiering van de daarop volgende onderzoeksprojecten. Ook dank aan het **Dr. C.J. Vaillant Fonds** en **Stichting Voorzorg Utrecht** voor een financiële bijdrage voor de aanschaf van activiteitenmonitors en slaapmatjes ten behoeve van de 24-uurs studie.

Promotieteam

Prof. dr. Visser Meily, beste **Anne**, bedankt voor je prettige begeleiding en het vertrouwen dat je mij hebt gegeven tijdens mijn promotieonderzoek, ook (en met name) wanneer ik het op momenten even niet meer zo rooskleurig inzag. Ik bewonder je gedrevenheid en passie voor wetenschappelijk onderzoek in de revalidatiegeneeskunde, en hoe je die twee werelden combineert en met elkaar verbindt in jouw rol als promotor, hoofd van het Kenniscentrum Revalidatiegeneeskunde Utrecht, en als revalidatiearts. Dankzij jouw heldere visie en het stellen van kritische vragen (of opmerkingen in de kantlijn) leerde je mij om net weer even anders te kijken. Ondanks je drukke agenda was je altijd goed bereikbaar, had je oog voor mijn persoonlijk welbevinden, maakte je tijd om mee te denken, en stelde je zelfs je huis open om met elkaar te sparren over mijn Discussie (waarna je mij tevens wist te verbazen – in de positieve zin van het woord – over jouw tafeltennistalenten!). Jij motiveerde mij om ook de kleine feestjes te (leren) vieren, manuscripten veel eerder op te sturen (ja, ook als ze nog niet perfect zijn), en nors reviewcommentaar met een korreltje zout te nemen. Dank voor je aanmoediging, sturing, en betrokkenheid de afgelopen jaren.

Dr. Verschuren, beste **Olaf**, wat ben ik blij om jou als co-promotor te hebben gehad tijdens mijn promotietraject. Ik bewonder jouw altijd, werkelijk áltijd positieve attitude, enthousiasme, flauwe (woord)grappen en jouw vermogen om mensen om je heen te motiveren over… anything (van slaap, tot de beste strategie om een tompoes te eten)! Als ik weer eens vervelende beren op de weg zag staan, wist jij met een positieve kijk, relativerende grap, of pragmatische oplossing ervoor te zorgen dat ik 'weer goed kon slapen'. Bedankt dat je mij altijd het gevoel hebt gegeven dat ik bij je kon aankloppen voor advies of een luisterend oor. De kleine gebaren, bijvoorbeeld een kort belletje of berichtje vlak na een belangrijke of moeilijke meeting, heb ik als heel fijn ervaren. Een hoogtepunt dat ik niet gauw zal vergeten was onze eendagsvliegcarrière als filmster op de set van MVOTV in het UMC Utrecht, waar we samen hebben mogen schitteren in de mooie video over beter slapen in het ziekenhuis (voor wie 'm nog niet kent, zie https://onlinedocu.nl/slaap). Bedankt dat jij, naast me door mijn schrijfhekel heen te motiveren, vervolgens mijn 5000+ woorden ongegeneerd wist weg te schrappen tot 3000 woorden (of iedere andere woordlimiet waar ik me aan te houden had). Gelukkig kent dit Dankwoord geen woordlimiet en had jij dit vast veel beknopter gedaan, maar juist dát vond ik zo prettig aan samenwerken met jou: jij gaf mij altijd het vertrouwen dat ik het zelf het beste wist en kón (ondanks mijn onzekerheden die op de loer lagen) en liet me vrij om te groeien. Heel veel dank voor jouw 'egg-cellente' begeleiding en simultaan bekleden van de functies van supervisor, mental coach, tafeltenniskoning, inspirator én scheerbaas 2.0.

Dr. Voorman, beste **Jeanine**, dank voor je begeleiding en samenwerking tijdens mijn promotietraject, mede dankzij jou als co-promotor ben ik gegroeid als zelfstandig onderzoeker. Dank dat je mij, toen ik net begon in de kinderrevalidatie, een blikje in de praktijk hebt gegeven door het organiseren van meeloopdagen bij jou en je collega's. Hoewel de implementatie van het CP Zorgpad project wat tegenslagen kende binnen het WKZ, vind ik het mooi om te zien dat jij als kinderrevalidatiearts slaap een standaard onderwerp van gesprek hebt gemaakt in jouw patiëntencontact in het WKZ en PMC. Het was verhelderend om jouw praktijkvoorbeelden mee te kunnen nemen in het onderzoek en de artikelen.

Dr. Pillen, beste **Sigrid**, ik heb jou altijd als onderdeel van mijn promotieteam beschouwd. Ik ben je ontzettend dankbaar voor het delen van je expertise en enthousiasme, maar ook zeker voor je betrokkenheid en prettige begeleiding vanaf de start van het zorginnovatieproject tot aan het laatste artikel. Ik heb waanzinnig veel van je mogen leren door mee te lopen in Kempenhaeghe, het organiseren (en bijwonen) van de slaapscholing en casuïstiekbesprekingen voor zorgprofessionals in Utrecht, maar ook door de inhoudelijke discussies en brainstormsessies, en jouw kritische en meedenkende blik op de manuscripten. Prachtig om te zien hoe jij het jouw missie hebt gemaakt om de kinderslaapzorg in Nederland te versterken via onderwijs, advies en supervisie – ik ben trots dat we dankzij jou als slaapexpert binnen ons zorginnovatieproject een goed functionerend slaapteam hebben opgezet en daarmee bijdragen aan betere slaap voor kwetsbare kinderen én hun gezin. Bedankt voor de fijne samenwerking de afgelopen jaren.

Prof. dr. Gorter, beste **Jan Willem**, jij bent later aangeschoven tijdens mijn PhD-traject maar vrijwel direct een onmisbare speler geworden van mijn promotieteam. Heel veel dank voor je betrokkenheid, oprechte interesse, en uiterst behulpzame feedback op de artikelen die we samen hebben mogen schrijven. Jij wist, ook met een overvolle agenda, toch altijd tijd vrij te maken om mee te denken, kritische vragen te stellen of eventjes te sparren, zodat ik vervolgens weer verder kon. Ik kan me nog goed herinneren dat we in de pauze van de Brain Center Research Day stonden te kletsen en jij me vroeg naar mijn Discussie. Volgens mij gaf ik jou toen een alleszeggende blik die zich vertaalde naar: "Mjèh..". De zoemer was al gegaan om weer plaats te nemen in de zaal, maar jij griste nog snel een servetje van de tafel en begon alles wat ik zei of dacht driftig mee te tekenen en schrijven met de woorden 'Hora Est!' eronder. Jij wist me altijd een oprecht gevoel van betrokkenheid en vertrouwen te geven, en ik ben je daar heel erg dankbaar voor. Ondanks dat je tijdens mijn promotietraject voor het grootste deel van de tijd nog in Canada woonde, heeft dit onze samenwerking nooit belemmerd – de verschillende tijdzones bleken juist ontzettend efficiënt: 's avonds stuurde ik mijn versie rond en nog voor ik opstond lag jouw feedback al in mijn inbox! Sluit wederom mooi aan op onze 24-uurs visie :-)

Beoordelingscommissie

Graag wil ik de leden van mijn beoordelingscommissie, **prof. dr. Buizer**, **prof. dr. Veenhof**, **prof. dr. Benders**, **dr. Nijhof** en **prof. dr. Corkum**, bedanken voor het lezen en beoordelen van mijn proefschrift. **Prof. dr. Grootenhuis**, hartelijk dank voor uw aanwezigheid bij de verdediging van dit proefschrift. Dear **Penny**, when I impulsively approached you about a year ago after watching one of your inspiring talks on sleep in children with NDDs, to discuss possible collaborations in the future, I never imagined you taking part in the assessment committee of my PhD thesis – thank you for taking the time to review my dissertation and for attending the defense ceremony.

Onderzoeksteam

Aan alle **co-auteurs** die hebben bijgedragen aan de artikelen in dit proefschrift: bedankt voor de fijne samenwerking, inhoudelijke discussies, en waardevolle feedback die de stukken alleen maar sterker hebben gemaakt.

Dank aan alle **collega's en projectpartners** met wie ik de afgelopen jaren heb mogen samenwerken binnen de diverse projecten. Jullie hebben mij stuk voor stuk veel geleerd over slaap, voeding, ouderparticipatie, accelerometrie, statistiek, kwalitatief onderzoek, en veel meer onderwerpen. Een aantal van jullie wil ik in het bijzonder bedanken:

OuderInzicht (Nicole en Karen), heel veel dank voor het inbrengen van het ouderperspectief. Jullie hebben daarmee een belangrijke bijdrage geleverd aan het CP Zorgpad project en ik ben blij om te zien hoe de samenwerking tussen ouders en onderzoekers een steeds grotere rol speelt bij de opzet, uitvoering en evaluatie van onderzoeks- en zorginnovatieprojecten van het Kenniscentrum en daarbuiten. **Nicole**, ik bewonder je enorm en ben je ontzettend dankbaar voor onze fijne samenwerking de afgelopen jaren. Ondanks je drukke leven waarin je 101 ballen weet hoog te houden (en hoe!), wist je altijd tijd vrij te maken voor een telefoontje, een oproep uit te zetten in jullie 'achterban', mee te schrijven als co-auteur, een presentatie te geven op ons landelijke symposium, en op de valreep heb je zelfs nog een mooie bijdrage geleverd over Ouderparticipatie voor dit proefschrift. Bedankt voor alles!

Collega's van **CP Nederland** – voorheen BOSK – (in het bijzonder **Martijn**, **Johannes**, **Corine** en **Marike**) dank voor de prettige samenwerking. We hebben met elkaar een prachtige pagina opgezet (www.cpnederland.nl/fysieke-gezondheid) waarmee we vele ouders en zorgprofessionals hebben weten te informeren over het belang van slaap, voeding en beweging bij kinderen met CP, o.a. door middel van infographics, video's, en slaapkaarten (met dank aan **Hjalmar**).

Collega's van **Nutricia Research – Ana**, **Pavel**, **Margien** en **Anne-Marie**. Dank voor de fijne en persoonlijke samenwerking vanaf de opstart tot implementatie van het CP Zorgpad op ons mooie landelijke symposium. Zonder jullie was dit nooit zo'n groot succes geweest.

Dear **Joyce**, where would I have been without you as our 'actigraphy guru'. Thank you for sharing your knowledge, SOPs, accelerometers, and advise from the start (design) of the sleep study and for supporting me all the way until the end (as English grammar check). Your input and constructive feedback were invaluable to me (and I'm sure I am speaking on behalf of the entire 'dream team'), and I am grateful for all the time and energy you have put into this project.

Dr. Dudink, beste **Jeroen**, jouw enthousiasme en passie voor alles rondom slaap is zo aanstekelijk! Jij bruist werkelijk van de ideeën. Dank voor de fijne samenwerking.

Floryt, bedankt voor de sparmomenten en het delen van je expertise en adviezen waar ik veel van heb geleerd en toegepast in het kwalitatieve onderzoek van dit proefschrift.

Renate, dank voor de fijne samenwerking rondom de vormgeving van het binnenwerk van dit proefschrift. **Joch**, bedankt voor je hulp bij de vormgeving van mijn cover, en **Wendy** voor de 'finishing touch'. Ik ben superblij met het eindresultaat.

And last, but definitely not least: lieve **student-stagiaires**, zonder jullie harde werk was mijn promotietraject een stuk zwaarder en vooral een stuk minder gezellig geweest. Ik ben jullie allen dan ook ontzettend dankbaar dat jullie jullie Masterthesis/afstudeerprojecten (variërend van 6 maanden tot 1,5 jaar) bij ons wilden komen doen: **Nicole**, **Barbara**, **Neele**, **Eveline**, **Ilse**, **Anke** en **Lieke**, jullie hebben stuk voor stuk waanzinnig werk geleverd. In het bijzonder wil ik het ***SleeCP dreamteam*** bedanken voor jullie tomeloze inzet, enthousiasme en gezelligheid gedurende het slaaponderzoek. Wat ben ik trots op ons dat we, ondanks alle tegenslagen (waaronder corona en lockdowns die alle metingen platlegden, vervelende slaapmatjes die maar rood bleven knipperen, dozen die vertraagd waren waardoor de hele planning weer overhoop lag, een script dat wéér de hele nacht opnieuw moest runnen, en nog meer argh!) toch alle brandjes met elkaar wisten te blussen en alle metingen – zoals vooraf gepland – alsnog hebben kunnen uitvoeren in een jaar tijd. Ik kon werkelijk altijd op jullie rekenen! **Anke**, mocht je je als docent of bijna-mama (!) nog vervelen, weet ik zeker dat ze je bij ActiLife zouden omarmen – dank voor alles (ook aan Wibe). **Lieke**, ondanks dat ik je maar kort heb gekend, ben ik overtuigd dat jij met jouw dosis enthousiasme en ambitie een prachtige scriptie gaat afleveren. **Ilse**, cheffin logistiek: na jouw afstuderen ben je vrijwillig bij ons gebleven want je moest en zou de laatste inclusie meemaken en dat heeft geresulteerd in een welverdiende aanstelling tot junior-onderzoeker en een prachtig artikel dat ik met trots heb opgenomen in dit proefschrift.

Collega's

Collega-artsen, therapeuten, managers en secretariaten van **De Hoogstraat Revalidatie** locaties Rembrandtkade en De Kleine Prins: dank voor jullie hulp en inzet tijdens zowel de implementatie van het CP Zorgpad als jullie betrokkenheid tijdens mijn promotietraject op wat voor manier dan ook. In het bijzonder mijn lieve collega's van de afdeling Kind & Jeugd: **Sanne**, **Eveline** en **Sigrid**, dank dat jullie altijd voor mij klaar stonden als ik weer op de stoep stond met een nieuw verzoek voor doorverwijzingen, gegevens nodig had uit het EPD, mijn hart wilde luchten of juist taart kwam langsbrengen om te vieren dat we de volgende mijlpaal hadden bereikt. **Eef**, dank voor alle steun van begin tot eind, ik heb genoten van onze koffie(en-gebak)-bijklets(en-spui)-momentjes; never a dull moment indeed. Mede dankzij jullie geduw en getrek is het zorginnovatieproject zo succesvol geïmplementeerd en uitgerold. **Dorien**, **Irene**, **Marie-Anne** en **Kitty**, ik heb jullie betrokkenheid erg gewaardeerd. **Geke** en **Monique** wat een prachtig slaapteam, bedankt voor jullie inzet. **Chris** en **Marloes**, medeslaapfanaten, fijn dat we met elkaar konden sparren over de Emfit matjes. **Tiny**, dank voor alle digitale hulp en gezellige berichtjes. Dames van de **receptie**, bedankt voor de tig brieven, dozen, en bedankjes waar jullie netjes zorg voor droegen. Ook veel dank aan alle deelnemende centra en collega's van het **Wilhelmina Kinderziekenhuis** (UMC Utrecht, in het bijzonder **Jeanine** en **Corine**), **Revalidatiecentrum Tolbrug** (Jeroen Bosch Ziekenhuis, in het bijzonder **Rick**) en **Rijndam Revalidatie** (in het bijzonder **Mirjam** en **Nicole**). Dankzij jullie inspanningen hebben we de beoogde N=60 inclusies voor het slaaponderzoek weten te halen binnen een (lastig corona)jaar, dank!

Mijn collega's van het **Kenniscentrum Revalidatiegeneeskunde Utrecht**: senior-onderzoekers **Olaf**, **Marcel**, **Anita**, **Tanja** en in het bijzonder **Marjolijn**: heel veel dank voor je fijne coaching en adviezen, je was altijd bereid om mee te denken, even te sparren of wijze inzichten te delen en dat heb ik als ontzettend waardevol ervaren. **Carlijn**, bedankt voor al je hulp, 101 ZOOM-linkjes, en het organiseren van gezellige (digitale) KC-uitjes. **Eline S**, wij waren een tijdje PhD-roomies van 'het stiltehok' (R.I.P.) en ik wil je bedanken voor de fijne sfeer die je bracht, het meedenken met statistiekvraagstukken of scriptjes, en je support in de laatste fase van mijn promotietraject. Junior-onderzoekers (mede-promovendi en alumni): **Aline**, **Eline**, **Heleen**, **Ilona**, **Isabe**, **Lauriane**, **Mette**, **Maren**, **Nicole**, **Remko**, **Tanja**, **Tijn**, **Vincent**, dankjulliewel voor de gezelligheid, koffiepauzes, borrels, Summer Schools en uitjes. Ondanks dat we allemaal op ons eigen onderzoekseilandje zaten, heb ik me altijd een team gevoeld met jullie. **Jochem** en **Joris**, ik heb genoten van de geslagen bruggen en verhelderende 'inzichten' binnen het Research Network. **Jord**, wij zijn het voorbeeld dat collega's en vriendschap samengaan, dank voor alle fijne gesprekken, koffiewandelingen, boulderlesjes, buikpijn en filosofische verwonderingen tijden onze 'tripjes'.

Ook veel dank aan de collega's van andere afdelingen (in het bijzonder de **mannen van de ICT Helpdesk**) met wie we pre-corona iedere dinsdagavond in de gymzaal stonden voor een fanatiek potje badminton (ik neem terug dat ik ooit dacht dat dit een suffe, saaie sport was!). Ook de donderdagmiddag potjes tafeltennis in de pauze waren een groot genot. We hebben ons onbaatzuchtig ingezet voor het thema Fysieke Gezondheid.

Vrienden en familie

Daarnaast was dit promotietraject ook absoluut niet mogelijk geweest zonder de liefde, gezelligheid en steun van mijn lieve familie, vriendjes en vriendinnetjes. Ik voel me gezegend met zo'n warm bad van vriendschappen om mij heen – of we elkaar nu 1x per week of 1x per jaar zien – en een aantal wil ik in het bijzonder noemen.

I'd like to start off by saying how grateful I am to have such crazy wonderful people standing beside me as my paranymphs: **Renan** – eight years ago I entered the AMC as Nicolab's first employee but I'm still pretty sure you had no idea who I was until we discovered a soulmate in each other through the bizarre philosophical yet hilarious 5am conversations on (our by now annual trip to) Mallorca. When everybody cancelled last year, we had the villa for ourselves and I enjoyed every bit of it; especially having Panettone for breakfast, lunch ánd diner, the 'open wound' on your heel, not to mention those silly clouds trying to fool us. Thanks for being my best friend, for putting up with my goods and bads, for always being willing to lose against me with whatever game we play (and then blaming the alcohol), for being the smartest yet dumbest person I know, and for making my belly hurt from laughing because of the random things you say. Your positivity and nerdy thoughts about life are a true inspiration to me and I still hope one day we will finish creating our own board game. You are already the best dad Vesper and Mika could dream of, and I wish you all the happiness in the world. Lieve **Eline**, wat ben ik dankbaar en blij dat je van collega tot (vegetarisch) vleesje bent gepromoveerd en straks naast mij staat als paranimf. Wij zijn het levende bewijs dat een vriendschap voor het leven op werk kan ontstaan. Naast onze eindeloze gesprekken en werelddilemma's, cappuccino's met en zonder havermelk, synchroon geschaterlach, peptalks en dronken vlogs, en zelfs een filosofische retraite op de heide, koester ik de vele uurtjes die wij samen tot diep in de nacht dansend hebben doorgebracht op Fiesta Macumba feestjes. Ik bewonder je onuitputtelijke energie en ambitie om jezelf altijd verder te willen ontwikkelen en ben trots op alles wat je doet. Ik hoop nog veel onvergetelijke, leerzame, en 'vunzige' avonturen met je te beleven.

Mijn lieve oud-klasgenootjes van de middelbare school aka 'het Ig'. **Charley**, **Vera**, **Eva** en **Marthe**, wat fijn dat we elkaar na al die jaren nog steeds zien en dat het dan altijd zo warm en vertrouwd voelt. Lieve **Talie**, ik ben je eeuwig dankbaar dat je je paleisje op de JV voor mij hebt opengesteld toen ik net begon met mijn PhD; ik heb genoten van de 1^e klas piston koffie, omgekeerd verstoppertje in het kasteel, en vooral van de dierbare herinneringen die ik daar heb opgebouwd. Buppeduppankjuppewuppel. Lieve **veerbeer**, jij bent één bolletje liefde en vertier met je gekke fratsen en dansjes (die we vrijwel iedere pauze aan elkaar opdroegen in jouw oude kamertje), ik heb je lief! Lieve **eefaap**, ik zal onze kleurrijke verkleedpartijtjes, Bunnik-avonturen en ritjes achterop de fiets vanaf Buitenveldert nooit vergeten, keep ya kinn-up! Lieve **Marty**, ik ben je zo dankbaar voor alle fijne gesprekken en dat je mij het gevoel geeft dat je me écht begrijpt. Je bent een powervrouw en ik kan niet wachten om samen met je te motormuizen! Lieve **joenie**, als ik er weer eens doorheen zat stond jij alweer (letterlijk) voor de deur met een maaltijd of andere welkome afleiding. Zelfs toen ik zo koppig was om mijn PhD-werk niet te back-uppen voor de vakantie, na meermaals aandringen van jou om dat wél te doen, stond jij voor me klaar toen mijn laptop inderdaad crashte na de vakantie. Dankjewel voor je onvoorwaardelijke vriendschap. Lieve **Ez**, wat fijn om je weer in mijn leven te hebben. Vanaf het moment dat we samen 1C-nerdjes werden, waren wij onafscheidelijk, en ik ben je dankbaar voor alles wat je mij sindsdien hebt gebracht.

Andy, mijn levensguru: ik ben je oneindig dankbaar voor al je wijsheid, inspiratie, en veels te domme appjes die mij keer op keer wisten te raken en versterken. Bedankt dat ik zoveel van je heb mogen leren en met veelvuldig vallen en opstaan ben gegroeid tot een betere versie van mezelf. Dank ook voor het connecten met **Camiël**, ik waardeer hoe we af en toe inchecken bij elkaar.

Lieve **Lotte**, wat bijzonder hoe onze vriendschap is ontstaan op de dag dat jij mijn oude baan bij de UvA overnam: het voelde vanaf het eerste moment alsof je het zusje was dat ik nooit heb gehad! Ik ben je dankbaar voor alle fijne gesprekken en etentjes, je peptalks en oprechte interesse. Je bent een prachtig mens en ik ben zo supertrots op hoe je alles doet.

Peter, ik koester de herinneringen aan onze *chillings* in Amsterdam (van Koningsdag tot je voor de zoveelste keer kwijtraken op één of ander leip festival) maar ook zeker aan Nepal waar we samen op één motor klommen inclusief backpacks (hoe dan?) en je me huilend op je rug naar de overkant van het moeras moest dragen door al die bloedzuigers :') Ik heb bewondering (of onbegrip? Misschien toch gewoon jaloezie) voor hoe jij met gemiddeld 4 uur per week een PhD weet te doen doordat je meer tijd besteedt aan crypto's en gamen dan aan papers schrijven. Maar tegelijkertijd ben ik megatrots op alles wat je de afgelopen jaren hebt neergezet met je bedrijfje, huisje en mooie gezinnetje in Almere (wie had dat gedacht?).

Dear Lorena, little **Tachita**, thanks for being one of my biggest professional cheerleaders throughout this PhD journey. I've enjoyed all the silly games and Brain Tonics that we shared together, and I'm so happy to have you back in NL so we can continue celebrating the geeky life.

PD, als ik zie hoe jij mijn leven hebt weten te verrijken in slechts een half jaar tijd, kan ik niet anders dan uitkijken naar alle (motor)avonturen die ons nog te wachten staan.

Sportieve tennisvrienden van TC IJburg. **MC** (**Erwin** aka **motormaat**) jij bent de grootste trofee uit mijn hele ladder-carrière. Bedankt dat je mij zo vaak laat winnen, voor het verzorgen van warme maaltijden, en bovenal voor onze vriendschap. Laten we gauw nog een *pikketanussie* pakken voor je onder het mes gaat en sterker terugkomt dan ooit! Tijdens de start van mijn promotietraject was ik nog onderdeel van het mix-team (ome **Jack** (tachtig procent!!), **Simon** en **Claudia**, **Nicolette**, **Sander**, **Renie** en **Aad** dank voor deze toptijd!) tot ik werd gekaapt en 'promoveerde' naar de 1^{e} klasse in het gezelligste damesteam van Amsterdam (**Alexandra**, **Anne**, **Marije**, **Alexandra**, **Daniëlle**, **Birgitta** en **Esther**) onder leiding van de beste captain die we ons kunnen wensen. Lieve **slopende DD-dubbel toppers**, dank voor alle bloedstollende 3-setters, het is elke woensdagavond een feestje met jullie. **Marije**, met jou op de baan voel ik me één, aanvallend en direct, wij lusten ze rauw. **Alex**, het is bewonderenswaardig hoe jij je captain-schap en reisleidster-schap (wat voor verborgen talenten heb jij nog meer?) zo nauwkeurig uitvoert! Dank voor de gezelligheid en moge **Sarah** en Nala nog veel avonturen beleven. **Anne**, naast tennisvriendin ben je ook mijn Diemenaresse, geliefde oppas van Nala, en exclusief lid van Team QuWi (spaar ze allemaal). Ik kijk nu alweer uit naar de volgende fanatieke spelletjesavond met **Niek** en **Sebas**. **Dolf**, dank voor je inzet voor de club en hoe je altijd trouw mijn bardiensten bijwoont (aan de betere kant van de bar). Ook dank aan de leukste trainers van de club, **Raúl** en **Nico**, ik kan me voorstellen dat lesgeven aan mij soms een vermoeiende taak is, maar ik bewonder hoe jullie het beste uit mij(n spel) naar boven weten te halen!

Heel veel dank aan de beste bokstrainers van Gym020: **Charro** en **Jermaine**, dankzij jullie trainingen kan ik mijn hoofd leegmaken en ga ik elke keer gegarandeerd met méér energie naar huis, ondanks dat ik volledig word afgemat door jullie. Ook een box aan alle mede-boksfanaten op en met wie ik mijn frustraties op de maandag-, dinsdag-, en/of donderdagavond kan uiten en delen! Hardcore!

Lieve, maar vooral stoere(!) **EnduroCats**, waar was ik geweest zonder jullie!? Ik ben zo blij dat ik de eerste WoW in Portugal met jullie heb mogen beleven, wat een blote-billen-pret op en naast de motor hebben we gehad; de blauwe plekken zijn dan wel bijgetrokken maar de buikpijn van het lachen is nog altijd aanwezig als ik terugdenk aan die week! Hoe vaak ik ook onderuit ging, jullie gaven mij de kracht en het vertrouwen om weer op te staan en mijn grenzen nog weer verder te verleggen; ik ben jullie zo ongelooflijk dankbaar voor jullie support en alles wat ik van jullie heb mogen leren. In het bijzonder **Fré**, van jou mocht ik een allereerste enduro-pakje lenen en kijk waar dat toe heeft geleid! **Chris**, bijzonder hoe jij altijd voor me klaar staat ondanks dat je zo ver weg woont, hopelijk slepen wij samen nog eens die Dirtride Queen trophy binnen, girlpowerrr! **Bouwie** je bent een inspiratie en ik rijd het liefste in jouw spoor (lees: achter je billen aan). **Natas**, bedankt voor al je 'wijze lessen', jij bent veruit de leukste (en ondeugendste) roomie! **Nathalie**, ik heb veel bewondering voor jou en ik hoop dat dat wij samen nog eens de HAT gaan rijden. Lieve wify **Eszy**, bedankt voor alle in- en ontspanning op de crossbaan, wakeboardbaan, of waar we dan ook onze adrenaline kwijt konden! Ik hoop dat we samen ooit nog eens meedoen met die zijspanrace. **A3N**, dank voor je vertrouwen dat ik elke keer weer (iets kapot) mocht rijden op je KTM ;) Braap!

Opperaap **Daan**, dank dat ik als gymjuffie mijn energie kwijt kon en verdrievoudigd terugkreeg door alle toffe lesjes en mooie mensen die ik heb leren kennen bij ApenkooiGym. Ook ben ik je oneindig dankbaar dat jij dacht dat enduro 'wel iets voor mij zou zijn' (inmiddels is het mijn grootste hobby!) en me meesleepte naar de cursus van Alex en niet veel later naar Santa Cruz – wat ben ik daar in het diepe (of beter in de hoogte) gegooid door je, thanks voor je support, oprapen van de grond, en bovenal voor alle lol. Lieve **Maus**, jij blijft de allerleukste gymdocent die ik ken. Ik zal onze 'funky monkey fitness' gymles samen nooit vergeten, evenals onze sicke Afrodance moves (Burna Boy zou trots op ons zijn) bij het USC én in de woonkamer tijdens de lockdown. **Han**, dank voor de fijne gesprekken en apenstreken. **Maarten**, thanks voor alle gezellige lesjes die we hebben gedraaid in de Apollohal. De tegeltjeswijsheid "*Doe niet zo stroperig, het waffelt er vanzelf wel uit*" hangt nog altijd boven mijn WC-pot. **Job**, dank voor het verzorgen van de spellen tijdens mijn promotieborrel! Gekke **ApG aapjes**, bedankt voor alle fanatieke potjes trefbal, bloedneuzen, en veels te gezellige borrels na afloop. Ik kom gauw weer een keertje meedoen.

Mijn (motor)buurmannen **Jerry** en **Vitor**, altijd gezellig om met jullie te ouwehoeren, met de dogs te wandelen of een blikje enerydrink te vinden in mijn brievenbus als blijk van support. Dank buuf **Suzan** voor je getoonde interesse en dat je mij uit de brand hebt geholpen met je Wifi.

Romy, mijn alleroudste vriendinnetje vanaf groep 4, ik ben zo blij dat we elkaar weer af en toe zien. Lieve **Jesse**, ik heb jou altijd beschouwd als mijn tweede mama en ik ben je nog steeds dankbaar dat je ons na onze wereldreis een dak hebt geboden.

Shout out naar de **JVT-crew** (**B-licious**, **Wesley** en **Murly**)! Inmiddels allemaal Doctor (zo volwassen, zo beleefd) en hopelijk zetten we ons jaarlijks tripje naar Mallorca voort zodat we onze opblaasflamingo's kunnen blijven gebruiken voor de drijvende GT-bar, we kunnen dansen rondom ons zwembad (of jacuzzi? Merel betaalt dit jaar toch, hierbij zwart op wit ;)) en rappen in de auto op de deuntjes van de Jeugd. **Haryadi** and other **BMEP** party people, thanks for showing me how awesome PhD students can be, you probably inspired me to become one too.

Lieve **Ken**, bedankt voor het organiseren van sportieve uitjes, de veels te zware bootcamp-trainingen, en de leerzame kooklesjes. Ik ben trots dat ik samen met jou mocht jureren bij het Open NK X-Diving van **Indoor Cliff Diving**. **Yorick** and **Elizabeth**, thanks for the lovely board game evenings. **Melvin**, dank voor de gezelligheid en het opgooien van meerdere (pingpong)balletjes. **Roy** – ik hoop nog veel te Groeien met Mezelf dankzij jouw verrijkende workshops!

Erwin wibibol, mede dankzij jou heb ik de baan bij De Hoogstraat gevonden, dank voor je hulp, potjes poolen, droge GIFjes en natuurlijk voor het oppassen op de bol. Lieve **Hilda**, dankjewel dat jij een warm gasthuis bood voor Nala toen ik in een moeilijke periode zat, en haar leven verrijkte met **Sulo** als allerbeste vriendje en eindeloze wandelingen door het Amsterdamse Bos. Lieve **Ivonne**, bedankt dat jij op een dag aanbelde om te vragen of je Nala mocht lenen en dat sindsdien regelmatig hebt gedaan.

Lieve **Rut**, als er iemand was die me wel weer op sleeptouw nam dan was jij het wel – wat hebben wij onvergetelijke avonturen beleefd, samen en met spruitje. Van onze safari door Sri Lanka, stiekeme peukies uit het raam van het JV kasteel, plantjes potjes en nog meer plantjes, op de scooter door Rome sjeezen, tot de vrijdagmiddag geetjes die eindigden op in plaats van aan de bar. Hoeveel Zeebonkjes wij hebben weggetikt, en hoeveel kringloopwinkels wij hebben afgespeurd op zoek naar één of andere schatkist... hopelijk moeten we de grootste nog vinden! Lieve **Fenna**, bedankt dat je altijd interesse bleef tonen in mij en mijn onderzoek (zelfs toen ik al een tijdje uit beeld was). Jouw 'support op afstand' gaf mij elke keer een warm gevoel en ik hoop dat je voor altijd zo'n knettergek mens blijft.

Lieve **schoonfamilie**, dank voor jullie gastvrijheid. Ik heb me altijd erg welkom gevoeld bij jullie over de vloer. **Catootje**, bedankt voor het graveren van alle streepjes in de zijkant van mijn hoofd, en dat je dat gekke beestje zoveel liefde geeft en zelfs hebt omgedoopt tot klimpark-hond – jullie zijn een leuk stel samen!

Allerliefste **pappie & mammie**, bedankt voor jullie onvoorwaardelijke liefde en support. Ondanks dat jullie zeker niet altijd achter de keuzes in mijn leven staan, staan jullie toch altijd voor mij klaar en steunen jullie mij op elke mogelijke manier, of dat nu is in de vorm van een gevulde koelkast, financieel advies, de zoveelste verhuizing, een overheerlijke Indonesische rijsttafel, stalling voor mijn motorfiets, het repareren of vermaken van mijn kleding, of gewoon een stevige knuffel. Ik hou van jullie! Lieve **Rocher** en **Mariëlla**, ook al zien we elkaar niet vaak, het is fijn om te weten dat jullie er altijd voor mij zijn. Lieve **Tante Alice**, bedankt voor al je lieve, vrolijke berichtjes waarmee je mij altijd het gevoel geeft dat je er bent.

Wellicht wat ongebruikelijk maar het is tenslotte míjn dankwoord, dus mag ik ook een klein momentje om **mezelf** te bedanken? Je maakt je het leven soms niet gemakkelijker door zo streng te zijn naar jezelf en je werk: je was vaak je meest kritische reviewer, maar dan wel eentje die jou altijd de mogelijkheid gaf voor een revisie. Maar lief strebertje, ik ben zo ongelooflijk TROTS op je dat je dit hebt gedaan – ik omarm al je imperfecties, emoties, impulsiviteit, en gekkigheid.

Lieve **Seb**, we kenden elkaar al een tijdje maar de vlam sloeg over tijdens de groepsvakantie in Portugal: jij op je surfplank, ik op de enduro. Inmiddels delen we beide hobby's met elkaar, maar niks gaat boven onze offroad adventures. Met jou valt er altijd wel wat (sportiefs) te beleven, of je me nu meesleept naar de 10-metertoren bij ICD, samen met Nala de brug af skaten, handstand-challenges, (illegale) sporttoernooitjes in de gymzaal, of hartjesbillen met octopusbenen. Bedankt voor je altijd vrolijke koppie, onuitputtelijke positiviteit, en grenzeloze liefde. Ik kan niet wachten op de avonturen die we straks gaan beleven op onze motor-wereldreis naar… Mongolië?!

Allerliefste **Nala**, bolletje wol, dikke spruit, maar bovenal: lief mopperkontje van mij. Wat een warmte en geluk dat ik jou vanaf maart 2019, toen thuiswerken de nieuwe normaal werd, weer fulltime in huis kon hebben en je alle aandacht en liefde kon geven die je verdient. Je was het beste excuus om tijdens de lockdown en avondklok tóch nog een wandeling te mogen maken. Bedankt voor je onvoorwaardelijke liefde, jij staat me altijd kwispelend of zelfs zingend(!) op te wachten. Zo fijn dat je mij dagelijks uitlaat, mijn voeten warm houdt onder mijn bureau of op de bank, en dat je me (terecht) om de zoveel tijd kwam afleiden of oppeppen met Meneer de Schildpad en muzikaal gebrom precies wanneer ik mijn laptop uit het raam wilde gooien. Werkelijk iedereen is gek op je. Je bent het liefste, gekste, eigenwijste, wolligste knuffeldier dat ik me ooit kon wensen en ik hoop dat ik nog heel veel jaartjes van je gestink mag genieten ♡

CURRICULUM VITAE

Raquel Yvette Hulst was born on 17 January 1990 in Utrecht, The Netherlands. After completing secondary school at St. Ignatius Gymnasium Amsterdam in 2008, she started a Bachelor's degree in Psychobiology at the University of Amsterdam. Her interest in the pathophysiology of the brain was sparked, and in 2012 she continued a Master's degree in Biomedical Sciences with a focus on Neuroscience. As part of her studies, Raquel performed a research internship at the Biomedical Primate Research Centre and at the Netherlands Institute for Neuroscience. In her second year, she was awarded two scholarships to pursue her final research project abroad at the Cognitive and Behavioural Neuroscience lab at the University of Wellington, New Zealand. In 2014, Raquel obtained her Master's degree (cum laude) from the University of Amsterdam.

The following two years Raquel continued on a world trip, working and travelling in several continents from Australia to South America. Upon return to The Netherlands in 2016, Raquel worked at the Biomedical Engineering & Physics department and thereafter at the Psychiatry department at the former Academic Medical Center Amsterdam. She then spent a year in education coordinating the Master's program Forensic Science at the University of Amsterdam.

Since April 2018, Raquel has fulfilled the role of Project Coordinator at De Hoogstraat Rehabilitation on a 3-year healthcare innovation project, which aimed to increase attention to the importance of lifestyle factors (sleep, nutrition, and physical activity) in the rehabilitative care for children with cerebral palsy (CP). In parallel, Raquel conducted scientific research studying sleep among children with CP and their families, which resulted in this dissertation. During her PhD, she participated in the research educational program 'Clinical and Experimental Neuroscience' at the Graduate School of Life Sciences at the Utrecht University.

In the spring of 2022, Raquel will embark on a round-the-world journey by motorcycle, chasing dreams and adventures!

LIST OF PUBLICATIONS

International publications

Hulst RY, Voorman JM, Pillen S, Ketelaar M, Visser-Meily JMA, Verschuren O. Parental perspectives on care for sleep in children with cerebral palsy: A wake-up call. Disabil Rehabil. 2022;44(3):458–67.

Hulst RY, Gorter JW, Voorman JM, Kolk E, Van Der Vossen S, Visser-Meily JMA, Ketelaar M, Pillen S, Verschuren O. Sleep problems in children with cerebral palsy and their parents. Dev Med Child Neurol. 2021;63(11):1344–50.

Verschuren O, **Hulst RY**, Voorman JM, Pillen S, Luitwieler N, Dudink J, Gorter JW. 24-hour activity for children with cerebral palsy: A clinical practice guide. Dev Med Child Neurol. 2021;63(1):54–9.

Hulst RY, Pillen S, Voorman JM, Rave N, Visser-Meily JMA, Verschuren O. Sleep health practices and sleep knowledge among healthcare professionals in Dutch paediatric rehabilitation. Child Care Health Dev. 2020;46(6):703–10.

Smit DJM, Zwinkels M, Takken T, **Hulst RY**, de Groot JF, Lankhorst K, Verschuren O. Sleep quantity and its relation with physical activity in children with cerebral palsy; insights using actigraphy. J Paediatr Child Health. 2020;56(10):1618–22.

Manuscripts under review

Hulst RY, Gorter JW, Obeid J, Voorman JM, Van Rijssen IM, Gerritsen A, Visser-Meily JMA, Pillen S, Verschuren O. Accelerometer-measured physical activity, sedentary behaviour, and sleep in children with cerebral palsy and their adherence to the 24-hour activity guidelines.

Van Rijssen IM, **Hulst RY**, Gorter JW, Gerrtisen A, Visser-Meily JMA, Dudink J, Voorman JM, Pillen S, Verschuren O. Device-based and subjective measurements of sleep in children with Cerebral Palsy: A comparison of sleep diary, actigraphy, and bed sensor data.

Manuscripts in preparation

Uelkes BU, **Hulst RY**, Bloemen M, van Es J, Verschuren O. Parental perceptions and needs regarding the physical activity management of their child with cerebral palsy: a comparison between ambulatory vs. non-ambulatory children.

Gerritsen A, **Hulst RY**, Van Rijssen IM, Obeid J, Pillen S, Gorter JW, Verschuren O. The temporal and bi-directional relationship between physical activity and sleep in ambulatory children with cerebral palsy.

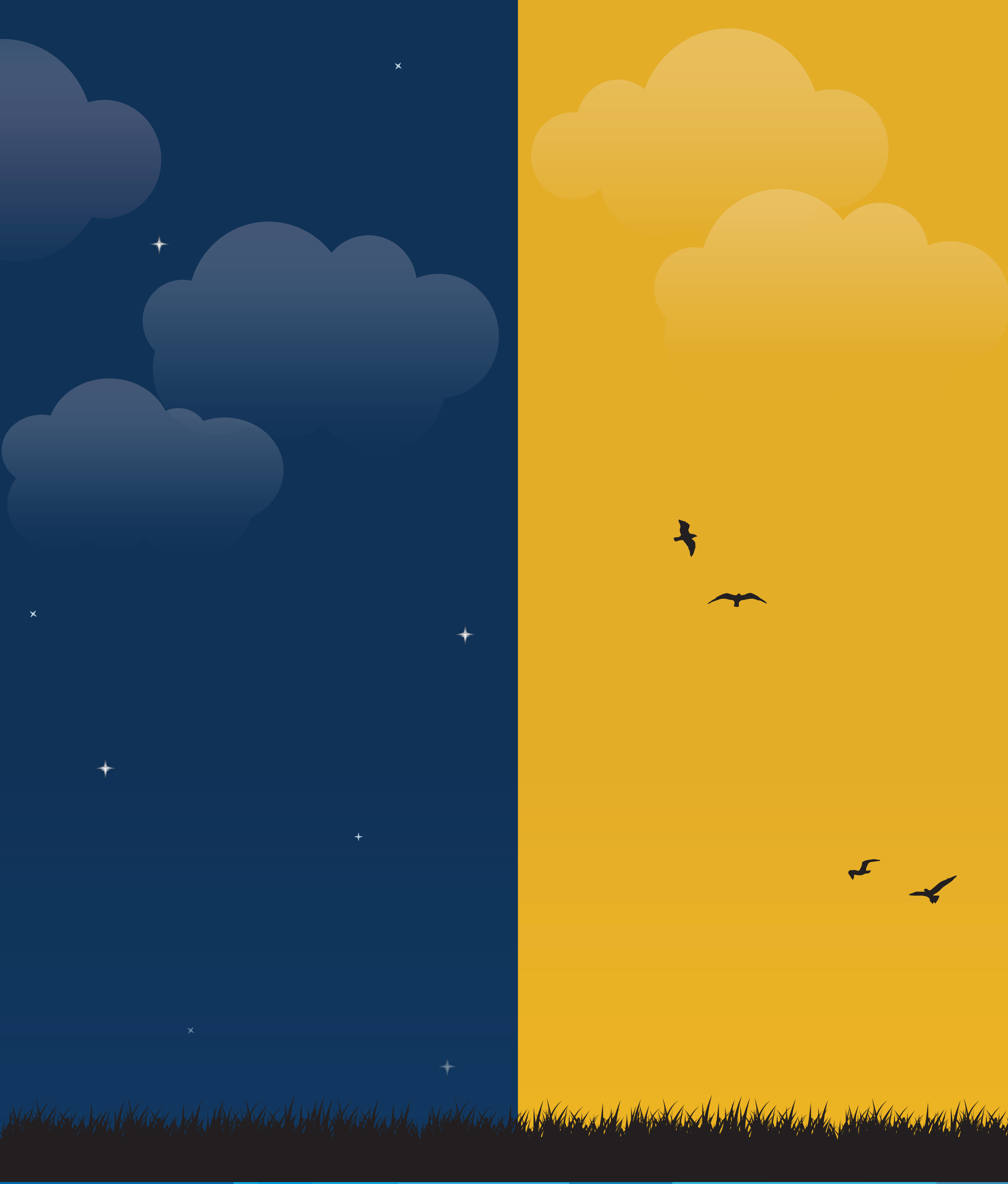
Breij LM, Ven van de RAH, **Hulst RY**, Bonovrié-Smorenburg A, Gorter JW, Verschuren O. Large inter-subject variability in body composition described by body mass index and fat mass in children with cerebral palsy.

Jansen L, **Hulst RY**, van Rijssen IM, Gerritsen A, Mulders H, Verschuren O. The interplay between sleep and behaviour problems in preschool and school-aged children with cerebral palsy.

National publications

Kolk E, **Hulst RY**, van der Vossen S, Medema D, Kuijper M, Heeneweer H, Verschuren O. Slaap bij kinderen met cerebrale parese: een onderbelicht probleem. Nederlands Tijdschrift voor Revalidatiegeneeskunde. December 2020.

Hulst RY, Verschuren O, Dudink J. Het grote belang van slaap voor patiënten. V&VN Magazine Kinderverpleegkunde. August 2019.



UMC Utrecht

Universiteit Utrecht

ISBN 978-90-393-7448-1