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Publication date

2023

Document Version

Final published version

[Link to publication](#)

Citation for published version (APA):

Schouwenaars, I. M. G. (2023). *The interplay between sleep and daytime functioning in autistic teenagers*. [Thesis, externally prepared, Universiteit van Amsterdam].

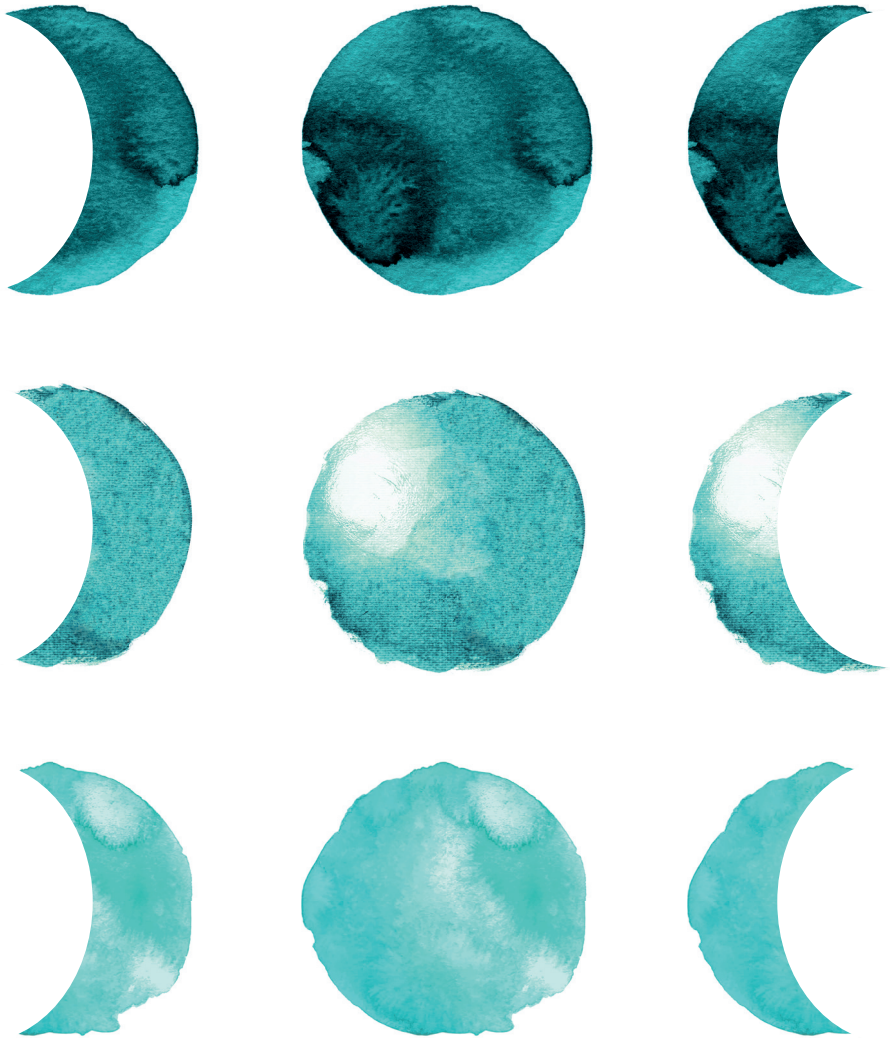
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THE INTERPLAY BETWEEN SLEEP AND DAYTIME FUNCTIONING IN AUTISTIC TEENAGERS



ILONA SCHOUWENAARS

The interplay between sleep and daytime functioning in autistic teenagers

Ilona Schouwenaars

ISBN: 978-94-6473-162-0

Cover and inside layout by: Bregje Jaspers | www.ProefschriftOntwerp.nl

Printed by: Ipskamp Printing

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The interplay between sleep and daytime functioning in autistic teenagers

ACADEMISCH PROEFSCHRIFT

er verkrijging van de graad van doctor

aan de Universiteit van Amsterdam

op gezag van de Rector Magnificus

prof. dr. ir. P.P.C. Verbeek

ten overstaan van een door het College voor Promoties ingestelde commissie,

in het openbaar te verdedigen in de Agnietenkapel

op donderdag 28 september 2023, te 13.00 uur

door Ilona Maria Geertruida Schouwenaars

geboren te Heusden

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CHAPTER 1

General Introduction

"I am a poor sleeper. I especially have problems with falling asleep and waking up. Before I can fall asleep I need time to process the day, and this often takes an hour or longer. The most challenging aspect for me is getting up in the morning. My alarm goes off at 7 AM, but it takes me a long time to wake up and I require encouragement from my parents. Furthermore, in the morning I am irritable and have difficulties concentrating at school." (A 17-year-old boy with autism spectrum disorder who participated in our study)

Autism spectrum disorder (ASD) is a condition that involves problems with social communication and social interaction across several contexts. People with ASD also exhibit restricted and repetitive patterns of behavior, interests, or activities. The symptoms range from mild to severe, and while they usually start in early childhood, they can be recognized at a later age (American Psychiatric Association, 2013). ASD is also a heterogeneous condition that includes individuals with autistic¹ traits and individuals with prototypical autism (people who experienced clinicians recognize as autistic) (Motttron & Bzdok, 2020). Co-occurring conditions often accompany autism spectrum disorder (Simonoff et al., 2008), and sleep problems are common (Mannion et al., 2013). In this thesis, I follow the ASD classification in the Diagnostic and Statistical Manual of Mental Disorders, fourth edition, text revision (DSM-IV-TR; American Psychiatric Association, 2000) or fifth edition (DSM-5; American Psychiatric Association, 2013). I focused on autistic teenagers (aged 12 – 20) with co-occurring sleep problems. All participating teenagers attend schools for special education and have no significant limitations in intellectual functioning.

This thesis project originated from interactions with teachers at Dutch schools for special education who expressed a need for an evidenced-based sleep intervention to adequately address autistic teenagers' sleep problems at school. The aim of this thesis was twofold: (i) to better understand the relationship between sleep and daytime functioning in autistic teenagers (Chapters 2 and 3), and (ii) to develop a school-based intervention to improve sleep in autistic teenagers and test the efficacy of that intervention (Chapters 4 and 5).

1.1 Sleep and daytime functioning in autistic teenagers

In general, the teenage years are characterized by insufficient sleep and daytime sleepiness (Moore & Meltzer, 2008). As children enter adolescence, their sleep pattern changes to include later bedtimes and waking times (e.g., a phase delay). Mostly this is caused by physiological changes (puberty) and psychosocial factors such as an after-school job, homework, or time spent with peers (Dahl & Lewin, 2002; Moore & Meltzer, 2008). As a result, their total sleep duration decreases, bedtimes are later, and there are increasing disparities between sleep schedules on school and weekend nights (Garipey et al.,

¹ There is no clear agreement about how to describe autism (Buijsman et al., 2022). In this paper we use identity-first language instead of person-first language, without the intention to disregard the various terms used to describe autism.

2020). Although sleep problems are common among all teenagers, they are even more prevalent among autistic teenagers: reported prevalence rates range from 58% to 88% in autistic teenagers compared to 7% to 38% in typically developing teenagers (Hodge et al., 2014; Paavonen et al., 2008). Hence, greater insight into these sleep problems is a necessary precursor to effective intervention.

Research has shown that autistic teenagers have poorer sleep quality than typically developing teenagers. Autistic teenagers have a longer sleep onset latency, longer night time awakenings, a shorter sleep duration, and a lower sleep efficiency (percentage of total time in bed spent asleep) (Tse et al., 2020). While we have known for some time that sleep problems are prevalent among autistic teenagers (Allik et al., 2006; Cortesi et al., 2010; Hodge et al., 2014; Paavonen et al., 2008; Tse et al., 2020), the underlying cause is not yet clear. However, researchers have identified several possible factors related to the ASD diagnosis in addition to associations with adolescence. For example, biological abnormalities (e.g., disturbed production of melatonin), medical or psychological co-occurring conditions (e.g., epilepsy, anxiety), and use of medication to treat the co-occurring conditions (Johnson & Malow, 2008) are factors that might have additional effects on sleep quality in autistic individuals. Thus, sleep problems can relate to many factors. Independent of which factor, poor sleep quality in itself can have a negative impact on daytime behavior (Dahl & Lewin, 2002; Gregory & Sadeh, 2012).

No one is surprised when one argues that sleep problems and daytime functioning are intertwined. Extensive research into typically developing teenagers has found that sleep problems can have significant consequences for daytime functioning, such as daytime sleepiness, fatigue, mood disturbances, difficulties with attention, and performance and behavioral problems (Dahl & Lewin, 2002; Gregory & Sadeh, 2012). Among autistic individuals, it is additionally found that the sleep problems also negatively affect the severity of autism traits, internalizing behavior, externalizing behavior, and problems with social behavior (Allik et al., 2006; Gabriels et al., 2005; Mazurek & Sohl, 2016; Phung et al., 2018; Richdale et al., 2014; Saré & Smith, 2020). However, knowledge about how sleep problems affect the lives of people with an ASD diagnosis remains rather fragmented. Even less research has focused on autistic teenagers.

Since the nature (Tse et al., 2020) and underlying causes (Cortesi et al., 2010) of sleep problems among autistic teenagers might differ from those of typically developing teenagers, the results of studies performed with typically developing teenagers cannot automatically be applied to autistic teenagers. Moreover, since sleep changes when children enter adolescence (Moore & Meltzer, 2008) and the nature of sleep problems changes with age (Goldman et al., 2011), it is important to focus on this specific population.

To gain a better understanding of the relationship between sleep and daytime functioning in autistic teenagers, we conducted a systematic review (Chapter 2). The research question for this review was: *What is the relationship between sleep and daytime functioning in autistic teenagers?* Specifically, we focused on sleep problems in general and on four specific sleep parameters that are known to be

commonly disturbed in autistic teenagers (Tse et al., 2020) and could be improved by behavioral sleep interventions (Johnson & Zarrinnegar, 2021). The parameters were total sleep time, sleep onset latency, sleep efficiency, and sleep timing.

Based on the results of the systematic review, we conducted a second study (Chapter 3) to gain a comprehensive picture of the relationship between specific sleep parameters and daytime functioning. The research question for this study was: *To what extent are characteristics of sleep (sleep onset latency, total sleep time, wake time after sleep onset, and sleep quality) related to aspects of daytime functioning (mood, concentration, daytime sleepiness)?* We used a network approach (Costantini et al., 2015) to identify the network structure between sleep variables (self-reported and objectively measured) and aspects of daytime functioning reported by the teenagers themselves and their parents and teachers. The network structure could reveal the complex pattern of direct and indirect relationships of sleep and factors influencing sleep, and identifying these patterns could give us relevant information about which variables stimulate or inhibit others. A network analysis can unravel the network structure between sleep variables and aspects of daytime functioning, which reveals the complexity of interacting variables (Borsboom & Cramer, 2013). We included both objective measurements and self-reports of sleep because self-reports and objectively measured sleep could reveal a different association with daytime functioning (Richdale et al., 2014). In addition, different informants' reports of a person's daytime functioning often show discrepancies (Youngstrom et al., 2001). These discrepancies contribute relevant information that can be used to draw a comprehensive picture of a person's daytime functioning (Achenbach, 2006). Therefore, we included different informants' perspectives of the teenagers' daytime functioning.

1.2 School-based sleep intervention

Behavioral sleep interventions have been shown to be effective in reducing sleep problems (Johnson & Zarrinnegar, 2021). However, those are mostly delivered in clinical practice (e.g., McCrae et al., 2020; Papadopoulos et al., 2019). Sleep interventions that could be delivered at schools are thought to be more easily accessible for school-aged youth and to therefore have a potentially wider reach. Moreover, easy accessibility of school-based sleep interventions might allow sleep problems to be treated earlier, thus avoiding exacerbation of them (Morin & Benca, 2012). Therefore, creating and testing a school-based intervention is the focus of this thesis.

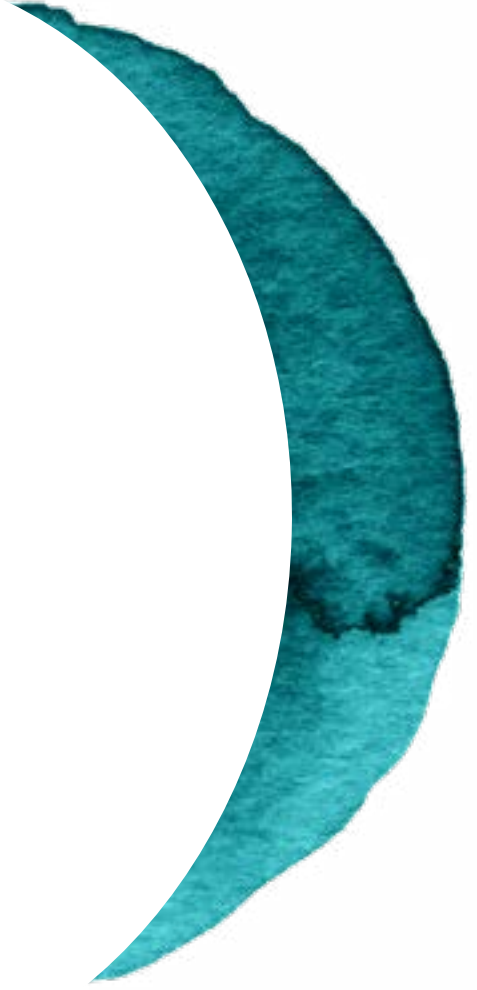
However, it is challenging to successfully implement evidence-based interventions in schools (Parsons et al., 2013). This is also true for sleep interventions, although we have seen that schools are willing to support their students in reducing sleep problems (Cassoff et al., 2013). Therefore, it is important to consider five domains during the development process to increase the likelihood of successful implementation (Damschroder et al., 2009). These domains refer to the characteristics of the intervention (e.g., the ability to test it, its perceived advantages, and the ability to adapt it to specific needs); the inner

setting (e.g., characteristics and culture of the organizations); the outer setting (e.g., alignment with future users' needs); characteristics of individuals involved with the intervention (e.g., knowledge and skills, identification with the organization); and the planning and executing of the implementation process itself (Damschroder et al., 2009). Together, these factors determine the adoption of an intervention. Interventions developed in co-creation pay attention to these issues and could increase the likelihood that an intervention is accepted and sustained (Robertson & Simonsen, 2012). Therefore, we co-created a school-based sleep intervention together with autistic teenagers, their parents and teachers, and relevant professionals.

In Chapter 4, we describe the co-creation of a school-based sleep intervention. In the development, we used the first three phases of the Centre for eHealth & Wellbeing Research (CeHReS) roadmap (van Gemert-Pijnen et al., 2011): contextual inquiry, value specification, and design. Each phase had specific goals and activities that led to iterative development with the active involvement of autistic teenagers, their parents and teachers, and relevant professionals. Contextual inquiry involves an investigation of the future users' daily routines and habits, how the intervention could improve them, and the weak and strong points of the current situation. The contextual inquiry provides general insight into the added value of a specific intervention. Value specification elaborates on the previous phase by identifying and prioritizing specific requirements for the intervention. In the design phase, the intervention is designed based on those requirements. The findings described in Chapters 2 and 3 were used as input for this co-created school-based sleep intervention that may reduce sleep problems and consequential effects on daily life in such a crucial life phase for autistic teenagers.

Since this sleep intervention was developed in co-creation and based on knowledge about evidence-based sleep interventions, we have good indications that it could be effective in reducing sleep problems. Thus, the next step was to investigate the efficacy of our co-created sleep intervention (Chapter 5). The research questions for this study were: (1) *What are the effects of our co-created school-based sleep intervention on the sleep quality of autistic teenagers?* And (2) *What are the effects of our co-created school-based sleep intervention on the daytime functioning of autistic teenagers?* We used a multiple baseline design to evaluate the efficacy of the intervention. Like a randomized controlled trial, a multiple baseline design can show the occurrence of change in outcome measures over time, but it requires a limited number of participants (Hawkins et al., 2007). The duration of the baseline was two to three weeks, and the intervention period was ten weeks for each participant. Follow-up data were collected eight weeks after the intervention during one entire week. The teenagers completed a daily web-based sleep diary and wore a sleep tracker during the night. They also answered questions about their daytime functioning, as did a parent and a teacher of each teenager. The teenagers, parents, and teachers also completed questionnaires (about sleep problems [ISI and CSHQ], and behavioral problems [SDQ]) at pretest (baseline), post-test (end of intervention period), and follow-up. In Chapter 6, I summarize the results of this thesis and offer practical and research recommendations.





Chapter 2

Sleep problems and daytime functioning in autistic teenagers: A systematic review

Schouwenaars, I. M. G., Magnée, M. J. C. M., Geurts, H. M., van Bennekom, C. A. M., Pillen, S., & Teunisse, J. P. (2021). Sleep problems and daytime functioning in adolescents with an autism spectrum disorder diagnosis: A systematic review. PsyArXiv. <https://doi.org/10.31234/osf.io/hjy6p>

2.1 Abstract

Sleep problems are common among autistic teenagers. However, there is only fragmented knowledge about how sleep problems affect those teenagers' lives. This review focuses on the relationship between sleep and daytime functioning in autistic teenagers. We performed an electronic database search (in APA PsycINFO, Cochrane, ERIC, PubMed, and Web of Science) and a hand search (INSAR congress archive and included studies reference lists) in September 2020 that resulted in 2561 studies. Studies were included if they related sleep to daytime functioning, were available in English, used original data, and included autistic participants between the ages of 10 and 19 without intellectual disability. Ultimately, we included nine studies in this review ($N_{\text{participants}} = 674$; $M_{\text{number of participants}} = 75$, $SD = 98$, range from 8 to 314). The most prominent finding was the association of all sleep parameters with internalizing problems, and sleep problems in general and sleep efficiency had the most significant associations with daytime functioning. The results provide input for tailored sleep interventions. However, more research is needed to get a clearer picture of the relationship between specific sleep parameters and daytime functioning and to ensure that this knowledge will provide better input for sleep interventions. It is important to focus on limited age ranges, larger samples sizes, multiple measurement methods, and longitudinal approaches.

2.2 Introduction

Sleep problems are common among teenagers with an autism spectrum disorder diagnosis (ASD) (Cortesi et al., 2010): the prevalence rate for autistic teenagers is 88% compared to 38% for their typically developing (TD) peers (Hodge et al., 2014). A growing number of studies show that sleep problems have a major impact on daytime functioning in TD teenagers, as disturbed sleep is related to mood disturbances, anxiety, school performance, difficulties with attention, and behavioral problems (Curcio et al., 2006; Dahl & Lewin, 2002; Dewald et al., 2010; Gregory & Sadeh, 2012; Orchard et al., 2020). However, little research has investigated the impact of sleep problems on daytime functioning in autistic teenagers. Although we might suspect that sleep problems would have a similar impact on autistic teenagers, the results of studies on TD teenagers cannot be automatically applied to autistic teenagers because the nature and underlying causes of their sleep problems differ. In this review, we summarize what is known about the relationship between sleep and daytime functioning in autistic teenagers.

The underlying cause of the increased prevalence of sleep problems in autistic people has not been clarified, but it seems to be associated with several ASD-related factors (Cortesi et al., 2010). These include biological abnormalities (e.g., disturbed production of melatonin), medical or psychological co-occurring conditions (e.g., epilepsy, anxiety), and use of medication to treat the co-occurring conditions (Johnson & Malow, 2008). All these factors lead to different expressions of sleep problems. Autistic teenagers have difficulties initiating and/or maintaining sleep and both a shorter sleep duration and a lower sleep efficiency than TD teenagers (Tse et al., 2020). Researchers also have identified developmental factors that contribute to sleep problems. Developmental factors specific to teenagers further interfere with sleep-wake patterns at this age because adolescents make a prominent sleep phase shift that results in later sleep timing and increasing differences in rise time between weekdays and weekends (Moore & Meltzer, 2008). All these factors may have different effects on aspects of daytime functioning, and they need to be considered when studying the relationship between sleep problems and daytime functioning in specific age groups with ASD.

The first steps taken to reduce sleep problems are behavioral sleep interventions (Crowe & Salt, 2015). On a group level, total sleep time can be increased by creating awareness about the importance of sleep and sleep hygiene (Keogh et al., 2019). Other interventions, such as faded bedtime and sleep restriction, are also effective in initiating and maintaining sleep (Johnson & Zarrinigar, 2021). Problems with sleep-wake patterns require sleep timing interventions that may involve light exposure and melatonin supplements (Johnson & Zarrinigar, 2021). Therefore, we need a better understanding of the relationship between specific sleep parameters and the impact on the lives of those with ASD to determine which intervention may be the most successful at improving both sleep and daytime functioning.

We conducted a systematic review to improve our understanding of the relationship between sleep and daytime functioning in autistic teenagers. We focused on sleep problems in general, and on four specific sleep parameters known to be commonly disturbed in autistic teenagers: total sleep time, sleep onset latency, sleep efficiency (percentage of total time in bed actually spent asleep), and sleep timing (Tse et al., 2020). Daytime functioning is broadly operationalized and includes aspects associated with disturbed sleep in TD teenagers: school functioning, attention, memory, behavioral problems, mood disturbances, sleepiness, and anxiety (Curcio et al., 2006; Dahl & Lewin, 2002; Dewald et al., 2010; Gregory & Sadeh, 2012; Orchard et al., 2020). Taken together, this resulted in the following research question: *What is the relationship between sleep (sleep problems in general, total sleep time, sleep onset latency, sleep efficiency, and sleep timing) and daytime functioning (externalizing behavior, internalizing behavior, autism symptom severity, social behavior, attention, daytime sleepiness/fatigue) in autistic teenagers?*

2.3 Methods

We investigated the relationship between sleep and daytime functioning in autistic teenagers following the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines (Moher et al., 2009).

2.3.1 Inclusion criteria

The following inclusion criteria were applied to select studies relevant to this review. The study must:

- include autistic participants: people diagnosed with autistic disorder, Asperger syndrome, or pervasive developmental disorder, not otherwise specified (PDD-NOS) following the DSM-IV (American Psychiatric Association, 1994) or DSM-IV-TR (American Psychiatric Association, 2000), diagnosed with ASD following DSM-5 (American Psychiatric Association, 2013), or autism following the ICD-10 (World Health Organization, 1993);
- include teenagers: defined as people aged 10 – 19 (World Health Organization, n.d.). We included studies where at least one of the following three conditions was met: 1) all participants were between the ages of 10 and 19; 2) the participants' mean age +/- 1 SD was 10 - 19 (in many studies the overall age range is not mentioned); 3) it reported on a subgroup of this age range;
- examine sleep problems in general, total sleep time, sleep onset latency, sleep efficiency, or sleep timing (which refers to sleep-wake patterns) related to daytime functioning;
- include teenagers without intellectual disability: defined by an IQ of 70 or higher (American Psychiatric Association, 2021) or by typical to high adaptive functioning concerning language. We focused on teenagers without significant limitations in intellectual functioning because the degree of impairment differs greatly between individuals with and without intellectual disability which would introduce heterogeneity (Kanne et al., 2011). We also included studies reporting on a subgroup of teenagers without intellectual disability;
- be available in English;

- be based on original data.

We set no restrictions on publication date.

2.3.2 Search strategy

We performed a combined electronic and hand search to obtain relevant studies. The electronic search was performed in September 2020 using the following databases: APA PsycINFO (OVID interface), PubMed, ERIC (EBSCO interface), Web of Science, and Cochrane. Search terms were various forms and combinations of ASD (see above), sleep, adolescence, school functioning, attention, memory, stress, behavioral problems, mood, and sleepiness (see supplementary material in Chapter 10.1 for details). The search strategy was developed in collaboration with the institutional information specialists who had expertise in systematic review searching. A hand search was then performed to identify additional relevant studies. This included the reference lists of included studies and the congress archive (2004 – 2020) of the International Society for Autism Research (INSAR), the largest worldwide scientific conference on ASD. The INSAR congress archive was searched for the word 'sleep.'

2.3.3 Study selection

We conducted the screening on inclusion criteria in three phases. The first screening (IS) was on language, population, and publication type; we excluded books, reviews, dissertations, and editorials.

Second, the title and abstract of the remaining studies were screened independently by four reviewers (IS, MM, JPT and a research assistant). Each study was screened by at least two reviewers. Half of the studies screened by IS and the research assistant were then randomly screened as an additional check by a senior researcher (MM or JPT). When disagreements arose, reviewers discussed them to reach consensus about the inclusion of studies. If the relevance of a study was difficult to assess based on the title and abstract, the full text of the study was screened. We emailed the authors of congress abstracts (N = 86) to ask whether their research had been published as a journal article. The study was excluded if the research was not published (N = 26) or if there was no response to the email within three weeks (N = 47; no reminders were sent).

Third, the full texts of the remaining studies were screened following the same procedure as in the second phase. If questions arose, we emailed the authors (N = 7) with a request for more information. If the research did not meet our inclusion criteria after clarification (N = 4) or we received no response to our request after three weeks (N = 3), we excluded the study based on the doubts about the inclusion criteria.

2.3.4 Data collection

The included studies were summarized by the first author. All authors agreed on the guidelines used for these summaries: the research problem, characteristics of participants, investigated sleep parameters and aspects of daytime functioning, instruments, and main results. If any uncertainties arose about the

interpretation of the study results, these were discussed by the authors of this review. Since the variables measuring daytime functioning were diverse, we combined the extracted information in the following categories:

- externalizing behavior (aggression, rule-breaking behavior, externalizing behavior in general);
- internalizing behavior (anxiety, depressive symptoms, somatic complaints, affective problems, internalizing behavior in general);
- autism symptom severity (social awareness, social cognition, social communication, social anxiety/avoidance, autistic traits, autism symptom severity in general). We use the term 'autism symptom severity' because it is commonly used in the studies included in the review. However, one could argue that if we do not embrace the medical model of ASD, we should rename these 'autism traits' or 'autistic characteristics.' Since we included the DSM terminology in our literature search, we opted for consistency in terminology and used the term 'autism symptom severity' instead of 'autistic traits/characteristics';
- social behavior (social problems, closeness and discord in relationships);
- memory;
- attention;
- daytime sleepiness/fatigue.

We included the following in this review: 1) characteristics of the group participants (age, diagnosis, IQ), 2) investigated sleep parameters and aspects of daytime functioning, and 3) the relationships between sleep and daytime functioning and the effect sizes. First, we described significant associations ($p \leq .05$) between sleep and daytime functioning, followed by large and medium-sized ($\geq .3$) but non-significant correlations.

If we observed overlap in authors, sample size, mean age, diagnosis of the participants, or outcome measures, we verified whether the included studies reported on the same study samples. The method section was also compared with the results section of each study for indications of selective report bias. If we discovered any differences in the method and result sections, the missing information was reported as a non-significant result and the reference was marked with an asterisk.

The quality of the included studies was evaluated independently by three reviewers (IS, MM, JPT) using the Standard Quality Assessment Criteria for Evaluating Primary Research Papers (Kmet et al., 2004). Each study was reviewed by two reviewers (IS and MM or IS and JPT). The quality assessment included 14 criteria to evaluate each study on the following aspects: research question and design, group selection, sample size and description of the participants' characteristics, outcome measures, analytic methods, description of results and whether the conclusions were supported by the results. The criteria were rated depending on the degree to which each criterion was met (fully = 2, partially = 1, not at all = 0). Disagreements between reviewers were discussed at the item level to reach consensus. The quality score was then calculated by summing the ratings and dividing that by the total possible score. Criteria

that were not applicable to a specific study were excluded from the quality score calculation. A higher score indicates a higher quality.

2.4 Results

2.4.1 Study selection

The electronic database search resulted in 2561 studies: 655 from APA PsycINFO, 651 from PubMed, 139 from ERIC, 202 from Cochrane, and 914 from Web of Science. In addition, 565 studies were identified by the hand search. After following the steps described in the Methods section (Figure 2.1), we included nine studies in the review. Disagreements about inclusion were discussed and consensus was reached in all cases.

2.4.2 Study characteristics

The nine studies were published between 2007 and 2019 and included a total of 674 participants ($M_{\text{number of participants}} = 75$, $SD = 98$, range 8 – 314). Table 2.1 gives an overview of all study characteristics.

The quality of each study was assessed. The summary scores range from .68 to 1.00, with a mean score of .85. The main weaknesses of the studies were: 1) incomplete or lack of controlling for confounding (two studies did not control for potential confounding variables and four only partially) and 2) limited information used to assess the sample size or small sample sizes (seven studies provided partial details). Despite these weaknesses, we concluded that, in general, the quality of the studies was good. Accordingly, we treated all studies equivalently regarding the description of the relationship between sleep and daytime functioning.

2.4.3 Different aspects of daytime functioning in relation to sleep

We investigated a broad range of daytime functioning variables in relation to sleep problems in general, total sleep time, sleep onset latency, sleep efficiency, and sleep timing (Table 2.1). For each aspect of sleep, we described the relationship with the selected aspects of daytime functioning (Table 2.2).

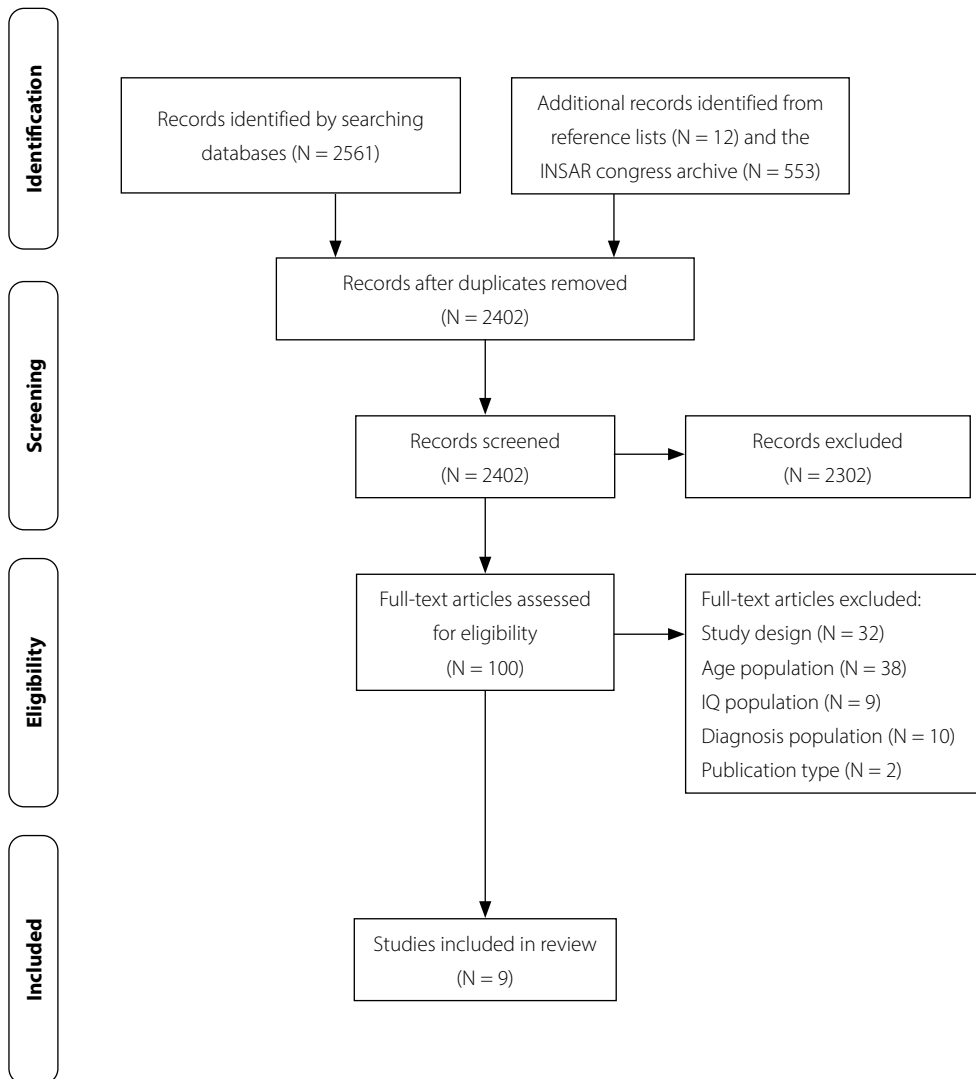
Figure 2.1 PRISMA Flow Diagram – Selection of Studies

Table 2.1 Overview of Characteristics of Included Studies

Citation	Design	Sample ^a	Diagnosis	IQ	Mean age (SD)	Measures of sleep ^b	Measures of daytime functioning ^b	Quality appraisal ^c
Baker et al., 2013 ^d	Cross-sectional	27	ASD confirmed by a score >11 on the SCQ (parent report), diagnostic report, or being a client of an autism network	≥70 or read at (above) average level	15.5 (1.3), range: NR	SOL (ACT), TST, SE (sleep diary)	Daytime sleepiness (paediatric daytime sleepiness scale), fatigue (Flinders Fatigue Scale)	.82
Brown et al., 2019	Cross-sectional	120	ASD confirmed by ADOS and ADI-R	N = 110 with full-scale IQ ≥ 70	14.2 (2.1) Range: 11-17	Sleep problems total score (CSHQ)	Physical and verbal aggression (C-SHARP)	.86
Bruni et al., 2007	Cross-sectional	8	AS diagnosed based on comprehensive and multidisciplinary assessments and ADOS, confirmed by interview with parent	NR	12.7 (2.6), range: 7-15	TST, SOL, SE (PSG)	Internalizing and externalizing behavior (CBCL)	.68
Calhoun et al., 2020	Cross-sectional	96	ASD diagnosed by psychologist based on comprehensive test	Mean full scale IQ = 101, SD = 14	12.5 (1.6), range: 11-17	Sleep problems total score (PBS)	Learning problems (PBS), working memory (WISC-IV WMI, WISC-V WMI, WAIS-III WMI), autism symptom severity (CASD)	.91
Johansson et al., 2018	Cross-sectional	314	A(S)D based on observation, medical history, chart review, ADOS, ADI-R	68.2% NVIQ >70	Late childhood: 15.9 (1.1), range: NR	Sleep duration problems (Simons Simplex Collection Sleep Interview)	Autism symptom severity (ADOS)	1.0
Phung & Goldberg, 2017	Cross-sectional	Study 1: 28 Study 2: 20	ASD based on parent-reported clinical diagnosis and confirmed by SCQ or ADOS-2 modules 3 and 4	Study 1: Mean full scale IQ = 95, SD = 19 Study 2: NR	Study 1: 14.6 (2.0), range: 12-18 Study 2: 16.7 (2.5), range: 11-20	Study 1: Sleep problems total score (CSHQ) Study 2: TST, SE (ACT)	Study 1: Daytime sleepiness (SHS), affective problems, anxiety problems, externalizing problems (CBCL) Study 1 and 2: Depressive symptoms (CES-D), closeness and discord in relationship (NRI-RQV)	.86

Table 2.1 Continued

Phung et al., 2018	Cross-sectional	19	ASD confirmed by SCQ and ADOS-2 modules 3 and 4	NR	16.9 (2.5), range: 11–20	TST, SE, SOL (ACT)	Closeness and discord in relationships (NRI-RQV)	.82
Richdale et al., 2014 ^a	Cross-sectional	27	ASD confirmed by SCQ, diagnostic report, or being a client of an autism network	≥ 70 or read at (above) average level	15.5 (1.3), range: NR	Bedtime, SE (sleep diary), sleep onset time (ACT), wake time, sleep length, SOL (sleep diary and ACT)	Depressed mood (CES-D), anxiety (DASS-21).	.82
Thenhausen et al., 2017	Cross-sectional	15	AS or ASD diagnosed by psychiatrist and confirmed by psychologist	NR	14.3 (3.0), range: NR	Sleep problems total score (Sleep Disturbance Scale for Children)	Autism symptom severity (SRS), rule-breaking, aggressive, internalizing and externalizing behavior, attention and social problems, anxious/depressed, withdrawn/ depressed, somatic complaints (CBCL)	.85

NR = not reported, ASD = autism spectrum disorder, SCQ = Social Communication Questionnaire, ADOS = Autism Diagnostic Observation Scale, ADI-R = Autism Diagnostic Interview-Revised, AS = Asperger's disorder, AD = autistic disorder, NVIQ = nonverbal Intelligence Quotient, SOL = sleep onset latency, ACT = actigraphy, TST = total sleep time, SE = sleep efficiency (percentage of total time in bed actually spent asleep), CSHQ = Children's Sleep Habits Questionnaire, PSG = polysomnography, PBS = Pediatric Behavior Scale, C-SHARP = Children's Scale for Hostility and Aggression: Reactive/Proactive, CBCL = Child Behavior Checklist, WISC-IV/WI WMI = Wechsler Intelligence Scale for Children-IV Working Memory Index, WAIS-III WMI = Wechsler Adult Intelligence Scale-III Working Memory Index, CASD = Checklist for Autism Spectrum Disorder, SHS = Sleep Habits Survey, CES-D = Center for Epidemiological Studies Depression Scale, NRI-RQV = Network of Relationships Inventory-Relationship Qualities Version, DASS-21 = Depression Anxiety Stress Scale, SRS = Social Responsiveness Scale

^a Sample size of the teenage (subgroup) included in the review.

^b Only outcome measures included in this review.

^c Quality scores ranging from .00 to 1.0, with a higher score indicating a higher quality.

^d Two studies investigated the same population. Since the studies' outcome measures are different, there is no risk of multiple publication bias.

Table 2.2 Overview of Associations for the Relationship between Sleep and Daytime Functioning

A. Sleep problems total score	Externalizing behavior	Internalizing behavior	Autism symptom severity	Social behavior	Attention	Daytime sleepiness/fatigue
Parent- or self-report						
Brown et al., 2019	$\eta^2 = .16$					
Calhoun et al., 2020			$ES = .73$			
Phung & Goldberg, 2017	$r = .25$	$r = .20$ to $.42$		$r = -.35$ to $.21$		$r = -.12$
Thenhausen et al., 2017	$r = .26$ to $.51$	$r = .06$ to $.18$	$r = .23$ to $.64$	$r = -.04$	$r = .33$	
B. Total sleep time	Externalizing behavior	Internalizing behavior	Autism symptom severity	Social behavior	Attention	Daytime sleepiness/fatigue
Objective measure						
Bruni et al., 2007	$r = .00$	$r = -.58$				
Phung & Goldberg, 2017 *		$\beta = .73$		$\beta = .60$ and ns		
Phung et al., 2018				$r = -.24$ and $.17$		
Richdale et al., 2014		$r = -.27$ and $.37$				
Parent- or self-report						
Baker et al., 2013						ns
Johansson et al., 2018			OR = .87			
Richdale et al., 2014		$r = -.43$ to $-.04$				
C. Sleep onset latency	Externalizing behavior	Internalizing behavior	Autism symptom severity	Social behavior	Attention	Daytime sleepiness/fatigue
Objective measure						
Baker et al., 2013						$r = .57$ and ns
Bruni et al., 2007	$r = .46$	$r = .38$				
Phung et al., 2018				$r = -.18$ and $.12$		
Richdale et al., 2014		$r = .17$ and $.35$				
Parent- or self-report						
Richdale et al., 2014		$r = .51$ and $.59$				



Table 2.2 Continued

D. Sleep efficiency	Externalizing behavior	Internalizing behavior	Autism symptom severity	Social behavior	Attention	Daytime sleepiness/ fatigue
Objective measure						
Bruni et al., 2007	$r = -.95$	$r = -.32$				
Phung & Goldberg, 2017*		$\beta = -.94$		$\beta = -.62$ and ns		
Phung et al., 2018				$r = -.00$ and .19		
Parent- or self-report						
Baker et al., 2013						$r = -.54$ and .24
Richdale et al., 2014		$r = -.46$ and $-.43$				
E. Sleep timing	Externalizing behavior	Internalizing behavior	Autism symptom severity	Social behavior	Attention	Daytime sleepiness/ fatigue
Objective measure						
Richdale et al., 2014		$r = .22$ to .54				
Parent- or self-report						
Richdale et al., 2014		$r = .17$ to .40				

Non-significant association(s) Both significant and non-significant associations Significant association(s)

In the included studies, associations were reported in several ways and subsequently reported in this table: correlation (r), standardized regression coefficients (β), eta-squared (η^2), effect size (ES) and odds ratio (OR).

ns = not significant (value not reported)

* A comparison of method and result section revealed missing information about significance. This is reported as non-significant.

Sleep problems in general. Four studies reported on the relationship between sleep problems in general and daytime functioning. All used self-reports or parent reports of sleep. Sleep problems were significantly associated to rule-breaking behavior ($r = .51$), affective problems ($r = .42$), and autism symptom severity (social anxiety/avoidance and impairments in social communication; $r = .64$ and $.55$) (Calhoun et al., 2020; Phung & Goldberg, 2017; Thenhausen et al., 2017). One study investigated the role of sleep problems in the relationship between working memory and learning problems. Deficits in working memory were significantly associated with increased learning problems only in those teenagers with severe sleep problems (Calhoun et al., 2020). Medium-sized but non-significant correlations were found with anxiety ($r = .32$), closeness in relationship ($r = -.35$), autism symptom severity (autistic traits and impairments in social cognition; $r = .32$ and $.46$), and problems with attention ($r = .33$) (Phung & Goldberg, 2017; Thenhausen et al., 2017).

Total sleep time. Six studies investigated the relationship between total sleep time and one or more aspects of daytime functioning. A study using an objective measure of total sleep time (actigraphy) reported significant associations with internalizing behavior (depressive symptoms; $\beta = .73$) and social behavior (discord in relationships; $\beta = .60$) (Phung & Goldberg, 2017). Medium to large but non-significant correlations were also found with internalizing behavior (using polysomnography (PSG); $r = -.58$ and using actigraphy; $r = -.43$) (Bruni et al., 2007; Richdale et al., 2014). A study using self-reported measures of total sleep time revealed a medium-sized but non-significant correlation with anxiety ($r = -.37$) (Richdale et al., 2014).

Sleep onset latency. Four studies investigated the relationship between sleep onset latency and daytime functioning. The studies that used an objective measure of sleep (actigraphy) reported a longer sleep onset latency associated with more daytime sleepiness ($r = .57$) (Baker et al., 2013). Moreover, medium-sized but non-significant correlations were found with externalizing behavior (using PSG; $r = .46$) and internalizing behavior (using actigraphy; $r = .35$ and using PSG; $r = .38$) (Bruni et al., 2007; Phung & Goldberg, 2017). A study using self-reports found a significant correlation with depressive symptoms ($r = .51$) and anxiety ($r = .59$) (Richdale et al., 2014).

Sleep efficiency. Five studies reported on the relationship between sleep efficiency and daytime functioning. Studies using objective assessments of sleep revealed that a lower sleep efficiency was associated with more externalizing behavior (using PSG; $r = -.95$) and internalizing behavior (depressive symptoms; using actigraphy; $\beta = -.94$) (Bruni et al., 2007; Phung & Goldberg, 2017). A medium-sized but non-significant correlation was also found with internalizing behavior (using PSG; $r = -.32$) (Bruni et al., 2007). A study that used self-reports established a significant relationship with internalizing behavior (anxiety and depressive symptoms; $r = -.43$ and $-.46$) (Richdale et al., 2014). Using self-reports, significant relationships were also found with daytime sleepiness ($r = -.54$) (Baker et al., 2013).

Sleep timing. We found no studies that used specific measurements for sleep timing, such as chronotype questionnaires or DLMO (Dim Light Melatonin Onset). However, one study (Richdale et al., 2014) investigated bedtime and wake time measurements. Objective measures (actigraphy) showed that sleep onset time was significantly related to depressive symptoms ($r = .54$). In addition, for wake time, medium-sized but non-significant correlations were found with depressive symptoms ($r = .41$). A medium-sized but non-significant correlation was also found for the relationship between wake time and depressive symptoms ($r = .40$) (Richdale et al., 2014).

2.5 Discussion

This systematic review resulted in an overview of the literature on the relationship between sleep and daytime functioning in autistic teenagers. We started with the relationship between sleep problems in general and daytime functioning. Then, to differentiate between aspects of sleep and their effects on daytime functioning that might direct future interventions, we studied the association between daytime functioning and total sleep time, sleep onset latency, sleep efficiency, and sleep timing. The most prominent finding was the association with internalizing problems. All sleep parameters, measured both by objective measures and self-reports, showed a relationship with internalizing behavior, in which more sleep and a better sleep quality were associated with fewer internalizing behavioral problems such as depressive symptoms and anxiety. Associations with other aspects of daytime functioning (e.g., externalizing behavior, internalizing behavior, severity of autism symptomology, and daytime sleepiness) were most prominent in studies that investigated sleep problems in general and sleep efficiency.

Although it is clear that more studies are needed to unravel the relationship between sleep and daytime functioning in autistic teenagers, these findings are in line with research on TD teenagers, which also show an association between sleep and daytime functioning. Research with a TD teenage population revealed that sleep problems were also associated with internalizing behavioral problems (Gregory & Sadeh, 2012). In addition, increased externalizing behavior and impaired school performance were related to sleep problems in the TD population (Dewald et al., 2010; Gregory & Sadeh, 2012). First indications show that the presence of sleep problems is also related to more externalizing behavior in autistic teenagers. However, more research is needed to better understand the association between sleep and daytime functioning in autistic teenagers and to improve our understanding of the impact of sleep problems on more specific domains, such as school functioning.

In comparison to research among TD teenagers, sleep timing has been little studied in the autistic population. Studies with TD teenagers show that sleep timing is related to daytime functioning (Short et al., 2013). Since melatonin plays an important role in the sleep-wake cycle and its production is commonly disturbed in autistic teenagers (Johnson & Malow, 2008), it is surprising that no specific measurement for sleep timing has been used. Only one study investigated sleep timing in relation to

daytime functioning, and it used no specific measures (Richdale et al., 2014). This study also found that sleep problems and aspects of daytime functioning were related, although these relationships need to be confirmed in future research.

As this review only included cross-sectional studies, we can draw no conclusions about the causality of the relationship between sleep and daytime functioning. However, we expect this relationship to be bidirectional, even triggering vicious circles: as noted in studies with TD teenagers, sleep affects daytime functioning, which can further exacerbate sleep problems (Dahl & Lewin, 2002; Gregory & Sadeh, 2012). This effect might be amplified in autistic teenagers because sleep loss exacerbates autism symptomatology. Longitudinal research designs are needed to shed more light on this issue and what this means for autistic individuals.

2.5.1 Limitations and further research

We reviewed selected findings to improve our understanding of the relationship between sleep and daytime functioning and provide input for sleep interventions for autistic teenagers. We used a total score for the assessment of the quality of the included studies. In general, the quality was good, although we included some studies with low quality on a limited number of aspects (e.g., small sample size and lack of controlling for confounding). These were included because only a few studies were available and we aimed to explore possible relationships. We focused on well-defined parameters of daytime functioning for the description of the results. For some studies, we also investigated less well-defined parameters; these could be included in future research.

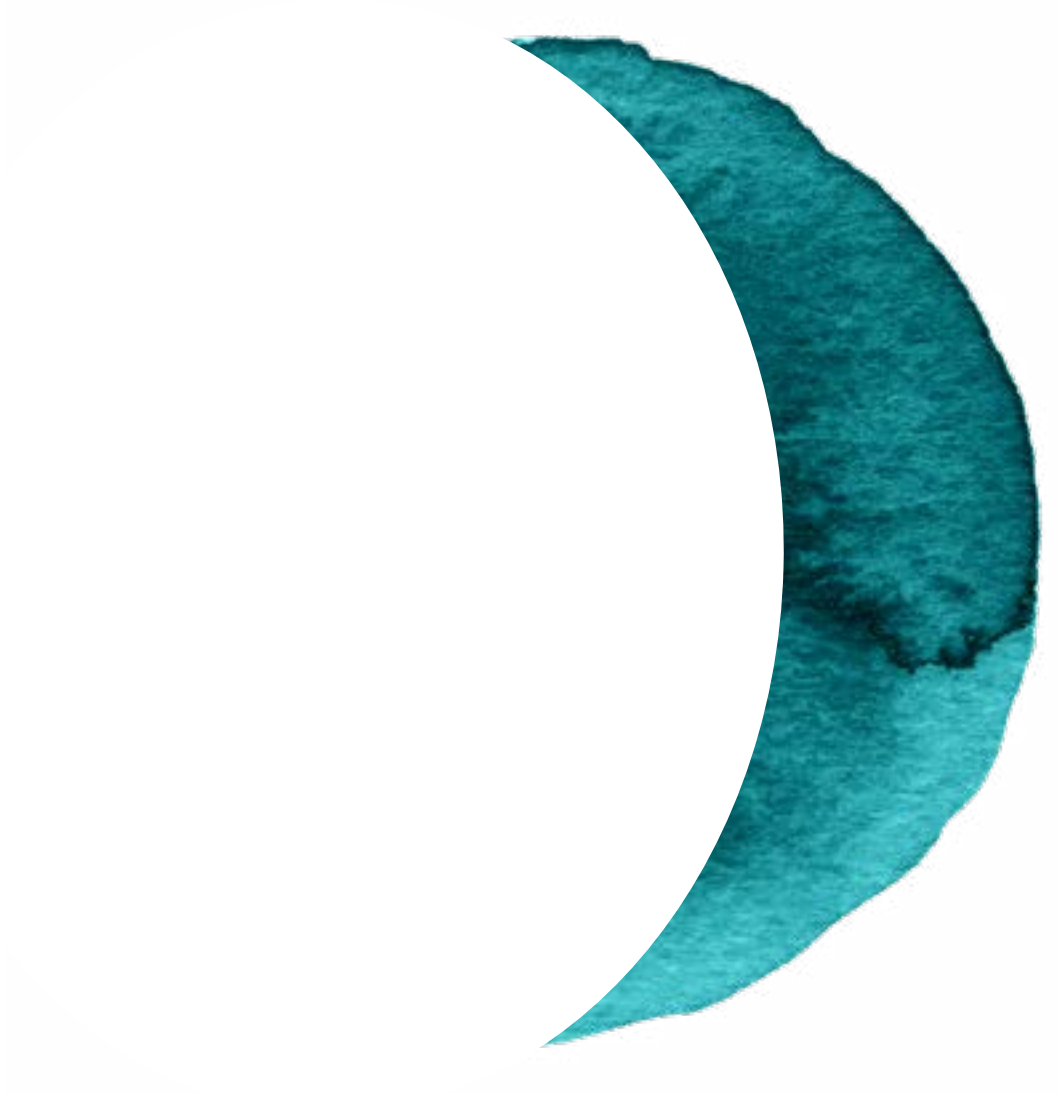
To further understand the relationship between sleep and daytime functioning, we encourage researchers to include several factors. First, most relationships between specific sleep parameters and specific aspects of daytime functioning have only been studied once, so replication is needed. In doing so, it is important to focus on the teenage population specifically. We had to exclude many studies because the age range was too broad. Given that sleep changes during adolescence (Moore & Meltzer, 2008) and the nature of sleep problems changes with age (Goldman et al., 2011), it is important to limit the age range or at least take the effect of age into account when analyzing data.

Second, we suggest a need for more research using both objective measurements and self-reports of sleep to get a clear picture of the relationship between specific sleep parameters and daytime functioning. Such research would give insights into (1) the relationship between objectively measured and self-reported sleep in autistic teenagers and (2) whether objective measures or self-reports are more important for detecting associations with impairments in daytime functioning. This review included no studies that addressed the relationship between objective measures and self-reported sleep alongside the relationship between sleep and daytime functioning. One study related objective measures and self-reported measures of sleep to daytime functioning (Richdale et al., 2014), but it is not yet possible to draw conclusions about this most important measure for detecting relationships with daytime functioning.

The last factor relates to the design of the studies. Most used a cross-sectional design to assess the relationship between sleep and daytime functioning. As mentioned, this makes it impossible to draw conclusions about causality. Moreover, cross-sectional designs provide no information about the long-term relationships between sleep problems and daytime functioning, which again stresses the need for longitudinal research.

2.5.2 Implications

In addition to implications for further research, our review has implications for clinical practice. Sleep problems may be overlooked in treatment because other issues, such as behavioral problems, are prioritized (Reynolds & Malow, 2011). Our review shows the importance of prioritizing sleep problems because they can have a severe impact on daytime functioning. In combination with our finding that anxiety is related to sleep problems, the knowledge that ASD is often associated with co-occurring conditions, of which anxiety is the most prevalent (Simonoff et al., 2008), stresses the importance of early intervention to reduce sleep problems and prevent negative impacts on daytime functioning. Our results provide input for tailored sleep interventions for autistic teenagers.



Chapter 3

Sleep and daytime functioning in autistic teenagers: A psychological network approach

Submitted as:

Schouwenaars, I. M. G., Magnée, M. J. C. M., Geurts, H. M., van Bennekom, C. A. M., Pillen, S., Waldorp, L.J., Blanken, T.F., & Teunisse, J. P. A Psychological Network Approach to Sleep and Daytime Functioning in Autistic Teenagers. [Manuscript submitted for publication]. Research Centre for Social Support and Community Care, HAN University of Applied Sciences
Available on PsyArXiv: <https://doi.org/10.31234/osf.io/sgvz2>

3.1 Abstract

Although sleep problems in autistic teenagers are associated with impaired daytime functioning, it remains unclear how sleep and daytime functioning are related. We used a network approach to disentangle patterns between sleep, sleep hygiene, and daytime functioning. Over a three-week period, 31 autistic teenagers answered daily questions about sleep and daytime functioning. Sleep tracker data were collected from 14 of the teenagers. We preregistered the analysis plan for this study at AsPredicted (#34594; <https://aspredicted.org/blind.php?x=3c4t65>). Perceived sleep quality seemed to be the most important sleep variable in relation to daytime functioning (self/parent/teacher reports). We also found that sleep onset latency, total sleep time, and wake time after sleep onset were related to daytime functioning, but mostly indirectly through sleep quality. These findings are important for developing sleep interventions because perceived sleep quality would be a logical target for increasing the likelihood of actually improving daytime functioning.

3.2 Introduction

Sleep problems are common among autistic teenagers: 40% to 80% experience them (Cortesi et al., 2010), compared to 16% to 31% of their typically developing peers (Calhoun et al., 2014). Autistic teenagers have a shorter sleep duration and longer nighttime awakenings, and they take longer to fall asleep than typically developing teenagers (Tse et al., 2020). Sleep problems are associated with disturbed daytime functioning in typically developing teenagers (Gregory & Sadeh, 2012). However, as the nature (Tse et al., 2020) and underlying causes (Cortesi et al., 2010) of autistic teenagers' sleep problems seem to differ from those of typically developing teenagers, the results of studies performed on typical teenagers cannot automatically be applied to autistic teenagers. In addition, though many studies have investigated the relationship between sleep, sleep hygiene, and daytime functioning based on bivariate correlations, correlations cannot disentangle patterns between multiple variables. Thus, we will use a network approach to investigate the relationship between aspects autistic teenagers' sleep, sleep hygiene, and daytime functioning to learn more about the patterns between them.

Sleep and factors that influence sleep form a complex pattern of direct and indirect relationships between variables (Goelema et al., 2018). Identifying these patterns would provide relevant information about which variables stimulate or inhibit others (Costantini et al., 2015). For example, one could find that caffeine intake and electronic device use are associated with a longer sleep onset latency. To shorten that latency, one might prefer an intervention that addresses caffeine intake because this seems easier than addressing electronic device use. However, the pattern of variables might show that electronic device use is directly related to sleep onset latency while caffeine use is indirectly related through electronic device use. Thus, addressing electronic device use could be more effective in improving sleep onset latency than addressing caffeine intake because of the direct relationship.

Here, identifying the network structure between variables could offer new insights into the associations between aspects of sleep, sleep hygiene, and daytime functioning. To validate the complexity of interacting variables (Borsboom & Cramer, 2013) and learn more about interventions that may be effective, we will use network analyses to identify the network structure between aspects of sleep, sleep hygiene, and daytime functioning.

Several aspects should be considered when unraveling the relationship between sleep, sleep hygiene, and daytime functioning. First, sleep can be measured in different ways: for instance, it can be self-reported or reported by the caregiver (sleep diaries and questionnaires) or measured objectively (actigraphy and polysomnography). Self- or caregiver reports and objectively measured sleep could reveal different outcomes regarding daytime functioning (Richdale et al., 2014).

Second, there are often discrepancies between different informants' reports of a person's daytime functioning (Youngstrom et al., 2001). For example, different contexts could reveal a different

presentation of one's functioning, or informants might have different perspectives on the same functioning. Using these different perspectives could reveal relevant information that contributes to creating a comprehensive picture of a person's daytime functioning (Achenbach, 2006).

To address these issues, we investigated the relationship between sleep, sleep hygiene, and daytime functioning using both (i) self-reported and objective measures of sleep, and (ii) multiple informants' perceptions of the teenagers' daytime functioning. We used a limited age range and focused on teenagers specifically because age-related changes in sleep patterns occur from childhood to adolescence (Moore & Meltzer, 2008).

We included four sleep variables: sleep onset latency (the time between trying to sleep and falling asleep), total sleep time (total amount of sleep during the night), wake time after sleep onset (total time of wakefulness after sleep onset), and perceived sleep quality (how well someone slept, their feeling of being rested at waking, and ease of waking). We chose these four sleep variables based on Buysse's (2014) recommendation that they should be included in sleep research because of their association with health outcomes.

For daytime functioning, we focused on three variables: daytime sleepiness, mood, and concentration problems (Buysse et al., 2006). The choice of these variables was based on research that showed that daytime sleepiness is a major consequence of disturbed sleep (Buysse et al., 2006; Dahl & Lewin, 2002). Moreover, mood and concentration are other aspects of daytime functioning that are affected by sleep problems (Buysse et al., 2006; Dahl & Lewin, 2002).

Since proper sleep hygiene contributes to the quality of sleep (Driver & Taylor, 2000; LeBourgeois et al., 2005), we also included aspects of sleep hygiene (caffeine intake, gaming, (intensive) physical exercise, electronic device use). Although stress and mental effort are not traditionally mentioned as aspects of sleep hygiene, we included those variables because of their impact on sleep (Åkerstedt et al., 2012; Irish et al., 2015). Moreover, including these aspects is relevant because autistic individuals perceive higher levels of stress than typically developing individuals (Hirvikoski & Blomqvist, 2015).

Our research question is: To what extent is there a relationship between characteristics of sleep (sleep onset latency, total sleep time, wake time after sleep onset [objectively measured and self-reported], perceived sleep quality, and sleep hygiene) and daytime functioning (mood, concentration, daytime sleepiness) in autistic teenagers from the perspective of different groups of informants (teenagers, parents, teachers)?

3.3 Methods

3.3.1 Participants

The participants in this study were 32 autistic teenagers and their teachers. Additionally, 27 parents of those 32 teenagers participated (5 teenagers participated without a parent). We recruited teenagers aged 12 to 20 years from four schools for special education. Inclusion criteria for the teenagers were: (a) diagnosed with an autism spectrum disorder by a psychiatrist or psychologist, based on the criteria of the DSM-IV (American Psychiatric Association, 2000) or DSM-5 (American Psychiatric Association, 2013), (b) presence of sleep problems, reported by the teenager and/or parents, (c) medication free or at a stable dose during participation in the study, and (d) absence of an intellectual disability, defined by an IQ of 70 or higher (American Psychiatric Association, 2021).

We obtained written informed consent from all participants (teenagers, teachers, and parents) before study entry. For teenagers aged 12 to 15 years, we also obtained written permission from both parents or legal representative(s). We collected data in two phases because we did not succeed to include the intended number of participants in the initial inclusion phase of the study (January -February 2019; 15 participants). Therefore, we added a second data collection phase (November -December 2019; 17 participants).

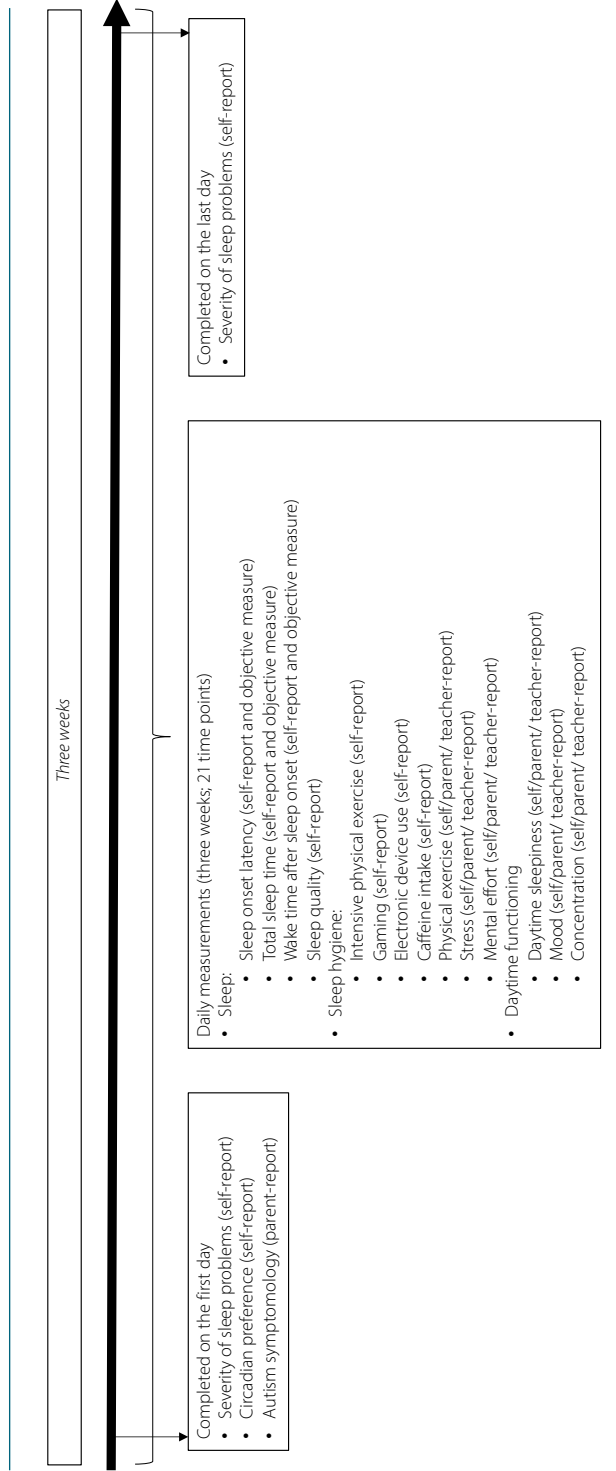
Approval for this study was obtained from the Ethics Review Board of the Faculty of Social and Behavioral Sciences at the University of Amsterdam (2018-EXT-9650) and the Internal Committee Biomedical Experiments of Philips Research (ICBE-2-26243).

3.3.2 Procedure

This study used a repeated measures design with daily measurements over a three-week period. The teenagers completed a web-based sleep diary every morning and answered questions about their daytime functioning at the end of the day (Figure 3.1). Teachers and parents answered the same daily questions about the teenagers' daytime functioning from their own perspective.

In addition to the daily measurements, teenagers completed the Insomnia Severity Index (ISI) (Bastien et al., 2001) at the beginning of the research period to measure the severity of their sleep problems. Parents completed the Social Responsiveness Scale-Second Edition (SRS-2) (Roeyers et al., 2015) at the beginning of the research period to measure deficits in their teenager's social behavior. We also used a sleep tracker to measure sleep in a randomly chosen subgroup of teenagers throughout the study period (N = 16; because of limited availability of the devices).

Figure 3.1 Overview of Study Design



3.3.3 Measures

Self-reported sleep. Self-reported sleep was measured using a sleep diary. It was based on the Consensus Sleep Diary (Carney et al., 2012), which has been found to be valid and useful for measuring self-reported sleep (Maich et al., 2018). The sleep diary for our study was developed in collaboration with a sleep expert who is a clinical pediatric neurologist (SP) with expertise in sleep treatment for autistic children and teenagers. To prevent the teenagers from completing multiple sleep diary entries retrospectively for several days, the sleep diary for a given day could be completed only until midnight. However, teenagers were instructed to complete the sleep diary as soon as possible after waking.

The sleep diary asked questions about bedtime, sleep onset time, nighttime awakenings, and wake time. The answers were used to calculate sleep onset latency (difference between time trying to fall asleep and sleep onset time), wake time after sleep onset (sum of time awake between sleep onset time and wake time), and total sleep time (difference between sleep onset time and wake time minus the wake time after sleep onset). The teenagers were also asked to rate (1 - 10, ranging from 1 = *very bad* to 10 = *very good*) (a) how well they slept, (b) how rested they felt upon waking, and (c) how easily they woke up. We took the mean of the answers to these last three questions to indicate their perceived sleep quality.

Sleep hygiene. The teenagers were asked questions about sleep hygiene at two moments. The first was in the sleep diary, which also asked them about the previous night's activities that might have affected their sleep: intensive physical exercise, gaming, use of electronic devices, and caffeine intake. The teenagers were asked if they performed those activities and, if so, at what time. Second, at the end of each day, the teenagers were asked to rate on a scale from 1 to 10 (1 = *very little* to 10 = *very much*) their amounts of physical exercise, stress (explained as tension or pressure), and mental effort (explained as the extent to which you have to perform tasks that require continual focus). The teenagers could answer the questions at any time between 2:00 p.m. and midnight, but they were instructed to do so in early evening (before 8:00 p.m.) to avoid interference with their bedtime routines.

One parent and one teacher of each teenager also were asked to answer the questions about the teenager's physical exercise, stress, and mental effort from their own perspective. Parents were asked to answer the questions in the same time range as the teenagers. However, since most teachers saw their students only once a day (which could be in the early morning), they were free to answer the questions at any time of day so they could do so immediately after seeing the teenager in question.

Objective sleep. Objective sleep variables were captured using a sleep tracker, a wrist-worn reflective plethysmography and accelerometry device (prototype developed by Philips Research). It was intended to obtain and store reflective, green-spectrum photoplethysmography data from the skin (64 Hz). Moreover, acceleration data was obtained using an internal 3D accelerometer (128 Hz). These data were converted to inter-beat intervals (the time between two consecutive pulses), which were used to

calculate sleep. A trained algorithm was used to measure sleep onset latency, total sleep time, and wake time after sleep onset (Wulterkens et al., 2021).

The teenagers were instructed to put on the sleep tracker when they went to bed and to remove it when they got up in the morning. To start and finish the measurement, they pressed a button on the sleep tracker. They also were asked to press a button when they intended to sleep, to mark sleep onset latency. Parents were not instructed to monitor whether their child pressed the sleep intent button because this would disturb the sleep.

Daytime functioning. Daytime functioning was measured by asking the teenagers to rate the following aspects on a scale from 1 to 10: daytime sleepiness (1 = *not at all sleepy* to 10 = *very sleepy*), concentration, and mood (1 = *very bad* to 10 = *very good*). This resulted in three variables. The questions were developed in collaboration with the sleep expert who also co-developed the sleep diary (clinical neurologist SP). The teenagers could answer the questions at any time between 2:00 p.m. and midnight, but they were instructed to do so in early evening (before 8:00 p.m.).

One parent and one teacher of each teenager also were asked to answer the questions about the teenager's daytime functioning from their own perspective. Parents were asked to answer the questions in the same time range as the teenagers. However, since most teachers saw their students only once a day, they were free to answer the questions at any time of day so they could do so immediately after seeing the teenager in question.

Severity of sleep problems. The Insomnia Severity Index (ISI) (Bastien et al., 2001) was used to assess the severity of insomnia and its consequences in the two weeks before assessment. The ISI consists of seven items measuring the following aspects: perceived severity of problems with sleep onset, sleep maintenance and early awakenings, satisfaction with sleep pattern, interference with daily functioning, notability of the consequences of disturbed sleep for others, and the degree of concerns related to disturbed sleep. A five-point Likert scale (ranging from 0 = *no problem* to 4 = *very severe problem*) was used to rate each item. The ISI was originally developed to measure insomnia in adults, but it has been found to be reliable and valid in teenaged populations (Chung et al., 2011). The sum score of all items was used in the analyses.

Severity of autism symptomatology. The Social Responsiveness Scale-Second Edition (SRS-2), version for 4- to 18-year-olds, was used to measure deficits in social behavior. The SRS-2 consists of 65 items covering two subscales: social communication and interaction, and stereotype behavior and interests (Roeyers et al., 2015). A four-point Likert scale (ranging from 1 = *not true* to 4 = *almost always true*) is used to rate each item. The reliability and validity of the SRS-2 in a Dutch population is good (Roeyers et al., 2015). The questionnaire was completed by one parent of the participating teenager. The sum score of all items was used in the analyses.

3.3.4 Statistical analyses

We preregistered the analysis plan for this study at AsPredicted (#34594; <https://aspredicted.org/blind.php?x=3c4t65>). In this paper, we focus on the analysis of the relationship between aspects of sleep (objectively measured sleep and self-reported sleep [including sleep hygiene]) and daytime functioning in autistic teenagers from the perspective of groups of informants (teenagers, parents, teachers). To reduce complexity and increase comprehensibility, we chose to focus on the group findings and to exclude the preregistered individual participant analyses.

We used a network approach to investigate the relationship between sleep and daytime functioning. A network contains nodes (the variables) that are connected by edges (the association among the variables). An edge represents an association between two variables given (conditional on) all other variables in the network; so, an edge represents the association when all other variables cannot account for this association. A positive association is illustrated by a blue edge and a negative association by a red edge. The thickness of the edge shows the strength of the association: the thicker the edge, the stronger the association (Costantini et al., 2015).

To determine the number of sleep variables used in the network analysis, we calculated the correlations between objectively measured (i.e., through the sleep tracker) and self-reported (i.e., in the sleep diary) sleep onset latency, total sleep time, and wake time after sleep onset. These correlations were based on data from the teenagers who both wore the sleep tracker and completed the sleep diary. If the observations of objectively measured and self-reported sleep correlated highly ($\geq .80$) (Field, 2009), the mean of each measurement for that variable was used in the subsequent analyses. If the correlation was $<.80$ for that specific relationship, both the objectively measured and self-reported observations of sleep were used.

As mentioned earlier, one teenager's data could not be used because they did not answer the questions about sleep and daytime functioning on the same day during the research period. Sleep tracker data from another two teenagers could not be used due to technical reasons. Thus, our analyses include self-reported data from 31 teenagers and sleep tracker data from 14 teenagers.

Instead of computing three networks (for teenagers, parents, and teachers separately) in which we were planning to include both self-reported and objectively measured sleep, we computed six networks (for teenagers, parents, and teachers separately combined with self-reported or objectively measured sleep). This deviation from the preregistered analysis plan allowed us to exclude those teenagers that did not wear a sleep tracker from the sleep tracker data analysis. Of the six networks we obtained, we included all 31 teenagers in the first three networks. These were based on their self-reported sleep combined with (i) their self-reported daytime functioning, (ii) the parent-reported daytime functioning, and (iii) the teacher-reported daytime functioning. In networks four to six, we included the 14 teenagers who wore

the sleep tracker. Thus, we included the teenagers' objectively measured sleep together with the same variables of daytime functioning reported by the teenager, parent, or teacher.

The nodes in networks represented the variables for that specific network: number of minutes for sleep onset latency, total sleep time, wake time after sleep onset (self-reported or objectively measured), sleep quality score (ranging from 1 to 10), the number of minutes before bedtime an activity was performed (intensive physical exercise, gaming, electronic device use, and caffeine intake; if the activity was not performed, the score was set to the highest reported number of minutes for that specific variable), score given for physical exercise, stress, and mental effort, and the scores given for aspects of daytime functioning (reported by the teenager, parent, or teacher).

Since we found more missing values than expected (missing at random; ranging from 18.1% [objectively measured sleep] to 52.6% [parent-reported daytime functioning]), we deviated from the preregistered analysis plan to solve problems with these missing values. First, we used the R-package Multivariate Imputation by Chained Equations (*mice*) (van Buuren & Groothuis-Oudshoorn, 2011) to impute our missing data using ten and 20 iterations and compared the results. Since the results of ten and 20 iterations were comparable (which showed the stability of the imputations), we used 20 iterations to obtain robust imputations. Imputations were conducted separately for self-reported and objectively measured sleep combined with each group of informants (see supplementary material in Chapter 10.2 for the imputation code). Thus, six imputations were conducted. Each imputation model used all the variables included in that specific network. Using all variables provides a more accurate imputation by preserving the relationships between variables.

We then used the R-package Mixed Graphical Model (*mgm*; Haslbeck & Waldorp, 2020) to estimate the networks with the imputed datasets. To reduce the likelihood of including false-positive edges, we used the Least Absolute Shrinkage and Selection Operator (LASSO) including the default threshold of *mgm*. We used the Extended Bayesian Information criterion (EBIC) to determine the penalty parameter of the LASSO (see supplementary material in Chapter 10.2 for the network estimating code). The stability of each network was assessed using the *resample* function of the *mgm* package. We ran 80 bootstrap samples for each network; with the *argument* quantiles, we specified the lower (.05) and upper (.95) quantiles of the sampling distribution. We included the edge in the network if the proportion of inclusion in each resample was at least 90%. This stability check confirms that the relationships we found are robust, so we have confidence in those relationships. The networks were illustrated using the R-package *qgraph* (Epskamp et al., 2012).

In addition to what was preregistered, we also computed the networks without the data from the first and last day of the research period because most of the missing data was from those days. After visually comparing the results, we concluded that the networks of the teenagers and parents (both with self-reported and objectively measured sleep) were stable. Since we did not want to remove useful data, we

included data about all the research days in the networks of the teenagers and parents. However, the teachers' networks were less stable. Since their percentage of missing values was much higher on the first research day than on the other days, we excluded that day from our network analyses.

3.4 Results

3.4.1 Background characteristics of participants

In total, 24 boys and seven girls with a mean age of 14.87 years ($SD = 1.88$, age range = 12 - 18) participated. The mean ISI score was 17.20 ($SD = 6.43$), which is an indication of clinical insomnia with moderate severity (Bastien et al., 2001). Their mean score on the SRS-2 was 78.74 ($SD = 27.46$). The majority of the participants (78%) scored above the autism cut-off score of 51.

There were no significant difference between the ISI and SRS scores for teenagers that did (ISI: $M = 17.38$, $SD = 6.24$ and SRS: $M = 85.09$, $SD = 16.41$) or did not wear the sleep tracker (ISI: $M = 16.09$, $SD = 6.01$ and SRS: $M = 72.92$, $SD = 34.43$); for the ISI and SRS respectively $t(22) = 0.52$, $p = .61$ and $t(21) = 1.07$, $p = .30$.

3.4.2 Relationship between self-reported and objectively measured sleep

We found small to moderate correlations between self-reported and objectively measured sleep. Self-reported and objectively measured total sleep time ($r = .46$, $p < .001$) and wake time after sleep onset ($r = .32$, $p < .001$) were moderately correlated. The correlation between self-reported and objectively measured sleep onset latency was small ($r = .28$, $p < .001$). Based on these correlations, we included objectively measured and self-reported sleep separately in the following analysis.

3.4.3 Relationship between sleep and daytime functioning

To estimate the networks, we used the six imputations described in the Methods section. In the presentation of the networks, the node's color reflects the type of question: green nodes relate to aspects of sleep hygiene, blue nodes to aspects of sleep, and orange nodes to aspects of daytime functioning. We describe the associations that are most relevant to our research question: (i) between sleep and daytime functioning, and (ii) between sleep hygiene and sleep. It is important to consider that it is difficult to interpret the absence of relationships with this type of analysis. Therefore, we focus on the presence of relationships rather than their absence.

An overview of the characteristics of the included variables and the correlations for the reports of daytime functioning completed by teenagers, parents, and teachers can be found in the supplementary materials in Chapter 10.2.

Teenagers. Figure 3.2 shows the networks of teenagers' self-reported sleep together with their self-reported daytime functioning (3.2a) and their objectively measured sleep together with their self-reported daytime functioning (3.2b). In network 3.2a, we found that better sleep quality was directly associated with better concentration and mood and less daytime sleepiness. Importantly, it appeared that sleep onset latency and wake time after sleep onset were indirectly related to daytime functioning variables through sleep quality. Network 3.2b revealed that several aspects of daytime functioning were connected to each other and to two sleep hygiene variables (mental effort and stress).

Parents. Figure 3.3 depicts the results of the networks with parents as informants for daytime functioning. Network 3a shows the results of the teenagers' self-reported sleep together with parent-reported daytime functioning, and network 3b shows the teenagers' objectively measured sleep together with parent-reported daytime functioning.

In network 3.3a, we found that better sleep quality was directly related to less daytime sleepiness. Moreover, it appeared that sleep onset latency and wake time after sleep onset were indirectly related to daytime sleepiness through sleep quality. In addition, this network showed that a shorter sleep onset latency and a shorter total sleep time were associated with better concentration. Wake time after sleep onset was indirectly related to concentration through total sleep time.

Our analysis of objective sleep variables (3.3b) showed that several connections remained, such as a relationship between total sleep time and concentration. This network also revealed connections between sleep hygiene and sleep. The longer the time between caffeine intake and bedtime, the longer the total sleep time, and the more stress during the day, the shorter the sleep onset latency.

Teachers. Figure 3.4 shows the networks of teenagers' self-reported sleep together with teacher-reported daytime functioning (3.4a) and objectively measured sleep together with teacher-reported daytime functioning (3.4b). As found in other networks, network 3.4a showed that better sleep quality was related to less daytime sleepiness and a better mood. Simultaneously, we found that a longer sleep onset latency was related to more daytime sleepiness. Total sleep time was indirectly associated with daytime sleepiness through sleep onset latency. This network also showed connections between sleep hygiene and sleep; the more physical exercise, the shorter the total sleep time, and the more mental effort, the shorter the sleep onset latency.

Our analysis of objective sleep variables (3.4b) showed that the longer the total sleep time, the less daytime sleepiness. We also found a connection between sleep hygiene and sleep: the longer the time between caffeine intake and bedtime, the longer the total sleep time. Moreover, this network showed that several sleep hygiene variables are interconnected.

Figure 3.2 Graphical Network with Teenagers' Self-Reported (a; N = 31) and Objectively Measured (b; N = 14) Sleep Together with their Self-Reported Daytime Functioning

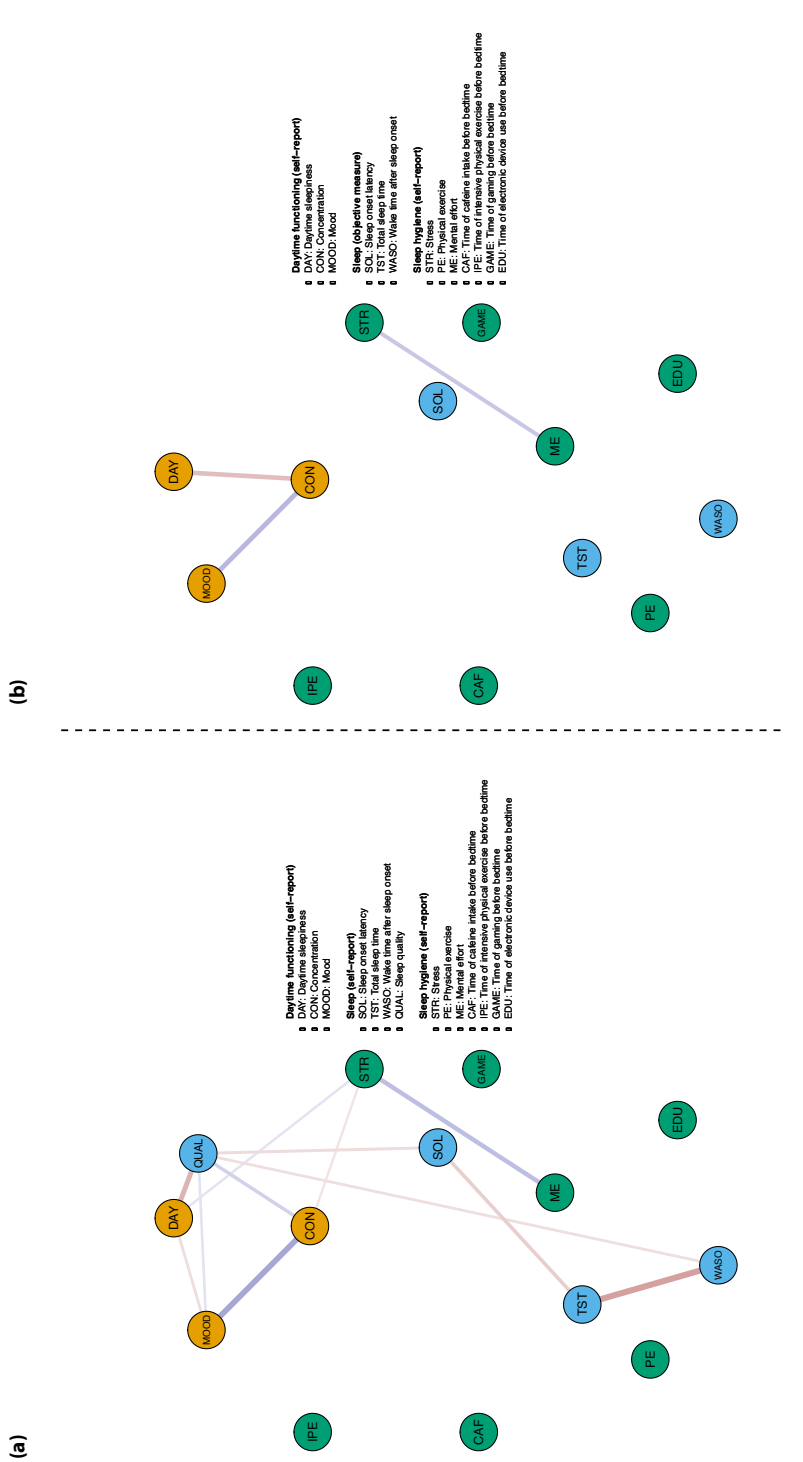
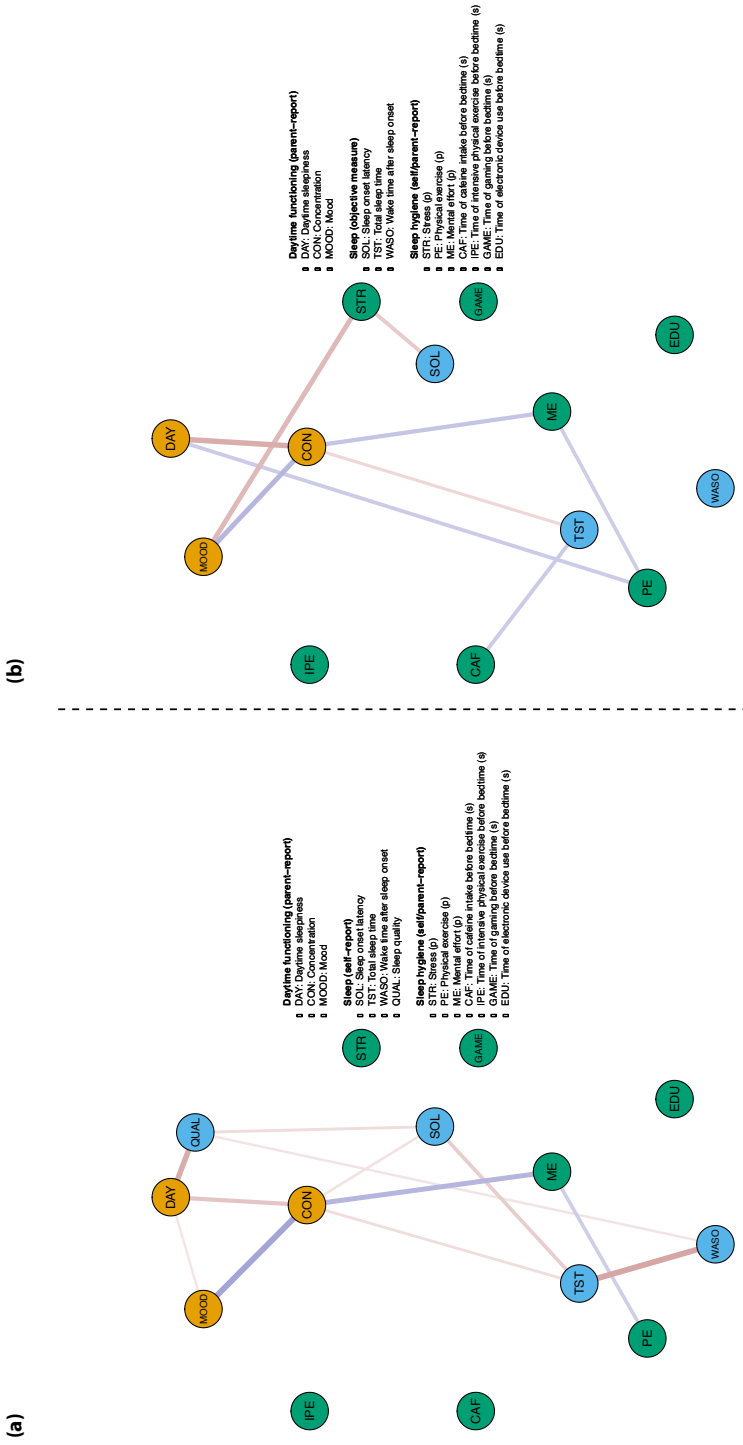
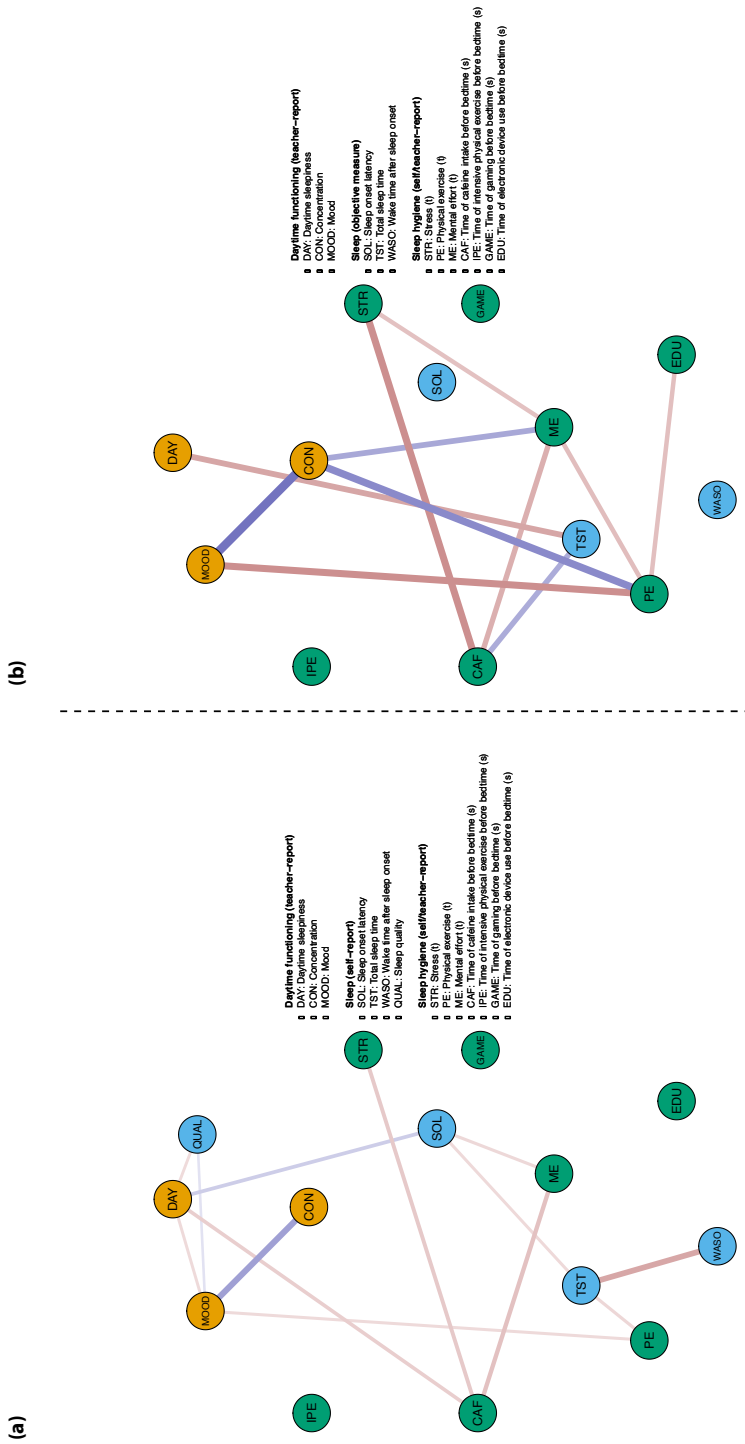


Figure 3.3 Graphical Network with Teenagers' Self-Reported (a; N = 31) and Objectively Measured (b; N = 14) Sleep Together with Parent-Reported Daytime Functioning



Note. s = self-reported, p = parent-reported.

Figure 3.4 Graphical Network with Teenagers' Self-Reported (a; N = 31) and Objectively Measured (b; N = 14) Sleep Together with Teacher-Reported Daytime Functioning

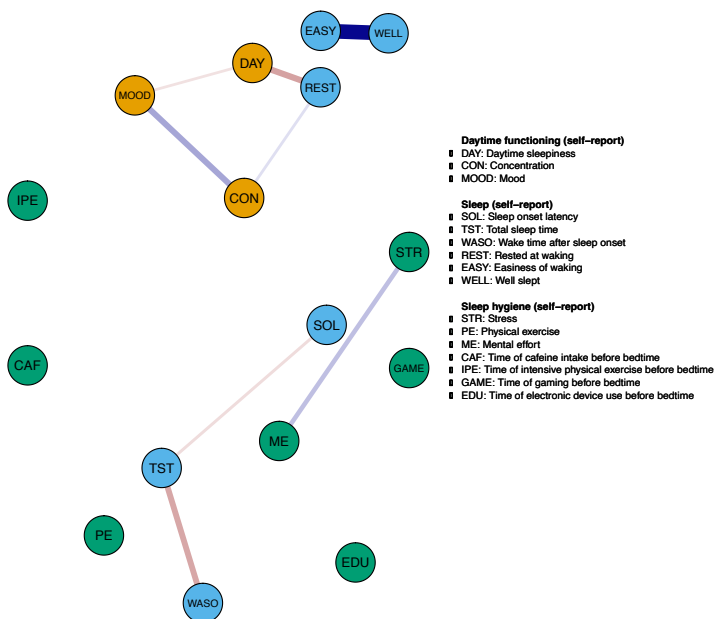


Note. s = self-reported, p = parent-reported.



Additional analysis. All networks revealed that sleep quality is related to one or more aspects of daytime functioning. We combined three items (how well the teenager slept, feeling of being rested at waking, and ease of waking up) in one node to evaluate variable sleep quality. To determine whether the aspects of sleep quality were related to daytime functioning in different ways, we also created an exploratory network in which we included those three items separately. This network (Figure 3.5) showed that of the three aspects of sleep quality, how rested the teenagers feel at awakening was related to daytime functioning.

Figure 3.5 Graphical Network with Teenagers' Self-Reported (a; N = 31) Sleep (including separate items of sleep quality*) Together with Their Self-Reported Daytime Functioning



*The sleep quality items are (a) how well the teenagers slept (WELL), (b) feeling of being rested at waking (REST), and (c) ease of waking up (EASY).

3.5 Discussion

In this study, we used a network approach to investigate the relationship between specific sleep variables and daytime functioning. The results of our analysis of the networks with self-reported sleep revealed that perceived sleep quality was, as expected, related to all aspects of daytime functioning (daytime sleepiness, mood, and concentration) for the teenage informants. Associations between sleep quality and daytime functioning also were found in the parents' and teachers' networks, but to a lesser extent. We also found that sleep onset latency, total sleep time, and wake time after sleep onset were related to daytime functioning, but mostly indirectly through sleep quality. This implies that sleep quality may act as gateway for other sleep variables in relation to daytime functioning.

Our study also showed that self-reports and objective measures of sleep were related to daytime functioning in different ways. Information about how someone perceives their sleep is crucial because perceived sleep quality is the most important aspect of sleep for daytime functioning. This finding is in line with previous research by Edinger et al. (2000), which found that people who slept worse according to objective measures but do not experience sleep problems reported better moods and less anxiety than people who experienced sleep problems while sleeping normally according to objective measures. Our finding, combined with the findings of previous research, highlights the importance of focusing on sleep quality in sleep interventions.

The term 'sleep quality' is widely used but not well defined (Krystal & Edinger, 2008). For example, sleep quality has been defined as how rested or restored people feel at awakening based on whether people feel tired at awakening and during the day, based on number of nighttime awakenings, and based on their state of mind before bedtime (Goelema et al., 2018; Harvey et al., 2008). While we agree that circumstances before sleep or during the day are related to sleep, we argue that it is problematic to use elements beyond sleep or waking to define perceived sleep quality. To avoid interference with daytime functioning, we defined sleep quality only using aspects that refer to sleep and waking (how well someone slept, feeling of being rested at waking, and the ease of waking up).

One could also argue that people may rate their sleep quality differently in the morning than later in the day. Such a rating may be a function of several variables or events, and it may mirror how the teenagers felt on a particular day instead of a sleep quality measure. Therefore, we instructed the teenagers to answer the questions as soon as possible after waking. Thus, one should separate daytime aspects from aspects related to sleep and waking when drawing conclusions about sleep quality.

The most prominent finding of this study was the relationship between sleep quality and daytime functioning. To determine whether aspects of sleep quality are related to daytime functioning in different ways, we included an exploratory network with each individual item instead of a sum score for sleep quality. This network showed that the item 'feeling of being rested at waking' was related to daytime

functioning. Surprisingly, this network also showed that only the items about how well someone slept, and the ease of waking were related to each another. Thus, being rested at waking seems to be different than the other two items. This again raises questions about the definition of perceived sleep quality. Perhaps the item “feeling rested at waking” is not solely a function of sleep or daytime functioning, so we are not certain whether it should be considered an aspect of sleep quality. Regardless, this finding invites future researchers to further elaborate on what exactly defines perceived sleep quality.

We not only found evidence for the importance of sleep quality in the teenagers’ network, but also in the networks of their parents and teachers with the same positive or negative direction (though to a lesser extent). Since discrepancies between reports of different informants are common (Youngstrom et al., 2001), these differences are not surprising. Moreover, these differences could contribute to the comprehensive picture of the relationships between sleep and daytime functioning. Although parents and teachers had less insight into the teenagers’ daytime functioning, they also noticed associations between sleep quality and daytime functioning that emphasize the importance of sleep quality. However, as we found most associations in the teenagers’ network, it should be sufficient to only include teenagers as informants when evaluating daytime functioning.

3.5.1 Strengths and limitations

While the relationship between sleep and daytime functioning has been studied extensively in typically developing teenagers (Short et al., 2013) and to a lesser extent in autistic populations (Richdale et al., 2014), this is, to our knowledge, the first attempt to investigate this relationship using a network approach. This approach enabled us to identify the network structure between sleep variables and aspects of daytime functioning, which let us take the first step toward disentangling the complexity of interacting variables. To unravel the complex relationships, we used both daily objective and self-reported measures of sleep, together with measures of daytime functioning, for a period of three weeks. This gave us a better understanding of relationships between specific sleep variables and daytime functioning in autistic teenagers. In addition, using multiple informants gave us insight into the experiences of the relationship between sleep and daytime functioning from multiple perspectives. For example, some relationships were visible in most networks, while others were less pronounced.

It is also worth noting several limitations of this study. First, the study design was vulnerable: for the network analysis it was needed that teenagers completed the sleep diary and questions about daytime functioning on the same day. This means that when teenagers forgot to answer the questions about their sleep, we could not use the answers to the questions about daytime functioning given by the teenagers, their parent, or their teacher.

Second, we instructed informants (teenager, parent, teacher) to answer the questions before a certain time of day. We asked the teenagers to complete the sleep diary as soon as possible after waking and no later than upon arrival at school, and we asked them to answer the questions about daytime functioning,

stress, mental effort, and physical exercise no later than 8 p.m. Despite these instructions, we noticed that questions were sometimes answered at later times (to avoid severe problems with recall bias, we limited the time the questions could be completed).

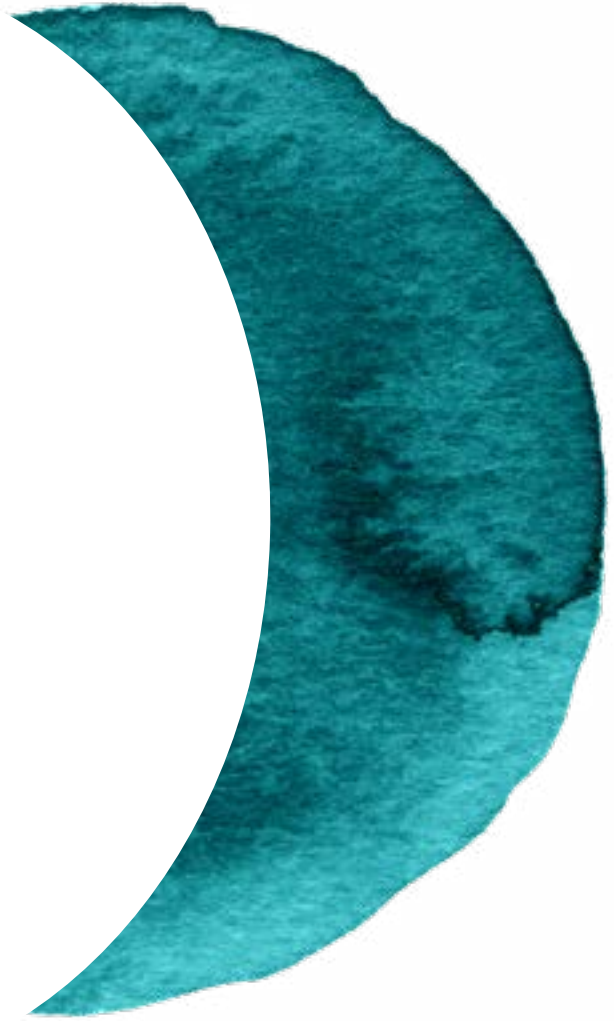
Third, since we had a limited number of sleep trackers available, we could not measure objective sleep in all participants. As a result, the network based on the objective measure of sleep included fewer participants than the network based on self-reported sleep. Certain relationships may not exceed the threshold due to the limited number of participants in this specific network, which would exceed the threshold in a larger group. Therefore, we should not assume an absence of relationships.

Fourth, we used the SRS-2 to identify the presence and severity of difficulties in social behavior on the autism spectrum. However, 22% of the participating teenagers did not score above the autism cut-off score. Still, one of our inclusion criteria for the teenagers was a diagnosis of an autism spectrum disorder by a psychiatrist or psychologist, based on the criteria of the DSM-IV (American Psychiatric Association, 2000) or DSM-5 (American Psychiatric Association, 2013). To determine whether the teenagers met this inclusion criteria, we asked parents if their child had an autism spectrum disorder diagnosis, in which year it was diagnosed, and by which authority this diagnosis was established. All parents declared their child has a formal diagnosis of an autism spectrum disorder that matched our inclusion criteria. Thus, we have no doubts about the diagnosis even though some participants scored below the autism cut-off score on the SRS-2.

3.5.2 Implications

In this study, we showed how the network approach provided insights into the interplay between specific sleep variables, aspects of sleep hygiene, and daytime functioning. The associations showed that sleep quality was the most important aspect of sleep in relation to daytime functioning because it was directly related to all aspects of daytime functioning. However, sleep onset latency and wake time after sleep onset were related to sleep quality. This means those variables also are indirectly related to daytime functioning.

The knowledge that sleep quality is the most important aspect related to daytime functioning provides input for developing sleep interventions, which should focus on improving sleep quality to have a widespread reach. Moreover, since sleep problems are often overlooked in treatment for autistic teenagers (Reynolds & Malow, 2011), this study highlights the importance of focusing on sleep problems to prevent a negative impact on daytime functioning.



Chapter 4

Using the CeHReS roadmap to develop a school-based sleep intervention for autistic teenagers

Schouwenaars, I., Magnée, M., Geurts, H., van Bennekom, C., Pillen, S., & Teunisse, J. (2022).
Using the CeHReS roadmap to develop a sleep intervention for autistic students.
PsyArXiv. <https://doi.org/10.31234/osf.io/h3a4b>

4.1 Abstract

Teachers at schools for special education observe that their autistic students experience sleep problems that negatively affect their daytime functioning. Although schools are willing to support these teenagers in reducing sleep problems, implementing sleep interventions in schools seems to have been unsuccessful. In this paper, we describe the lessons we learned from the school-partnered development of a school-based sleep intervention for autistic teenagers. We will focus on the first three phases of the Centre for eHealth & Wellbeing Research (CeHReS) roadmap: contextual inquiry, value specification, and design. Each phase had specific goals and activities that led to iterative development with the active involvement of stakeholders (future users and professionals). These stakeholders indicated that four factors, with corresponding operationalizations, were important to include in the sleep intervention: limited burden, sense of usefulness, clear guidance and structure, and support cues. The iterative approach with an emphasis on stakeholder participation enabled us to identify possible barriers to implementing the intervention during its development. Identifying those barriers and applying lessons to adapt the sleep intervention during development will likely increase its successful implementation in schools.

4.2 Introduction

Teachers at schools for special education observe that their autistic students experience sleep problems (Cortesi et al., 2010) that negatively impact their daytime functioning (Schouwenaars et al., 2021). Behavioral sleep interventions have been shown to be effective in reducing sleep problems (Johnson & Zarrinnegar, 2021), and they are mostly delivered by clinicians (e.g., McCrae et al., 2020; Papadopoulos et al., 2019). However, sleep interventions that could be delivered by teachers are more easily accessible for school-aged youth and therefore have a greater reach. Moreover, the easy accessibility of school-based sleep interventions might allow sleep problems to be treated earlier. Early interventions to reduce sleep problems are important to prevent exacerbating them (Morin & Benca, 2012), and reducing sleep problems would also have cost benefits related to reduced health care utilization (Daley et al., 2009).

Although we have seen that schools are willing to support their students in reducing sleep problems, the implementation of sleep interventions in schools remains unsuccessful because of the lack of a tailored approach to sleep problems (Cassoff et al., 2013). Some schools have offered programs to enhance sleep knowledge or taught strategies to improve sleep habits, without the intended result (Cassoff et al., 2013). Developing interventions in co-creation could solve that problem because it would involve future users in early stages of the development process, allowing them to express their needs and wishes (Frauenberger et al., 2016). In this study we describe the co-creation of a school-based sleep intervention.

Co-creation is an approach in which stakeholders with different fields of expertise collaborate in a development process, each contributing their own knowledge and experience (Roschelle & Penuel, 2006). Important stakeholders are the people for whom an intervention is designed or who are indirectly affected by the intervention, such as the school administration. They are seen as “experts of their experience” (Sanders & Stappers, 2008, p. 12), and they are allowed to voice their needs and wishes during the development process (Bratteteig & Wagner, 2012). The intention of co-creation is to improve the meaningfulness of the intervention and to gain an understanding of the future users’ needs, wishes, and contextual circumstances to enhance the match between the future users and the intervention (Steen et al., 2011). Thus, co-creation results in an intervention that better matches the future users’ needs, wishes, and circumstances and that already tackles important implementation issues. This increases the likelihood that such an intervention will be implemented successfully.

During the development process, it is important to consider five domains that could increase the likelihood of successful implementation (Damschroder et al., 2009). These domains refer to the characteristics of the intervention (e.g., the ability to test it, its perceived advantages, and the ability to adapt it to specific needs); the inner setting (e.g., characteristics and culture of the organizations); the outer setting (e.g., alignment with future users’ needs); characteristics of individuals involved with the intervention (e.g., knowledge and skills, identification with the organization); and the planning and executing of

the implementation process itself (Damschroder et al., 2009). These factors together determine the adoption of an intervention. The Centre for eHealth & Wellbeing Research (CeHReS) roadmap offer hands-on guidelines based on co-creation to plan, coordinate, and execute development processes of interventions with the intention to increase the likelihood of their successful implementation (van Gemert-Pijnen et al., 2011). The CeHReS roadmap was developed to design and implement eHealth and it is successfully used in health care (e.g., Stuij et al., 2020). Although, to our knowledge, the CeHReS roadmap has not been used in an educational setting before, we expect it could be also useful in this setting.

For this study, we used the first three phases of the CeHReS roadmap: contextual inquiry, value specification, and design. Contextual inquiry involves an investigation of the future users' daily routines and habits, how the intervention could improve them, and the weak and strong points of the current situation. The contextual inquiry provides general insight about the added value of a specific intervention. Value specification elaborates on the previous phase by identifying and prioritizing specific requirements for the intervention. In the design phase, the intervention is designed based on those requirements. All three phases are connected by formative evaluation to verify that the activities include the future users' perspectives and are related to the outcomes of previous phases (van Gemert-Pijnen et al., 2011). The iterative approach and stakeholder participation combined in the CeHReS roadmap enhance the intervention's applicability to its future users, which increases the chance of a successful implementation.

As noted before, Cassoff et al. (2013) showed that the consideration for motivation could be the key factor for a reduction in sleep problems. To enhance sustained motivation, they recommend a personalized approach to sleep problems. To address this recommendation, we co-created a school-based sleep intervention with these main principles: 1) daily monitoring of the teenagers' sleep and 2) an individual sleep consultation based on the results of the monitoring. In this paper, we report on the co-creation in an educational setting of a school-based sleep intervention for autistic teenagers.

4.3 Methods

Each of the CeHReS roadmap phases (contextual inquiry, value specification, and design) has specific goals and activities that led to iterative development involving the important stakeholders, which are described here.

4.3.1 Research context

The research was initiated by schools for special education that perceive the negative impact of sleep problems on their autistic students' daytime functioning. We prepared and conducted this research within a partnership consisting of four schools for special education, two universities, a sleep medicine

center, and a digital agency. They were united in the Sleep Better, Learn Better consortium (founded before the start of the research project). Approval for this study was obtained from the Ethics Review Board of the Faculty of Social and Behavioral Sciences at the University of Amsterdam (2019-EXT-10811).

4.3.2 Stakeholders: future users and professionals

To gather information in each phase of the co-creation process, we consulted both future users (autistic teenagers, their parents, and teachers) and professionals (researchers, teachers, a somnologist, and software developers).

Future users. The participating teenagers (N = 29) each attended one of the four schools for special education in the Netherlands and were between 12 and 20 years old. Inclusion criteria for teenagers were: (a) diagnosis of an autism spectrum disorder made by a psychiatrist or psychologist following the DSM-IV-TR (American Psychiatric Association, 2000) or DSM-5 criteria (American Psychiatric Association, 2013), (b) presence of sleep problems, reported by the teenager and/or the parents, (c) medication free or at a stable dose during participation in the study, (d) absence of an intellectual disability, defined by an IQ of 70 or higher (American Psychiatric Association, 2021), (e) access to a smartphone, PC, or tablet, and (f) access to their own email address. Before the study began, we obtained written informed consent from each participant. If a future user was between the ages of 12 and 15 years, we also obtained written permission from both parents or legal representative(s). Teenagers received a small reward for their participation in each iteration of the design phase.

Professionals. The professionals were teachers (N = 8, including 7 teachers that also participated as future users), researchers (N = 5, including MM, CvB, HG, JPT), a somnologist (N = 1, SP), and software developers (N = 2). The participating teachers work in special education with autistic teenagers, and they were the key figures for this research project at the participating schools. The researchers have extensive knowledge about autism and/or experience with technology in health and education. The somnologist was involved because of her broad clinical experience with sleep treatment for autistic teenagers. Finally, the software developers added their practical and technological experience.

4.3.3 Research approach

Contextual inquiry. The goal of the contextual inquiry was to gain an understanding of the future users and identify weak and strong points of the current situation regarding sleep problems. This was achieved by examining existing knowledge, holding focus groups with future users, and consulting with the professionals (Table 4.1).

Table 4.1 Goals and Activities of the CeHReS Roadmap Phases

Phase	Goal	Activities
Contextual inquiry	Understand future users Identify weak/strong points of the current situation	<ul style="list-style-type: none"> • Systematic review of literature about sleep and daytime functioning • Focus groups (6 teenagers, 5 parents, 5 teachers) • Professional consultation (N = 14)
Value specification	Identify, prioritize, and translate requirements into a design plan	<ul style="list-style-type: none"> • Focus groups (5 teenagers, 6 parents, 5 teachers) • Professional consultation (N = 15)
Design	Iteratively design the sleep intervention	<ul style="list-style-type: none"> • Usability test of web app (9 teenagers, 10 parents, 3 teachers) • Usability test of sleep intervention (10 teenagers, 11 parents, 10 teachers) • Usability questionnaire (9 teenagers) • Log files (8 teenagers) • Professional consultation (N = 11)

Note. In total, seven teenagers, eight parents, and five teachers participated in the focus groups; most participated in the focus groups in both phases. The usability tests included different teenagers and parents, and some teachers participated in both the usability tests and the focus groups. In total, 16 professionals participated in the consultations.

Existing knowledge. To learn more about the impact of sleep problems on autistic teenagers, we conducted a systematic literature review to investigate the relationship between sleep and daytime functioning. This review provided insight into the impact of sleep problems on autistic teenagers, and its results were described in a separate paper (Schouwenaars et al., 2021).

Focus groups. Focus groups made up of autistic teenagers (N = 6, all male; $M_{\text{age}} = 17.5$ years, $SD = 0.6$), their parents (N = 5), and teachers (N = 5, including four teachers that also participated in the professional consultations) identified: 1) the current situation relating to the teenagers' sleep problems from the perspectives of the teenagers themselves, their parents, and teachers and 2) weak and strong points of this current situation to gain a greater understanding of what a sleep intervention could contribute in terms of improving sleep. The video-recorded focus groups were organized at each participating school. The recordings were transcribed and analyzed to identify how a sleep intervention could help.

Professional consultation. During the consultation with the professionals (N = 14, two individuals could not attend), we established what had previously been used at schools to reduce teenagers' sleep problems and the successful aspects of those previous attempts. Minutes of this session were taken and analyzed to identify potential aspects that could contribute to the success of a sleep intervention.

Value specification. Based on the issues and opportunities identified in the previous phase, we identified the future users' requirements for a school-based sleep intervention with technology, prioritized those requirements, and translated them into a design plan. This was done through two activities: focus groups with the future users at the participating schools and a professional consultation (Table 4.1).

Focus groups. The goal of the focus groups in this phase was to determine the requirements of the sleep intervention and their added value according to the autistic teenagers ($N = 5$, all male; $M_{\text{age}} = 17.3$ years, $SD = 0.3$), their parents ($N = 6$), and teachers ($N = 5$, including 3 teachers that also participated in the professional consultations). Most of the participants (4 teenagers, 3 parents, and all teachers) had also participated in the contextual inquiry. Each focus group met for approximately 150 minutes. They formulated requirements for the content of the sleep intervention, its usability, and persuasive elements. In the next step, these requirements were prioritized, and the recordings of these focus groups were transcribed. The transcripts were used to identify the requirements for the sleep intervention from the different perspectives.

Professional consultation. During the consultation with the professionals ($N = 15$, one individual could not attend), we presented the conclusions from the focus groups. This was followed by a discussion about the requirements from different perspectives: being in line with existing knowledge about sleep interventions, being in line with the existing research plan, and being (technologically) feasible. Based on consensus decisions, the requirements then were translated into a design plan. In addition, the somnologist (SP) was consulted to translate existing knowledge about effective sleep interventions into usable requirements for the purpose of this intervention.

Design. The goal of this phase was to iteratively design the sleep intervention based on the requirements formulated in the value specification. We started with the development of a prototype for the web app to measure sleep and daytime functioning. This prototype was tested in a first usability test (usability test of the web app).

Usability test of the web app. We developed a prototype for a web app to measure sleep and daytime functioning. This prototype was tested by autistic teenagers ($N = 9$, 7 males; $M_{\text{age}} = 14.7$ years, $SD = 2.1$), their parents ($N = 10$), and teachers ($N = 3$, all teachers also participated in the professional consultations). The usability of the web app was evaluated in interviews with each participant. The teenagers and parents who took part in the usability test did not participate in the focus groups for the contextual inquiry and value specification; the teachers did.

After the usability test of the web app, we developed the sleep intervention in which the web app was one of three parts. We completed the design of the sleep intervention in three design iterations, each of which contained a usability test (usability test of the sleep intervention), completion of log files, and completion of a usability questionnaire. A professional consultation was organized after the first and third iterations.

Usability test of the sleep intervention. The sleep intervention was tested by a new group of autistic teenagers and their parents and then evaluated in focus groups. Each focus group met for approximately one hour after each iteration and included the participating autistic teenagers ($N = 10$, 5 males; $M_{\text{age}} = 14.7$

years, $SD = 1.7$), their parents ($N = 11$), and teachers ($N = 10$, including 6 teachers who also participated in the professional consultations). The goal of these focus groups was to gain information about successes, aspects in need of attention or improvements, and what was learned. We organized two focus groups in the design phase at the participating schools. Due to restrictions related to the COVID-19 pandemic, the other 16 focus groups were conducted digitally, individually, or in smaller groups (for approximately 30 minutes). The recordings of the focus groups and interviews were transcribed. These transcripts were used to identify further requirements for the sleep intervention.

Log files. Teenagers ($N = 8$, 5 males; $M_{\text{age}} = 15.6$ years, $SD = 1.6$) also completed log files to give us more information about the implementation of the sleep intervention. The log files consist of questions about successes and difficulties with applying the intervention.

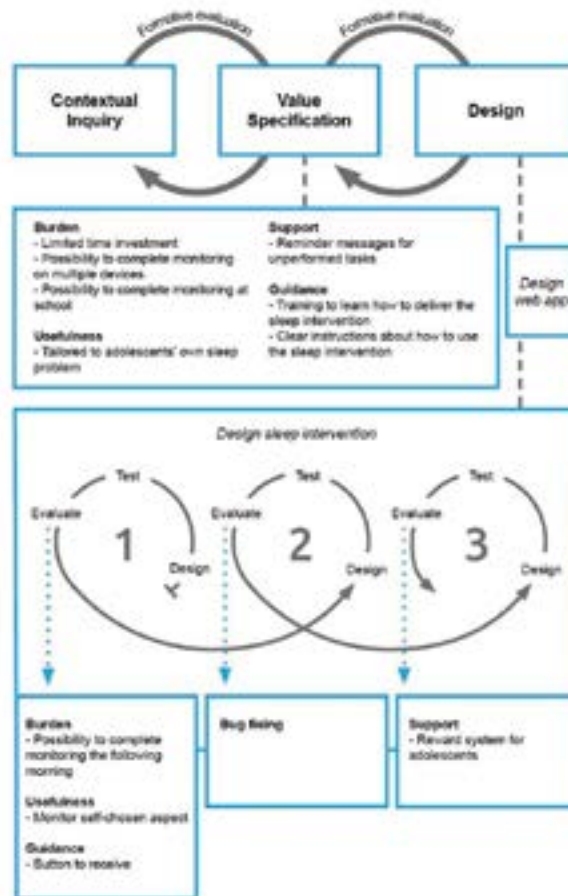
Usability questionnaire. Additionally, in the design phase, the sleep recommendations from the sleep intervention were evaluated by a usability questionnaire. This questionnaire was based on International Organization for Standardization ISO 9241-11 (Abran et al., 2003) and contained six items about effectiveness, efficiency, and satisfaction. We also added an item about ease of continuing to use the co-created sleep intervention. After each iteration, the participating teenagers ($N = 9$, 5 males; $M_{\text{age}} = 15.0$ years, $SD = 1.8$) were asked to rate each item on a scale from 1 (totally disagree) to 10 (totally agree). The questionnaires were analyzed using descriptive statistics (mean, standard deviation) to get an overview of the acceptance of the sleep intervention. The results of the usability questionnaires were applied to further improve the intervention.

Professional consultations. During the consultation with the professionals ($N = 11$; five people were unable to attend), we presented conclusions from the focus groups. This presentation was followed by a discussion about the requirements from different perspectives: being in line with existing knowledge about sleep interventions, being in line with the existing research plan, and being (technologically) feasible. Based on consensus, decisions were made to further improve the sleep intervention. Additionally, the somnologist (SP) was consulted to translate the future users' feedback into usable requirements for the purpose of this intervention.

4.4 Results

Based on the previously described activities, we developed a school-based sleep intervention that consists of three parts: 1) monitoring the teenagers' sleep and daytime functioning, 2) holding a sleep consultation with a teacher based on the monitoring, and 3) implementing the sleep recommendations (see supplementary material in Chapter 10.3 for an expanded description of the sleep intervention). The future users and professionals indicated that it was important to include four factors with corresponding operationalizations in the sleep intervention: limited burden, sense of usefulness, clear guidance and structure, and additional support cues (Figure 4.1).

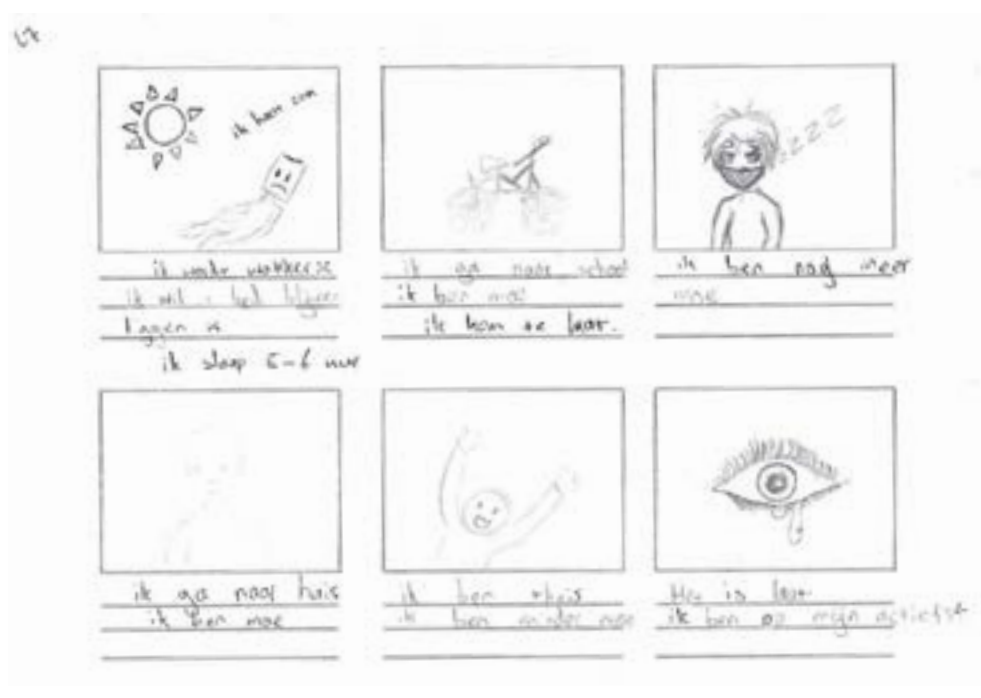
Figure 4.1 Requirements for the sleep intervention



4.4.1 Contextual inquiry

The focus groups identified the tools and circumstances that were related to better sleep for the participants (Figure 4.2). In general, teenagers, parents, and teachers mentioned that “sleep problems can be handled better in periods with a clear routine, without unexpected situations”, and when the teenagers “feel at ease with themselves”. The teenagers added that sleep could be promoted by avoiding certain foods, doing physical exercise or getting fresh air during the day, and taking melatonin. Participants also mentioned things that did not help them sleep (e.g., sleep-well tea or soothing music).

Figure 4.2. Contextual Inquiry Worksheet



Note. This teenager describes a day after a night of poor sleep. When he wakes up and feels tired, he wants to stay in bed. He arrives at school late and still feels tired. After a day at school, he goes home and feels more awake. Finally, he feels his best late in the evening.

During the professional consultation, the professionals stressed that investing in a strong teenager-parent-teacher alliance is the key element for a successful sleep intervention. Such a strong alliance would make it possible to start a conversation about sleep habits and daytime functioning to determine the cause of sleep problems and create awareness about the influence of certain activities on sleep quality. The professionals also noticed that it is important to consider the teenagers' personal situation (e.g., give them the opportunity to do their schoolwork in a quieter environment). Furthermore, the professionals

mentioned that important parts of the sleep intervention should focus on psychoeducation, sleep hygiene, alignment of the biological clock, dysfunctional thoughts and behaviors, and stress.

4.4.2 Value specification

Burden. During the focus groups and the professional consultation, future users and professionals mentioned that the sleep intervention should be as unburdensome as possible. Future users indicated that it should be possible to monitor the teenagers' sleep and daytime functioning with a limited number of questions that could be answered in a few minutes. Moreover, parents and teachers shared their desire for options to complete the monitoring on multiple devices (smartphone, tablet, PC) to increase the likelihood of completing it. The parents felt that it is important for teenagers to have the opportunity to complete the monitoring at school. This avoids interference with their families' daily morning routines, which also increases the likelihood that monitoring will be completed.

Sense of usefulness. Another important aspect of a sleep intervention is its sense of usefulness. In the focus groups, teenagers and teachers stated that it would be important to experience the benefits of the sleep intervention shortly after it began. In line with this, a sleep intervention should be tailored to personal needs. Sleep recommendations need to align with one's specific sleep problems and motivation to implement certain sleep recommendations. The importance of the sense of usefulness was confirmed by the professionals in the professional consultation.

Simplicity and guidance. Focus group participants mentioned the importance of simple and clear guidance about the sleep intervention. During the professional consultation, the professionals mainly stressed the importance of training the teachers who deliver the sleep intervention. Since teachers generally are not equipped to support teenagers in solving their sleep problems, this training provides the necessary knowledge and is of great value for the teachers' self-efficacy. We developed an e-learning program to address this issue.

Support cues. During the focus groups, parents indicated that it is important to receive reminder messages about unperformed tasks.

4.4.3 Design

Burden. In the usability test of the web app, teachers mentioned the importance of paying attention to the monitoring, especially in the first days, to implement a routine for completing the monitoring. Some teachers organized a daily group for this purpose with the participating teenagers during the usability test. The usability test showed that completing the monitoring required a limited amount of time and effort. The teenagers mentioned that there "were not a lot of questions [...] it took me about four minutes each time, so eight minutes a day" and "the questions were clear; they were not long or complicated".

During the usability test of the first iteration of the sleep intervention, parents and teachers expressed their desire to complete the monitoring questions (see Figure 4.3 for an example) later to increase the likelihood that they could do so. As one parent mentioned: "I sometimes only had time to complete the monitoring after midnight, when the monitoring for that day was already closed." Teachers perceived similar problems: if they forgot to complete the monitoring one day, it was closed the following morning.

Figure 4.3. Example of Monitoring Questions

The figure shows a digital monitoring interface with five sections, each with a 10-point Likert scale. The scales are color-coded from green (low) to red (high). The sections are:

- Daytime sleepiness**: (1 = not at all, 10 = very much). The scale shows a gradient from green to red, with a blue circle '1' next to the first point.
- Stress**: (1 = not at all, 10 = very much). The scale shows a gradient from green to red, with a blue circle '1' next to the first point.
- Mental effort**: (1 = not at all, 10 = very much). The scale shows a gradient from green to red, with a blue circle '1' next to the first point.
- Concentration**: (1 = very bad, 10 = very good). The scale shows a gradient from red to green, with a blue circle '1' next to the first point.
- Mood**: (1 = very bad, 10 = very good). The scale shows a gradient from red to green, with a blue circle '1' next to the first point.

As in the value specification, the usability questionnaire revealed that a sleep intervention should require as little burden as possible. This implies that sleep recommendations should be easy and feasible for the teenager. The results from the usability questionnaire showed that it was challenging to apply and continue to use the recommendations (Table 4.2). Teenagers and parents mentioned that it was "sometimes difficult to change their habits and fit the sleep recommendations into daily routines". This emphasizes the need for optimal guidance of teenagers in implementing the recommendations and motivating them for long-term adherence. The log files confirmed these findings.

Table 4.2. Descriptive Statistics of the Usability Questionnaire about Sleep Recommendations

	Mean (SD) ^a	Range	N ^b
Beneficial to apply	7.64 (1.8)	5 - 10	11
Enjoyable to apply	5.83 (1.9)	3 - 9	12
Easy to apply	5.17 (1.9)	2 - 7	12
Useful to apply	7.00 (3.0)	2 - 10	11
Easy to continue using	5.50 (1.9)	3 - 9	12
Easy to understand	8.00 (1.4)	5 - 10	11
Resources were available	7.75 (2.4)	3 - 10	8

^a Measured on a scale from 1 (totally disagree) to 10 (totally agree).

^b In total, nine teenagers participated in the design phase. Some participated in multiple iterations. Since the usability questionnaire was completed after each iteration, some teenagers completed the questionnaire multiple times.

Sense of usefulness. Overall, the usability questionnaire showed that the teenagers experienced the sleep recommendations as beneficial and useful (Table 4.2). Especially the personalized approach seemed to aid in this aspect. Teenagers and their parents indicated that the opportunity to monitor a self-chosen factor that they expected to influence their own/their child's sleep could further enhance this personalized approach. This possibility was therefore added after the first iteration. These ways of providing a sense of usefulness by creating a personalized approach seem promising.

However, some teenagers also gave low scores to usefulness. Two teenagers explained their low score during the usability test of the sleep intervention. One teenager mentioned: "I did not notice changes in my sleep pattern". However, during the conversation she also mentioned that "One of the sleep recommendations was to change my bedtime to a later hour. Moving my bedtime gave me more spare time in the evening without making me sleepy the following day". Hence, while this advice might be helpful to others, it might not have suited her needs.

The other teenager mentioned that the sleep recommendations were not immediately suitable. For example, one recommendation was that he not take his smartphone into his bedroom when going to bed, but he uses his smartphone to play music to relax before falling asleep. Since he participated in both the second and third iterations of the usability test of the sleep intervention, the focus in the third iteration was on fine-tuning the sleep recommendations. His evaluation of the third iteration showed that this fine-tuning was successful. The scores on the third questionnaire were much higher, and in the focus group the teenager and parent expressed their positive experiences with the sleep recommendations.

Simplicity and guidance. During the usability test of the web app, we saw that expectations were not completely clear. Before implementing such an intervention, expectations and time investments should be clear to those who deliver and receive it. For example, it should be clear how and when the monitoring should be completed and how questions should be interpreted.

The usability questionnaire and log files showed that, overall, the instructions for the sleep recommendations were clear and most teenagers perceived that they had support for applying them (Table 4.2). However, one teenager gave a low score to available resources. This again stresses the need to guide the teenagers. To best prepare them, we paid specific attention in the sleep consultation to how the sleep recommendations could be implemented.

Support cues. During the third iteration of the sleep intervention’s usability test and the professional consultation, future users and professionals mentioned that in addition to reminder messages it would be desirable for the sleep intervention to be accompanied by a rewards system for teenagers. These rewards could be different for the various components of the sleep intervention. For instance, teenagers could earn points and messages like “well done” for the monitoring. But all felt that the most effective reward would mainly be “the experienced benefits to sleep quality or daytime functioning”.

Bug fixing. After the second iteration of the usability test of the sleep intervention, the adaptations only entailed fixing bugs in the web app. This was mainly focused on sending the reminder messages, completing the monitoring at another time, and increasing the readability of the output (Figure 4.4).

Figure 4.4. Example of Output from the Monitoring



4.5 Discussion

It is challenging to successfully implement school-based sleep interventions (Cassoff et al., 2013). We tried to address this issue by developing a school-based sleep intervention in co-creation using the first three phases of the CeHReS roadmap (van Gemert-Pijnen et al., 2011). Due to this roadmap's iterative development approach with an emphasis on stakeholder participation, we noticed possible barriers to the intervention's application during its development. This approach enabled us to anticipate those barriers and adapt the sleep intervention during its development, which increases the likelihood of successful implementation.

We paid attention to five domains (characteristics of the individual, intervention characteristics, inner setting, outer setting, implementation process) that could increase the likelihood of successful implementation (Damschroder et al., 2009). First, we involved relevant stakeholders to consider the characteristics of the individual, intervention characteristics, inner setting, and outer setting. Our development process included autistic teenagers, their parents, and teachers as future users, each with their own role and perspective on the sleep problems. The autistic teenagers contributed their own experience with sleep problems and expressed their needs and wishes for the intervention. In addition, parents and teachers contributed their own perspectives on the sleep problems of their child/student and on the requirements for the intervention.

We found that teenagers, parents, and teachers sometimes had different requirements. For example, teenagers preferred to complete the monitoring on their smartphone, while some parents and teachers preferred to do this on a PC. In such cases, we tried to search for a solution that met the different requirements. In this example, we decided to develop a web app which could be reached using a web browser and was therefore available on smartphone, tablet, and PC.

Besides future users, it was also crucial to involve the somnologist during the development phase. The somnologist added scientific knowledge about effective sleep interventions as well as broad clinical experience with sleep treatment for autistic children, which we used in developing the intervention. For sustainability purposes, it is important to develop an intervention that can be used without involving experts. Thus, we developed an e-learning that could be used after this study to train new teachers to deliver the sleep intervention.

Second, since teachers deliver this sleep intervention, involving teachers gave us an understanding of their attitudes, skills, and beliefs about their own competence to do so (characteristics of the individual). One finding from the value specification phase was the importance of training teachers to deliver the sleep intervention. Despite this training, teachers were initially less confident about their ability to deliver the intervention, but their confidence grew during the development of the intervention. Although we trained teachers, their prior education had not prepared them to treat teenagers with multiple and

interacting problems. Therefore, it is crucial that teachers can recognize when problems need more specialized treatment. When it is known that a teenager has multiple or interacting problems (e.g., depression, anxiety disorder, substance dependence, or game addiction), these problems should be treated before teachers attempt to address sleep problems.

We also found that other existing problems could emerge while treating sleep problems. For instance, despite his good intentions, one teenager was unable to follow the sleep recommendations. An informal conversation with that teenager led the teacher to suspect he might have a game addiction. As this case illustrates, the inability to implement the sleep recommendations could be a sign of multiple or interacting problems. When a teacher notices such an inability, this observation should be addressed in a sleep consultation to gain further understanding. In addition, colleagues and parents should be asked for their view on the teenager's problems and, if necessary, help to refer the teenager to suitable treatment.

Third, the intervention characteristics were also considered through the iterative approach in which we tested the intervention on small scales. In every development phase, we considered the needs and wishes of the future users (autistic teenagers, parents, teachers), evaluated them, and applied our findings to further develop the sleep intervention. Another aspect of the intervention characteristics is the costs. On the one hand, developing an intervention in co-creation is a process that requires a substantial time investment from those who are involved, with associated costs. On the other hand, a sleep intervention delivered by teachers could have cost benefits because sleep problems might be treated earlier, which might reduce health care utilization. These last aspects should also be taken into account when considering the development of an intervention in co-creation.

Finally, we considered the implementation process by involving representatives from the schools' administrations. The role of those representatives is crucial (Thoonen et al., 2011). They could motivate teachers to get involved in the process of developing the sleep intervention and promote its planning and execution. Moreover, they know who is well-suited to deliver the sleep intervention and could adapt the school's organization to implement it.

During the development process, the representatives from the schools' administrations organized the development and implementation of the sleep intervention in different ways. One school, for example, arranged a work group for teachers and facilitated them by providing sufficient time. This enhanced the collaboration and collective responsibility for implementing the intervention (Stoll et al., 2006).

Taken together, this study stresses the added value of using the CeHReS roadmap in an educational setting to increase the chance that an intervention will be implemented successfully. Although this roadmap was initially developed to design and implement technologies in health care, we demonstrated that it can also be applied in other settings. Regardless of the setting and subject, we

recommend this approach for developing interventions because including relevant stakeholders and using an iterative approach provided us meaningful information that otherwise probably would not have emerged. For this study, the most prominent added value of using the CeHReS roadmap was the stakeholders' information about contextual factors. This development process led us to address the recommendation of Cassoff et al. (2013) and develop a personalized sleep intervention specific to a school-based setting that shows promise in allowing teachers to support their students in reducing sleep problems. We also experienced that teachers became more enthusiastic about and supportive of the sleep intervention during its development. They proposed suggestions and took time to engage with the sleep intervention. Moreover, this development process led us to design a sleep intervention that better matches the future users' needs, wishes, and circumstances, which also benefits teachers who were not involved in its development.

4.5.1 Strengths and limitations

The greatest strengths of this study are the inclusion of multiple stakeholders' perspectives and the user-centered approach from the beginning of the intervention's development. In every development phase, we considered the needs and wishes of the future users (autistic teenagers, parents, teachers), evaluated them, and applied our findings to further develop the sleep intervention.

Engaging autistic people in co-creation can be challenging (Benton et al., 2012) for both the autistic and non-autistic participants in such a process. Co-creation involves collaborating with others, creating new ideas, and communicating those ideas, all of which could be challenging for autistic people (Benton et al., 2012). Benton et al. (2014) offer recommendations for implementing a successful co-creation process with autistic teenagers: these include maintaining a similar structure for each session, conducting the sessions in a familiar environment, and involving adults who are familiar to the teenagers to engage with and support them in putting their thoughts into words. We applied these recommendations and successfully engaged autistic teenagers in this study. Despite these recommendations, some teenagers were not engaged enough to participate in all activities. However, this problem is not specific to autistic teenagers; it also appears in co-creation with other populations (Penuel et al., 2007). As a result, we strongly recommend that future researchers who want to engage in meaningful co-creation with autistic people consider applying the recommendations of Benton et al. (2014).

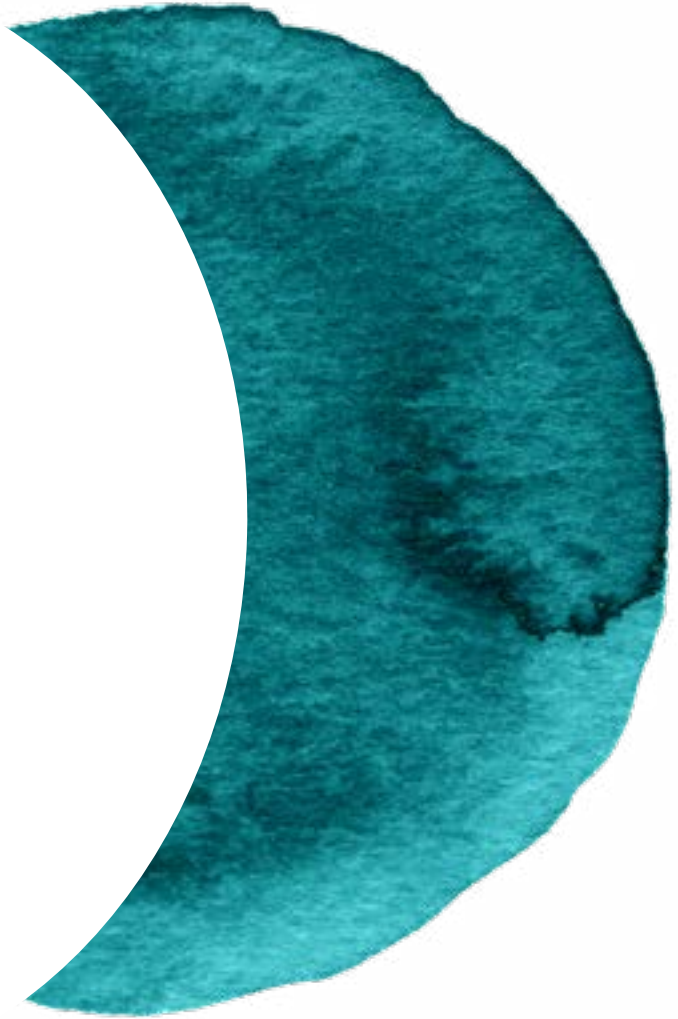
One limitation of this study was the exclusively male and relatively older participant group in the initial phase of the study. Because there are potential differences in the needs and wishes of male and female and younger and older children (Chow et al., 2012; Kinzie & Joseph, 2008), we made sure to include female and younger participants in subsequent phases of the development process.

After the third iteration, we decided that the development of the sleep intervention was complete. This decision was based on a professional consultation that looked at input from future users. Although we still found some low scores on the usability questionnaire, overall, the usability tests showed that

future users were satisfied with the intervention. One could argue that the development was not yet finished because of these low scores and that saturation is a better approach to deciding whether the development process is complete (Saunders et al., 2018). But as most of the low scores were given for confidence regarding the application of the sleep recommendations, we were confident that the issues would be addressed because the final version of the intervention contains a longer cyclical process of monitoring the teenagers' sleep and daytime functioning, holding a sleep consultation, and advising teenagers and parents about how to implement the sleep recommendations. This offers opportunities to evaluate and adapt those recommendations.

4.5.2 Future directions

We used the CeHReS roadmap to develop a school-based sleep intervention. Since it was developed in co-creation and based on knowledge about evidence-based sleep interventions, we expect a test of its efficacy to be successful. Therefore, the next step is to investigate the school-based sleep intervention's efficacy and sustainability.



Chapter 5

A school-based sleep intervention for autistic teenagers: Effects on sleep quality and daytime functioning

Schouwenaars, I.M.G., Magnée, M.J.C.M., Geurts, H.M., van Bennekom, C.A.M., Pillen, S., Deen, M., & Teunisse, J. (2023). A School-Based Sleep Intervention for Autistic Teenagers: Effects on Sleep Quality and Daytime Functioning. PsyArXiv. <https://doi.org/10.31234/osf.io/dz5uf>

5.1 Abstract

We co-created a school-based sleep intervention based on the premises that such interventions are likely to be more easily accessible than intervention delivered in clinical practice. In this study, we primarily investigated the efficacy of this intervention on sleep quality and daytime functioning using a multiple baseline design with 23 teenagers ($M_{\text{age}} = 14.8$, $SD = 1.7$, range = 12 - 18 years). The teenagers completed a daily web-based sleep diary and wore a sleep tracker overnight. They also answered questions about their daytime functioning, which were also answered by a parent and a teacher. Additionally, the teenagers, parents, and teachers completed questionnaires (sleep problems [ISI and CSHQ], and behavioral problems [SDQ]) at pretest, post-test, and follow-up. We found that the intervention had no significant effect on sleep quality or daytime functioning, however, the majority of the teenagers showed clinically significant improvement on the self-reported sleep problems questionnaire (ISI). Although our study population had mild sleep problems, which offers little room for improvement, this result suggests that the school-based sleep intervention has a positive effect on the severity of insomnia symptoms.

5.2 Introduction

The majority of sleep interventions for autistic teenagers are delivered in clinical practice (e.g., Loring et al., 2018; McCrae et al., 2020; Papadopoulos et al., 2019). These interventions successfully reduce sleep problems, however, school-based sleep interventions could be more easily accessible than interventions delivered in clinical practice (WHO, 1997). Easy accessibility is important for the early treatment of sleep problems, which could prevent exacerbation of these problems (Morin & Benca, 2012). This is not only beneficial for those with sleep problems and their families, however, improving sleep has cost benefits derived from reduced health care utilization (Daley et al., 2009). A school-based sleep intervention could be more easily accessible for teenagers and thus have a greater reach. In this study, we investigate the effectiveness of a co-created school-based sleep intervention for autistic teenagers.

Sleep problems are common among autistic teenagers: 40 - 80% experience them (Cortesi et al., 2010) compared to 16 - 31% of typically developing teenagers (Calhoun et al., 2014). The most prevalent sleep problems among autistic teenagers include difficulties initiating and maintaining sleep, a shorter sleep duration, and a lower sleep efficiency (Tse et al., 2020). Those sleep problems have negative effects on daytime functioning, such as more severe autism traits, internalizing behavior, externalizing behavior, and problems with social behavior (Allik et al., 2006; Gabriels et al., 2005; Mazurek & Sohl, 2016; Phung et al., 2018; Richdale et al., 2014). Thus, interventions to improve sleep could also have a positive impact on daytime functioning.

In general, the initial approach to reducing sleep problems is starting a behavioral sleep intervention (Crowe & Salt, 2015). This type of intervention consists of creating awareness about the importance of sleep and sleep hygiene, faded bedtime, sleep restriction, and sleep timing interventions (Johnson & Zarrinnegar, 2021; Keogh et al., 2019). For autistic teenagers, it is important to personalize those general sleep recommendations to fit their personal preferences, since factors that facilitate sleep in autistic teenagers could differ from generalized sleep recommendations (Pavlopoulou, 2021). Therefore, a collaborative and individual approach is crucial to developing personalized sleep interventions to reduce autistic teenagers' sleep problems.

Based on the aforementioned knowledge, we co-created a school-based sleep intervention (Schouwenaars et al., 2022) with three parts: 1) monitoring the teenagers' sleep and daytime functioning, 2) holding a sleep consultation with a teacher based on the monitoring, and 3) implementing the sleep recommendations. The aim of this study is to determine whether that intervention improves sleep quality and daytime functioning in autistic teenagers.

5.3 Methods

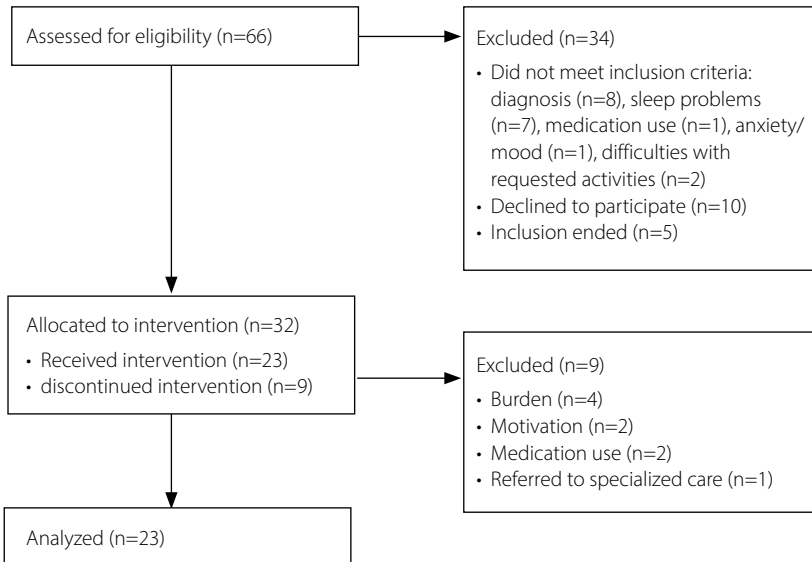
5.3.1 Participants

The study participants were autistic teenagers, each with one parent and one teacher. Eligible teenagers attended one of four schools for special education in the Netherlands and were between 12 and 20 years old.

In total, 66 teenagers were screened to determine whether they met our inclusion criteria. Those criteria were: (a) being a student at one of the participating schools; (b) being diagnosed with an autism spectrum disorder by a psychiatrist and/or psychologist, based on the criteria of the DSM-IV-TR (American Psychiatric Association, 2000) or DSM-5 (American Psychiatric Association, 2013); (c) a score of >7 (which means sub-threshold insomnia) on the Insomnia Severity Index (ISI); and (d) being medication free or at a stable dose during this study.

Exclusion criteria were: (a) intellectual disability, defined by an IQ < 70 (American Psychiatric Association, 2021); (b) diagnosed or suspected depression, anxiety disorder, substance dependence, or game addiction; (c) disturbed parent-child relationship that could hinder the sleep intervention; and (d) indications that the teenagers (and parent) would have difficulties performing the requested activities. Of the 66 teenagers screened for participation, 34 were excluded, resulting in 32 participants (Figure 5.1).

Figure 5.1. Inclusion Flow Diagram



We obtained written informed consent from all participants before study entry. For children aged 12 to 15, we also obtained written permission from both parents or legal representative(s). The first group of teenagers participated from September 2021 to February 2022, and the second from February 2022 to July 2022. The teenagers received a small reward for their participation. Study approval was obtained from the Ethics Review Board of the Faculty of Social and Behavioral Sciences at the University of Amsterdam (2021-EXT-13604).

5.3.2 Design

We used a multiple baseline design to investigate the effectiveness of our school-based sleep intervention. Like a randomized controlled trial, a multiple baseline design can show the occurrence of change in outcome measures over time (Hawkins et al., 2007). The duration of the baseline was two to three weeks. The intervention period was ten weeks for each participant, and follow-up data were collected eight weeks after the intervention during one entire week (Figure 5.2).

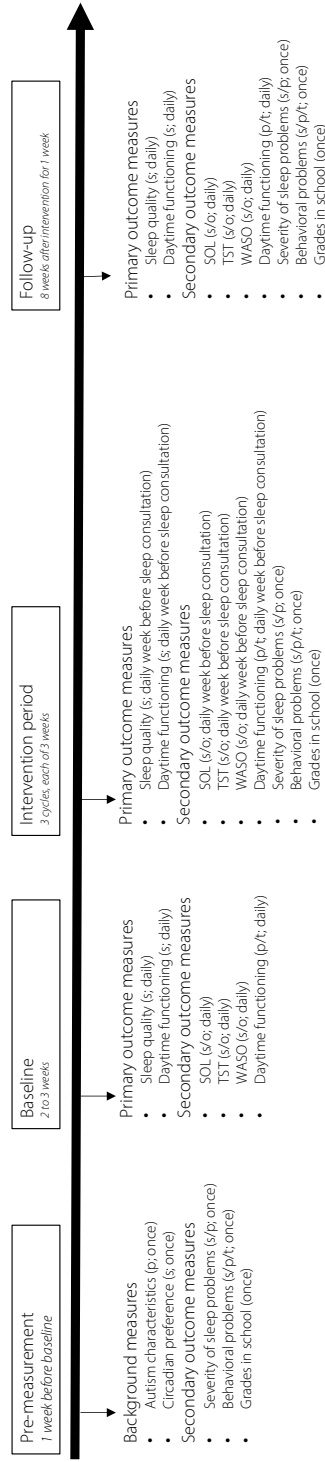
5.3.3 Intervention

The school-based sleep intervention started one week after pre-measurement. It consists of three repeating parts: 1) monitoring the teenagers' sleep and daytime functioning, 2) holding a sleep consultation with a teacher based on the monitoring, and 3) implementing the sleep recommendations (Schouwenaars et al., 2022).

To gain insight into each teenager's specific sleep problems, we monitored their sleep for two to three weeks using a sleep diary. This sleep diary contains questions about bedtime, sleep onset time, nighttime awakenings, wake time and last night's activities: intensive physical exercise, gaming, use of electronic devices, and caffeine intake (Carney et al., 2012). To evaluate the association between sleep and daytime functioning, the teenagers rated the following aspects: daytime sleepiness, stress, concentration, mood, mental effort, and physical exercise.

After the sleep and daytime functioning monitoring, the teenagers had a sleep consultation with a teacher. Each teacher had completed a one-and-a-half hour e-learning course containing basic information about healthy sleep and sleep disturbance. The e-learning consisted of six modules: one with information about normal sleep followed by five about possible causes of sleep problems: psychoeducation, sleep hygiene, alignment of the biological clock, dysfunctional thoughts and behaviors, and stress. The course gave teachers basic knowledge about how to evaluate sleep diaries, identify possible causes of sleep problems, and use case examples to develop sleep recommendations related to the cause of the sleep problems.

Figure 5.2. Overview of Study Design



s = self-reported, p = completed by parents, t = completed by teachers, o = objectively measured, SOL = sleep onset latency, TST = total sleep time, WASO = wake time after sleep onset

During the initial sleep consultation, the teenager and teacher discussed the teenager's sleep pattern and daytime functioning. In line with suggestions from Pavlopoulou (2021), the teacher made sleep recommendations that aligned with the teenager's interests, abilities, and preferences. The teenager and teacher then planned the implementation of the sleep recommendations.

During the second and third sleep consultations, the teenager and teacher evaluated the effects of previous plans based on new sleep patterns from one week of monitoring. They also discussed facilitators and barriers to adopting the sleep recommendations, and they modified the recommendations when necessary.

5.3.4 Measures

Background data measures. We gathered information on autism characteristics and circadian preferences to describe the background characteristics of the study participants.

Autism characteristics. We used the Social Responsiveness Scale-Second Edition (SRS-2), version for 4- to 18-year-olds, to measure difficulties in social behavior. The SRS-2 comprises 65 items and calculates a total score for autism characteristics, including two subscales: social communication and interaction and stereotype behavior and interests (Roeyers et al., 2015). A four-point Likert scale (1 = *not true* to 4 = *almost always true*) is used to rate each item. The reliability and validity of the SRS-2 in a Dutch population is good (Roeyers et al., 2015). The questionnaire was completed by one of the teenager's parents. For this study, we used the total score. A higher score implies more autism characteristics.

Circadian preference. We used the Morningness-Eveningness Questionnaire for Children and Adolescents (MEQ-CA) to measure each teenager's circadian preference. The MEQ-CA comprises 19 items that indicate preferred times for several activities and how easy or difficult certain activities are at given times. The total score was used as measure for circadian preference, with higher scores indicating greater morningness.

The MEQ-CA is based on the MEQ, a questionnaire for measuring circadian preference in adults. The items on the MEQ refer to work activities, while the items on the MEQ-CA refer to academic activities to align with the experiences of children and teenagers (Tonetti et al., 2015). The MEQ-CA has been found to be valid in the teenager population (Tonetti, 2007).

To translate the English version of the MEQ-CA into Dutch, we did the following: 1) the first author translated the English version into Dutch, 2) a near native speaker of English (teacher educator and senior lecturer of English) translated the Dutch version back into English, 3) the English and Dutch versions were compared to reveal minor differences, and 4) the Dutch version was slightly modified to account for those differences.

Primary outcome measures. We have two primary outcome measures.

Sleep quality. Teenagers were asked for daily reports of how well they slept, their feeling of being rested when waking up, and difficulties with waking up (1 = very bad to 10 = very good). We calculated the mean of the answers to these three questions and used it as a measure of sleep quality, with a higher score indicating a better self-reported sleep quality.

Daytime functioning. Daytime functioning was measured by the teenagers' daily reports of the following aspects: mood (1 = *very bad* to 10 = *very good*), concentration (1 = *very bad* to 10 = *very good*), and sleepiness (1 = *not at all* to 10 = *very severe*). We summed the scores (with reversed scores for sleepiness) and used that total as a measure for daytime functioning. A higher score implies better daytime functioning.

Secondary outcome measures. Besides the two primary outcome measures, we have 14 secondary outcome measures.

Severity of sleep problems. We assessed the experience of sleep problems by asking the teenagers to complete the Insomnia Severity Index (ISI) (Bastien et al., 2001) and the parents to complete the Children's Sleep Habits Questionnaire (CSHQ) (Waumans et al., 2010). The ISI was originally developed to measure insomnia in adults, however, it has been found to be reliable and valid in a teenager population (Chung et al., 2011). The CSHQ has been validated for children aged 4 - 10 years, and it is also used for autistic children of secondary school age (see e.g., Mazurek & Sohl, 2016). For both questionnaires, we used the total score, with higher scores implying more sleep problems.

Sleep onset latency, total sleep time, and wake time after sleep onset. We used a sleep diary to measure self-reported sleep. It consists of questions about bedtime, sleep onset time, nighttime awakening, and wake time. Answers were used to calculate sleep onset latency (difference between time trying to fall asleep and sleep onset time), wake time after sleep onset (sum of time awake between sleep onset time and wake time), and total sleep time (difference between sleep onset time and wake time minus the wake time). We also captured objective sleep parameters using wrist-worn reflective plethysmography and accelerometry (prototype developed by Philips Research). Sleep onset latency, total sleep time, and wake time after sleep onset were measured using a trained algorithm (Wulterkens et al., 2021).

Behavioral problems. We used the Strengths and Difficulties Questionnaire (SDQ), version 4- to 17-years-old for parents and teachers and version 11- to 17-years-old for teenagers, to measure psychosocial problems (Goodman, 2001). For this study, we used the psychosocial problems total score. A higher score implies more behavioral problems.

Daytime functioning. A parent and teacher of each teenager completed questions about the teenager's daytime functioning from their own perspective. They gave daily scores to the following aspects: mood (1 = *very bad* to 10 = *very good*), concentration (1 = *very bad* to 10 = *very good*), and sleepiness (1 = *not at all* to 10 = *very severe*). We used the sum of the scores (with reversed scores for sleepiness) as a measure for daytime functioning. A higher score implies better daytime functioning.

Grades in school. We used the teenagers' actual grades for Dutch, English and math. A higher score implies better performance.

5.3.5 Statistical analyses

We preregistered the analysis plan for this study at AsPredicted (#66893; https://aspredicted.org/XFK_HHG). We slightly deviated from it regarding one exclusion criteria: we chose not to exclude teenagers who did not complete the sleep diary for at least three weekdays and one weekend day within each week for each measurement period to include all data.

First, we visually analyzed the data of the primary outcome measures, which involved analyzing trend and level data within and between subsequent phases for each participant. We then used the R-package nlme (Pinheiro et Bates, 2022) to evaluate the statistical significance of the time*intervention-interaction effect via a multilevel modeling approach. We computed a model with phase * time centered and imposed an AutoRegressive (AR1) residual covariance structure on it. We preregistered that if the p -value of the fixed effect of phase*time-interaction was $\leq .05$ (and the scores improved) on at least sleep quality, we would conclude that the sleep intervention is effective.

Missing data were imputed using the mice package (van Buuren & Groothuis-Oudshoorn, 2011) in R. We used five multiple imputations with 20 iterations to obtain robust imputations. Two imputation models were conducted: (i) we used all participants and all aforementioned variables to analyze sleep quality, daytime functioning (self- and parent-reported), sleep onset latency, sleep duration, and wake time after sleep onset (self-reported and objectively measured) and, (ii) we used all variables to analyze teacher-reported daytime functioning, however, we excluded one participant that had no scores for teacher-reported daytime functioning. We performed the analysis to evaluate the statistical significance of the intervention both without and with imputations to see if the results differed. When these results differ, we will describe the results with the imputations.

The secondary outcome measures (sleep onset latency, total sleep time, and wake time after sleep onset (self-reported and objectively measured) and daytime functioning reported by parents and teachers) were analyzed as described above. We analyzed the questionnaires (ISI, SDQ, CSHQ) using reliable change index (RCI) (Jacobson & Truax, 1991). The RCI is a measure for individual-level change and is calculated by dividing the individual's change score by the standard error of the difference of the tests. The psychometric data used to calculate the RCI were derived from the following studies: (i) Chung et

al. (2011) for the ISI, (ii) Goedhart et al. (2003) for the SDQ, and (iii) Owens et al. (2000) and Schlarb et al. (2010) for the CSHQ. For this last questionnaire, we used two studies because, as far as we know, no study has reported on both the SD and test-retest of the total score.

We used the results of the secondary outcome sleep measures and questionnaires to gain a more in-depth understanding of the results of the primary outcome measures. We also exploratively compared grades in school using a repeated measures ANOVA to see whether improvements were visible after the intervention.

5.4 Results

5.4.1 Background characteristics of participants

We included 32 teenagers in this study, each with one of their parents and teachers. Nine teenagers withdrew from the study during the research period (see Figure 5.1). As a result, 23 teenagers (19 boys) with a mean age of 14.8 years ($SD = 1.7$, age range = 12 - 18 years) participated.

The mean MEQ-CA score was 51.3 ($SD = 10.5$), which indicates the intermediate circadian preference type (not outspoken morning or evening type) (Tonetti et al., 2015). The majority of the teenagers ($n = 12$) scored in the range of the intermediate type, five teenagers scored in the range of the morning type, and three had a score which indicated evening types. Three teenagers did not complete the MEQ-CA.

The mean score on the SRS-2 was 78.6 ($SD = 23.5$). The majority of the teenagers (82%) scored above the autism cut-off score of 51. There were no significant difference in the MEQ-CA and SRS scores for teenagers who withdrew from the intervention (MEQ-CA: $M = 45.00$, $SD = 5.66$ and SRS: $M = 67.50$, $SD = 14.25$) and those who completed the intervention (MEQ-CA: $M = 51.25$, $SD = 10.48$ and SRS: $M = 78.59$, $SD = 23.52$); for the MEQ-CA and SRS respectively $t(24) = 1.39$, $p = .18$ and $t(26) = 1.09$, $p = .29$. Specific data on socioeconomic status and ethnicity were not recorded.

Overall, of the 23 teenagers, 10 attended all three sleep consultations and 13 attended two. There were no significant difference in the MEQ-CA and SRS scores for teenagers that attended three (MEQ-CA: $M = 49.11$, $SD = 9.01$ and SRS: $M = 87.56$, $SD = 17.60$) versus two sleep consultations (MEQ-CA: $M = 53.00$, $SD = 11.67$ and SRS: $M = 72.38$, $SD = 25.69$); for the MEQ-CA and SRS respectively $t(18) = 0.82$, $p = .42$ and $t(20) = -1.54$, $p = .14$.

5.4.2 Primary outcome measures

Table 5.1 presents descriptive data about sleep quality and self-reported daytime functioning during baseline, the intervention period, and at follow-up.

Table 5.1. Descriptive Data of Sleep Quality and Self-Reported Daytime Functioning

	Baseline		Intervention period		Follow-up	
	M (SD)	Range	M (SD)	Range	M (SD)	Range
Sleep quality (s)^a (no. of measurements = 241/248/57)	6.5 (1.8)	1.0 - 10.0	6.9 (1.7)	1.0 - 9.7	7.1 (1.7)	1.0 - 9.3
Daytime functioning (s)^b (no. of measurements = 173/206/46)	20.7 (5.7)	5.0 - 30.0	21.8 (5.5)	3.0 - 29.0	22.4 (4.9)	3.0 - 27.0

s = self-reported

^a Possible range 1 - 10

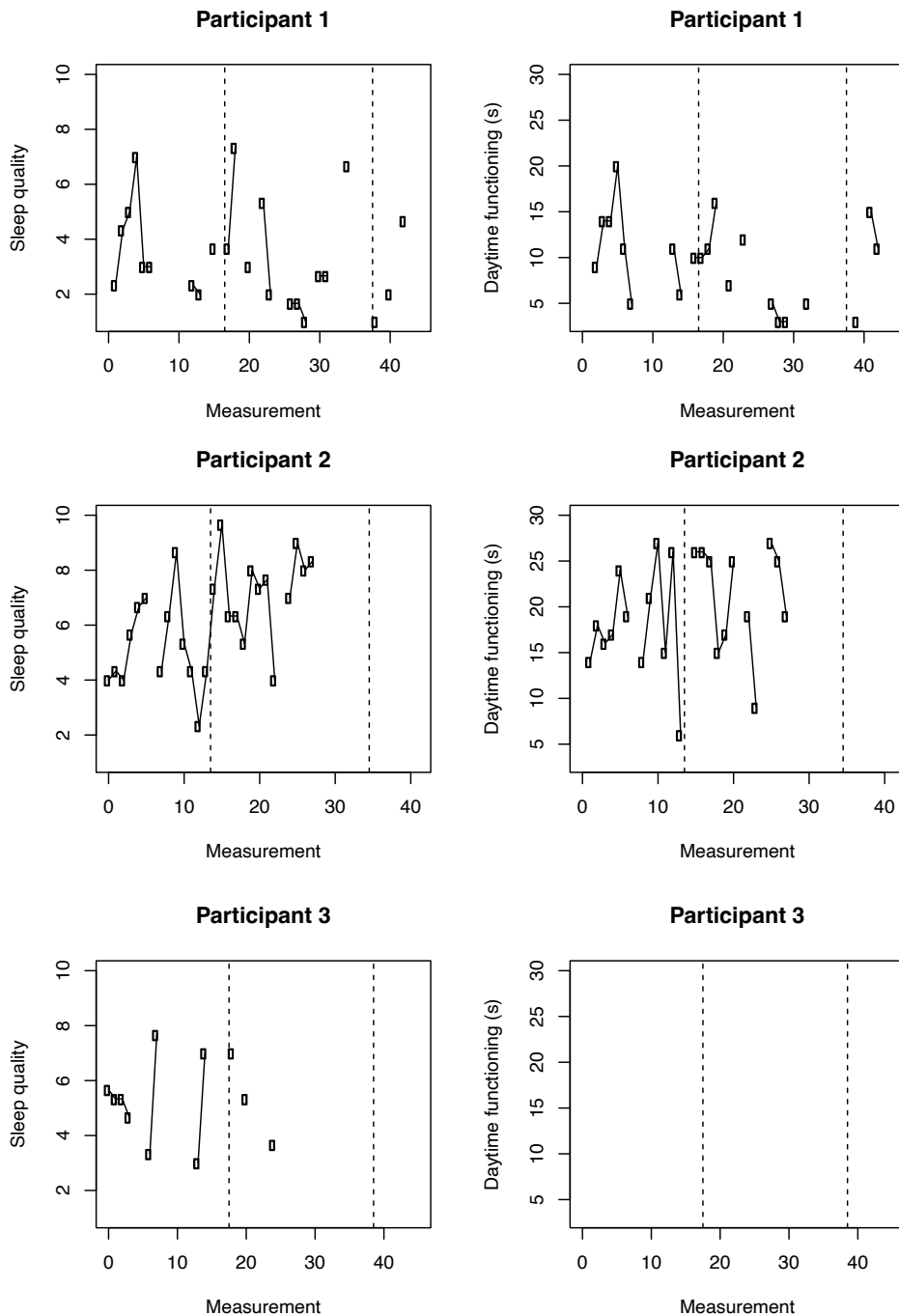
^b Possible range 3 - 30

Note. Number of possible measurements differed during each measurement period (see Figure 5.2)

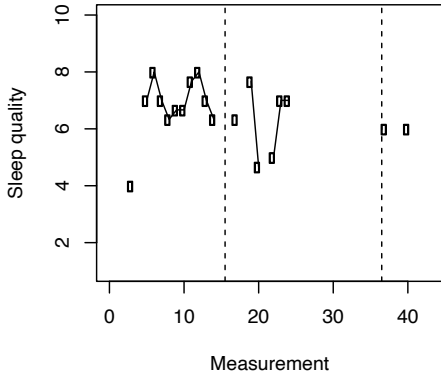
The individual scores for sleep quality and self-reported daytime functioning during baseline, the intervention period, and follow-up are shown in Figure 5.3. The visual analysis showed a slight improvement for participants 2, 6, and 23. Participant 10 showed a decrease in sleep quality. The other participants showed more or less unchanged scores or an irregular pattern.

Regarding self-reported daytime functioning, participant 10 also showed a slightly decrease, while the other participants showed more or less unchanged scores or an irregular pattern. Table 5.2 contains the parameter estimates for the multilevel model, showing that sleep quality and self-reported daytime functioning did not significantly improve.

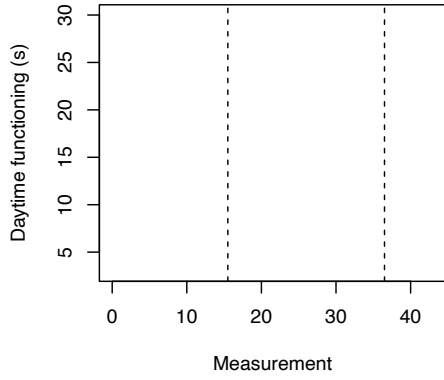
Figure 5.3. Sleep Quality and Self-Reported Daytime Functioning



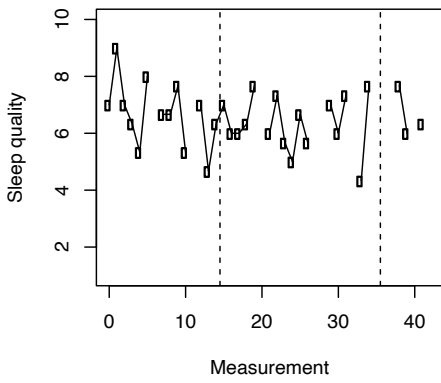
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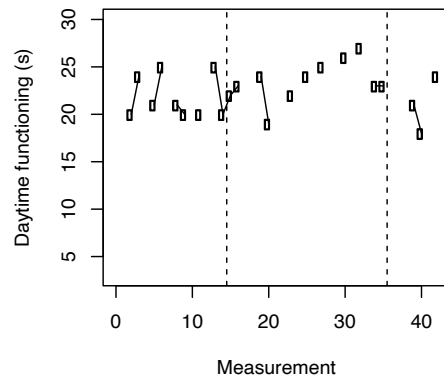
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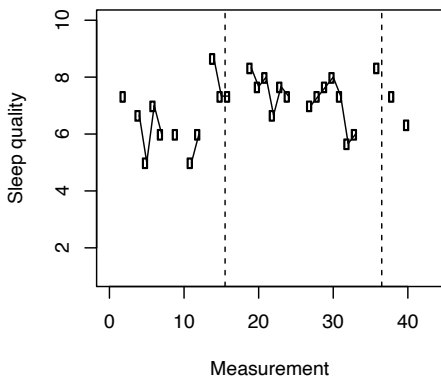
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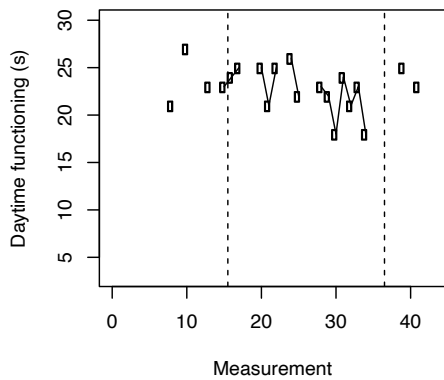
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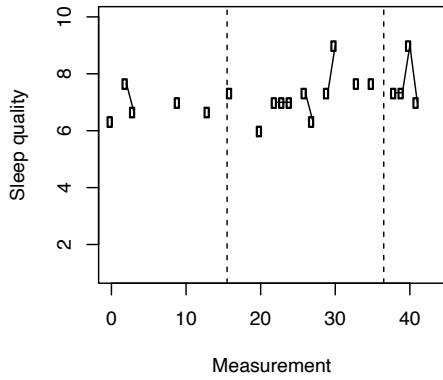
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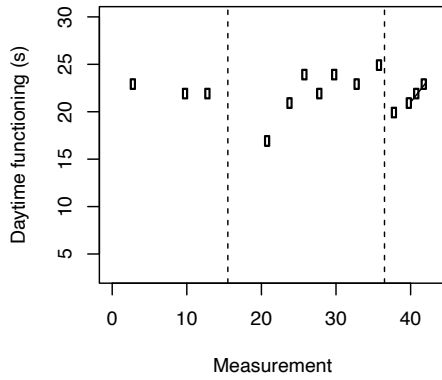
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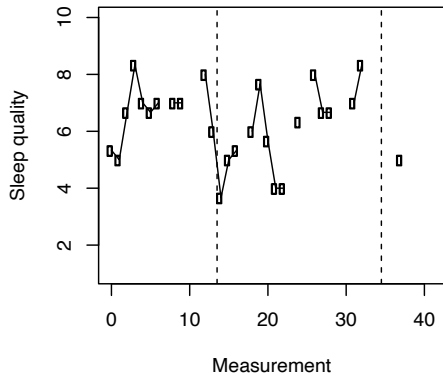
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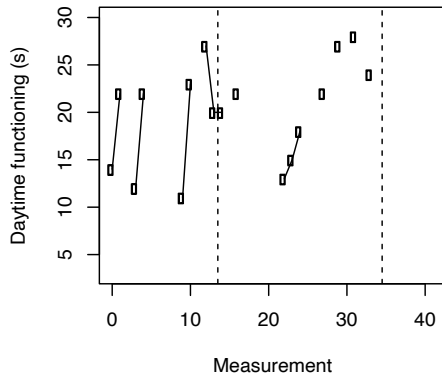
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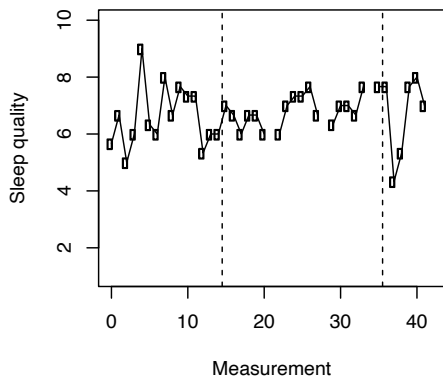
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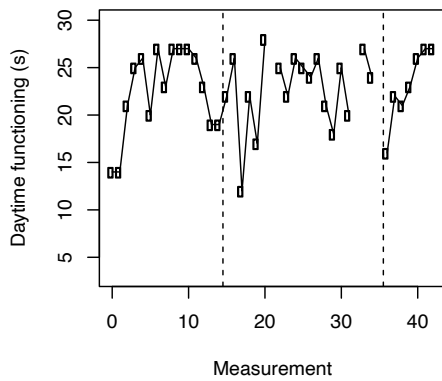
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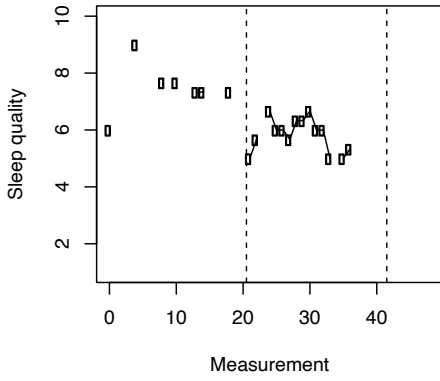
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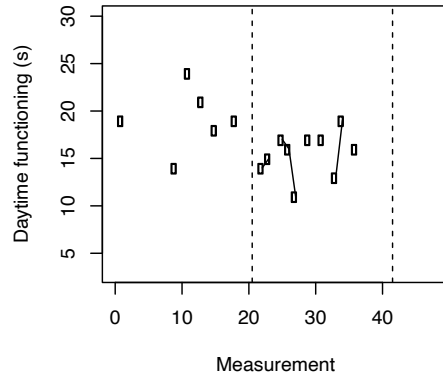
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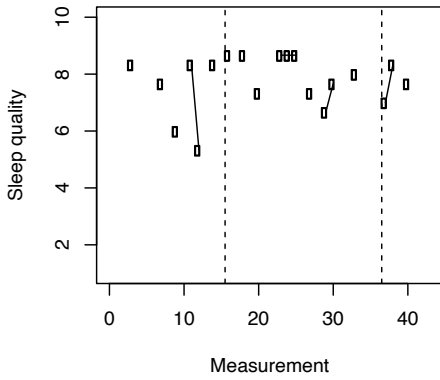
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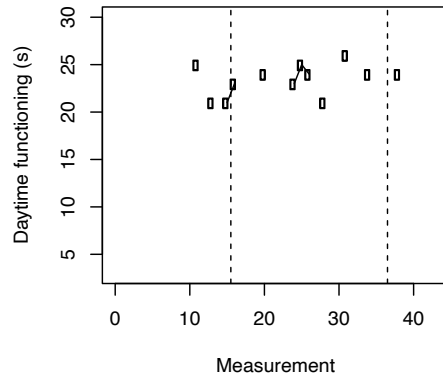
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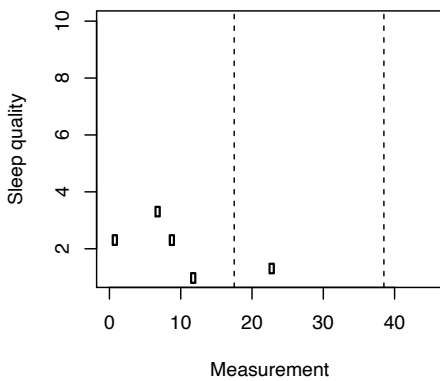
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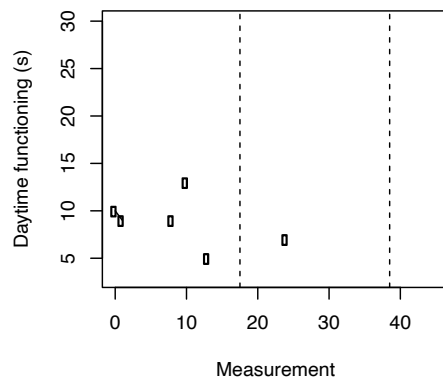
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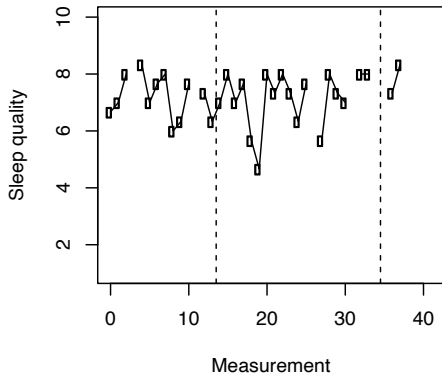
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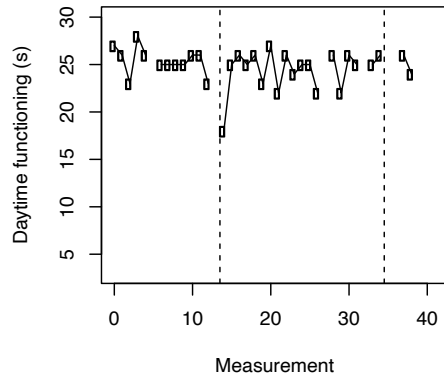
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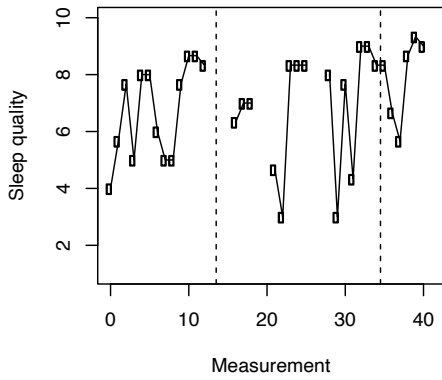
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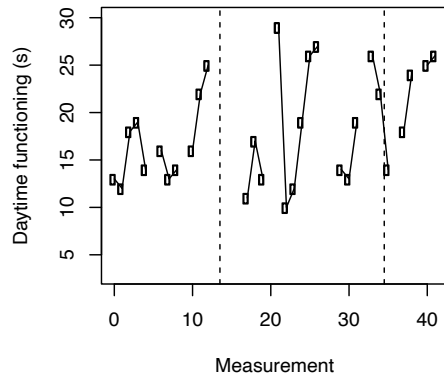
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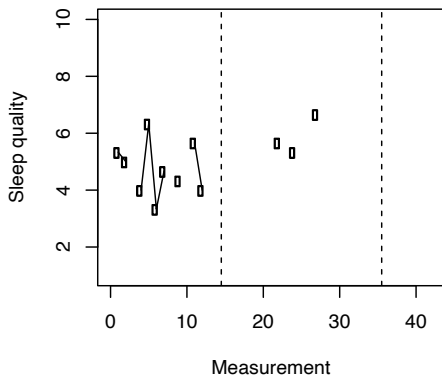
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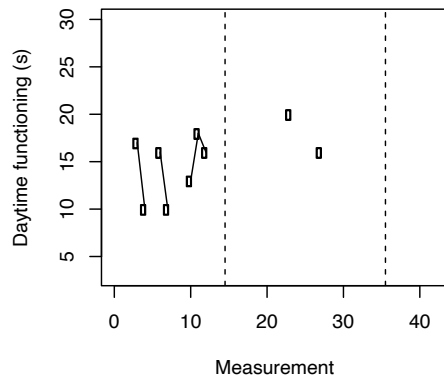
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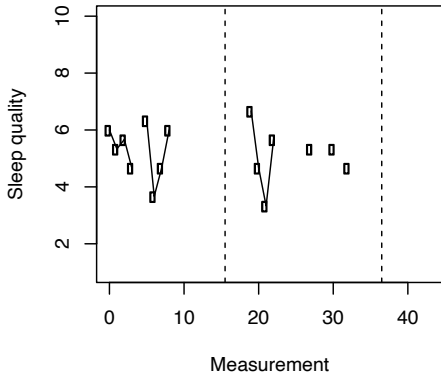
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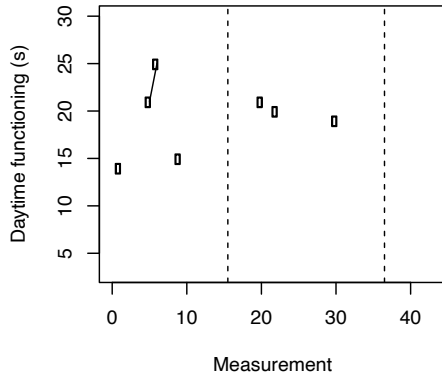
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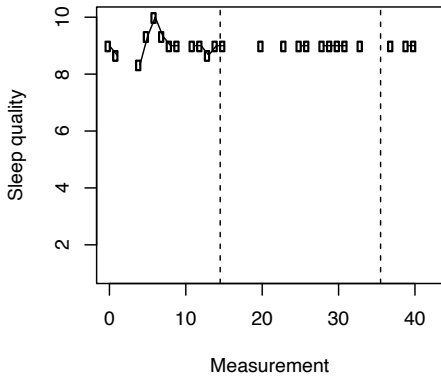
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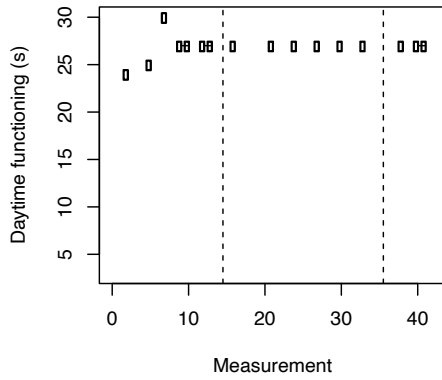
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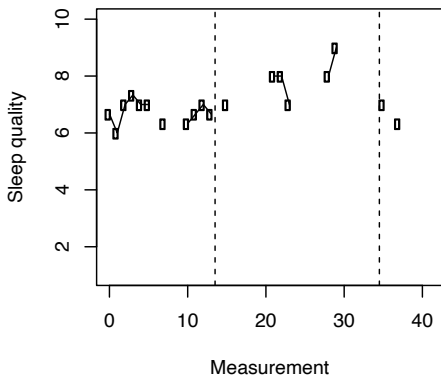
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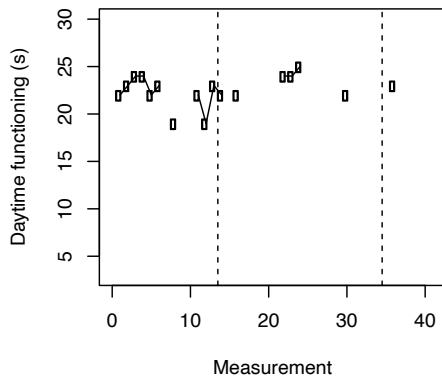
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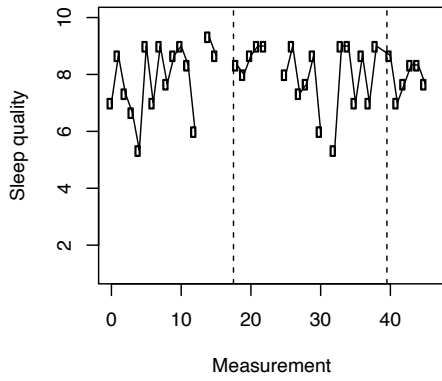
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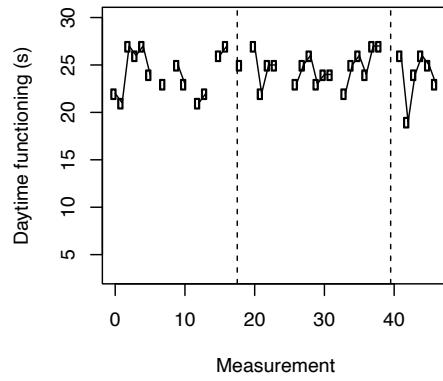
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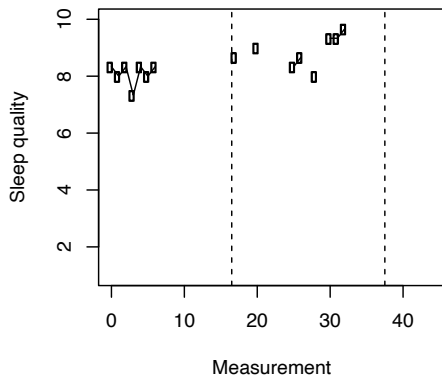
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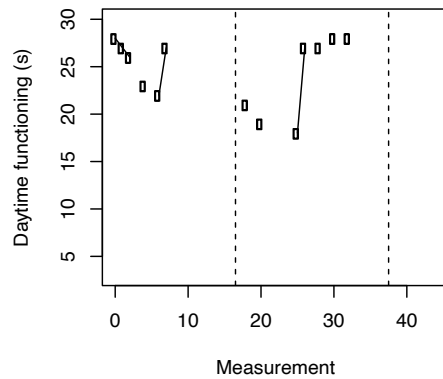
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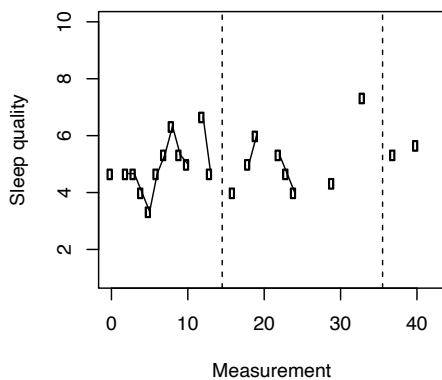
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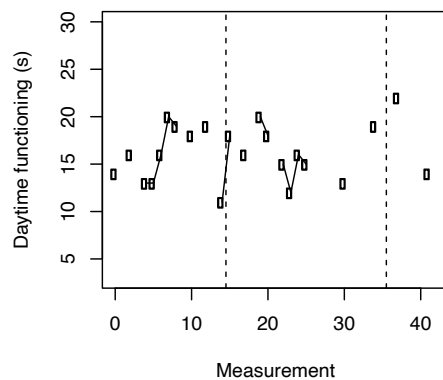
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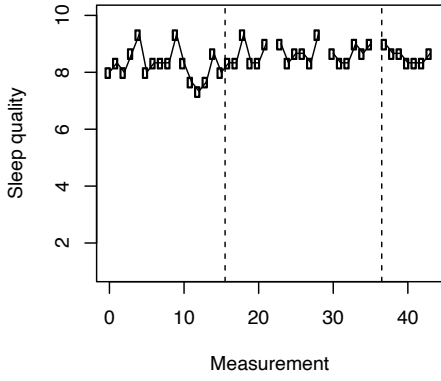
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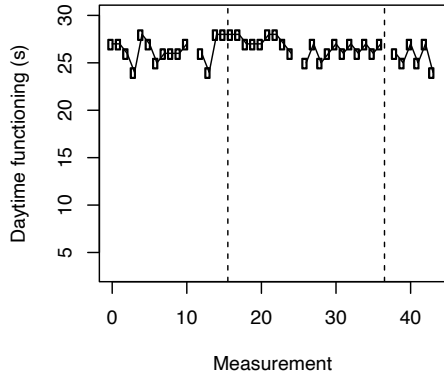
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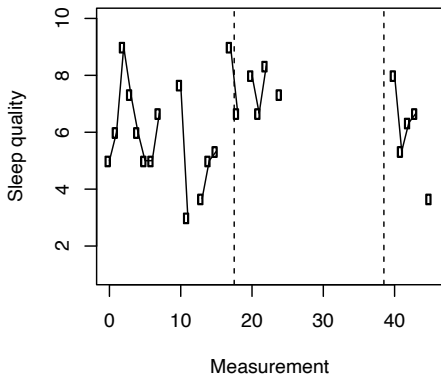
Participant 22



Participant 22



Participant 23



Participant 23

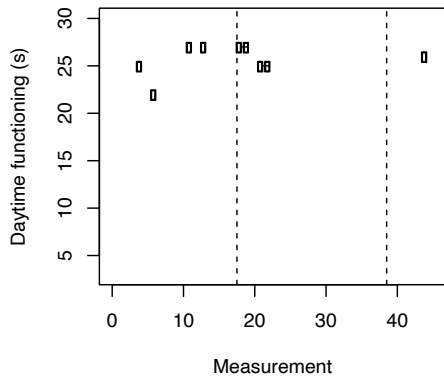


Table 5.2. Results for Sleep Quality and Self-Reported Daytime Functioning of the Multilevel Modeling Analyses

	b	SE	t	df	p
Sleep quality^a					
Intercept (baseline)	6.31	0.20	31.55	668.65	<.01
Intervention	0.25	0.24	1.02	895.22	.31
Follow-up	0.12	0.38	0.32	64.44	.75
Time centered	0.00	0.03	0.08	385.42	.93
Intervention * time centered	-0.00	0.05	-0.04	401.66	.97
Follow-up * time centered	0.00	0.11	0.04	43.92	.97
Daytime functioning (self-reported)^b					
Intercept (baseline)	20.23	0.78	25.38	19.66	<.01
Intervention	1.00	0.81	1.23	67.99	.22
Follow-up	0.38	1.22	0.31	19.64	.76
Time centered	-0.01	0.11	-0.05	111.40	.96
Intervention * time centered	-0.00	0.14	-0.03	289.09	.98
Follow-up * time centered	0.26	0.32	0.82	20.86	.42

^a possible range 1 - 10

^b possible range 3 - 30

5.4.3 Secondary outcome measures

Table 5.3 shows the reliable change index scores for the secondary outcome measures severity of sleep problems (ISI and CSHQ) and behavioral problems (SDQ). We found that seven of 15 participants showed clinically significant improvement on the ISI (severity of insomnia symptoms) from pretest to post-test, and ten of 19 participants showed clinically significant improvement on the ISI from pretest to follow-up. Teachers reported a clinically significantly greater number of behavioral problems, but this was not seen in the self- and parent-reports of behavioral problems.

The secondary outcome measures sleep onset latency, total sleep time, and wake time after sleep onset (self-reported and objectively measured) and daytime functioning reported by parents and teachers did not show significant changes (see supplementary material in Chapter 10.4 for descriptive data, visual analyses, and multilevel modeling analyses).

Table 5.3. Reliable Change Index Scores

	ISI			CSHQ			SDQ-Y			SDQ-P			SDQ-T		
	T1-T2	T1-T3	T1-T3	T1-T2	T1-T3	T1-T3	T1-T2	T1-T3	T1-T3	T1-T2	T1-T3	T1-T2	T1-T3	T1-T2	T1-T3
1	0.67	0.34		-1.05	-0.53	-0.92	-0.31	-0.92		-3.10	-2.79				
2		4.36			1.58					0.31					
3										-0.31					
4	1.68	1.01		0.79			0.31								
5		1.17			-0.26										
6	-1.34	0.00	1.46	0.26	-0.26	-0.31*	-0.31*	-0.31*		-1.24	-1.86				
7	2.01	1.68	0.32	-2.37	-0.53	0.61*	0.61*	0.61*		0.31	-0.31				
8		-1.34			-0.26					-1.55	-0.62				
9	1.68	1.34		0.26	1.84	1.53*	0.61*	1.53*		-0.62					
10	5.03	5.70	0.00	0.53	0.00	0.31*	1.22*	0.31*		0.31					
11	2.35	3.02		2.37	2.37	0.00*	1.22*	0.00*		-1.55	-2.17				
12	-0.34	-1.01	3.24	-1.05	-1.84		-0.92*			0.62					
13		1.68			-0.26					1.86	1.24				
14		0.00	-0.16	-0.26	1.32	0.31	0.00	0.31		-1.55	-1.55				
15	0.34			1.32			0.31*			-1.86	-1.86				
16	0.34	0.00	0.00	0.26	-0.26	0.31*	0.92*	0.31*		-3.41	-3.41				
17		5.03	0.65	0.53	0.53	-2.45	0.00*	-2.45		0.00	-2.17				
18	0.34		1.78	0.53	0.00		0.00*			-1.86*					
19	0.67	1.68	0.97	-0.26	0.53	3.36*	3.06*	3.36*		1.55	2.48				
20	2.01	2.01		1.05	0.79	1.83*	0.31*	1.83*		-0.31	0.00				
21										0.00	0.00				
22	3.02	2.35		0.00	-0.53					-0.93					
23	1.34	2.68	4.05	0.79*	1.05*	-0.92*		-0.92*		-1.24	-0.93				

T1 = pretest, T2 = post-test, T3 = follow-up

* scored above the behavioral problems cut-off score at pretest

RCI ≥ 1.65 indicates significant improvement

RCI ≤ -1.65 indicates significant decline

We exploratively analyzed grades in school (Dutch, English and math) using a one-way repeated measures ANOVA to investigate whether there was a change in the teenagers' grades at pretest, post-test, and follow-up. The results showed no significant effect (Dutch: Wilks' Lambda = .90, $F(2, 8) = .44$, $p = .66$, partial $\eta^2 = .10$; English: Wilks' Lambda = .79, $F(2, 8) = 1.08$, $p = .38$, partial $\eta^2 = .21$; Math: Wilks' Lambda = .97, $F(2, 8) = .12$, $p = .89$, partial $\eta^2 = .03$).

5.5 Discussion

In this study, we investigated the effect of a school-based sleep intervention on sleep quality and self-reported daytime functioning. We found that the intervention had no effect on the primary outcome measures (sleep quality and daytime functioning). However, we did find an effect on a secondary outcome measure about the severity of insomnia symptoms (the ISI questionnaire).

The ISI combines nighttime symptoms and daytime consequences, and it consists of items that measure symptoms of insomnia, its consequences, and the degree of distress caused by the sleep problem (Bastien et al., 2001). Although sleep diaries (as we used for our primary outcome measure sleep quality) are widely used in sleep research (Buysse et al., 2006), they do not measure the degree of impairment and distress triggered by the sleep problem (Bastien et al., 2001). This knowledge, in combination with our finding that the majority of the teenagers showed a clinically significant decrease in insomnia symptoms while showing no significant improvement in sleep quality, demonstrates that the ISI provides additional and relevant information about the perception of sleep problems.

At pretest, the ISI score was moderately elevated at the group and individual levels, while other sleep parameters were relatively normal. For example, the mean rates of sleep onset latencies and wake times after sleep onset at baseline were already close to the normal range of up to 30 minutes (Lichstein et al., 2003). This means that, in general, the teenagers in our study seemed to have, objectively, relatively mild sleep problems. This is not surprising since these were regular autistic teenagers rather than a clinical sample of teenagers with sleep disorders. Most studies that investigate the effectiveness of sleep interventions and find promising results include participants from a clinical population with a more severe starting point regarding sleep (e.g., de Bruin et al., 2015; McCrae et al., 2020; Papadopoulos et al., 2019). The relatively good starting point of our participants, in combination with the large observed ranges for our variables, made it less likely that we would see significant improvements in our primary outcome measures.

Although we seemed to include a population with mild sleep problems, the finding that the majority of the teenagers made clinically significant improvements on the ISI showed that a school-based sleep intervention could be effective for improving insomnia symptoms. Insomnia symptoms are associated with factors such as quality of life (Kyle et al., 2010) and anxiety and depressive symptoms

(Morin et al., 2011). This, combined with the knowledge that early treatment of sleep problems could prevent exacerbation of them (Morin & Benca, 2012), supports the importance of school-based sleep interventions that are easily accessible.

5.5.1 Strengths and limitations

An important strength of this study was that a different group of teachers delivered the intervention and completed the questionnaires. As a result, the teachers who delivered the intervention were not biased by the teenagers' behavior in class, which allowed them to approach the sleep consultation with open minds. This is especially important given that teachers reported more behavioral problems after the intervention. We were surprised that we did not see the increase in behavioral problems mentioned in the self- and parent reports, however, discrepancies between reports from different informants are common (Youngstrom et al., 2001). These discrepancies could arise because different contexts reveal a different presentation of teenagers' behavior, or because informants might have different perspectives on the same behavior (Achenbach, 2006).

A second strength of this study was its applied nature. Teachers delivered the sleep intervention without interference from researchers. This allowed us to investigate the intervention while approaching the educational practice as closely as possible. However, this also presented several challenges.

The first was that this study was conducted during the COVID-19 pandemic. When we began the study the spread of COVID-19 was low in the Netherlands, but infection rates rose as the study continued, resulting in many teenagers and teachers who were sick or in isolation. As infections increased, the Dutch government closed the schools. Although the pandemic seriously hindered the delivery of the intervention, teachers tried to continue as well as possible. Teenagers who were in isolation but not sick were encouraged to keep completing the sleep diary. Sleep consultations were also moved online. However, this did not solve all the issues, so some sleep consultations were rescheduled or missed. As a result, the number of missed consultations may be much larger than could be expected in a non-pandemic situation.

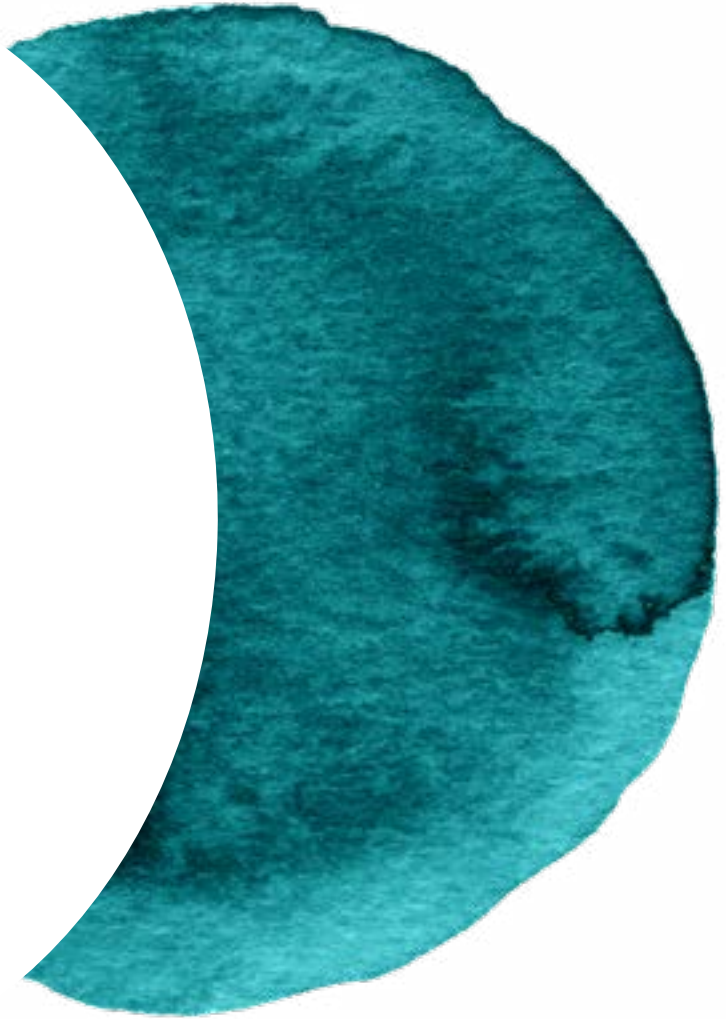
The second limitation is also related to the COVID-19 pandemic. After we completed this study, research revealed the impact of the COVID-19 pandemic lockdown on sleep patterns, with contrasting results. On the one hand, teenagers reported sleeping longer, having a better sleep quality and feeling less sleepy in the daytime during the pandemic (Gruber et al., 2020; Ramos Socarras et al., 2021). On the other hand, the stress they experienced during the pandemic has been related to sleep problems (Hyun et al., 2021). Since we took measurements across a wide range of months, we do not expect the pandemic to have had a major effect on this study. However, it could increase the observed ranges of variables, which could limit the likelihood of finding significant results.

Third, to ensure we could formulate proper sleep recommendations, we thought it was important that the teenagers completed the sleep diary at least three weekdays and one weekend day within each week for each measurement period. However, this was too difficult for some teenagers. We strived to collect sufficient data by sending automatic and personal reminders via the researcher or a teacher.

Fourth, we used the SRS-2 to identify the presence and severity of difficulties in social behavior on the autism spectrum. However, 18% of the participating teenagers did not score above the autism cut-off score. To determine whether the teenagers met our inclusion criteria of an autism spectrum disorder diagnosis, we asked parents if their teenager had such a clinical diagnosis and in which year and by which authority this diagnosis was established. All the participating parents declared their child had a formal autism spectrum disorder diagnosis.

5.5.2 Conclusion and future directions

This study shows that the co-created school-based sleep intervention has a positive effect on insomnia symptoms measured by the ISI, but not on sleep problems measured by the sleep diary and sleep tracker. We also found no effect of the sleep intervention on daytime functioning. Although the majority of the teenagers reported a decrease in insomnia symptoms, some showed no improvement. Future studies are needed to unravel the characteristics of the teenagers that benefit from the sleep intervention and to study the discrepancy between the results of the insomnia symptoms and other sleep parameters. This knowledge would allow us to focus on that specific group of autistic teenagers who would benefit from the school-based sleep intervention.



Chapter 6

General Discussion

This thesis project originates from a need of teachers at Dutch schools for special education for an evidence-based sleep intervention to adequately address sleep problems at school. To address this issue, the aim of this thesis was twofold: (i) to increase knowledge about the relationship between sleep and daytime functioning in autistic teenagers (Chapters 2 and 3), and (ii) to develop a school-based intervention to improve sleep in autistic teenagers and test its efficacy (Chapters 4 and 5). I would like to use this final chapter to summarize the main results, reflect on the strengths and limitations, and discuss the implications of the findings and suggestions for future research.

6.1 This thesis

We first focused on gaining more knowledge about the relationship between sleep and daytime functioning in autistic teenagers. Therefore, we did a systematic review on this topic (Chapter 2). We found that all included sleep variables were related to internalizing problems. In addition, we found that sleep problems in general and sleep efficiency had the most significant associations with daytime functioning. The systematic review also revealed several research gaps. First, few studies have focused specially on autistic teenagers. This is problematic because results of studies involving children or adults cannot automatically be applied to teenagers since sleep (Moore & Meltzer, 2008) and the nature of sleep problems (Goldman et al., 2011) change during adolescence. Second, few studies have related both objective measures and self-reported measures of sleep to daytime functioning. To disentangle the relationship between sleep variables and daytime functioning, it is recommended to include both objective measurements and self-reports of sleep.

That is why we conducted a second study (Chapter 3) in which we focused on the teenage age group and included both objective measures and self-reports of sleep. We used a network approach to identify the complex pattern of direct and indirect relationships between sleep (objectively measured and self-reported), factors influencing sleep, and aspects of daytime functioning. We used a repeated measures design with daily measurements, using multiple informants (self, parent, and teacher) of teenagers' daytime functioning. This study revealed that perceived sleep quality (as measured with a web-based sleep diary) was related to all aspects of daytime functioning for the teenage group. We also found associations between perceived sleep quality and daytime functioning in the parents' and teachers' networks, however, to a lesser extent. Other self-reported sleep variables were mostly indirectly related to daytime functioning through sleep quality. We also found that self-reports and objective measures of sleep were related to daytime functioning in different ways. As self-reported sleep had more (direct) associations with daytime functioning, this would be a logical target for a sleep intervention to increase the likelihood of improving daytime functioning.

Based on the results of Chapters 2 and 3 and the premise that school-based sleep interventions might be more easily accessible than interventions delivered in clinical practice, we developed a school-

based sleep intervention (Chapter 4). Previous school-based sleep interventions had been unsuccessful, although schools are willing to support their students in reducing sleep problems (Cassoff et al., 2013). Therefore, we co-created the sleep intervention with the intention to enhance the match with future users (Steen et al., 2011). We found that four factors (limited burden, sense of usefulness, clear guidance and structure, and support cues), with corresponding operationalizations, were important to include in a sleep intervention to benefit autistic teenagers.

In the final study, we investigated the efficacy of our co-created school-based sleep intervention (Chapter 5). It included both self-reports and objective measures of sleep: teenagers completed a daily web-based sleep diary and wore a sleep tracker overnight. The teenagers also answered questions about their daytime functioning, and a parent and a teacher of each teenager answered those same questions. Additionally, teenagers, parents, and teachers completed questionnaires about sleep problems (ISI; Bastien et al., 2001 and CSHQ; Waumans et al., 2010) and behavioral problems (SDQ; Goodman, 2001) at pretest, post-test, and follow-up.

Since we had learned that self-reported sleep – particularly perceived sleep quality – was important in relation to daytime functioning (Chapter 3), we used sleep quality and daytime functioning as our primary outcome measures. We found that the intervention had no effect on these measures. However, we did find an effect on the severity of insomnia symptoms (using the ISI questionnaire). The ISI is a measure that assesses both the symptoms of insomnia and the impact it has on daytime functioning (e.g., the consequences and distress caused by sleep problems). Sleep diaries, which were used as the primary outcome measure in this study to assess sleep quality, do not capture the extent of consequences and distress. However, the findings of this study, which showed a significant decrease in insomnia symptoms but no significant improvement in sleep quality, suggest the ISI may offer additional and relevant insights into the perception of sleep problems.

6.2 Strengths and limitations

One strength of this thesis was the development of a school-based sleep intervention for autistic teenagers in co-creation with the teenagers themselves, their parents, teachers, researchers, a somnologist, and software developers (Chapter 4). The process was guided by the Centre for eHealth & Wellbeing Research (CeHReS) roadmap, which provides practical guidelines for planning, coordinating, and implementing interventions through stakeholder participation. Throughout the development of the sleep intervention, we considered the needs and wishes of the teenagers, parents, and teachers and incorporated them through an iterative approach, ensuring that the intervention was both evidence-based and practical for implementation in schools (Parsons et al., 2013). By identifying potential barriers to implementation during the development phase and adjusting the intervention, we increased its feasibility and the chances of successful implementation in schools.

Moreover, we used different kinds of statistical techniques in this thesis which were relatively novel to this research field: network analysis (Chapter 3) and a multiple baseline design (Chapter 5). We used a network analysis to disentangle patterns between sleep, sleep hygiene, and daytime functioning in autistic teenagers. This approach allowed us to do justice to the complexity of interacting variables (Borsboom & Cramer, 2013) and gave us knowledge we could use for possible effective interventions. We used a multiple baseline design to assess the effectiveness of our school-based sleep intervention. This type of study can provide valuable insights into the effectiveness of an intervention at the individual level. In addition, a series of multiple baselines can be established to evaluate the efficacy of an intervention more broadly and provide evidence of its general effectiveness (Hawkins et al., 2007).

In addition, we used various measurements of sleep (different self-reports [sleep diary and questionnaire] and objective measures) and informants (teenagers, parents, teachers) to evaluate daytime functioning. These multiple measurement methods helped us draw conclusions about (i) the association between different measurements of sleep in relation to daytime functioning and (ii) the perspectives of different informants regarding the relationship between sleep and daytime functioning. The inclusion of multiple measurements methods and various informants contributed to the comprehensive picture of the relationships between sleep and daytime functioning (Chapter 3) and the effect of the intervention (Chapter 5) from different perspectives.

The studies described in Chapters 3, 4, and 5 were conducted in the actual context of the study samples: special education. Therefore, we have already experienced how the sleep intervention is delivered in that context, strengthening the relevance and generalizability of our results. Although our participants reflect the population at the participating schools for special education, we were not able to represent the entire autistic population.

This brings me to the first limitation: the sample size of part of our studies (Chapter 3) was limited. Moreover, to avoid heterogeneity because of great differences in impairments (Kanne et al., 2011), we excluded teenagers with intellectual disabilities. In addition, more boys than girls participated in our studies. Although there is a strong prevalence of ASD diagnosis among boys (Loomes et al., 2017), it is important to include a sufficient number of girls in such a study because of the potential sex-based differences in the relationship between sleep and daytime functioning (Saré & Smith, 2020). Thus, the small sample sizes, the exclusion of teenagers with intellectual disabilities, and the limited participation of girls mean that part of the autistic teenage population was not represented. As a result, the conclusions of this thesis cannot be generalized to the autistic youth population as a whole.

Another potential threat to external validity was the use of a self-selected sample throughout the studies in this thesis. The study participants were volunteers. Teachers at the participating schools promoted the studies with activities such as newsletters for parents, class visits, and introductory meetings with teenagers and parents. Our study participants might not reflect the general school populations because

of their dissatisfaction with their sleep and their intention to change their habits to improve their sleep. Nevertheless, the characteristics of our sample (dissatisfaction with sleep and willingness to change) also represent the individuals who are most likely to participate in a school-based sleep intervention in a non-research setting.

Furthermore, the studies described in Chapters 4 and 5 were (partly) conducted during the COVID-19 pandemic. This had some implications for the studies. As our study on the development of the school-based sleep intervention (Chapter 4) progressed, the COVID-19 pandemic began. Pandemic-related constraints forced schools to reorganize their teaching, which placed a greater emphasis on teaching and left less room for research. As a result, we had to temporarily postpone our research. When schools became available again, we resumed the development of the sleep intervention in a modified way. When our final study (Chapter 5) began, COVID-19 was spreading slowly in the Netherlands and there were no restrictions that would hinder the study. However, infection rates rose as the study continued and the Dutch government finally closed the schools.

Although the pandemic seriously hindered the delivery of the intervention, teachers tried to continue online as well as possible. However, this did not resolve all the issues and more consultations may have been missed than would be expected in a non-pandemic situation. Moreover, research has revealed the effects of pandemic-related lockdowns on sleep patterns, with contrasting results. On the one hand, during the pandemic, teenagers reported sleeping longer, having a better sleep quality, and feeling less sleepy in the daytime (Gruber et al., 2020; Ramos Socarras et al., 2021). On the other hand, the stress teenagers experienced during the pandemic has been related to sleep problems (Hyun et al., 2021). Since we took measurements across a wide range of months, we do not expect the pandemic to have had a major effect on this study. However, it could have increased the observed ranges of variables, which could limit the likelihood of finding significant results.

6.3 Implications

6.3.1 Importance of addressing sleep problems at school

This thesis showed that teachers are able to deliver a sleep intervention that reduces insomnia symptoms in autistic teenagers. Although we seemed to include a population with mild sleep problems, the finding that the majority of the teenagers experienced a clinically significant reduction in insomnia symptoms showed that a school-based sleep intervention could be effective in reducing insomnia symptoms. This is important because the school-based sleep intervention might be viewed as an initial step in a stepped-care approach and early treatment of sleep problems could prevent their exacerbation (Morin & Benca, 2012). Those teenagers who did not benefit enough from this intervention may require more specialized sleep treatment, such as cognitive behavioral therapy for insomnia (CBTI).

Additionally, implementing a sleep intervention in schools could increase teachers' awareness of the importance of sleep and the consequences of sleep problems. This increased awareness could lead to earlier intervention and potentially reduce the duration of sleep problems and associated consequences in teenagers. Therefore, it is important that teachers be able to recognize signs of sleep problems in their students and deliver an intervention so they can offer support when needed.

However, teachers are teachers and not clinicians. There are limits to what they can do to reduce teenagers' sleep problems. Teachers need guidance to understand for whom the school-based sleep intervention could be suitable, so I will now explain some factors that might define that group.

First, the teenagers should intend to change their sleep habits. This intention to change is affected by the teenagers' attitudes, subjective norms, and perceived behavioral control (McEachan et al., 2016). For example, it is known that sleep timing interventions are effective in reducing problems with sleep-wake patterns (Johnson & Zarrinegar, 2021), and a change in sleep timing is more likely to occur when the behavioral change is important to the teenager (attitude) and others (subjective norm). In addition, teenagers need to feel they are able to change their behavior (perceived behavioral control) (McEachan et al., 2016). Therefore, it is important to explain what participating in the sleep intervention entails and to pay attention to what is expected from the participants. The teenagers should be in control of their decision to participate. However, since the behavioral change should also be important to others (subjective norms), the teenager's family needs to be supportive.

Second, we excluded teenagers with interacting or multiple problems, such as depression, anxiety disorder, substance dependence, or game addiction. Although we trained teachers to deliver the sleep intervention, they do not have sufficient education to deliver the intervention to teenagers with multiple and interacting problems. It is important to recognize that teenagers with complex needs can be challenging and may require additional support. One should also be aware that interacting or multiple problems could emerge while delivering the sleep intervention. When teachers notice other problems, they should be addressed to gain further understanding. If necessary, the teenagers should be referred to clinical care for appropriate treatment.

Taken together, it is important that teachers be aware of the importance of sleep and of their limitations with regard to implementing the sleep intervention. Since teachers generally are not equipped to support teenagers in reducing their sleep problems, a dedicated teacher training is necessary to provide knowledge and skills.

Our studies do not clarify which elements of the sleep intervention contribute to its positive effect on insomnia symptoms. Moreover, the studies were not designed to determine why certain teenagers benefited from the sleep intervention while others did not. To obtain a better understanding of the effective elements, future research should identify the mechanisms and circumstances that explain

when, why, how, and for whom the sleep intervention has a positive effect on sleep. This will guide teachers further by suggesting which elements under which circumstances are most promising to improve sleep.

6.3.2 Implementation of school-based interventions

We co-created a school-based sleep intervention with the intention of getting a better understanding of the needs, wishes, and circumstances of future users and addressing key implementation issues upfront. Additionally, this approach demonstrates respect for the expertise and experience of teenagers, parents, and teachers, which can help us understand how interventions can be effectively carried out in varied and complex school settings (Parsons et al., 2013). Co-creating interventions can increase the probability of successful implementation (Robertson & Simonsen, 2012). However, one can argue whether this justifies the participants' substantial time investment, especially in an educational setting where workloads are high (den Brok et al., 2017).

We experienced the benefits of this approach. It helped us notice barriers that might hinder the use of the intervention during its development. For example, early in the development process, future users had already given us important information about contextual factors which we used for further development. In general, co-creation could be an attempt to narrow the gap between research and practice (Parsons et al., 2013).

However, there is no guarantee that co-created interventions will be implemented more successfully than other interventions (van Gemert-Pijnen et al., 2011). At the end of the study, we encountered differences in the extent to which schools adopted the sleep intervention. Therefore, it would be interesting to elaborate more on the aspects that contribute to the adoption of the intervention.

School administrations play an important role and should be involved in implementation (Damschroder et al., 2009). For example, they have to formulate a long-term vision for the intervention and allocate associated resources to implement it properly. A successful implementation requires one to see the implementation process as part of the development process (van Gemert-Pijnen et al., 2011). We saw that the schools that did so adopted the sleep intervention more easily after the research project ended. Moreover, research on the adoption of school-based mental health interventions has shown the promise of coaching and facilitation for adopting school-based interventions (Beidas et al., 2012).

Taken together, this shows us that developing an intervention that matches the users' needs, wishes, and contexts is promising, but it not enough to ensure a successful implementation. The school administration should also embrace the intervention and express its importance. Moreover, they should encourage, support, and facilitate their teachers.

6.3.3 Impact of sleep measurements on conclusions

In our studies, we combined self-reported and objective measures of sleep. In Chapter 3, we used both kinds of sleep measures to investigate the relationships between sleep and daytime functioning. We mainly found relationships from the self-reported sleep measures, the most important of which was perceived sleep quality. In Chapter 5, we used self-reported and objective sleep measures to investigate the effect of the intervention. We found that it had a significant effect on the severity of insomnia symptoms, which was also a self-reported measure. However, we also discovered differences among the various self-reported sleep measures (e.g., sleep diary and questionnaire). Although our studies demonstrated the value of self-reported sleep measures and not of objective measures, it is premature to discount the usefulness of objective measures since other studies have found that sleep interventions have significant effects on objective measures and other self-reported sleep measures (e.g., de Bruin et al., 2014; McCrae et al., 2020).

Many consumer-targeted wearable devices can be used to objectively monitor sleep, and they have become quite popular in recent years (Khosla et al., 2018). These sleep trackers have advantages, such as raising awareness about sleep patterns, being user-friendly, and providing a low-cost and unobtrusive way to collect sleep data. However, it is important to be aware of their limitations and drawbacks. One potential drawback is developing a preoccupation with having perfect sleep, known as orthosomnia (Aupetit et al., 2019). An increasing number of individuals are seeking treatment for sleep disturbances they have self-diagnosed using a sleep tracker, and some people rely more on the sleep tracker than on the consulted sleep expert (Baron et al., 2018). In addition, some studies have raised concerns about the validity of these devices (Baron et al., 2018; Scott et al., 2020; Tobin et al., 2021). It is important to consider that some teenagers participating in the sleep intervention may also use wearable devices to track their sleep, and the limitations of such devices should be explained to them (Khosla et al., 2018). Formal research should use validated sleep trackers to ensure the accuracy and reliability of the collected data. Thus, given the numerous advantages of consumer-targeted wearable devices, more research is needed to determine and improve their accuracy.

6.3.4 Other sleep interventions and generalizability

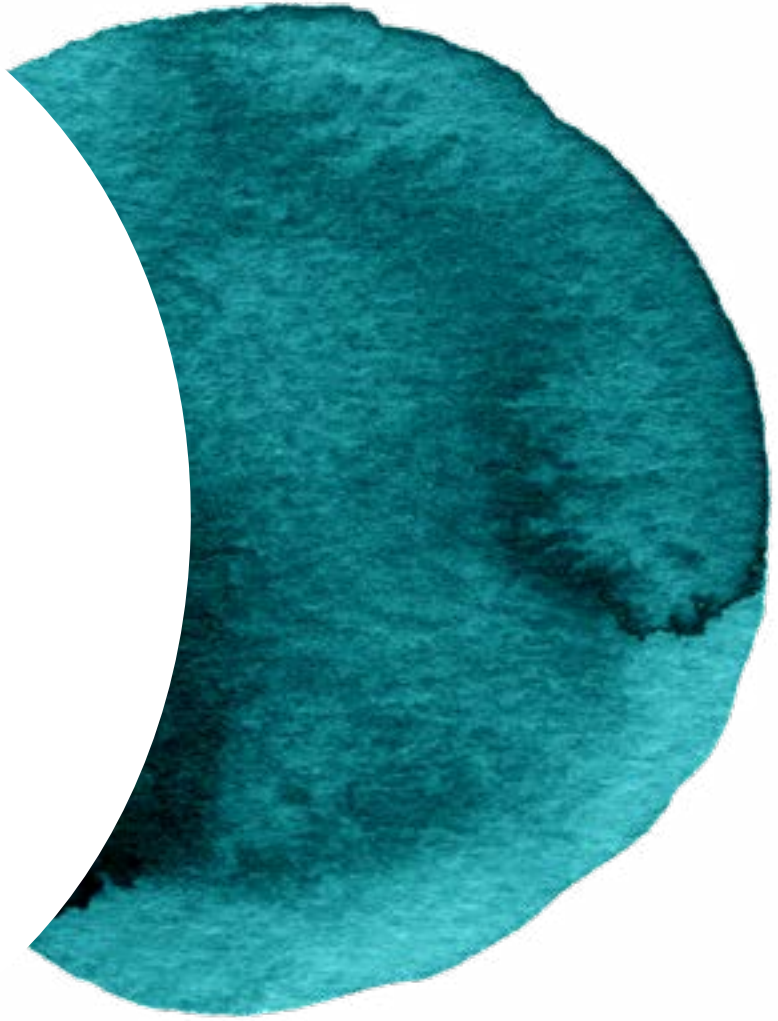
There is a range of possible sleep interventions for autistic children and teenagers, such as pharmacologic and behavioral interventions (Johnson & Zarrinegar, 2021; Keogh et al., 2019). In general, the initial approach to reducing sleep problems is beginning a behavioral sleep intervention (Crowe & Salt, 2015). Those sleep interventions could follow different approaches: (i) one-size-fits-all, where the sleep intervention is the same for everyone regardless of the sleep problem (e.g., de Bruin et al., 2014); (ii) a stepped-care approach where a fixed first step is followed by an evaluation that serves to determine additional steps tailored to the specific sleep problem (e.g., Papadopoulos et al., 2022); or (iii) a modular approach where the sleep intervention is focused from the beginning on each teenager's specific sleep problem, which means the sleep intervention could be different for every individual (e.g., Harvey et al., 2008).

Although most sleep interventions seem to follow a stepped-care approach, we developed an intervention based on a modular approach because, in the co-creation sessions, future users stressed the importance of a tailored approach. This aligns with research conducted by Pavlopoulou (2021), who emphasized that sleep interventions for individuals with autism should be tailored to their specific environmental and personal factors. For example, it is important to personalize general sleep recommendations to fit personal preferences, since factors that facilitate sleep in autistic teenagers could differ from generalized sleep recommendations.

Another important component we included in the intervention is the contribution teenagers made to the sleep recommendations, following the principles of motivational interviewing (Miller & Rollnick, 2009). This method for achieving behavior change in teenagers emphasizes collaboration between the therapist and client, evokes the client's own motivations for change, and respects the client's autonomy. It is a proven technique for working with teenagers (Martins & McNeil, 2009). Thus, instead of telling the teenagers what they should do to improve their sleep, the teacher and teenager discuss the sleep recommendations following the principles of motivational interviewing. The teenager can express their thoughts and preferences, and the teacher and teenager work together to come up with a plan the teenager is willing to follow. This collaborative approach ensures that the sleep recommendations are tailored to the teenager's needs and are more likely to be more successful.

It would be interesting to examine whether this sleep intervention is also suitable for other populations, such as teenagers with Attention Deficit Hyperactivity Disorder (ADHD) who also report high rates of sleep problems (Cortese et al., 2013) or typically developing teenagers. Although school-based sleep interventions for typically developing teenagers do exist, they do not follow tailored and modular approaches (Cassoff et al., 2013). We developed this sleep intervention with and for autistic teenagers. However, its starting point is everyone's own sleep pattern. As our sleep recommendations are not specific to autism, they might be helpful for other teenagers with sleep problems. The first step should be to investigate whether this intervention is also suitable for other populations by investigating their needs, wishes, and contextual circumstances. The second step should be to investigate the efficacy of the school-based sleep intervention. If the intervention is shown to be effective for other groups, it can be applied to a larger population. This means more teenagers with sleep problems could receive guidance, which is important because addressing sleep problems early can prevent them from becoming more severe.

"The prevalence of sleep problems among autistic teenagers was not previously recognized at school. To address this, we now ask how the teenagers sleep and continue to deliver the sleep intervention. Additionally, we offer workshops to educate colleagues on the importance of adequate sleep for this population. Raising awareness about the significance of sleep is crucial." (Participating teacher after the intervention)



Chapter 7

Summary

Sleep problems are common among teenagers with an autism spectrum disorder diagnosis. However, there is only fragmented knowledge about how sleep problems affect those teenagers' lives. Moreover, interventions to improve sleep are mainly delivered in clinical practice, while school-based sleep interventions could be more easily accessible for teenagers. The studies in this thesis focus on increasing knowledge about the relationship between sleep and daytime functioning in autistic teenagers (Chapters 2 and 3), developing a school-based sleep intervention (Chapter 4), and investigating its efficacy (Chapter 5).

In **Chapter 1**, we give an overview of autistic teenagers' sleep and interventions to improve sleep, and we introduce the studies in this thesis. We know sleep problems are common among autistic teenagers (up to 88%; Chapter 2) and these sleep problems are related to daytime functioning (Chapters 2 and 3). We also know that the informant and measurement method one uses can affect the relationship one finds between sleep and daytime functioning (Chapters 2, 3, and 5). Teachers can successfully deliver a co-created sleep intervention (Chapter 4), which reduces the insomnia symptoms of autistic teenagers (Chapter 5).

Sleep problems are much more prevalent among autistic teenagers than typically developing teenagers. Autistic teenagers have more difficulties initiating and maintaining sleep, a shorter sleep duration, and a lower sleep efficiency. The underlying cause of their sleep problems is not yet fully clear. It seems to be associated with several factors related to an ASD diagnosis (e.g., biological abnormalities, medical or psychological co-occurring conditions, and use of medication), but more general adolescence-related factors (e.g., changing sleep patterns with later bedtimes and rise times) may further interfere with sleep.

The first steps to reduce sleep problems are behavioral sleep interventions (e.g., creating awareness about the importance of sleep and sleep hygiene, faded bedtime and sleep restriction, sleep timing interventions). To determine which intervention may be the most successful in improving both sleep and daytime functioning, we need to know more about the relationship between specific sleep parameters and the impact on autistic teenagers' lives.

In addition, behavioral sleep interventions are mostly delivered in clinical practice. Sleep interventions that could be delivered at schools would be more easily accessible for school-aged youth and, therefore, have a greater reach. Although we have seen that schools are willing to support their students in reducing sleep problems, the implementation of sleep interventions in schools remains unsuccessful because of the lack of a tailored approach to sleep problems. Developing interventions in co-creation could solve that problem because it involves future users in early stages of the development process, allowing them to express their needs and wishes.

In **Chapter 2**, we present the results of a systematic literature review following the PRISMA guidelines (Moher et al., 2009). It answered the research question: What is the relationship between sleep (sleep

problems in general, total sleep time, sleep onset latency, sleep efficiency, and sleep timing) and daytime functioning (externalizing behavior, internalizing behavior, autism symptom severity, social behavior, attention, daytime sleepiness/fatigue) in autistic teenagers?

We performed an electronic database search (APA PsycINFO, Cochrane, ERIC, PubMed, Web of Science) and a hand search (INSAR congress archive and reference lists of included studies) in September 2020 that identified 2561 studies. Studies were included if they contained autistic participants aged 10 – 19 years with no intellectual disability, were related sleep to daytime functioning, were available in English, and used original data. Two reviewers evaluated the quality of each study and the first author extracted data following predefined guidelines. Ultimately, nine studies were included ($N_{\text{participants}} = 674$).

The most prominent finding was the association of all sleep parameters with internalizing problems. We also found that sleep problems in general and sleep efficiency (percentage of total time in bed actually spent asleep) had the most significant associations with daytime functioning. The results provide input for tailored sleep interventions. However, more research is needed to get a clearer picture of the relationship between specific sleep parameters and daytime functioning to ensure that this knowledge will provide better input for sleep interventions. It is important to focus on limited age ranges, larger samples sizes, multiple measurement methods, and longitudinal approaches.

In **Chapter 3**, we describe the network approach we used to disentangle patterns between sleep, sleep hygiene, and daytime functioning. Over a three-week period, 31 autistic teenagers ($M_{\text{age}} = 14.87$, $SD = 1.88$, range = 12 - 18) answered questions about sleep every morning and questions about daytime functioning at the end of every day. Sleep tracker data were collected from 14 of the teenagers. The results showed that perceived sleep quality seemed to be the most important sleep variable in relation to daytime functioning (self/parent/teacher reports). We also found that sleep onset latency, total sleep time, and wake time after sleep onset were related to daytime functioning, but mostly indirectly through sleep quality.

We also showed how the network approach gave us insights into the interplay between specific sleep variables, aspects of sleep hygiene, and daytime functioning. When developing sleep interventions, it is useful to know that sleep quality is the most important aspect of sleep for daytime functioning because perceived sleep quality would be a logical target for increasing the likelihood of actually improving daytime functioning.

In **Chapter 4**, we describe the co-creation in an educational setting of a school-based sleep intervention for autistic teenagers. We focused on the first three phases of the Centre for eHealth & Wellbeing Research (CeHReS) roadmap: contextual inquiry, value specification, and design. Each phase had specific goals and activities that led to iterative development with the active involvement of relevant stakeholders (autistic teenagers, their parents and teachers, and professionals). These stakeholders indicated that four factors

with corresponding operationalizations were important to include in the sleep intervention: limited burden, sense of usefulness, clear guidance and structure, and support cues. The iterative approach with an emphasis on stakeholder participation enabled us to notice possible barriers to implementing the intervention during its development. We could then identify those barriers and apply lessons to adapt the sleep intervention during development, which will likely increase successful implementation in schools.

In **Chapter 5**, we present the results of our study on the school-based intervention's efficacy in improving sleep quality and daytime functioning. We used a multiple baseline design with 23 teenagers ($M_{\text{age}} = 14.8$, $SD = 1.7$, range = 12 - 18 years). The teenagers completed a daily web-based sleep diary and wore a sleep tracker overnight. They also answered questions about their daytime functioning, which were also answered by a parent and a teacher. Additionally, the teenagers, parents, and teachers completed questionnaires (about sleep problems [ISI and CSHQ] and behavioral problems [SDQ]) at pretest, post-test, and follow-up. We found that the intervention had no significant effect on sleep quality or daytime functioning. However, the results of the self-reported sleep problems questionnaire (ISI) show that the majority of the teenagers made clinically significant improvement. Although our study population had mild sleep problems, which offers little room for improvement, this result suggests that the school-based sleep intervention has a positive effect on the severity of insomnia symptoms.

In **Chapter 6**, I summarize the main findings of the studies in this thesis. These findings emphasize the importance of addressing sleep problems in special education. It is important that teachers be aware that they can contribute to reducing their students' sleep problems. Delivering a school-based sleep intervention might be viewed as an initial step in a stepped-care approach, which is valuable because early treatment of sleep problems could prevent their exacerbation. Therefore, it is important for teachers to be able to recognize signs of sleep problems and deliver an intervention so they can offer support when needed. It also would be interesting to examine whether this sleep intervention is suitable for other populations who report high rates of sleep problems.

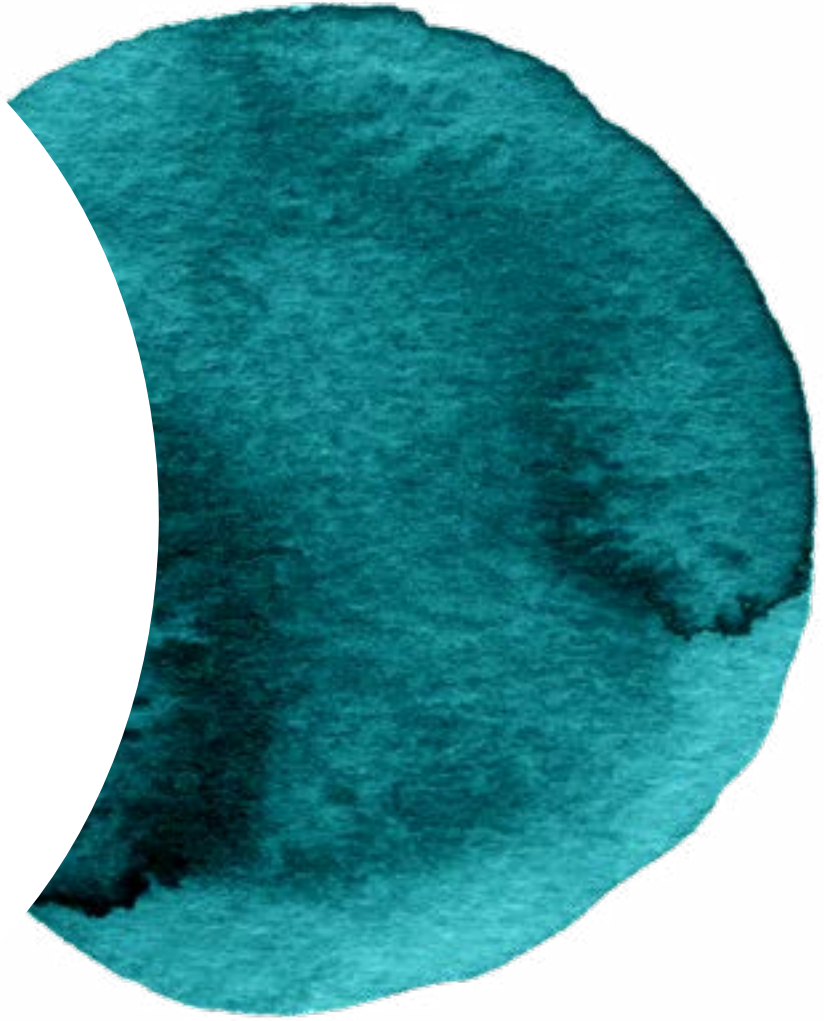
However, it is important to realize that teachers are not clinicians. There are limits to what they can do to reduce teenagers' sleep problems. Teachers need guidance to identify those teenagers for whom the school-based sleep intervention may be suitable. It is also important to recognize that teenagers with complex needs can be a challenge and may require additional support. Since teachers are generally not equipped to support teenagers in reducing their sleep problems, dedicated teacher training is necessary to provide knowledge and skills.

Furthermore, it is important that school administrators embrace and promote the use of the sleep intervention. School administrators play an important role in the successful implementation of educational innovations by encouraging, supporting, and facilitating their teachers.

For further research, it is important to determine the effective elements of the sleep intervention and identify the mechanisms and circumstances that explain when, why, how, and for whom the sleep intervention has a positive effect on sleep. This will give teachers further guidance on which elements under which conditions are most promising for improving their students' sleep.

It is also useful to continue to include self-reported and objective measures of sleep in future research. Although our studies have shown the value of self-reported sleep measures and not objective measures, it is premature to discount the usefulness of objective measures because other studies have used them to find significant effects of sleep interventions.

Further research can also provide more insight into the value of different sleep measures. In this regard, it is important to use validated sleep trackers to ensure the accuracy and reliability of collected data. Many consumer-targeted wearable devices can be used to measure sleep, and they have become quite popular in recent years. However, their accuracy and reliability is still a concern. Given the benefits of such devices (e.g., user-friendly, inexpensive, unobtrusive), more research is needed to determine and improve their accuracy.



Chapter 8

Samenvatting (summary in Dutch)

Slaapproblemen komen vaak voor bij jongeren met autisme. Echter, de kennis over hoe de slaapproblemen het leven van deze jongeren beïnvloedt is versnipperd. Daarnaast worden slaapinterventies voornamelijk aangeboden binnen de gezondheidszorg, waar helaas vaak wachtlijsten zijn. Wanneer we een interventie kunnen ontwikkelen die door docenten op school kan worden ingezet is een slaapinterventie mogelijk toegankelijker voor jongeren en heeft deze daardoor een groter bereik hebben. De studies in dit proefschrift richten zich op het vergroten van de kennis over de relaties tussen slaap en dagelijks functioneren bij jongeren met autisme (hoofdstuk 2 en 3) en de ontwikkeling (hoofdstuk 4) en toetsing (hoofdstuk 5) van een slaapinterventie die door docenten op school ingezet kan worden.

Hoofdstuk 1 geeft een overzicht van slaap bij jongeren met autisme, interventies om slaap te verbeteren en introduceert de studies in dit proefschrift. Slaapproblemen komen bij jongeren met autisme vaker voor dan bij niet-autistische jongeren. Jongeren met autisme hebben meer moeite met in- en doorslapen, hebben een kortere slaapduur en lagere slaapefficiëntie (percentage van de totale tijd in bed dat je daadwerkelijk slaapt). De onderliggende oorzaak van de slaapproblemen is nog niet helemaal opgehelderd. Het lijkt gerelateerd te zijn aan verschillende oorzaken die te maken hebben met het autisme (bv. biologische afwijkingen, medische of psychologische comorbiditeit of medicijngebruik), maar ook aan oorzaken die te maken hebben met de adolescentie (bv. veranderende slaappatronen). De eerste stap om slaapproblemen te verminderen zijn gedragsinterventies, bv. bewustzijn creëren over het belang van slaap en slaaphygiëne. Om te bepalen welke interventie het meest effectief is voor zowel het verbeteren van slaap als het verbeteren van dagelijks functioneren is meer kennis nodig over de relatie tussen specifieke slaapkenmerken en de impact daarvan op dagelijks functioneren van jongeren met autisme. Daarnaast worden gedragsinterventies vaak aangeboden binnen de gezondheidszorg. Slaapinterventies die ingezet kunnen worden op scholen zijn mogelijk toegankelijker voor schoolgaande jongeren en hebben daardoor een groter bereik. Hoewel scholen bereid zijn hun leerlingen te ondersteunen bij het verminderen van hun slaapproblemen, blijkt de implementatie van slaapinterventies op scholen weinig succesvol. Vooral door het gebrek aan een gepersonaliseerde aanpak. Het ontwikkelen van interventies in co-creatie kan dat probleem mogelijk oplossen door in een vroeg stadium toekomstige gebruikers te betrekken bij het ontwikkelproces.

In **hoofdstuk 2** beschrijven we de resultaten van een systematisch literatuuronderzoek, uitgevoerd volgens de PRISMA-richtlijnen, met als onderzoeksvraag: Wat is de relatie tussen slaap (slaapduur, inslaapduur, slaapefficiëntie, slaappatroon, slaapproblemen in het algemeen) en dagelijks functioneren (externaliserend gedrag, internaliserend gedrag, autismesymptomen, sociaal gedrag, geheugen, concentratie, vermoeidheid) bij jongeren met autisme? In september 2020 werd een elektronische database zoekopdracht uitgevoerd (in APA PsycINFO, Cochrane, ERIC, PubMed, Web of Science) en handmatig gezocht in het INSAR-congresarchief en referentielijsten van geïncludeerde studies. Dit resulteerde in 2561 studies. We includeerde studies als de deelnemers tussen de 10 en 19 jaar zijn en geen verstandelijke beperking hebben, slaap wordt gerelateerd aan dagelijks functioneren, beschikbaar

is in het Engels en empirisch onderzoek bevat. Elke studie werd beoordeeld op kwaliteit door twee onafhankelijke beoordelaars. In totaal werden negen studies geïncludeerd ($N_{\text{deelnemers}} = 674$). De relaties van alle slaapvariabelen met internaliserend gedrag was de meest opvallende bevinding. Verder werden voor meerdere aspecten van dagelijks functioneren significante relaties gevonden met slaapproblemen in het algemeen en slaapefficiëntie. Deze resultaten leveren belangrijke informatie voor slaapinterventies. Om slaapinterventies nog beter vorm te geven is echter meer onderzoek nodig naar de relatie tussen specifieke slaapvariabelen en dagelijks functioneren. In vervolgonderzoek is het belangrijk te focussen op specifieke leeftijdsgroepen, grotere steekproeven te betrekken, meerdere meetmethoden van slaap slaapefficiëntie te combineren en longitudinaal onderzoek uit te voeren. In de vervolgstudie (hoofdstuk 3) hebben we daarom drie weken lang slaap (zelf-gerapporteerd en objectief gemeten) en dagelijks functioneren gemeten bij jongeren in de leeftijd van 12 tot 20 jaar. Op deze manier hebben we de relatie tussen slaap en dagelijks functioneren verder onderzocht.

In **hoofdstuk 3** hebben we een netwerkanalyse uitgevoerd om de relatie tussen slaap en dagelijks functioneren bij jongeren met autisme te onderzoeken. Drie weken lang vulden 31 jongeren ($M_{\text{leeftijd}} = 14.87$, $SD = 1.88$, range = 12 - 18) dagelijks een digitaal slaapdagboek in. Aan het eind van de dag vulden de jongeren vragen in over hun dagelijks functioneren (vermoeidheid overdag, concentratie, humeur). De vragen over het dagelijks functioneren werden ook ingevuld door een ouder en docent van de jongeren. Daarnaast droegen 14 van de 31 jongeren 's nachts een *sleeptracker*. De resultaten lieten zien dat de zelf-gerapporteerde slaapkwaliteit (slaapdagboek) de belangrijkste slaapvariabele was in relatie tot dagelijks functioneren. Daarnaast vonden we dat de andere slaapvariabelen (inslaaptijd, slaapduur, en tijd wakker 's nachts) in de meeste gevallen indirect, via slaapkwaliteit, gerelateerd zijn aan dagelijks functioneren. In dit onderzoek hebben we door het in kaart brengen van ook indirecte relaties laten zien hoe een netwerkanalyse inzicht geeft in het samenspel van specifieke slaapvariabelen, slaaphygiëne en dagelijks functioneren. De kennis dat slaapkwaliteit de belangrijkste slaapvariabele was in relatie tot het dagelijks functioneren levert relevante informatie op voor de ontwikkeling van slaapinterventies. Door slaapinterventies te richten op het verbeteren van de ervaren slaapkwaliteit hebben deze waarschijnlijk het meeste effect.

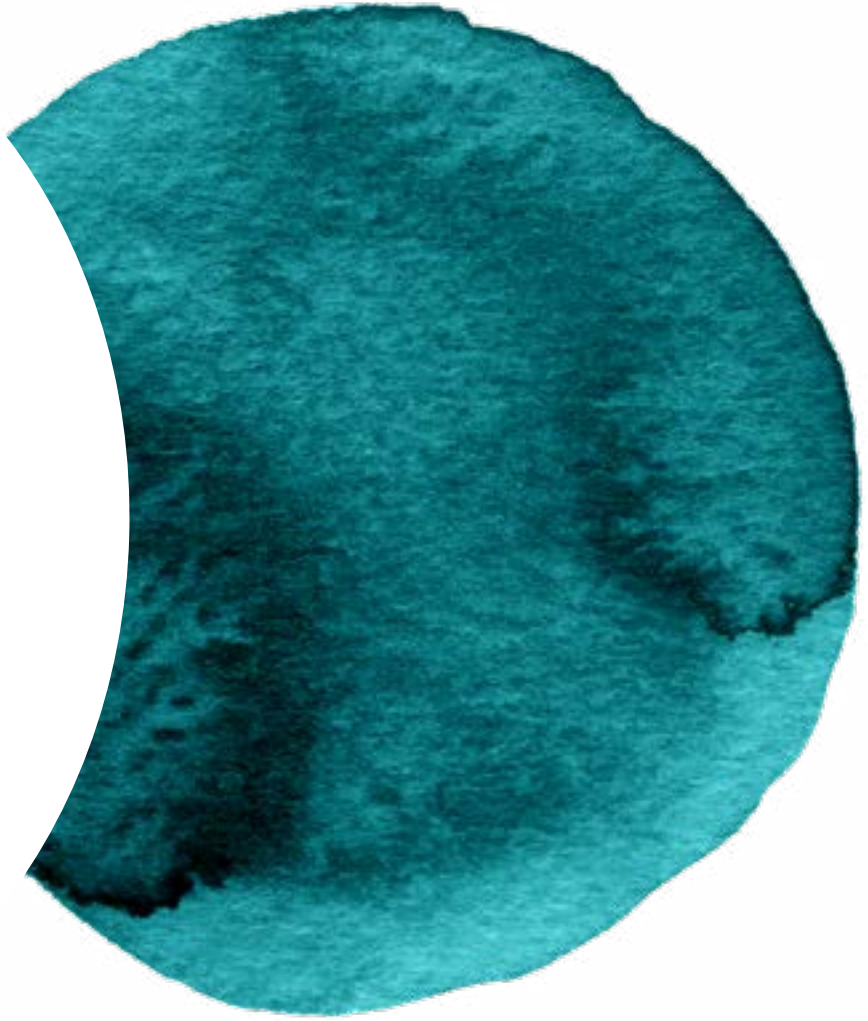
Hoofdstuk 4 is de beschrijving van het in co-creatie ontwikkelen van een slaapinterventie voor jongeren met autisme die kan worden ingezet op scholen. We hebben hiervoor gebruik gemaakt van de eerste drie fasen van de CeHReS roadmap: contextonderzoek, waardenspecificatie en ontwerp. Elke fase had specifieke doelen en bijbehorende activiteiten die leidden tot een iteratieve ontwikkeling. De ontwikkeling van de interventie deden we samen met relevante *stakeholders* (jongeren met autisme, hun ouders en docenten en professionals). Zij benoemden vier factoren, met bijbehorende specificaties, die belangrijk zijn om mee te nemen in de ontwikkeling van de slaapinterventie: beperkte belasting, nut, duidelijke begeleiding en structuur en een ondersteuningssysteem. Door de iteratieve aanpak en de participatie van *stakeholders* hebben we mogelijke belemmeringen voor het uitvoeren van de

slaapinterventie al tijdens de ontwikkeling ervan opgemerkt. We hebben de slaapinterventie hierop kunnen aanpassen wat een succesvolle implementatie op scholen waarschijnlijk vergroot.

In **hoofdstuk 5** presenteerden we de resultaten van de studie naar de effectiviteit van de slaapinterventie, waarbij we ons focusten op slaapkwaliteit en dagelijks functioneren. Door middel van een multiple baseline design met 23 jongeren met autisme ($M_{\text{leeftijd}} = 14.8$, $SD = 1.7$, range = 12 – 18 jaar) hebben we dit onderzocht. Jongeren vulden dagelijks een digitaal slaapdagboek in en droegen 's nachts een *sleeptracker*. Aan het eind van de dag vulden de jongeren vragen in over hun dagelijks functioneren. De vragen over het dagelijks functioneren werden ook ingevuld door een ouder en docent van de jongeren. Daarnaast vulden jongeren, ouders en docenten vragenlijsten in over slaapproblemen (ISI [jongeren] en CSHQ [ouders]) en gedragsproblemen (SDQ [jongeren, docenten, ouders]) tijdens de voormeting, nameting en follow-up. We vonden geen significant effect van de interventie op slaapkwaliteit en dagelijks functioneren. De meerderheid van de jongeren liet echter wel een klinisch significante verbetering zien op de zelf-gerapporteerde vragenlijst over slaapproblemen (ISI). Dit geeft aan dat de slaapinterventie een positief effect heeft op de ernst van insomniesymptomen.

Tot slot zijn in **hoofdstuk 6** de belangrijkste bevindingen van de studies in dit proefschrift samengevat. Deze bevindingen benadrukken het belang van aandacht voor slaapproblemen in het speciaal onderwijs. Voor docenten is het van belang dat zij zich bewust zijn dat zij kunnen bijdragen aan het verminderen van de slaapproblemen van hun leerlingen. Het inzetten van een slaapinterventie op school kan een eerste stap zijn in een *stepped care* benadering. Dit is waardevol omdat een vroege behandeling van slaapproblemen verergering kan voorkomen. Hiervoor is het belangrijk dat docenten in staat zijn signalen van slaapproblemen bij hun leerlingen te herkennen. Zo kunnen zij ondersteuning bieden wanneer dat nodig is. Anderzijds is het van belang te beseffen dat docenten geen klinici zijn. Er zijn grenzen aan hun kunnen om de slaapproblemen van hun leerlingen te verminderen. Docenten moeten inzicht hebben voor welke jongeren de slaapinterventie op school geschikt zou kunnen zijn. Ten eerste zijn dit jongeren die de intentie hebben om hun slaapgewoonten te veranderen. Ten tweede zijn dat jongeren zonder interacterende of meervoudige problemen, zoals depressie, angststoornis, middelenafhankelijkheid of gameverslaving. Het is belangrijk te erkennen dat jongeren met complexe behoeften mogelijk extra ondersteuning nodig hebben. Aangezien docenten over het algemeen niet zijn toegerust om jongeren te ondersteunen bij het verminderen van hun slaapproblemen, is een speciale training nodig om kennis en vaardigheden op te doen. Daarnaast is het belangrijk dat de schoolleiding de inzet van de slaapinterventie omarmt en het belang ervan uitdraagt. De schoolleiding speelt namelijk een belangrijke rol in een succesvolle implementatie. Dit kunnen zij doen door hun docenten aan te moedigen, te ondersteunen en faciliteren. Voor vervolgonderzoek is het van belang om zicht te krijgen op de effectieve elementen van de slaapinterventie en de mechanismen en omstandigheden in kaart te brengen die verklaren wanneer, waarom, hoe en voor wie de slaapinterventie werkt. Dit zal docenten handvatten bieden welke elementen onder welke omstandigheden het meest veelbelovend zijn om de slaap van hun leerlingen te verbeteren. Daarbij is het zinvol om zelf-gerapporteerde en objectieve

maten van slaap te blijven betrekken in toekomstig onderzoek. Ondanks dat onze studies de waarde van zelf-gerapporteerde slaapmaten hebben aangetoond en niet van objectieve maten, is het te voorbarig om het nut van objectieve maten af te schrijven. Andere studies hebben namelijk wel significante effecten van slaapinterventies gevonden op objectieve maten. Vervolgonderzoek kan op deze manier ook meer zicht geven op de waarde van de verschillende slaapmaten. Hierbij is het van belang om gevalideerde *sleeptrackers* te gebruiken om de nauwkeurigheid en betrouwbaarheid van de verzamelde gegevens te garanderen. Tegenwoordig zijn er veel *wearable devices* voor consumenten op de markt die gebruikt kunnen worden om de slaap te meten. De nauwkeurigheid en betrouwbaarheid van deze *devices* is echter nog een punt van aandacht. Gezien de voordelen (bv. gebruiksvriendelijk, goedkoop, onopvallend) van *wearable devices* is er ook meer onderzoek nodig om de nauwkeurigheid hiervan vast te stellen en deze te verbeteren.



Chapter 9

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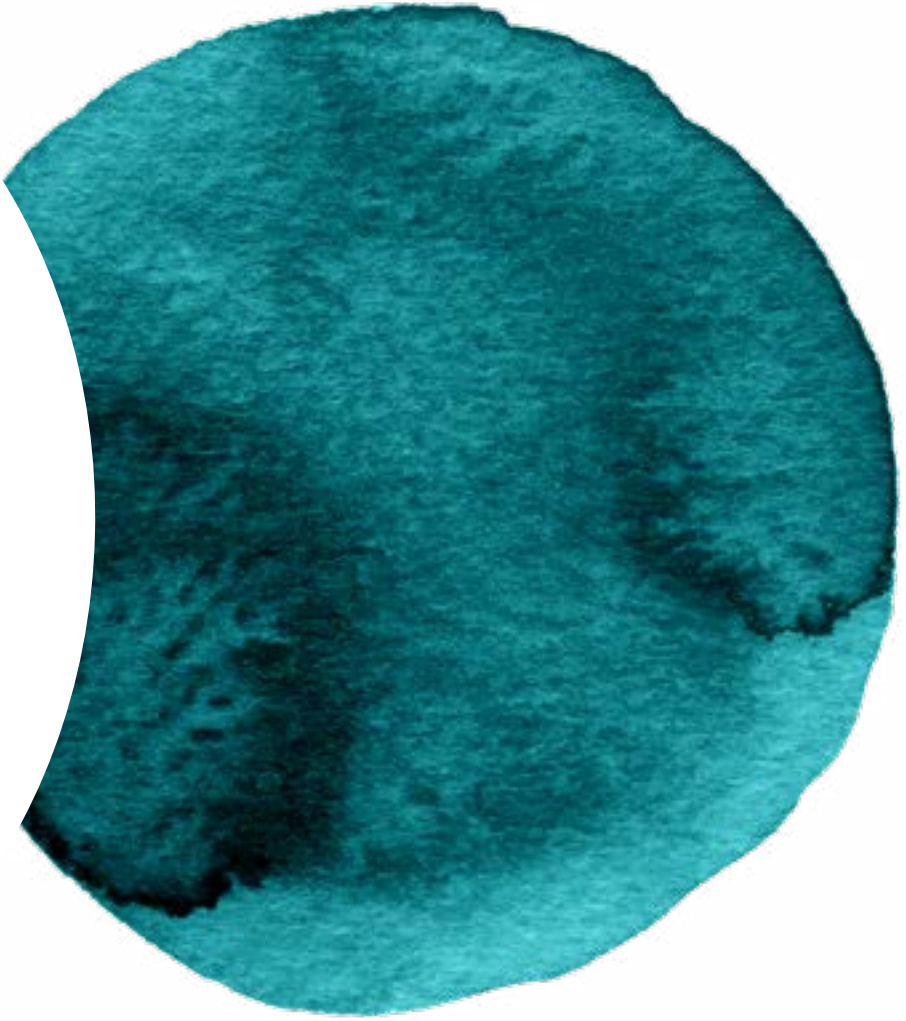
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Chapter 10

Supplementary materials

10.1 Supplementary material accompanying Chapter 2

10.1.1 Search strategy PsycINFO

#	Searches
1	autism spectrum disorders/ OR (autis* OR Asperger* OR (Pervasive ADJ1 development* ADJ1 disorder*) OR(Pervasive ADJ1 Child ADJ1 Development ADJ1 Disorder*) OR PDD OR PDDNOS OR ASDS OR ASD).ti,ab,id.
2	EXP sleep/ OR sleep deprivation/ OR EXP sleep disorders/ OR sleep onset/ OR sleep wake cycle/ OR (sleep* OR (early ADJ1 awake*) OR (Circadian ADJ1 rhythm*) OR Dyssonm* OR Parasomn* OR Hypersomn* OR insomn* OR (rest ADJ1 activit*) OR wakeful* OR chronotype OR (time ADJ2 bed) OR bedtime* OR nightmar*).ti,ab,id.
3	(child* OR kid OR kids OR prepuberty* OR teen* OR young* OR youth* OR minors* OR (under ADJ1 ag*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR (secondary ADJ1 education) OR (junior ADJ1 high*) OR highschool* OR student* OR (young ADJ1 adult*) OR infan* OR teen* OR (young ADJ1 people) OR school).ti,ab,id.
4	EXP academic achievement/ OR EXP truancy/ OR school refusal/ OR academic achievement prediction/ OR academic achievement motivation/ OR academic aptitude/ OR Academic failure/ OR school learning/ OR school graduation/ OR Educational attainment level/ OR school attendance/ OR (((academic* OR school* OR education* OR scholastic OR student* OR class* OR learn* OR cognitive) ADJ1 (fail* OR achiev* OR underachiev* OR overachiev* OR functioning OR performance* OR level* OR abilit* OR dropout* OR (drop* ADJ1 out*) OR success OR progress* OR result* OR readiness OR aptitude OR underperformance* OR aspiration)) OR ((learn* ADJ1 problem*) OR grade* OR GPA OR (Educational ADJ1 Test ADJ1 Score*) OR (school ADJ1 learn*) OR (school ADJ1 attendance) OR (school ADJ1 graduation) OR (Education* ADJ1 Attainment) OR (Student* ADJ1 Promotion*) OR truancy OR (Academic ADJ1 Test ADJ1 Score*) OR (school ADJ1 refusal))).ti,ab,id.
5	EXP attention/ OR EXP attention span/ OR concentration/ OR distraction/ OR distractibility/ OR (attenti* OR concentrat* OR distract*).ti,ab,id.
6	(Memory OR forget*).ti,ab,id.
7	Stress/ OR chronic stress/ OR psychological stress/ OR social stress/ OR academic stress/ OR stress reactions/ OR distress/ OR stress management/ OR relaxation/ OR (stress* OR distress* OR tension OR strain OR anxiety OR pressure OR relaxation).ti,ab,id.

- 8 behavior problems/ OR antisocial behavior/ OR aggressive behavior/ OR bullying/ OR (misbehavi* OR (disruptive ADJ1 behavi*) OR (behavi* ADJ1 problem*) OR (problem ADJ1 behavi*) OR (antisocial ADJ1 behavi*) OR (prosocial ADJ1 behavi*) OR (social ADJ1 behavi*) OR (self-destructive ADJ1 behavi*) OR (social ADJ1 problem*) OR irritab* OR aggressi* OR (obsessive ADJ1 compulsive ADJ1 behavi*) OR fidgeting OR (externali* ADJ1 behavi*) OR (internali* ADJ1 behavi*) OR (sensory ADJ1 problem*) OR (sensory ADJ1 over-responsivit*) OR (restricted ADJ2 repetitive ADJ1 behavi*) OR (stereotyp* ADJ1 behavi*) OR (deviant ADJ1 behavi*) OR bullying OR (addictive ADJ1 behavi*) OR (dysfunctional ADJ1 behavi*) OR hostility OR SDQ OR (Child ADJ1 Behavior ADJ1 Checklist) OR CBCL OR (Teacher ADJ1 Report ADJ1 Form) OR YSR OR (Youth ADJ1 Self-Report)).ti,ab,id.
-
- 9 Emotional regulation/ OR (mood* OR (frame ADJ1 mind) OR (state ADJ1 mind) OR temper OR tempered OR emotion* OR affect* OR nervousness OR frustration).ti,ab,id.
-
- 10 fatigue/ OR sleepiness/ OR (tired* OR fatigue OR exhaust* OR drows* OR sleepiness OR lethargy).ti,ab,id.
-
- 11 1 AND 2 AND 3 AND (4 OR 5 OR 6 OR 7 OR 8 OR 9 OR 10)
-

10.1.2 Search Strategy Cochrane

#	Searches
1	(autis* OR Asperger* OR (Pervasive NEAR/1 development* NEAR/1 disorder*) OR(Pervasive NEAR/1 Child NEAR/1 Development NEAR/1 Disorder*) OR PDD OR PDDNOS OR ASDS OR ASD):ti,ab,kw
2	(sleep* OR (early NEAR/1 awake*) OR (Circadian NEAR/1 rhythm*) OR Dyssomn* OR Parasomn* OR Hypersomn* OR insomn* OR (rest NEAR/1 activit*) OR wakeful* OR chronotype OR (time NEAR/2 bed) OR bedtime* OR nightmar*):ti,ab,kw
3	(child* OR kid OR kids OR prepuberty* OR teen* OR young* OR youth* OR minors* OR (under NEAR/1 ag*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR (secondary NEAR/1 education) OR (junior NEAR/1 high*) OR highschool* OR student* OR (young NEAR/1 adult*) OR infan* OR teen* OR (young NEAR/1 people) OR school):ti,ab,kw
4	((academic* OR school* OR education* OR scholastic OR student* OR class* OR learn* OR cognitive) NEAR/1 (fail* OR achiev* OR underachiev* OR overachiev* OR functioning OR performance* OR level* OR abilit* OR dropout* OR (drop* NEAR/1 out*) OR success OR progress* OR result* OR readiness OR aptitude OR underperformance* OR aspiration)) OR ((learn* NEAR/1 problem*) OR grade* OR GPA OR (Educational NEAR/1 Test NEAR/1 Score*) OR (school NEAR/1 learn*) OR (school NEAR/1 attendance) OR (school NEAR/1 graduation) OR (Education* NEAR/1 Attainment) OR (Student* NEAR/1 Promotion*) OR truancy OR (Academic NEAR/1 Test NEAR/1 Score*) OR (school NEAR/1 refusal)):ti,ab,kw
5	(attenti* OR concentrat* OR distract*):ti,ab,kw
6	(Memory OR forget*):ti,ab,kw
7	(stress* OR distress* OR tension OR strain OR anxiety OR pressure OR relaxation):ti,ab,kw
8	(misbehavi* OR (disruptive NEAR/1 behavi*) OR (behavi* NEAR/1 problem*) OR (problem NEAR/1 behavi*)OR (antisocial NEAR/1 behavi*) OR (prosocial NEAR/1 behavi*) OR (social NEAR/1 behavi*) OR (self-destructive NEAR/1 behavi*) OR (social NEAR/1 problem*) OR irritab* OR aggressi* OR (obsessive NEAR/1 compulsive NEAR/1 behavi*) OR fidgeting OR (externali* NEAR/1 behavi*) OR (internali* NEAR/1 behavi*) OR (sensory NEAR/1 problem*) OR (sensory NEAR/1 over-responsivit*) OR (restricted NEAR/2 repetitive NEAR/1 behavi*) OR (stereotyp* NEAR/1 behavi*) OR (deviant NEAR/1 behavi*) OR bullying OR (addictive NEAR/1 behavi*) OR (dysfunctional NEAR/1 behavi*) OR hostility OR SDQ OR (Child NEAR/1 Behavior NEAR/1 Checklist) OR CBCL OR (Teacher NEAR/1 Report NEAR/1 Form) OR YSR OR (Youth NEAR/1 Self-Report)):ti,ab,kw
9	(mood* OR (frame NEAR/1 mind) OR (state NEAR/1 mind) OR temper OR tempered OR emotion* OR affect* OR nervousness OR frustration):ti,ab,kw
10	(tired* OR fatigue OR exhaust* OR drows* OR sleepiness OR lethargy):ti,ab,kw
11	#1 AND #2 AND #3 AND (#4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10)

10.1.3 Search strategy ERIC

#	Searches
1	DE "Pervasive Developmental Disorders" OR DE "Asperger Syndrome" OR DE "Autism" OR TI (autis* OR Asperger* OR (Pervasive N1 Development* N1 Disorder*) OR (Pervasive N1 Child N1 Development N1 Disorder*) OR PDD OR PDDNOS OR ASDS OR ASD) OR AB (autis* OR Asperger* OR (Pervasive N1 Development* N1 Disorder*) OR (Pervasive N1 Child N1 Development N1 Disorder*) OR PDD OR PDDNOS OR ASDS OR ASD) OR SU (autis* OR Asperger* OR (Pervasive N1 Development* N1 Disorder*) OR (Pervasive N1 Child N1 Development N1 Disorder*) OR PDD OR PDDNOS OR ASDS OR ASD)
2	DE "Sleep" OR TI (sleep* OR (early N1 awake*) OR (Circadian N1 rhythm*) OR Dyssomn* OR Parasomn* OR Hypersomn* OR insomn* OR (rest N1 activit*) OR wakeful* OR chronotype OR (time N2 bed) OR bedtime* OR nightmar*) OR AB (sleep* OR (early N1 awake*) OR (Circadian N1 rhythm*) OR Dyssomn* OR Parasomn* OR Hypersomn* OR insomn* OR (rest N1 activit*) OR wakeful* OR chronotype OR (time N2 bed) OR bedtime* OR nightmar*) OR SU (sleep* OR (early N1 awake*) OR (Circadian N1 rhythm*) OR Dyssomn* OR Parasomn* OR Hypersomn* OR insomn* OR (rest N1 activit*) OR wakeful* OR chronotype OR (time N2 bed) OR bedtime* OR nightmar*)
3	DE "Adolescents" OR DE "Preadolescents" OR DE "Early adolescents" OR DE "Secondary School Students" OR DE "High School Students" OR DE "Junior High School Students" OR TI (child* OR kid OR kids OR prepuberty* OR teen* OR young* OR youth* OR minors* OR (under N1 ag*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR (secondary N1 education) OR (junior N1 high*) OR highschool* OR student* OR (young N1 adult*) OR infan* OR teen* OR (young N1 people) OR school) OR AB (child* OR kid OR kids OR prepuberty* OR teen* OR young* OR youth* OR minors* OR (under N1 ag*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR (secondary N1 education) OR (junior N1 high*) OR highschool* OR student* OR (young N1 adult*) OR infan* OR teen* OR (young N1 people) OR school) OR SU (child* OR kid OR kids OR prepuberty* OR teen* OR young* OR youth* OR minors* OR (under N1 ag*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR (secondary N1 education) OR (junior N1 high*) OR highschool* OR student* OR (young N1 adult*) OR infan* OR teen* OR (young N1 people) OR school)

- 4 DE "Academic Achievement" OR DE "School Attendance" OR DE "Educational Attainment" OR DE "Student Promotion" OR DE "Academic ability" OR DE "academic aptitude" OR DE "Academic failure" OR DE "Learning problems" OR DE "Academic Aspiration" OR DE "Grades (Scholastic)" OR DE "Grade Inflation" OR DE "Grade Point Average" OR DE "Underachievement" OR DE "Achievement Gap" OR DE "Overachievement" OR TI (((academic* OR school* OR education* OR scholastic OR student* OR class* OR learn* OR cognitive) N1 (fail* OR achiev* OR underachiev* OR overachiev* OR functioning OR performance* OR level* OR abilit* OR dropout* OR (drop* N1 out*) OR success OR progress* OR result* OR readiness OR aptitude OR underperformance* OR aspiration)) OR ((learn* N1 problem*) OR (school N1 attendance) OR grade* OR GPA OR (Educational N1 Test N1 Score*) OR (school N1 learn*) OR (school N1 graduation) OR (Education* N1 Attainment) OR (Student* N1 Promotion*) OR (Academic N1 Test N1 Score*) OR truancy OR (school N1 refusal))) OR AB (((academic* OR school* OR education* OR scholastic OR student* OR class* OR learn* OR cognitive) N1 (fail* OR achiev* OR underachiev* OR overachiev* OR functioning OR performance* OR level* OR abilit* OR dropout* OR (drop* N1 out*) OR success OR progress* OR result* OR readiness OR aptitude OR underperformance* OR aspiration)) OR ((learn* N1 problem*) OR (school N1 attendance) OR grade* OR GPA OR (Educational N1 Test N1 Score*) OR (school N1 learn*) OR (school N1 graduation) OR (Education* N1 Attainment) OR (Student* N1 Promotion*) OR (Academic N1 Test N1 Score*) OR truancy OR (school N1 refusal))) OR SU (((academic* OR school* OR education* OR scholastic OR student* OR class* OR learn* OR cognitive) N1 (fail* OR achiev* OR underachiev* OR overachiev* OR functioning OR performance* OR level* OR abilit* OR dropout* OR (drop* N1 out*) OR success OR progress* OR result* OR readiness OR aptitude OR underperformance* OR aspiration)) OR ((learn* N1 problem*) OR (school N1 attendance) OR grade* OR GPA OR (Educational N1 Test N1 Score*) OR (school N1 learn*) OR (school N1 graduation) OR (Education* N1 Attainment) OR (Student* N1 Promotion*) OR (Academic N1 Test N1 Score*) OR truancy OR (school N1 refusal)))
- 5 DE "Attention" OR DE "Attention Span" OR TI (attenti* OR concentrat* OR distract*) OR AB (attenti* OR concentrat* OR distract*) OR SU (attenti* OR concentrat* OR distract*)
- 6 TI (Memory OR forget*) OR AB (Memory OR forget*) OR SU (Memory OR forget*)
- 7 TI (stress* OR distress* OR tension OR strain OR anxiety OR pressure OR relaxation) OR AB (stress* OR distress* OR tension OR strain OR anxiety OR pressure OR relaxation) OR SU (stress* OR distress* OR tension OR strain OR anxiety OR pressure OR relaxation)

-
- 8 DE "Behavior Problems" OR DE "Antisocial Behavior" OR DE "Aggression" OR DE "Bullying" OR DE "Self Destructive Behavior" OR DE "Addictive Behavior" OR TI (misbehavi* OR (disruptive N1 behavi*) OR (behavi* N1 problem*) OR (problem N1 behavi*) OR (antisocial N1 behavi*) OR (prosocial N1 behavi*) OR (social N1 behavi*) OR (self-destructive N1 behavi*) OR (social N1 problem*) OR irritab* OR aggressi* OR (obsessive N1 compulsive N1 behavi*) OR fidgeting OR (externali* N1 behavi*) OR (internali* N1 behavi*) OR (sensory N1 problem*) OR (sensory N1 over-responsivit*) OR (restricted ADJ2 repetitive N1 behavi*) OR (stereotyp* N1 behavi*) OR (deviant N1 behavi*) OR bullying OR (addictive N1 behavi*) OR (dysfunctional N1 behavi*) OR hostility OR SDQ OR (Child N1 Behavior N1 Checklist) OR CBCL OR (Teacher N1 Report N1 Form) OR YSR OR (Youth N1 Self-Report)) OR AB (misbehavi* OR (disruptive N1 behavi*) OR (behavi* N1 problem*) OR (problem N1 behavi*) OR (antisocial N1 behavi*) OR (prosocial N1 behavi*) OR (social N1 behavi*) OR (self-destructive N1 behavi*) OR (social N1 problem*) OR irritab* OR aggressi* OR (obsessive N1 compulsive N1 behavi*) OR fidgeting OR (externali* N1 behavi*) OR (internali* N1 behavi*) OR (sensory N1 problem*) OR (sensory N1 over-responsivit*) OR (restricted ADJ2 repetitive N1 behavi*) OR (stereotyp* N1 behavi*) OR (deviant N1 behavi*) OR bullying OR (addictive N1 behavi*) OR (dysfunctional N1 behavi*) OR hostility OR SDQ OR (Child N1 Behavior N1 Checklist) OR CBCL OR (Teacher N1 Report N1 Form) OR YSR OR (Youth N1 Self-Report)) OR SU (misbehavi* OR (disruptive N1 behavi*) OR (behavi* N1 problem*) OR (problem N1 behavi*) OR (antisocial N1 behavi*) OR (prosocial N1 behavi*) OR (social N1 behavi*) OR (self-destructive N1 behavi*) OR (social N1 problem*) OR irritab* OR aggressi* OR (obsessive N1 compulsive N1 behavi*) OR fidgeting OR (externali* N1 behavi*) OR (internali* N1 behavi*) OR (sensory N1 problem*) OR (sensory N1 over-responsivit*) OR (restricted ADJ2 repetitive N1 behavi*) OR (stereotyp* N1 behavi*) OR (deviant N1 behavi*) OR bullying OR (addictive N1 behavi*) OR (dysfunctional N1 behavi*) OR hostility OR SDQ OR (Child N1 Behavior N1 Checklist) OR CBCL OR (Teacher N1 Report N1 Form) OR YSR OR (Youth N1 Self-Report))
-
- 9 TI (mood* OR (frame ADJ1 mind) OR (state ADJ1 mind) OR temper OR tempered OR emotion* OR affect OR nervousness OR frustration) OR AB (mood* OR (frame ADJ1 mind) OR (state ADJ1 mind) OR temper OR tempered OR emotion* OR affect OR nervousness OR frustration) OR SU (mood* OR (frame ADJ1 mind) OR (state ADJ1 mind) OR temper OR tempered OR emotion* OR affect OR nervousness OR frustration)
-
- 10 DE "fatigue" OR TI (tired* OR fatigue OR exhaust* OR drows* OR sleepiness OR lethargy) OR AB (tired* OR fatigue OR exhaust* OR drows* OR sleepiness OR lethargy) OR SU (tired* OR fatigue OR exhaust* OR drows* OR sleepiness OR lethargy)
-
- 11 S1 AND S2 AND S3 AND (S4 OR S5 OR S6 OR S7 OR S8 OR S9 OR S10)
-

10.1.4 Search strategy PubMed

#	Searches
1	"Child Development Disorders, Pervasive"[Mesh] OR autism*[tiab] OR Asperger*[tiab] OR Pervasive development disorder*[tiab] OR Pervasive Child Development Disorder*[tiab] OR PDD[tiab] OR PDDNOS[tiab] OR ASDS[tiab] OR ASD[tiab]
2	"Sleep"[Mesh] OR "Sleep Wake Disorders"[Mesh:NoExp] OR "Dyssomnias"[Mesh] OR "Parasomnias"[Mesh:NoExp] OR "Circadian Rhythm"[Mesh] OR sleep*[tiab] OR early awake*[tiab] OR Circadian rhythm*[tiab] OR Dyssomn*[tiab] OR Parasomn*[tiab] OR Hypersomn*[tiab] OR insomn*[tiab] OR rest activit*[tiab] OR wakeful*[tiab] OR chronotype[tiab] OR time in bed[tiab] OR bedtime*[tiab] OR nightmar*[tiab]
3	"Adolescent"[Mesh] OR "Child"[Mesh] OR "Minors"[Mesh] OR "Young Adult"[Mesh] OR child*[tiab] OR kid[tiab] OR kids[tiab] OR prepuberty*[tiab] OR teen*[tiab] OR young*[tiab] OR youth*[tiab] OR minors*[tiab] OR under ag*[tiab] OR underag*[tiab] OR juvenil*[tiab] OR preadolesc*[tiab] OR adolesc*[tiab] OR secondary education[tiab] OR junior high*[tiab] OR highschool*[tiab] OR student*[tiab] OR young adult*[tiab] OR infan*[tiab] OR teen*[tiab] OR young people[tiab] OR school[tiab]

4 "Academic Failure"[Mesh] OR "Aptitude"[Mesh] OR Academic fail*[tiab] OR Academic achiev*[tiab] OR Academic underachiev*[tiab] OR Academic functioning[tiab] OR Academic performance*[tiab] OR Academic level*[tiab] OR Academic abilit*[tiab] OR Academic dropout*[tiab] OR Academic success[tiab] OR Academic progress*[tiab] OR Academic result*[tiab] OR Academic readiness[tiab] OR Academic aptitude[tiab] OR Academic underperformance*[tiab] OR School fail*[tiab] OR School achiev*[tiab] OR School underachiev*[tiab] OR School functioning[tiab] OR School performance*[tiab] OR School level*[tiab] OR School abilit*[tiab] OR School drop out*[tiab] OR School dropout[tiab] OR School success[tiab] OR School progress*[tiab] OR School result*[tiab] OR School readiness[tiab] OR School aptitude[tiab] OR Education achiev*[tiab] OR Education performance*[tiab] OR Education level*[tiab] OR Education abilit*[tiab] OR Education success[tiab] OR Education progress*[tiab] OR Education result*[tiab] OR Educational fail*[tiab] OR Educational achiev*[tiab] OR Educational underachiev*[tiab] OR Educational functioning[tiab] OR Educational performance*[tiab] OR Educational level*[tiab] OR Educational abilit*[tiab] OR Educational dropout*[tiab] OR Educational success[tiab] OR Educational progress*[tiab] OR Educational result*[tiab] OR Educational readiness[tiab] OR Educational aptitude[tiab] OR Scholastic fail*[tiab] OR Scholastic achiev*[tiab] OR Scholastic underachiev*[tiab] OR Scholastic functioning[tiab] OR Scholastic performance*[tiab] OR Scholastic level*[tiab] OR Scholastic abilit*[tiab] OR Scholastic success[tiab] OR Scholastic progress*[tiab] OR Scholastic result*[tiab] OR Scholastic aptitude[tiab] OR Student fail*[tiab] OR Student achiev*[tiab] OR Student functioning[tiab] OR Student performance*[tiab] OR Student level*[tiab] OR Student abilit*[tiab] OR Student dropout*[tiab] OR Student drop out*[tiab] OR Student success[tiab] OR Student progress*[tiab] OR Student result*[tiab] OR Student readiness[tiab] OR Student aptitude[tiab] OR student aspiration*[tiab] OR students fail*[tiab] OR students achiev*[tiab] OR students functioning[tiab] OR students performance*[tiab] OR students level*[tiab] OR students ability*[tiab] OR students success[tiab] OR students progress*[tiab] OR students result*[tiab] OR students readiness[tiab] OR studens aspiration*[tiab] OR Class fail*[tiab] OR Class achiev*[tiab] OR Class performance*[tiab] OR Class level*[tiab] OR Class abilit*[tiab] OR Class progress*[tiab] OR Class result*[tiab] OR Class readiness[tiab] OR Learn ability*[tiab] OR learning fail*[tiab] OR learning achiev*[tiab] OR learning performance*[tiab] OR learning level*[tiab] OR learning ability*[tiab] OR learning success[tiab] OR learning progress*[tiab] OR learning result*[tiab] OR learning readiness[tiab] OR learning aptitude[tiab] OR Cognitive fail*[tiab] OR Cognitive achiev*[tiab] OR Cognitive functioning[tiab] OR Cognitive performance*[tiab] OR Cognitive level*[tiab] OR Cognitive abilit*[tiab] OR Cognitive success[tiab] OR Cognitive progress*[tiab] OR Cognitive result*[tiab] OR Cognitive readiness[tiab] OR Cognitive aptitude[tiab] OR Cognitive underperformance*[tiab] OR learning problem*[tiab] OR grade*[tiab] OR GPA[tiab] OR Educational Test Score*[tiab] OR Academic Test Score*[tiab] OR school learn[tiab] OR school graduation[tiab] OR education attainment[tiab] OR educational attainment[tiab] OR student promotion[tiab] OR school attendance[tiab] OR truancy[tiab] OR School refusal[tiab]

5	"Attention"[Mesh] OR Attenti*[tiab] OR concentrat*[tiab] OR distract*[tiab]
6	Memory[tiab] OR forget*[tiab]
7	"Stress, Psychological"[Mesh]OR "Relaxation"[Mesh] OR stress*[tiab] OR distress*[tiab] OR tension[tiab] OR strain[tiab] OR anxiety[tiab] OR pressure[tiab] OR relaxation[tiab]
8	"Problem Behavior"[Mesh] OR misbehavi*[tiab] OR disruptive behavi*[tiab] OR antisocial behavi*[tiab] OR prosocial behavi*[tiab] OR social behavi*[tiab] OR self-destructive behavi*[tiab] OR social problem*[tiab] OR irritab*[tiab] OR aggressi*[tiab] OR obsessive compulsive behavi*[tiab] OR fidgeting[tiab] OR externalising behavi*[tiab] OR externalizing behavi*[tiab] OR internalising behavi*[tiab] OR internalizing behavi*[tiab] OR sensory problem*[tiab] OR sensory over-responsivit*[tiab] OR restricted repetitive behavi*[tiab] OR stereotype behavi*[tiab] OR stereotypical behavi*[tiab] OR deviant behavi*[tiab] OR bullying[tiab] OR addictive behav*[tiab] OR hostility[tiab] OR SDQ[tiab] OR Child Behavior Checklist[tiab] OR CBCL[tiab] OR Teacher Report Form*[tiab] OR YSR[tiab] OR Youth Self-Report[tiab] OR behavior problem*[tiab] OR behaviour problem*[tiab] problem behav*[tiab] OR Behavioral Problem*[tiab] OR Behavioural Problem*[tiab] OR Dysfunctional Behav*[tiab]
9	"Affect"[Mesh] OR mood*[tiab] OR state of mind*[tiab] OR temper[tiab] OR tempered[tiab] OR affects[tiab] OR emotion*[tiab] OR nervousness[tiab] or frustration[tiab]
10	"Fatigue"[Mesh] OR "Sleepiness"[Mesh] OR "Lethargy"[Mesh] OR tired*[tiab] OR fatigue[tiab] OR exhaust*[tiab] OR drows*[tiab] OR sleepiness[tiab] OR lethargy[tiab]
11	#1 AND #2 AND #3 AND (#4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10)

10.1.5 Search strategy Web of Science

#	Searches
1	TS=(autis* OR Asperger* OR ("Pervasive" NEAR/1 Development NEAR/1 Disorder*) OR "PDD" OR "PDDNOS" OR "ASDS" OR "ASD")
2	TS=(sleep* OR ("early" NEAR/1 awake*) OR ("Circadian" NEAR/1 rhythm*) OR Dyssomn* OR Parasomn* OR Hypersomn* OR Insomn* OR ("rest" NEAR/1 activit*) OR wakeful* OR "chronotype" OR ("time" NEAR/2 "bed") OR bedtime* OR nightmar*)
3	TS=(child* OR "kid" OR "kids" OR prepuberty* OR teen* OR young* OR youth* OR minors* OR ("under" NEAR/1 age*) OR underag* OR juvenil* OR preadolesc* OR adolesc* OR ("secondary NEAR/1 education") OR junior high* OR highschool* OR student* OR ("young" NEAR/1 adult*) OR infan* OR teen* OR ("young NEAR/1 people") OR ("middle" NEAR/1 "school"))
4	TS=((((academic* OR school* OR education* OR "scholastic" OR student* OR class* OR learn* OR "cognitive") NEAR/1 (fail* OR achiev* OR underachiev* OR overachiev* OR "functioning" OR performance* OR level* OR abilit* OR dropout* OR (drop* NEAR/1 out*) OR "success" OR progress* OR result* OR "readiness" OR "aptitude" OR underperformance* OR "aspiration")) OR ((learn* NEAR/1 problem*) OR grade* OR "GPA" OR ("Educational" NEAR/1 "Test" NEAR/1 Score*) OR ("school" NEAR/1 learn*) OR ("school" NEAR/1 "attendance") OR ("school" NEAR/1 "graduation") OR (Education* NEAR/1 "Attainment") OR (Student* NEAR/1 Promotion*) OR "truancy" OR ("Academic" NEAR/1 "Test" NEAR/1 Score*) OR ("school" NEAR/1 "refusal"))))
5	TS=("attention" OR concentrat* OR distract*)
6	TS=("Memory" OR forget*)
7	TS=(stress* OR distress* OR "tension" OR "strain" OR "anxiety" OR "pressure" OR "relaxation")
8	TS=(misbehavi* OR ("disruptive" NEAR/1 behavi*) OR (behavi* NEAR/1 problem*) OR ("problem" NEAR/1 behavi*) OR ("antisocial" NEAR/1 behavi*) OR ("prosocial" NEAR/1 behavi*) OR ("social" NEAR/1 behavi*) OR ("self-destructive" NEAR/1 behavi*) OR ("social" NEAR/1 problem*) OR irritab* OR aggressi* OR ("obsessive" NEAR/1 "compulsive" NEAR/1 behavi*) OR "fidgeting" OR (externali* NEAR/1 behavi*) OR (internali* NEAR/1 behavi*) OR ("sensory" NEAR/1 problem*) OR ("sensory" NEAR/1 over-responsivit*) OR ("restricted" NEAR/2 "repetitive" NEAR/1 behavi*) OR (stereotyp* NEAR/1 behavi*) OR ("deviant" NEAR/1 behavi*) OR "bullying" OR ("addictive" NEAR/1 behavi*) OR ("dysfunctional" NEAR/1 behavi*) OR "hostility" OR "SDQ" OR ("Child" NEAR/1 "Behavior" NEAR/1 "Checklist") OR "CBCL" OR ("Teacher" NEAR/1 "Report" NEAR/1 "Form") OR "YSR" OR ("Youth" NEAR/1 "Self-Report"))
9	TS=(mood* OR ("frame NEAR/1 mind") OR ("state NEAR/1 mind") OR "temper" OR tempered OR emotion* OR affect* OR "nervousness" OR "frustration")
10	TS=("sleepiness" OR tired* OR "fatigue" OR exhaust* OR drows* OR "lethargy")
11	#1 AND #2 AND #3 AND (#4 OR #5 OR #6 OR #7 OR #8 OR #9 OR #10)

10.2 Supplementary material accompanying Chapter 3

10.2.1 Imputation code

```
###-----###
### Goal of script: Impute missing data                                     ###
###-----###

#-----#
# 1. Load packages & data                                               #
#-----#

# Load packages
require(mice)
require(lattice)
require(pan)
require(foreign)
require(multilevel)

# Set working directory
setwd(dirname(rstudioapi::getSourceEditorContext()$path))
data<-read.spss("WP2_ronde 1+2_totaal_def_long_netwerk.sav",to.data.frame = TRUE,
  use.value.labels = FALSE)

#-----#
# 2. Select data                                                         #
#-----#

dataIn_sh_max<-data[c("Respondentnr","SOLA","TSTA","WASOA","Slaapkwal",
  "Vermoedheid_L","Stress_L","Concentratie_L","Humeur_L","Beweging_L",
  "Mentaal_L","Recode_Cafeine_max","Recode_Sporten_max",
  "Recode_Gamen_max","Recode_Beeldscherm_max")]

#-----#
# 3. Impute data                                                         #
#-----#

set.seed(123)

# Imputation
```

```
ini <- mice(datalln_sh_max, maxit = 20)

# Change imputation method
meth <- ini$meth
meth
meth[2:15] <- "norm"
meth

# Predictor matrix (use all variables)
pred <- ini$pred

# Observed range for SOLA and WASOA, possible range for other variables
post <- ini$post

post["SOLA"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(0, 400))"
post["WASOA"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(0, 409))"
post["Slaapkwal"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Vermoeidheid_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Stress_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Concentratie_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Humour_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Beweging_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"
post["Mentaal_L"] <- "imp[[j]][, i] <- squeeze(imp[[j]][, i], c(1, 10))"

# Impute
restricted_imp_max_lln <- mice(datalln_sh_max, meth = meth, pred = pred, post = post, print = FALSE)
pred

# Inspect the trace lines
Tracelines_lln <- plot(restricted_imp_max_lln)

#pdf("30052022_Tracelines_lln.pdf")
plot(Tracelines_lln)
#dev.off()

# Densities of the observed and imputed data
Density_lln <- densityplot(restricted_imp_max_lln)

#pdf("30052022_Densityplot_lln.pdf")
```

```

plot(Density_IIIn)
#dev.off()

# Save
restricted_imp_max_IIIn_dataframe <- complete(restricted_imp_max_IIIn, include = FALSE)

write.csv(restricted_imp_max_IIIn_dataframe, 'Data_IIIn_imp.csv')

```

10.2.2 Network estimating code

```

###-----###
### Goal of script: Estimate network          ###
###-----###

# ----- #
# 1. Load packages & data                      #
# ----- #

#load packages
library("mgm")
library("qgraph")

# Set working directory
setwd(dirname(rstudioapi::getSourceEditorContext()$path))
data_IIIn_imp<-read.csv('Data_IIIn_imp.csv')

#select data
data_imp_max_IIIn<-data_IIIn_imp[,c("SOLA","TSTA","WASOA","Slaapkwal","Vermoeidheid_L",
  "Stress_L","Concentratie_L","Humeur_L","Beweging_L","Mentaal_L",
  "Recode_Cafeine_max","Recode_Sporten_max","Recode_Gamen_max",
  "Recode_Beeldscherm_max")]

# ----- #
# 2. Estimate network                          #
# ----- #

set.seed(123)

fit_mgm_imp_max_IIIn <- mgm(data = data_imp_max_IIIn, type = rep("g",14), levels = rep(1,14),
  k = 2, lambdaSel = "EBIC", ruleReg = "AND")

```

```
# weighted adjacency matrix
fit_mgm_imp_max_lln$pairwise$wadj

# ----- #
# 3. Visualize network                                #
# ----- #

# Visualize using qqgraph
names(data_imp_max_lln) <- c("Sleep onset latency (s)", "Total sleep time (s)",
  "Wake time after sleep onset (s)", "Sleep quality (s)", "Daytime sleepiness (s)",
  "Stress (s)", "Concentration (s)", "Mood (s)", "Physical exercise (s)",
  "Mental effort (s)", "Time of caffeine intake before bedtime (s)",
  "Time of intensive physical exercise before bedtime (s)",
  "Time of gaming before bedtime (s)",
  "Time of electronic device use before bedtime (s)")

Plot_Network_imp_max_lln <- qqgraph(fit_mgm_imp_max_lln$pairwise$wadj,
  edge.color = fit_mgm_imp_max_lln$pairwise$edgecolor,
  nodeNames = names(data_imp_max_lln), layout='spring',
  legend.cex = 0.3, title = "Netwerk leerlingen")

#pdf("07062022_netwerk_lln.pdf")
plot(Plot_Network_imp_max_lln)
#dev.off()

# ----- #
# 4. Resample network                                #
# ----- #

#resample
res_imp_max_lln <- resample(object = fit_mgm_imp_max_lln, data = data_imp_max_lln, nB = 80)

#Plot summary of all sampling distributions
pdf("14062022_resample_lln.pdf", width = 10, height = 50)
resample_lln<-plotRes(object = res_imp_max_lln, quantiles = c(0.05, .95),
  labels = colnames(data_imp_max_lln), layout.width.labels = .95)
dev.off()

# ----- #
```

```

# 5. Select edges                                     #
# ----- #

# weighted adjacency matrix
wadj_lln <- fit_mgm_imp_max_lln$pairwise$wadj
rownames(wadj_lln) <- colnames(wadj_lln) <- names(data_imp_max_lln)

# put edges <.90 to zero
wadj_lln["Physical exercise (s)","Time of intensive physical exercise before bedtime (s)"] = wadj_lln["Time of intensive physical exercise before bedtime (s)","Physical exercise (s)"] = 0
wadj_lln["Physical exercise (s)","Total sleep time (s)"] = wadj_lln["Total sleep time (s)","Physical exercise (s)"] = 0
wadj_lln["Mental effort (s)","Total sleep time (s)"] = wadj_lln["Total sleep time (s)","Mental effort (s)"] = 0
wadj_lln["Stress (s)","Total sleep time (s)"] = wadj_lln["Total sleep time (s)","Stress (s)"] = 0
wadj_lln["Concentration (s)","Time of electronic device use before bedtime (s)"] = wadj_lln["Time of electronic device use before bedtime (s)","Concentration (s)"] = 0
wadj_lln["Concentration (s)","Daytime sleepiness (s)"] = wadj_lln["Daytime sleepiness (s)","Concentration (s)"] = 0
wadj_lln["Stress (s)","Time of caffeine intake before bedtime (s)"] = wadj_lln["Time of caffeine intake before bedtime (s)","Stress (s)"] = 0
wadj_lln["Daytime sleepiness (s)","Time of caffeine intake before bedtime (s)"] = wadj_lln["Time of caffeine intake before bedtime (s)","Daytime sleepiness (s)"] = 0

# ----- #
# 6. Visualize after put edges to zero               #
# ----- #

Labels <- c("SOL","TST","WASO","QUAL","DAY","STR","CON","MOOD",
"PE","ME","CAF","IPE","GAME","EDU")

Groups <- c("Sleep","Sleep","Sleep","Sleep","Daytime functioning","Sleep hygiene",
"Daytime functioning","Daytime functioning","Sleep hygiene","Sleep hygiene",
"Sleep hygiene","Sleep hygiene","Sleep hygiene","Sleep hygiene")

edge_col_lln<-fit_mgm_imp_max_lln$pairwise$edgecolor
edge_col_lln[edge_col_lln=='darkgreen']<-'darkblue'
edge_col_lln[edge_col_lln=='red']<-'darkred'

Plot_Network_imp_max_lln$layout
Plot_layout <- Plot_Network_imp_max_lln$layout
Plot_layout[1,1]<- 0.30

```



```
Plot_layout[1,2]<- -0.12
Plot_layout[2,1]<- -0.5
Plot_layout[2,2]<- -0.65
Plot_layout[3,1]<- -0.30
Plot_layout[3,2]<- -1.15
Plot_layout[4,1]<- 0.27
Plot_layout[4,2]<- 0.76
Plot_layout[5,1]<- -0.06
Plot_layout[5,2]<- 0.85
Plot_layout[6,1]<- 0.7
Plot_layout[6,2]<- 0.15
Plot_layout[7,2]<- 0.35
Plot_layout[7,1]<- -0.10
Plot_layout[8,1]<- -0.63
Plot_layout[8,2]<- 0.73
Plot_layout[9,1]<- -0.78
Plot_layout[9,2]<- -0.90
Plot_layout[10,1]<- 0.07
Plot_layout[10,2]<- -0.55
Plot_layout[11,1]<- -1.15
Plot_layout[11,2]<- -0.27
Plot_layout[12,1]<- -1.15
Plot_layout[12,2]<- 0.34
Plot_layout[13,1]<- 0.85
Plot_layout[13,2]<- -0.28
Plot_layout[14,1]<- 0.44
Plot_layout[14,2]<- -1.00
```

```
Plot_Network_imp_max_lln_resample <- qgraph(wadj_lln, edge.color = edge_col_lln,
      nodeNames = names(data_imp_max_lln), layout= Plot_layout,
      legend.cex = 0.3, layoutOffset = -0.15, vsize = 5, groups = Groups,
      theme = 'colorblind', labels = Labels, maximum = 1,
      title = "Netwerk leerlingen")
```

```
pdf("1406022_netwerk_lln_na_resample.pdf")
plot(Plot_Network_imp_max_lln_resample)
dev.off()
```

10.2.3 Characteristics of included variables

	Mean (SD)	Observed range	Number of measurements ^c	Percentage missing values
Sleep				
Sleep onset latency (s) ^a	32.11 (38.7)	0 - 400	482	25.5
Sleep onset latency (o) ^a	15.88 (21.4)	0 - 162	177	39.8
Total sleep time (s) ^a	460.5 (103.3)	20 - 730	483	25.3
Total sleep time (o) ^a	452.57 (66.3)	207 - 567	177	39.8
Wake time after sleep onset (s) ^a	16.74 (46.1)	0 - 409	493	23.8
Wake time after sleep onset (o) ^a	17.24 (15.6)	2 - 134	177	39.8
Sleep quality (s) ^b	5.55 (1.8)	1 - 10	499	22.9
Sleep hygiene				
Physical exercise (s) ^b	4.75 (2.3)	1 - 10	382	41.0
Physical exercise (p) ^b	4.60 (2.2)	1 - 10	307	52.6
Physical exercise (t) ^b	4.93 (2.1)	1 - 10	192	48.5
Mental effort (s) ^b	5.73 (2.2)	1 - 10	382	41.0
Mental effort (p) ^b	5.43 (1.8)	1 - 10	307	52.6
Mental effort (t) ^b	5.68 (1.6)	1 - 9	192	48.5
Time of caffeine intake before bedtime (s) ^a	308.29 (114.5)	-120 - 360	495	32.5
Time of intensive physical exercise before bedtime (s) ^a	207.66 (18.3)	15 - 210	498	23.0
Time of gaming before bedtime (s) ^a	218.02 (63.64)	-30 - 240	500	22.7
Time of electronic device use before bedtime (s) ^a	36.25 (81.8)	-200 - 780	492	24.0
Daytime functioning				
Daytime sleepiness (s) ^b	5.59 (2.2)	1 - 10	391	39.6
Daytime sleepiness (p) ^b	5.77 (2.1)	1 - 10	313	51.6
Daytime sleepiness (t) ^b	4.84 (2.1)	1 - 10	194	48.1
Stress (s) ^b	4.41 (2.2)	1 - 10	392	39.4
Stress (p) ^b	4.74 (2.3)	1 - 10	313	51.6
Stress (t) ^b	3.89 (2.1)	1 - 9	193	48.3
Concentration (s) ^b	5.44 (2.1)	1 - 10	392	39.4
Concentration (p) ^b	5.49 (1.7)	1 - 9	313	51.6
Concentration (t) ^b	5.54 (1.8)	1 - 9	194	48.1
Mood (s) ^b	6.13 (2.0)	1 - 10	392	39.4
Mood (p) ^b	6.37 (1.8)	1 - 10	313	51.6
Mood (t) ^b	6.40 (1.5)	2 - 10	194	48.1

s = self-reported, o = objectively measured, p = parent-reported, t = teacher-reported.

^a Measured in minutes. For time of caffeine intake, gaming and, electronic device use before bedtime the value is negative when the teenagers does the activity in bed

^b Possible range 1 - 10

^c Based on different number of participants; N = 31 for self-reports of sleep, N = 14 for objectively measured sleep, N = 27 for parent-reports, and N = 31 for teacher-reports

10.2.4 Correlations for daytime functioning reports of teenagers, parents and teachers

	Day (s)	Con (s)	Mood (s)	Day (p)	Con (p)	Mood (p)	Day (t)	Con (t)	Mood (t)
Con (s)	-.32*								
Mood (s)	-.32*	.50*							
Day (p)	.40*	-.43*	-.39*						
Con (p)	-.15*	.35*	.23*	-.42*					
Mood (p)	-.08	.18*	.36*	-.32*	.43*				
Day (t)	.13	-.22*	-.37*	.35*	-.13	-.17			
Con (t)	-.16	.28*	.34*	-.05	.13	.08	-.44*		
Mood (t)	-.20*	.40*	.53*	-.21*	.30*	.13	-.39*	.55*	

Day = daytime sleepiness, Con = concentration, Mood = mood, s = self-reported, p = parent-reported, t = teacher-reported

* $p \leq .05$

10.3 Supplementary material accompanying Chapter 4

10.3.1 Description of the co-created sleep intervention

The sleep intervention consists of three parts: 1) monitoring the teenagers' sleep and daytime functioning, 2) holding a sleep consultation with a teacher based on the monitoring, and 3) implementing the sleep recommendations. The sleep intervention followed a modular approach which enabled us to focus on each teenager's specific sleep problem. To get insight into that problem, the teenager's sleep was monitored using a sleep diary. This sleep diary consists of questions about bedtime, sleep onset time, wake time after sleep onset, and wake time and last night's activities: physical exercise, gaming, use of electronic devices, and caffeine (Carney et al., 2012). To evaluate the association between daytime functioning/activities and sleep, the teenagers also rated the following aspects: daytime sleepiness, stress, concentration, mood, mental effort, and physical exercise.

After the monitoring, the teenager had a sleep consultation with a trained teacher during which they discussed the sleep pattern and daytime functioning/activities. The teacher offered sleep recommendations that aligned with the teenager's interests, possibilities, and preferences (Pavlopoulou, 2020). The sleep recommendations focus on psychoeducation, sleep hygiene, alignment of the biological clock, dysfunctional thoughts and behaviors, and stress. At the end of the sleep consultation, the teenager received a log file in which they were asked to note facilitators of and barriers to implementing the sleep recommendations. These facilitators and barriers were discussed in subsequent consultations – as were the new sleep patterns – to determine whether the sleep recommendations should be modified.

We trained the teachers to analyze the sleep diaries, identify the causes of sleep problems, and formulate sleep recommendations related to the cause. The training started with information about normal sleep and was followed by five modules: psychoeducation, sleep hygiene, alignment of the biological clock, dysfunctional thoughts and behaviors, and stress. Each module included background information and related sleep recommendations. The modules concluded with a case to apply the knowledge.

10.4 Supplementary material accompanying Chapter 5

10.4.1 Descriptive data of questionnaires and grades

Measure (n = pre/post/FU)	Pretest		Post-test		Follow-up (FU)	
	M (SD)	Range	M (SD)	Range	M (SD)	Range
ISI ^a (n = 23/15/19)	13.4 (4.2)	8 - 22	9.8 (6.0)	2 - 20	8.2 (6.1)	1 - 22
CSHQ ^b (n = 17/12/10)	48.8 (9.5)	40 - 68	44.6 (8.1)	33 - 59	42.3 (8.1)	33 - 56
SDQ-Y ^c (n = 23/16/19)	14.4 (5.2)	3 - 22	14.4 (5.5)	5 - 24	12.6 (5.9)	5 - 25
SDQ-P ^c (n = 22/14/11)	16.14 (6.9)	7 - 34	16.1 (6.4)	8 - 31	17.1 (7.1)	7 - 33
SDQ-T ^c (n = 22/20/14)	10.1 (5.6)	1 - 23	11.8 (6.9)	2 - 29	12.6 (7.4)	1 - 28
Grade Dutch ^d (n = 14/17/17)	7.2 (0.8)	5.8 - 9.0	7.4 (0.9)	5.8 - 9.1	7.2 (1.1)	4.9 - 9.1
Grade English ^d (n = 16/17/16)	7.5 (1.3)	5.0 - 9.4	7.6 (1.0)	6.2 - 9.3	7.0 (1.8)	2.1 - 9.1
Grade Math ^d (n = 15/15/16)	7.5 (1.0)	6.0 - 9.4	7.4 (1.3)	5.2 - 10.0	7.4 (1.3)	5.2 - 10.0

^a Possible range 0 – 28, higher score implies more sleep problems

^b Possible range 31 – 97, higher score implies more sleep problems

^c Possible range 0 – 60, higher score implies more behavioral problems

^d Possible range 1 – 10, higher score implies better performance

10.4.2 Descriptive data of secondary daily outcome measures

Measure (no. of measurements = pre/post/FU)	Baseline		Intervention period		Follow-up (FU)	
	M (SD)	Range	M (SD)	Range	M (SD)	Range
Sleep onset latency (s) ^a (no. of measurements = 230/245/57)	37.4 (48.8)	0.0 - 390.0	29.9 (34.0)	0.0 - 265.0	34.7 (52.2)	0.0 - 252.0
Sleep onset latency (o) ^a (no. of measurements = 203/146/20)	25.2 (46.4)	0.0 - 436.5	18.0 (17.7)	0.0 - 87.5	14.3 (11.5)	2.5 - 43.0
Total sleep time (s) ^a (no. of measurements = 231/246/56)	473.1 (91.2)	75.0 - 656.0	495.9 (88.0)	75.0 - 690.0	490.5 (85.2)	221.0 - 718.0
Total sleep time (o) ^a (no. of measurements = 203/146/20)	423.6 (107.0)	4.0 - 741.5	409.4 (119.0)	28.0 - 663.0	382.5 (89.9)	207.0 - 496.5
Wake time after sleep onset (s) ^a (no. of measurements = 241/248/57)	26.6 (95.3)	0.0 - 740.0	27.7 (12.5)	0.0 - 58.5	2.6 (9.6)	0.0 - 60.0
Wake time after sleep onset (o) ^a (no. of measurements = 203/146/20)	28.4 (20.7)	0.0 - 152.5	8.2 (34.4)	0.0 - 299.0	27.1 (10.6)	10.5 - 45.0
Daytime functioning (p) ^b (no. of measurements = 245/267/66)	18.7 (4.4)	6.0 - 30.0	19.3 (4.4)	4.0 - 30.0	21.2 (3.4)	14.0 - 27.0
Daytime functioning (t) ^b (no. of measurements = 134/84/26)	19.0 (4.0)	9.0 - 30.0	17.8 (4.5)	4.0 - 30.0	19.4 (5.3)	6.0 - 28.0

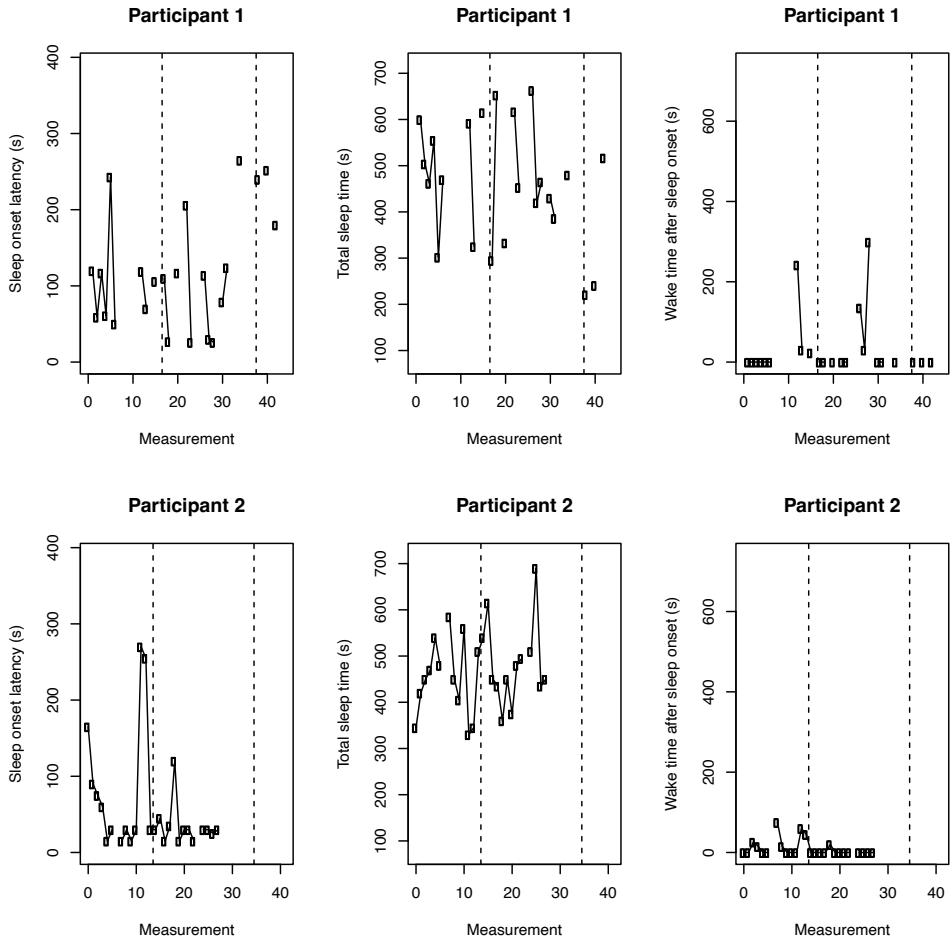
s = self-reported, o = objectively measured, p = parent-reported, t = teacher-reported

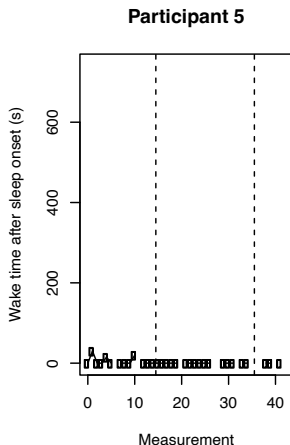
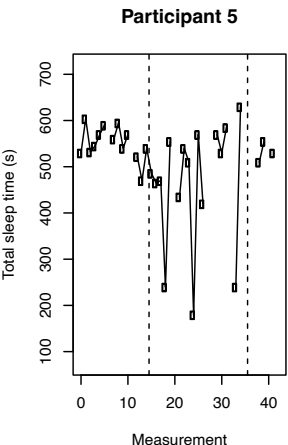
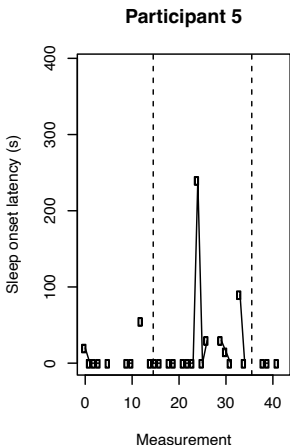
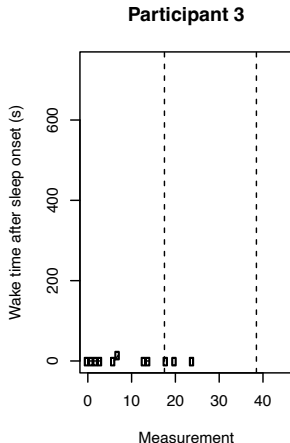
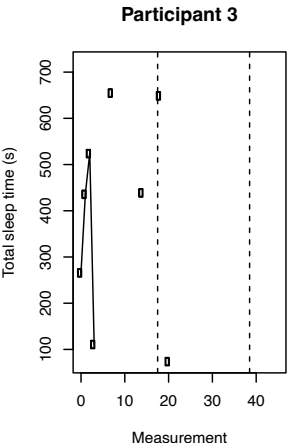
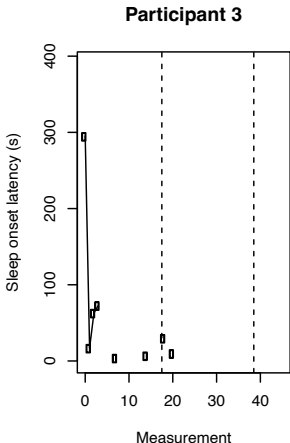
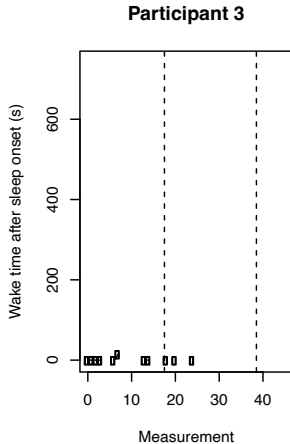
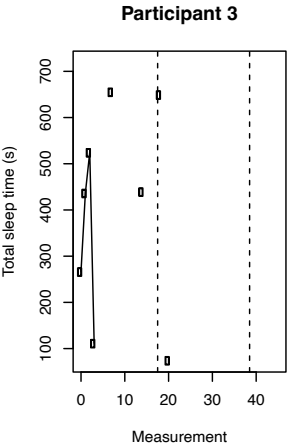
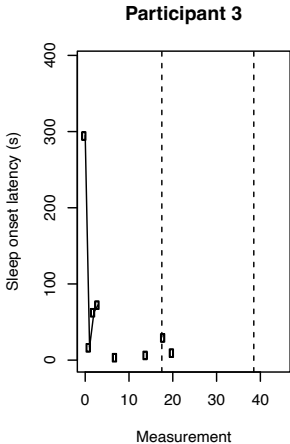
^a Measured in minutes

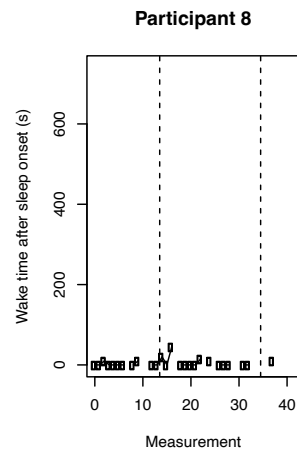
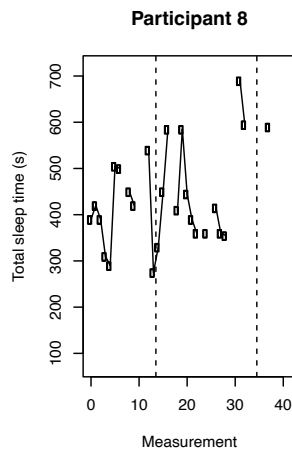
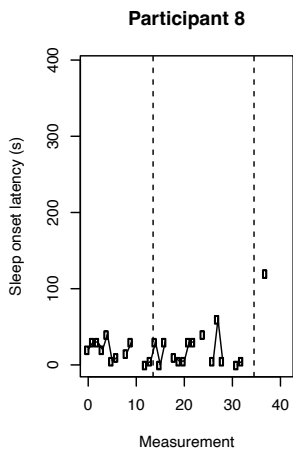
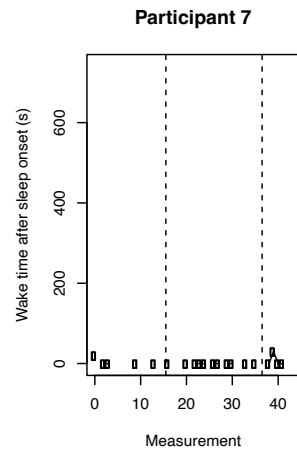
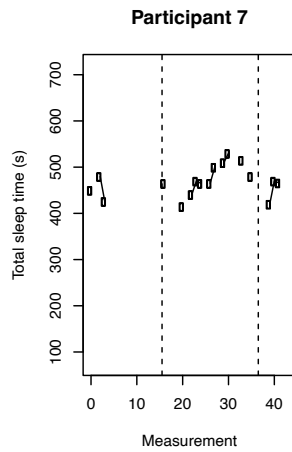
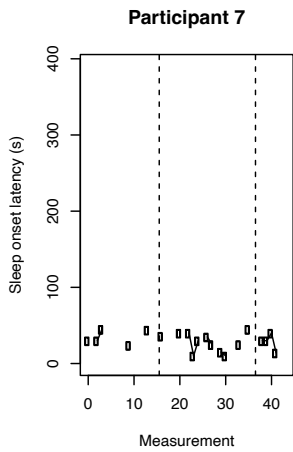
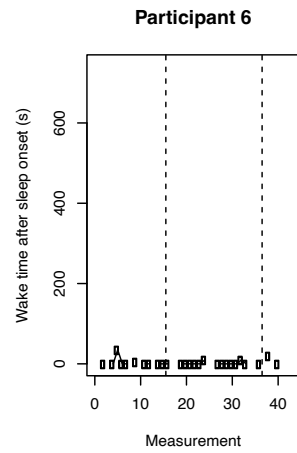
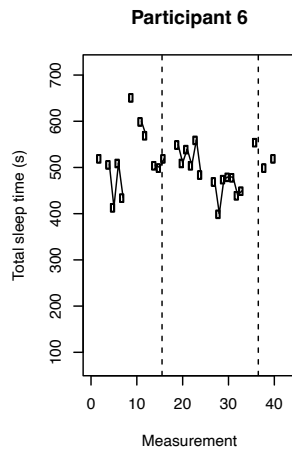
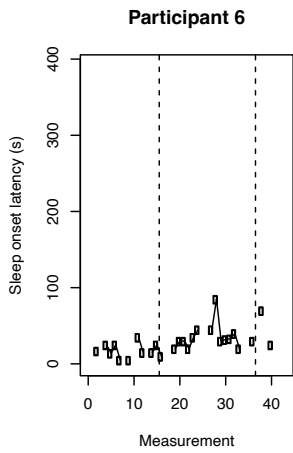
^b Possible range 3 – 30

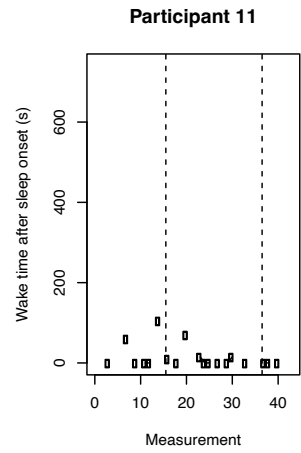
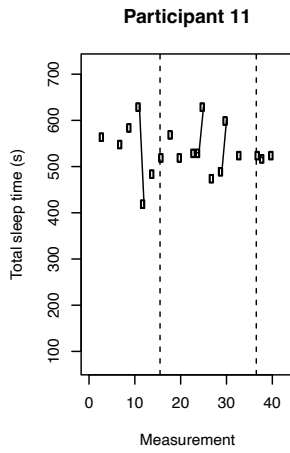
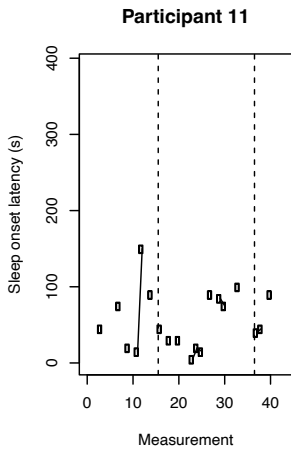
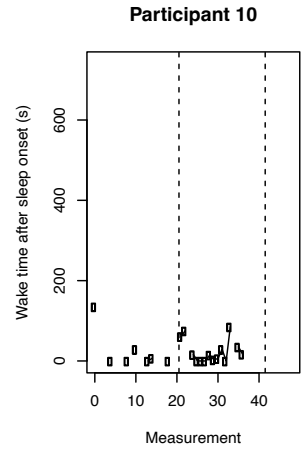
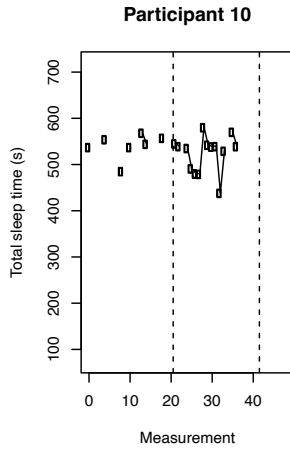
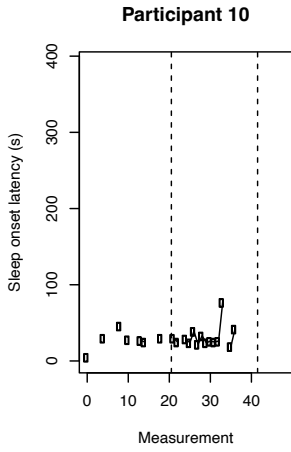
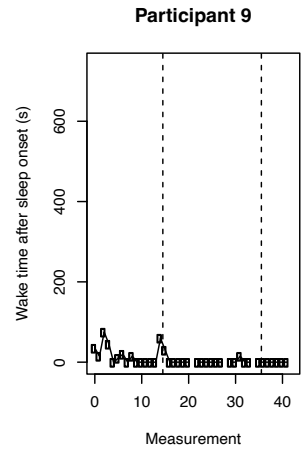
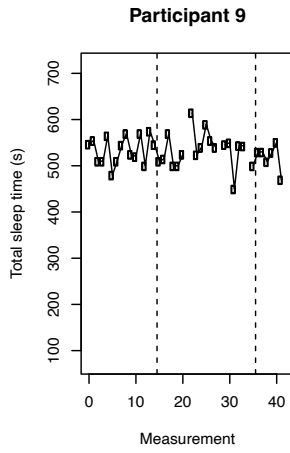
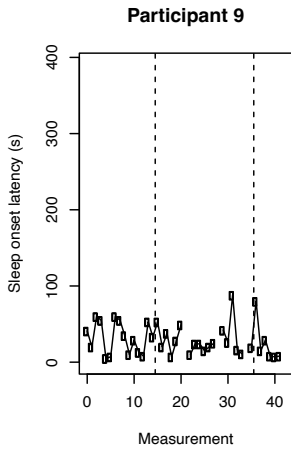
Note. Number of possible measurements differed during each measurement period (see Figure 5.2)

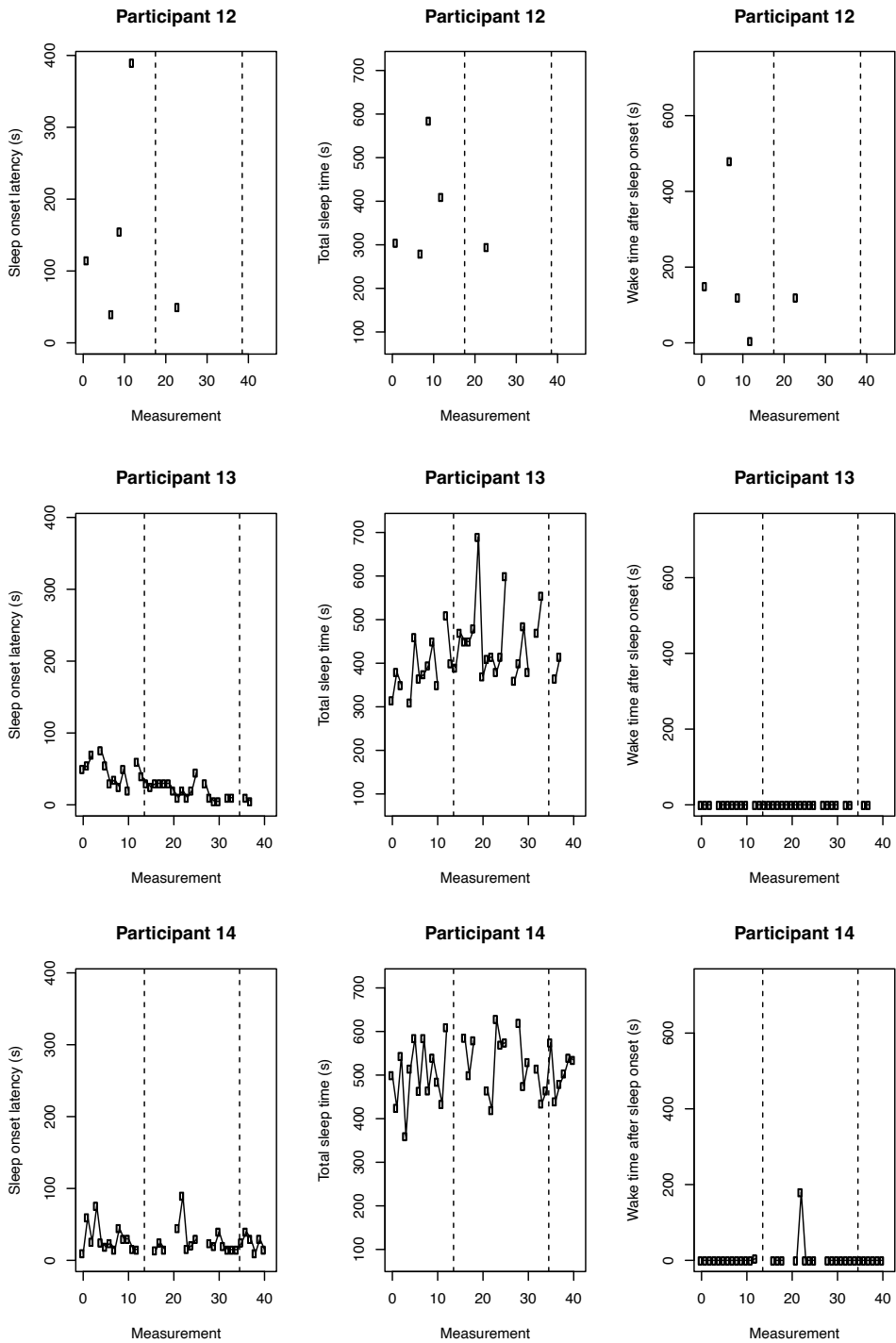
10.4.3. Self-reported secondary sleep measures

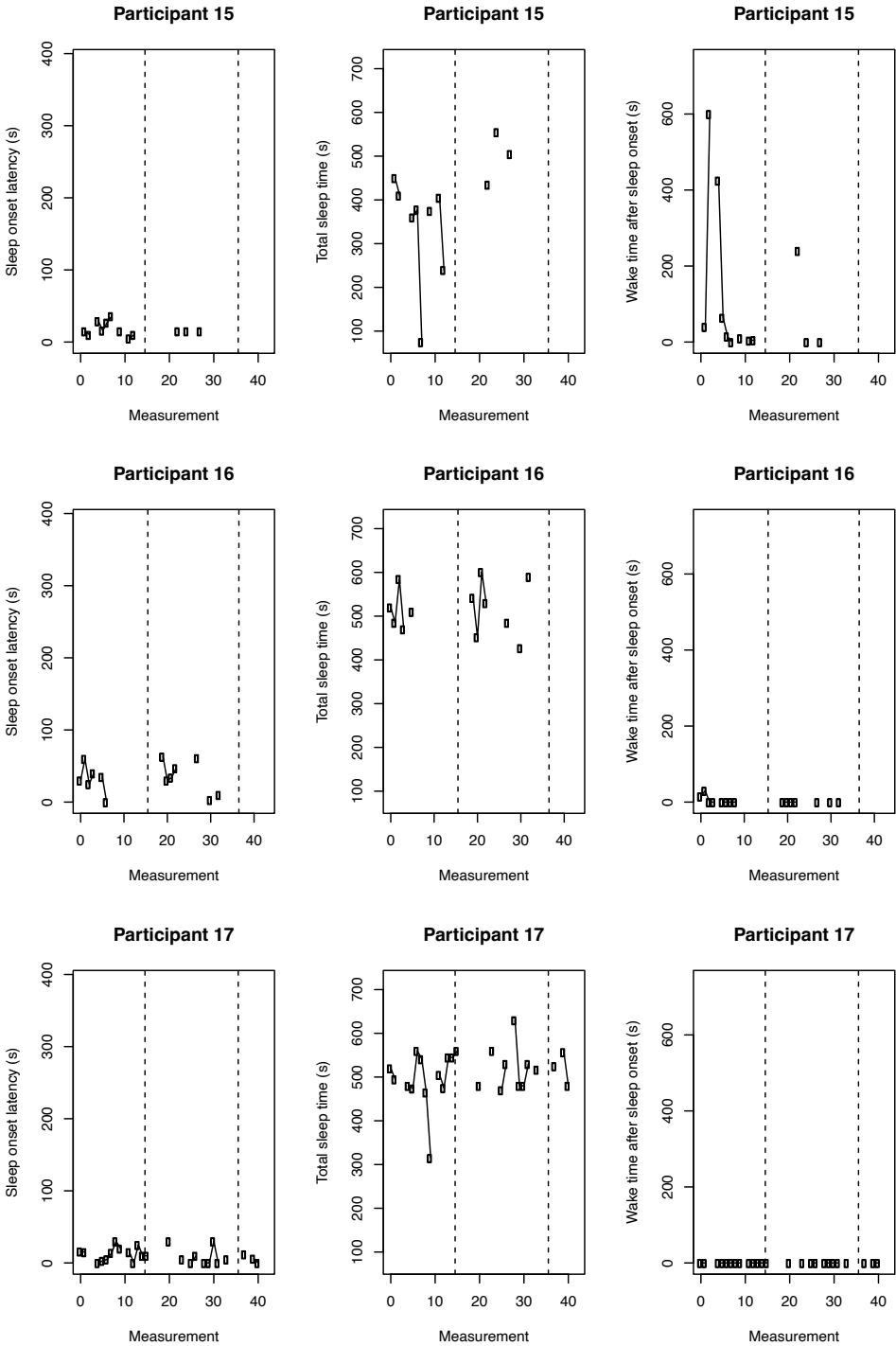


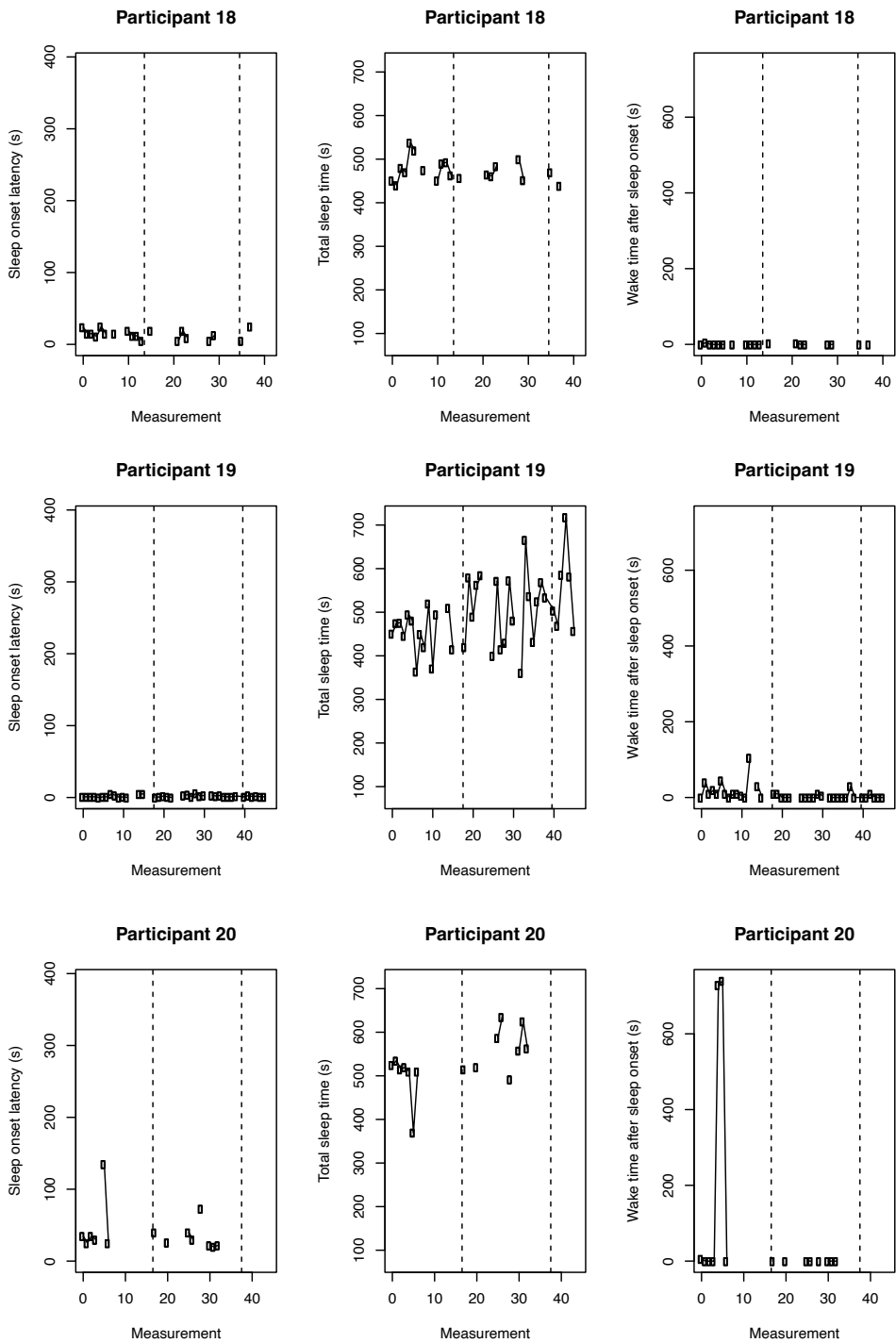


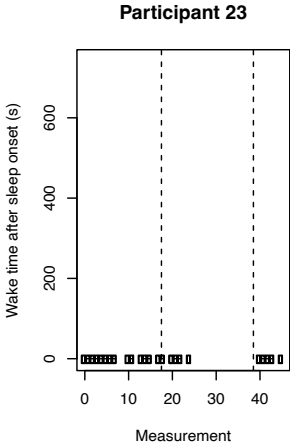
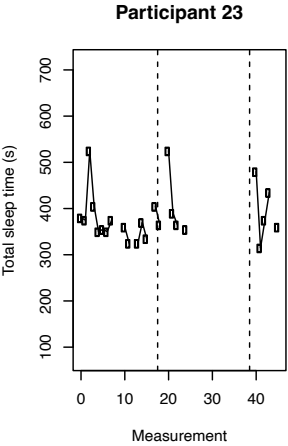
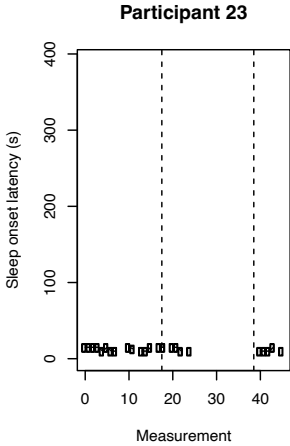
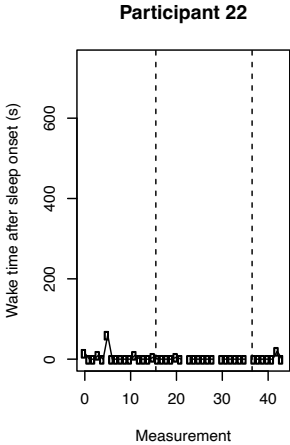
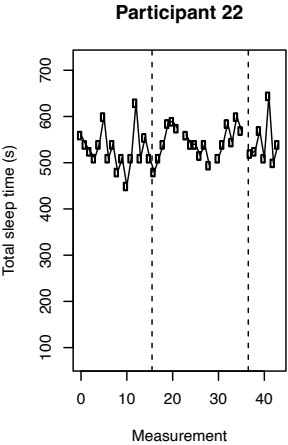
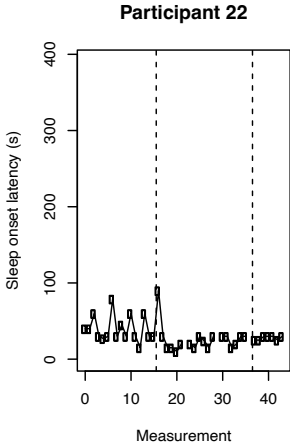
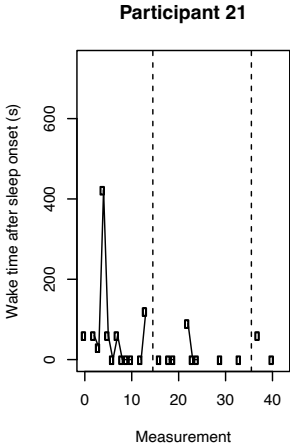
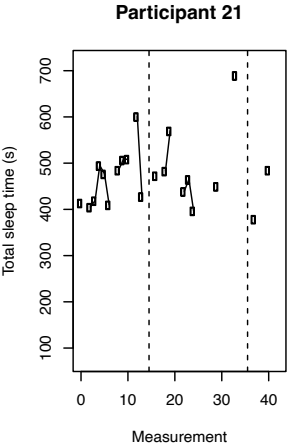
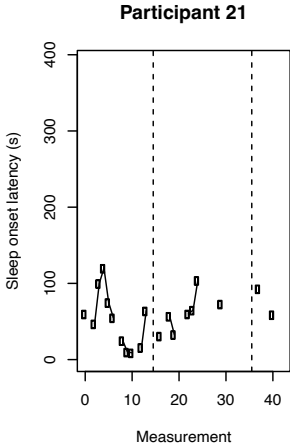




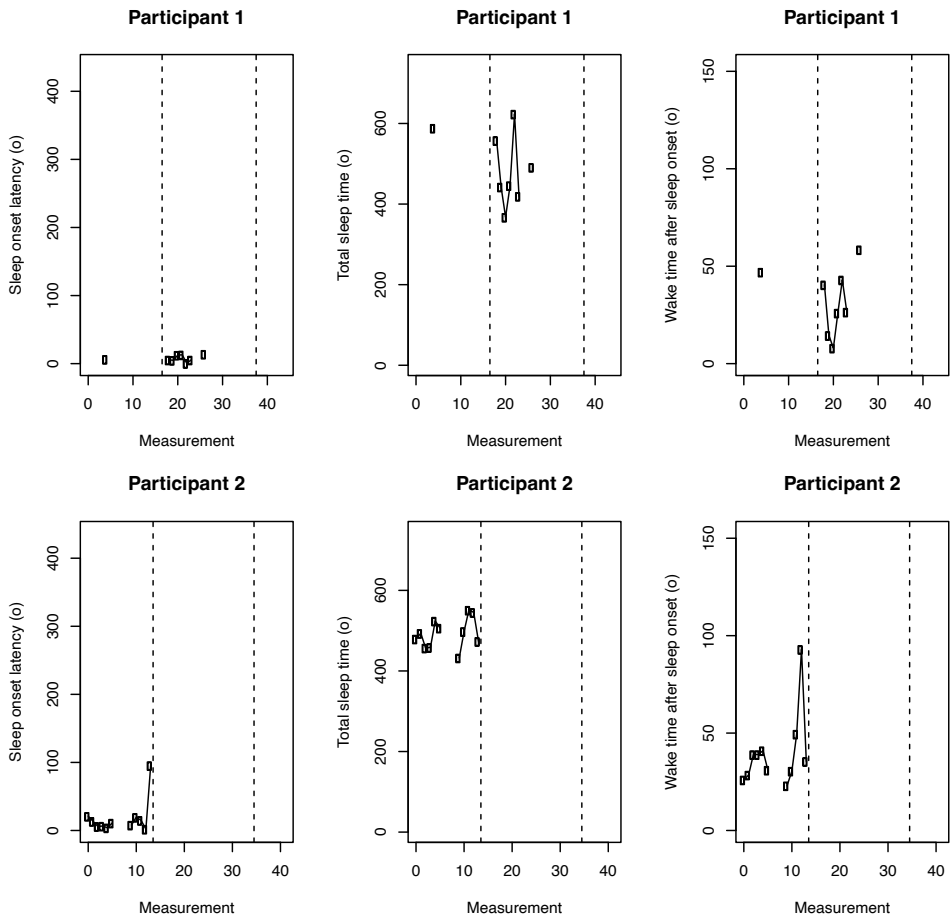


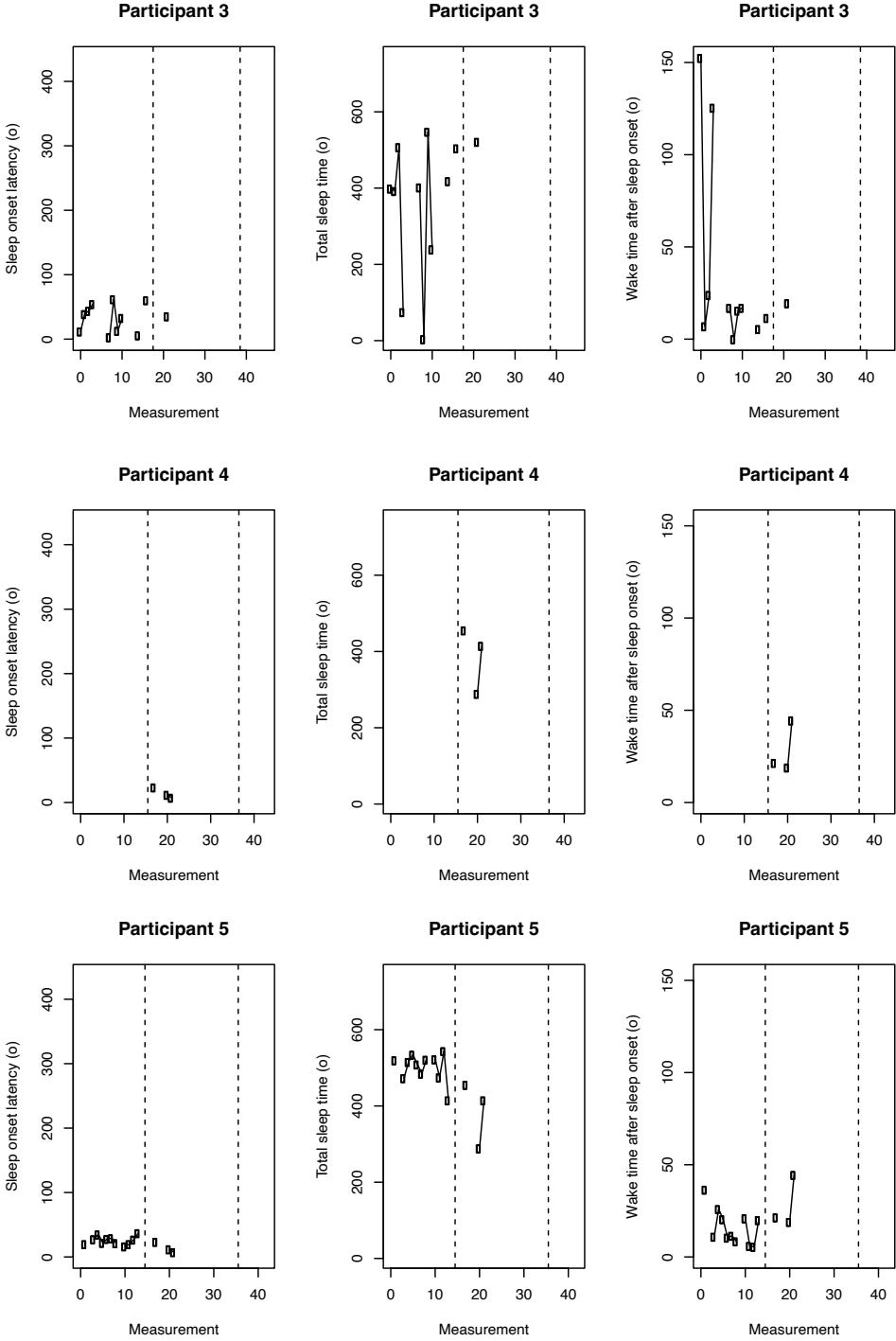


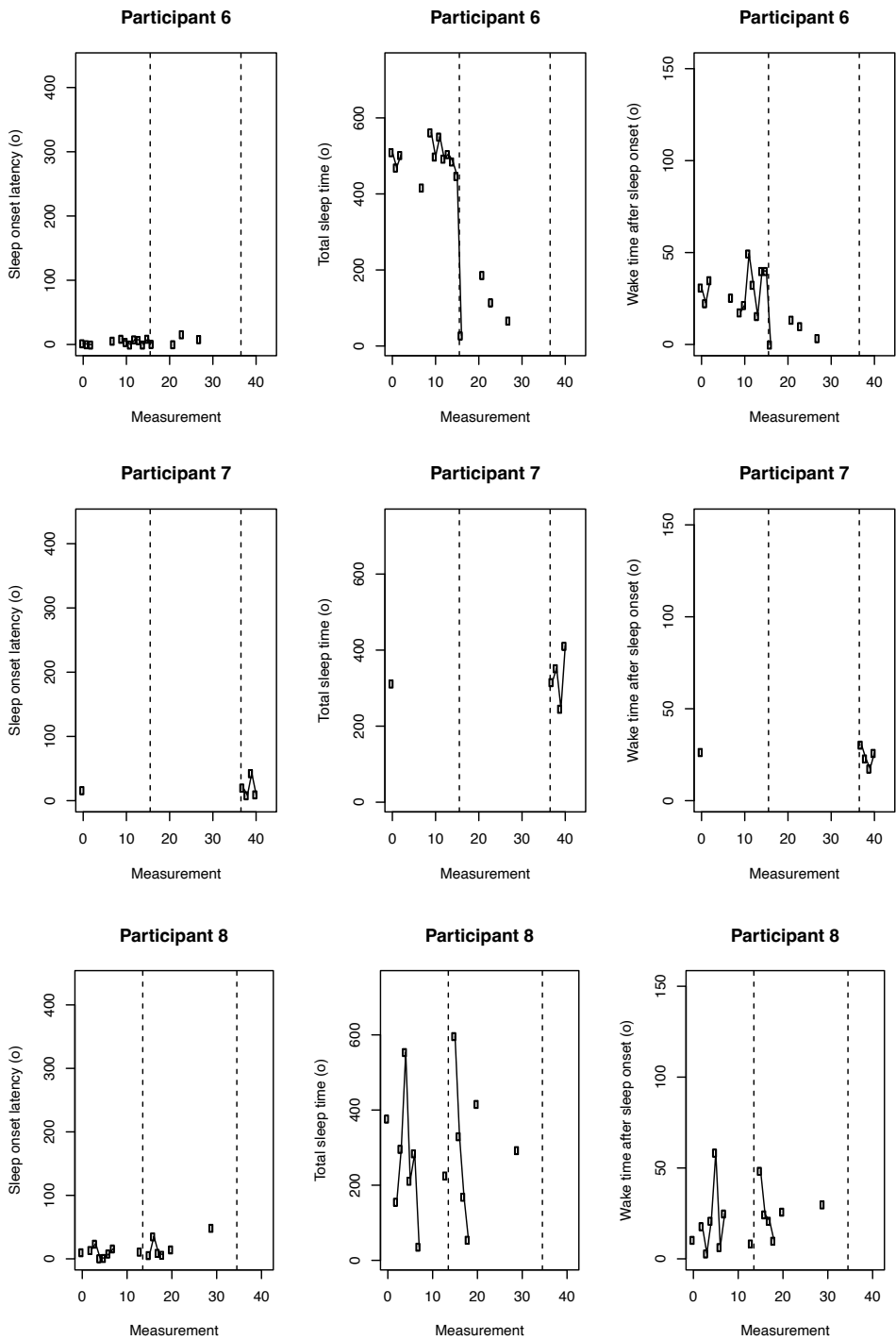




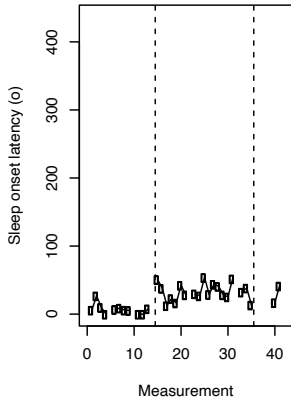
10.4.4. Objectively measured secondary sleep measures



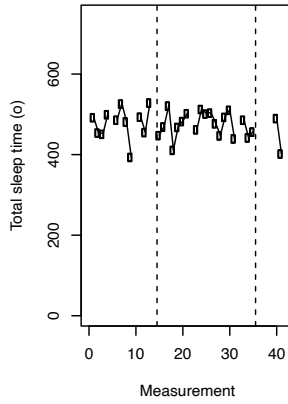




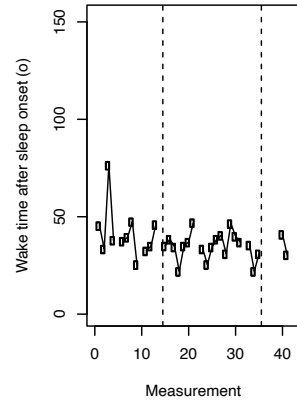
Participant 9



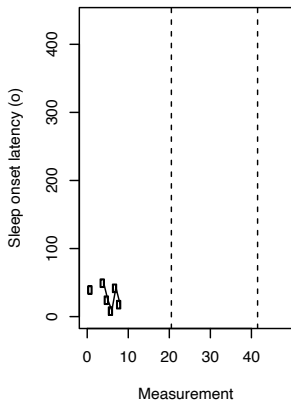
Participant 9



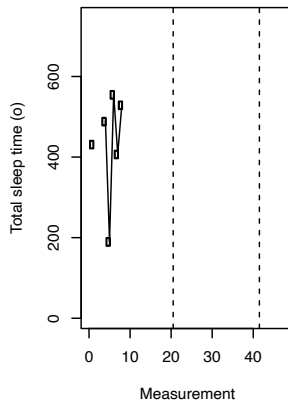
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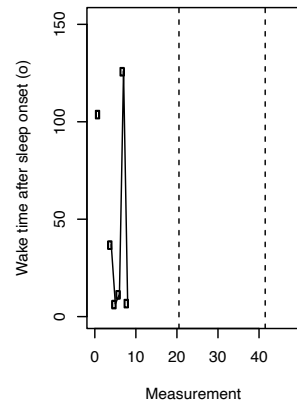
Participant 10



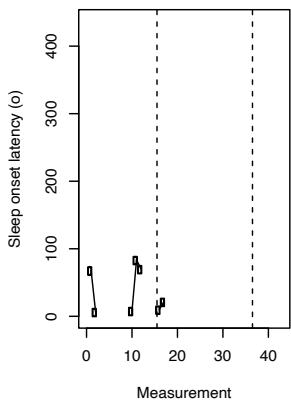
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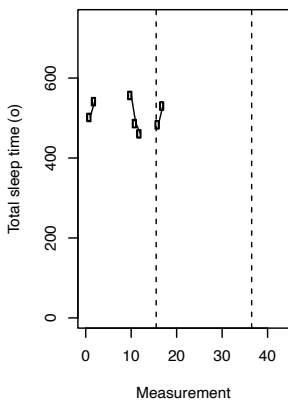
Participant 10



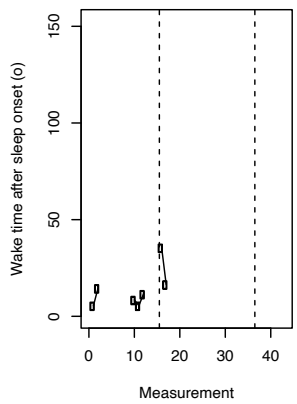
Participant 11

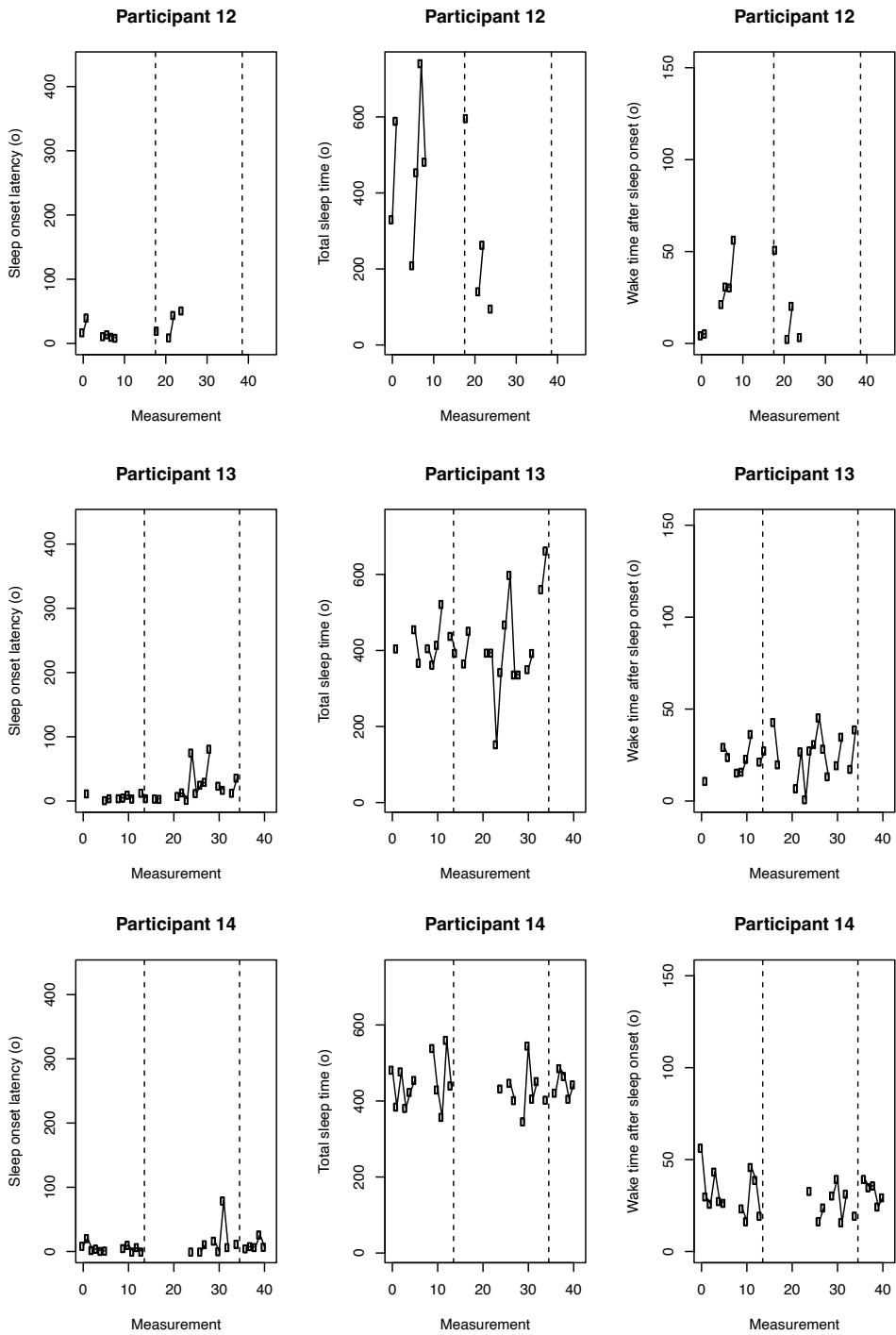


Participant 11

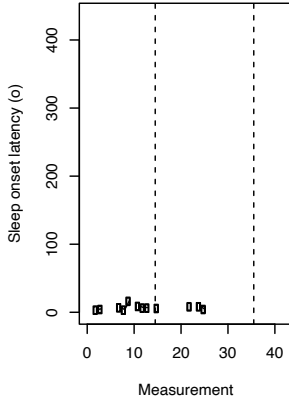


Participant 11

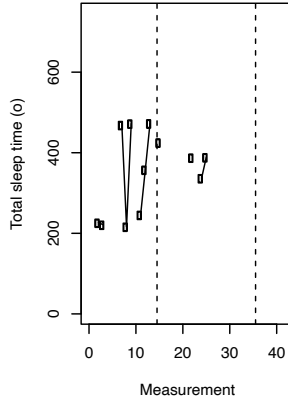




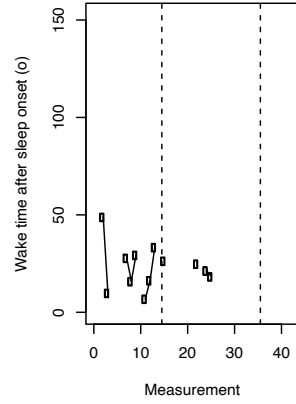
Participant 15



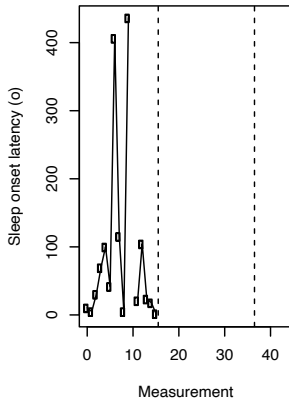
Participant 15



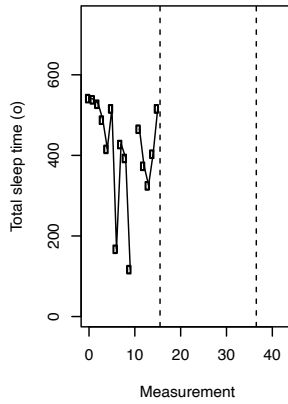
Participant 15



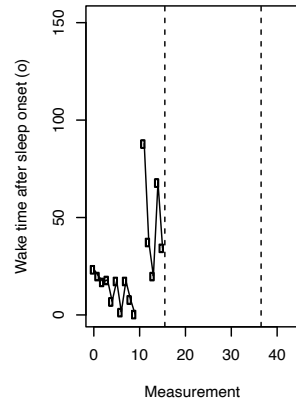
Participant 16



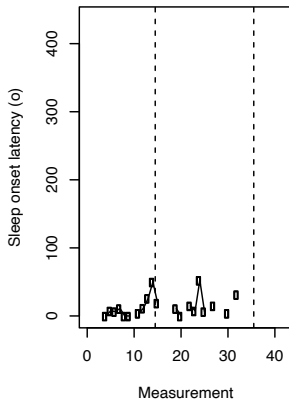
Participant 16



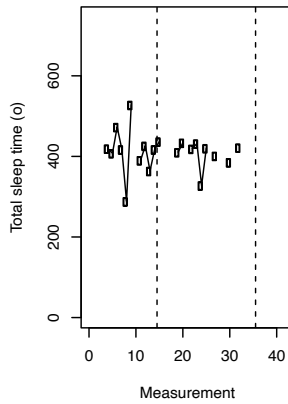
Participant 16



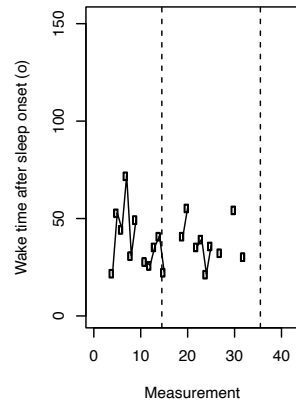
Participant 17

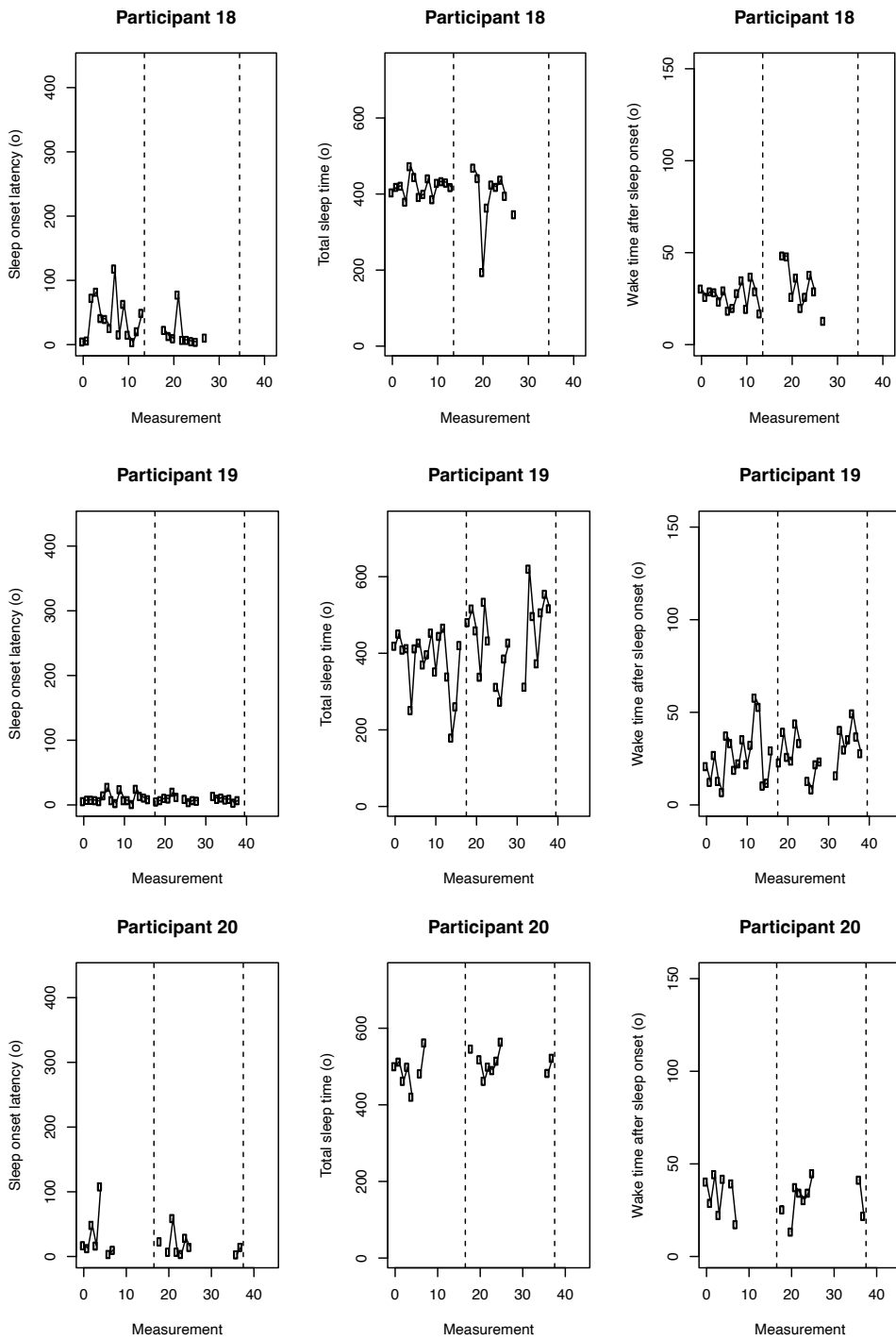


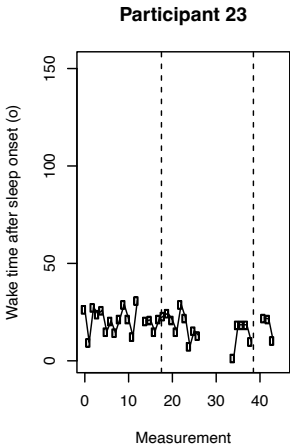
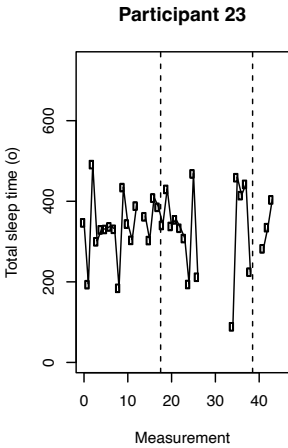
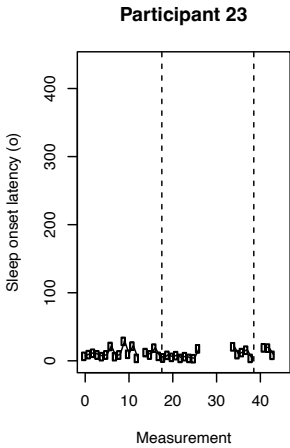
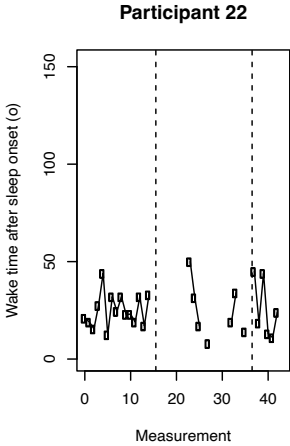
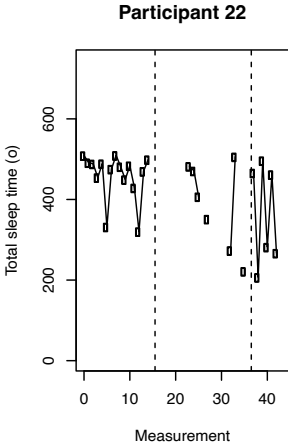
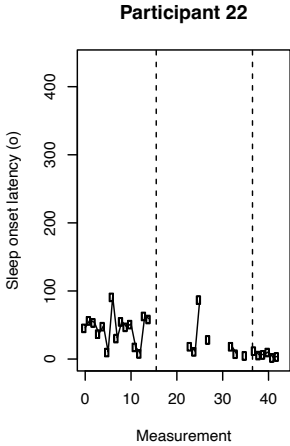
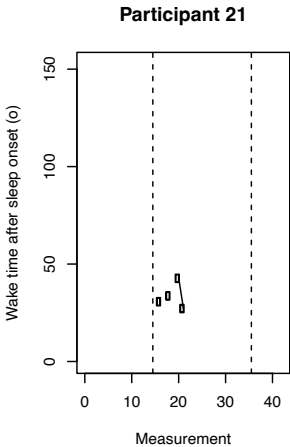
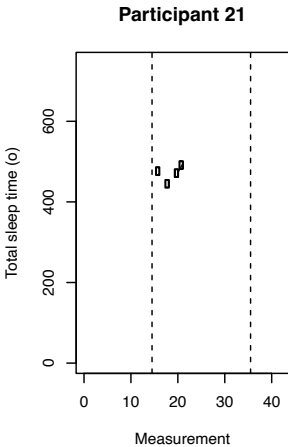
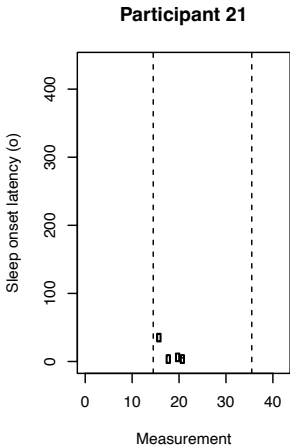
Participant 17



Participant 17

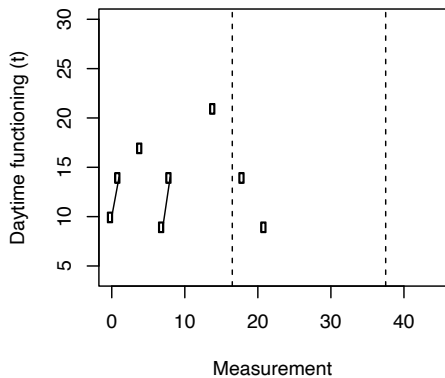




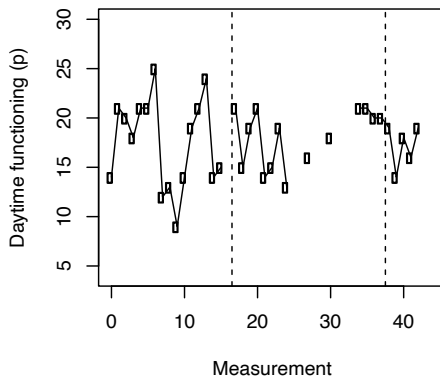


10.4.5. Parent- and teacher-reported daytime functioning

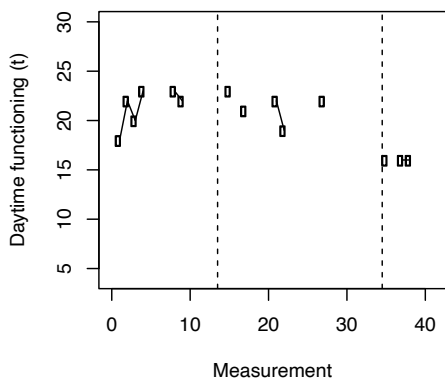
Participant 1



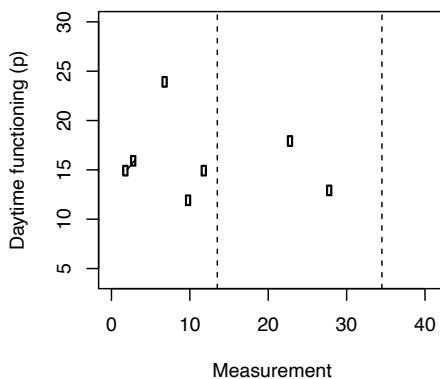
Participant 1



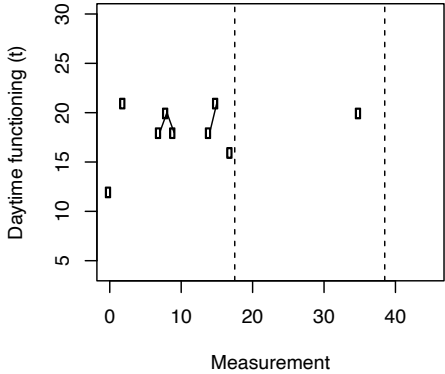
Participant 2



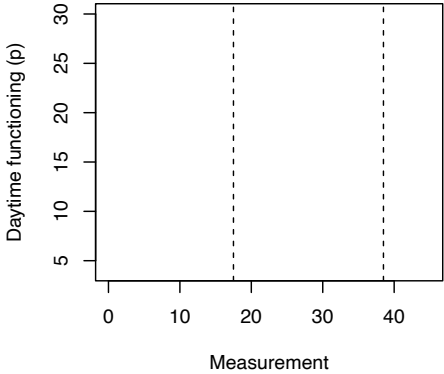
Participant 2



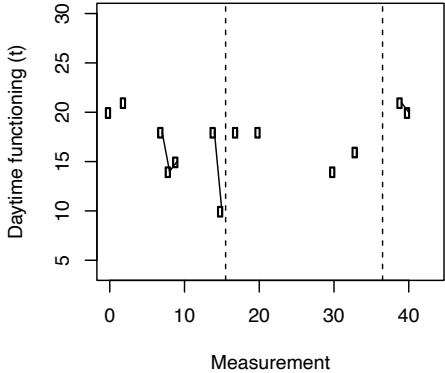
Participant 3



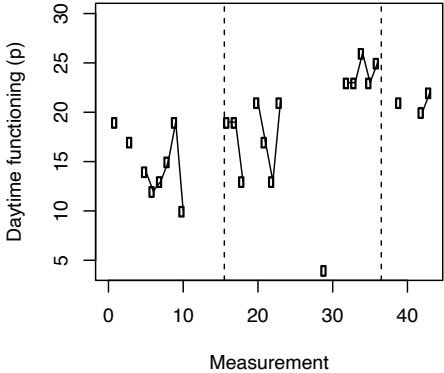
Participant 3



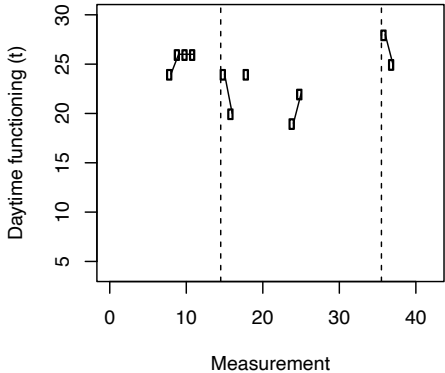
Participant 4



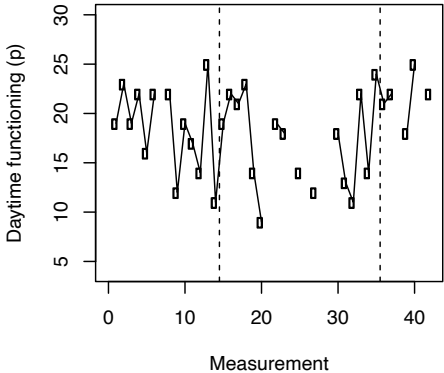
Participant 4



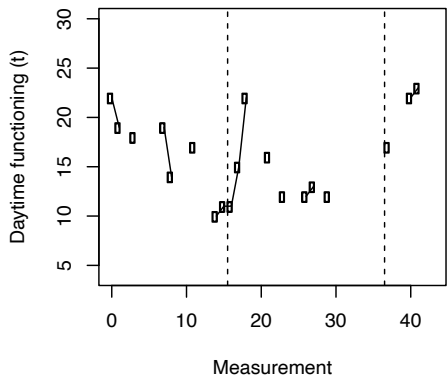
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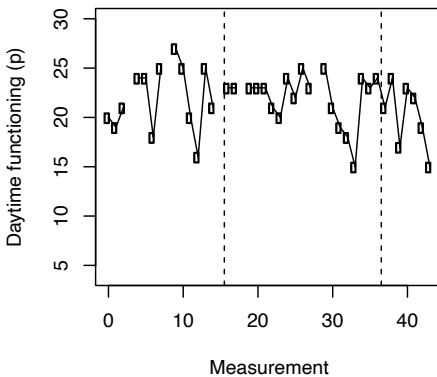
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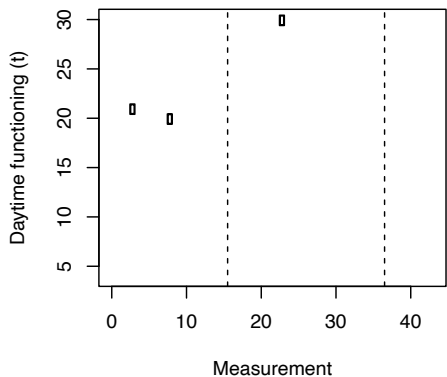
Participant 6



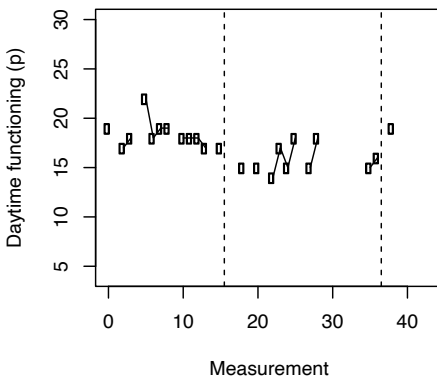
Participant 6



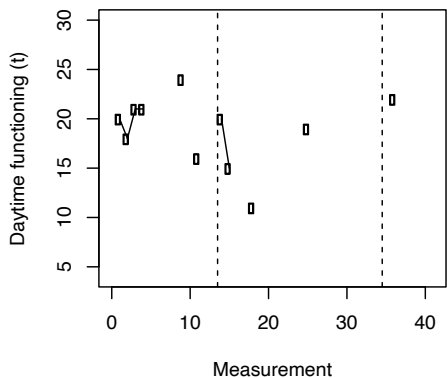
Participant 7



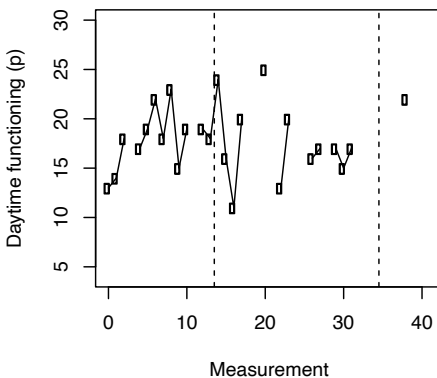
Participant 7



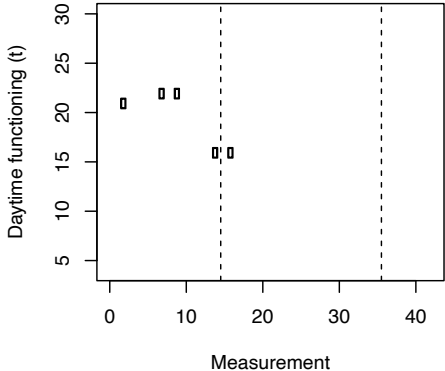
Participant 8



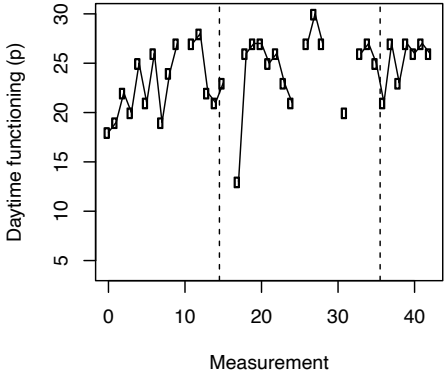
Participant 8



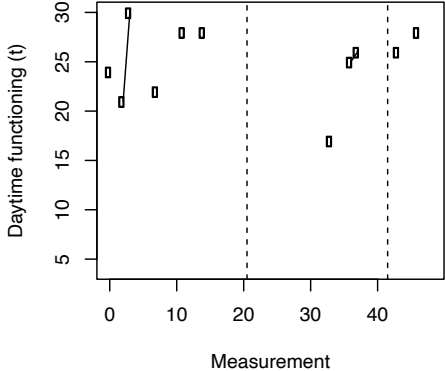
Participant 9



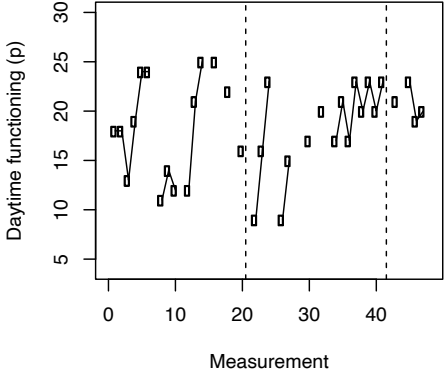
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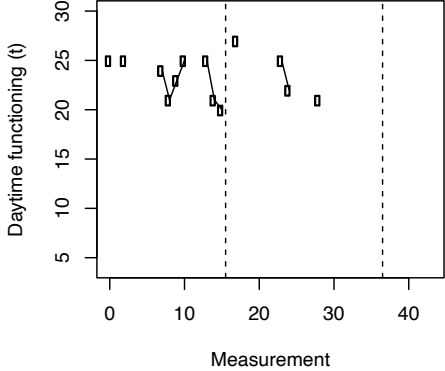
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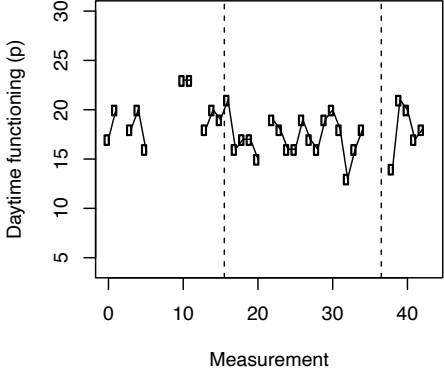
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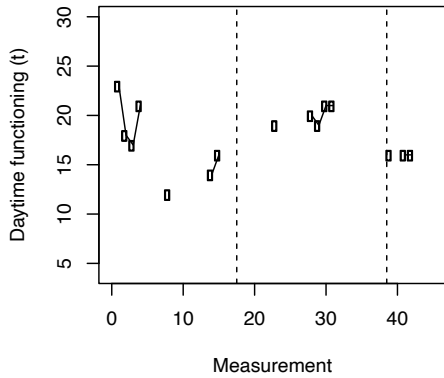
Participant 11



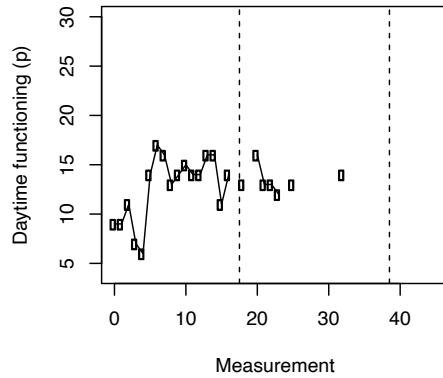
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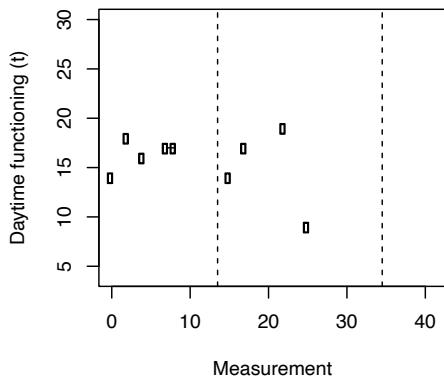
Participant 12



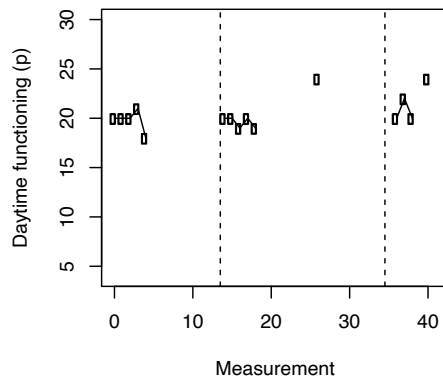
Participant 12



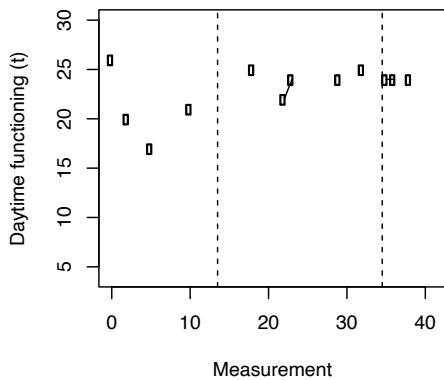
Participant 13



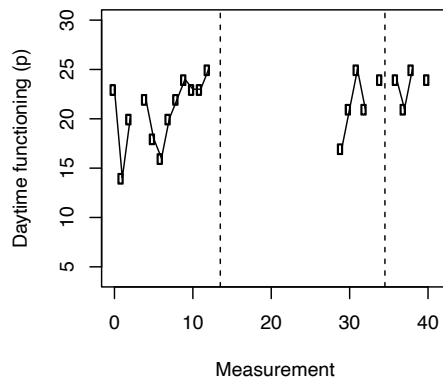
Participant 13



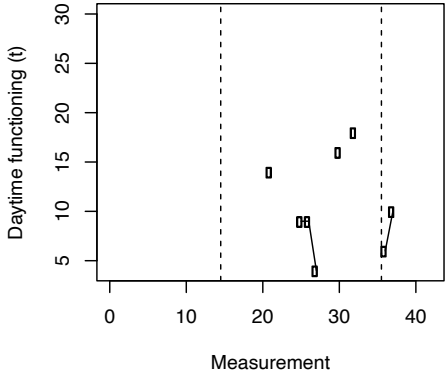
Participant 14



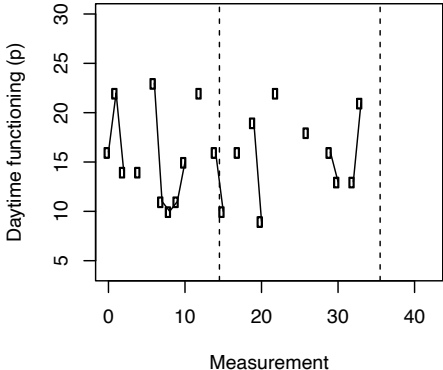
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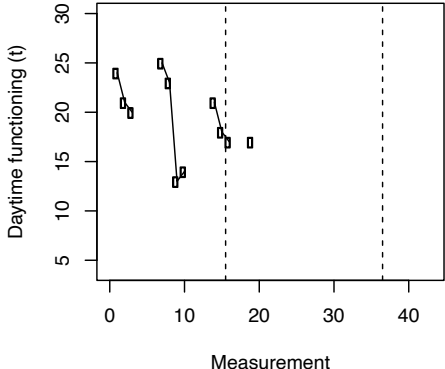
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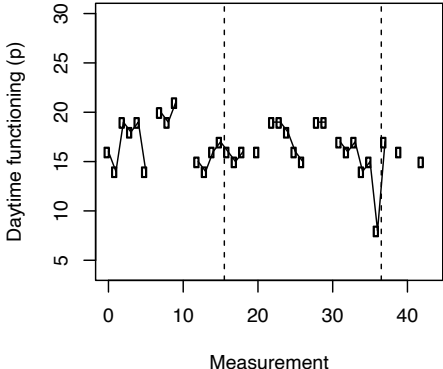
Participant 15



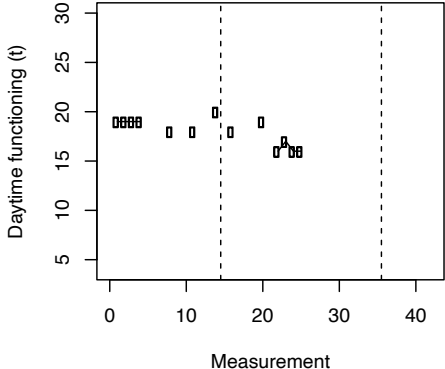
Participant 16



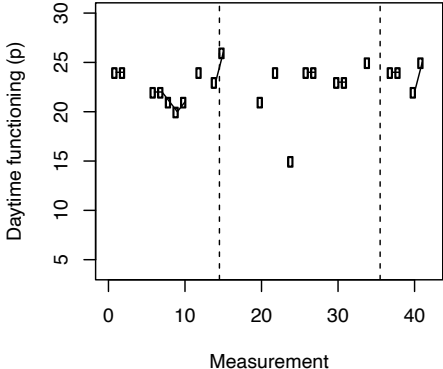
Participant 16



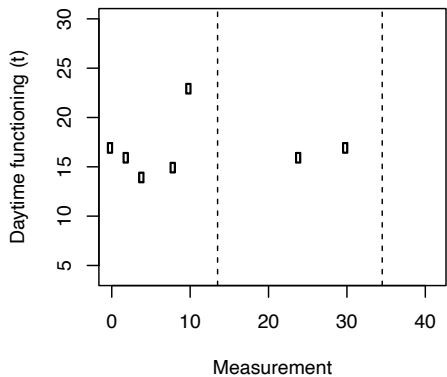
Participant 17



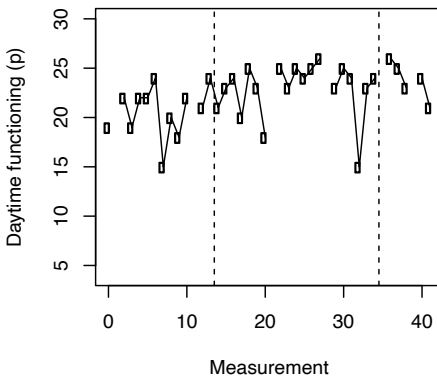
Participant 17



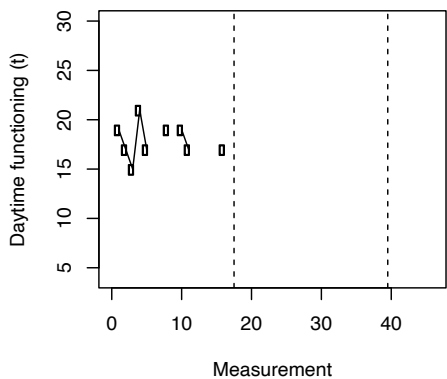
Participant 18



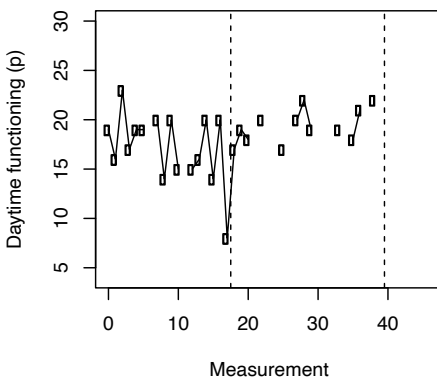
Participant 18



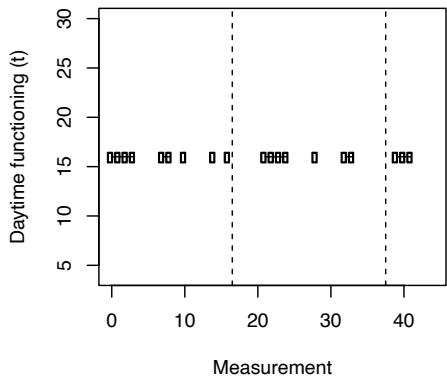
Participant 19



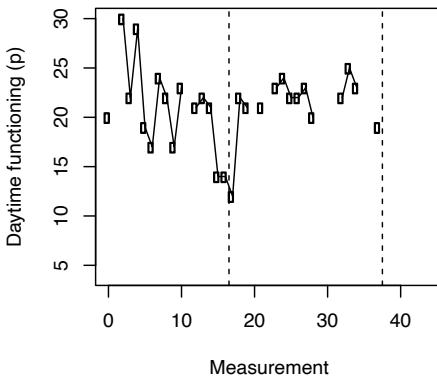
Participant 19



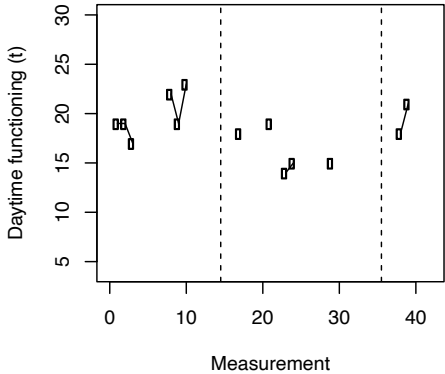
Participant 20



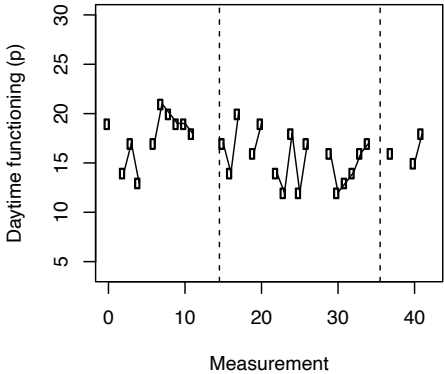
Participant 20



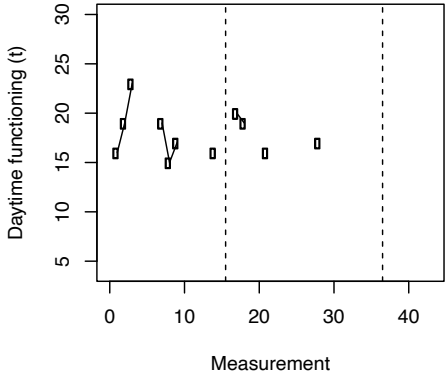
Participant 21



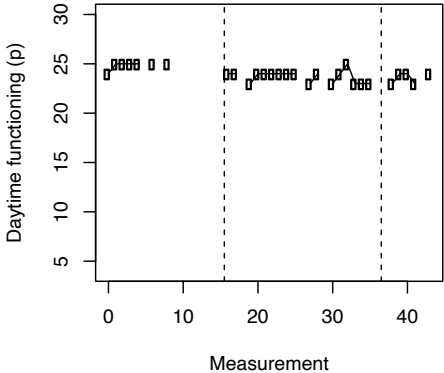
Participant 21



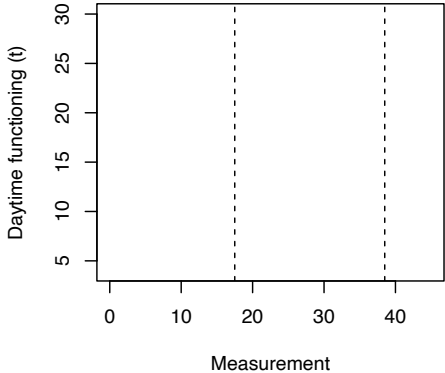
Participant 22



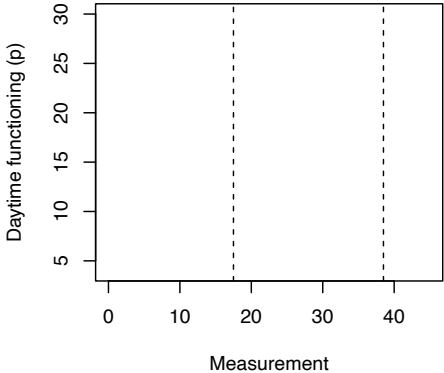
Participant 22



Participant 23



Participant 23



t = teacher-reported, p = parent-reported.



10.4.6 Results of multilevel modeling analyses of the secondary outcome measures

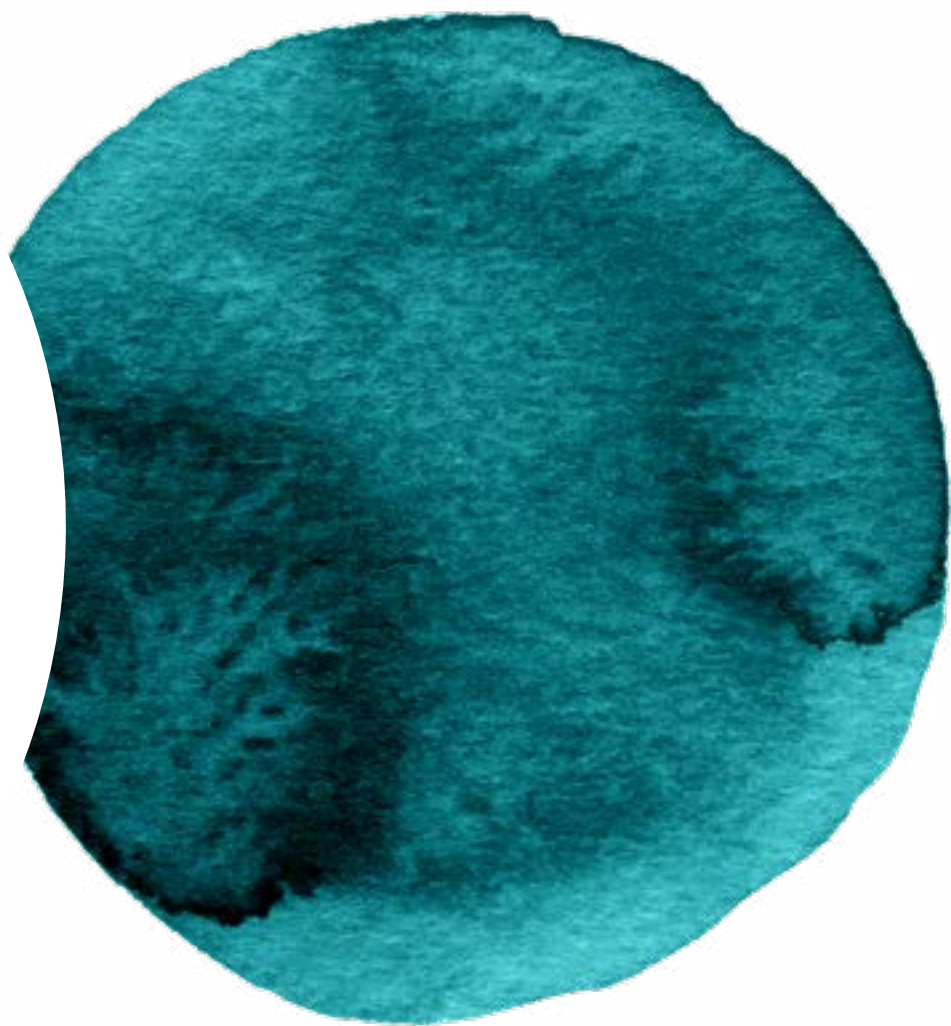
	<i>b</i>	<i>SE</i>	<i>t</i>	<i>df</i>	<i>p</i>
Sleep onset latency (s)^a					
Intercept (baseline)	39.61	3.82	10.36	55.45	<.01
Intervention	-4.38	4.50	-0.97	389.61	.33
Follow-up	2.84	6.95	0.41	43.73	.68
Time centered	-0.52	0.76	-0.69	71.91	.49
Intervention * time centered	0.62	0.99	0.63	44.58	.53
Follow-up * time centered	-0.64	2.68	-0.24	43.99	.81
Total sleep time (s)^a					
Intercept (baseline)	482.58	7.03	68.68	29.92	<.01
Intervention	2.02	8.91	0.23	62.16	.82
Follow-up	6.18	17.29	0.36	8.84	.73
Time centered	0.84	1.34	0.63	54.57	.53
Intervention * time centered	-0.32	2.09	-0.15	15.66	.88
Follow-up * time centered	1.01	6.48	0.16	9.88	.88
Wake time after sleep onset (s)^a					
Intercept (baseline)	30.25	5.47	5.53	440.47	<.01
Intervention	-9.37	7.09	-1.32	632.87	.19
Follow-up	-8.92	10.74	-0.83	64.15	.41
Time centered	-0.72	1.23	-0.59	65.83	.56
Intervention * time centered	1.13	1.41	0.80	265.30	.42
Follow-up * time centered	0.52	3.84	0.14	273.64	.89
Sleep onset latency (o)^a					
Intercept (baseline)	18.19	1.64	11.11	32.65	<.01
Intervention	0.19	1.90	0.10	121.39	.92
Follow-up	0.11	2.93	0.04	16.25	.97
Time centered	0.03	0.28	0.10	63.87	.92
Intervention * time centered	0.21	0.46	0.46	13.18	.65
Follow-up * time centered	0.46	1.78	0.26	5.88	.80
Total sleep time (o)^a					
Intercept (baseline)	429.58	14.05	30.58	6.56	<.01
Intervention	-5.43	13.01	-0.42	14.68	.68
Follow-up	-0.82	16.45	-0.05	16.53	.96
Time-centered	-0.36	2.19	-0.16	9.45	.87
Intervention * time centered	1.14	2.53	0.45	12.45	.66
Follow-up * time centered	1.81	6.52	0.28	15.14	.78
Wake time after sleep onset (o)^a					
Intercept (baseline)	27.64	1.02	27.21	195.32	<.01
Intervention	0.39	1.41	0.28	36.99	.78
Follow-up	0.34	1.96	0.17	30.55	.86
Time-centered	-0.19	0.30	-0.65	16.13	.53
Intervention * time centered	0.19	0.38	0.51	12.39	.62
Follow-up * time centered	0.53	0.69	0.77	185.62	.44

Daytime functioning (p)					
Intercept (baseline)	18.93	0.46	41.24	28.85	<.01
Intervention	0.25	0.48	0.52	325.02	.60
Follow-up	0.57	0.73	0.78	50.37	.44
Time-centered	-0.01	0.07	-0.08	671.71	.94
Intervention * time centered	0.03	0.10	0.30	101.23	.76
Follow-up * time centered	0.03	0.22	0.12	642.47	.90
Daytime functioning (t) ^b					
Intercept (baseline)	18.78	0.41	46.19	49.82	<.01
Intervention	-0.25	0.61	-0.41	18.90	.68
Follow-up	-0.24	0.77	-0.31	28.79	.76
Time-centered	-0.09	0.10	-0.91	14.96	.38
Intervention * time centered	0.12	0.14	0.87	11.64	.40
Follow-up * time centered	0.08	0.23	0.33	526.16	.47

s = self-reported, o = objectively measured, p = parent-reported, t = teacher-reported

^a Measured in minutes

^b Possible range from 3 – 30



Chapter 11

Funding and author contributions

This thesis project was supported by The Taskforce for Applied Research SIA (foundation for innovation) awarded to HAN University of Applied Sciences under Grant RAAK.PRO 02.150.

Chapter 2

Schouwenaars, I. M. G., Magnée, M. J. C. M., Geurts, H. M., van Bennekom, C. A. M., Pillen, S., & Teunisse, J. P. (2021). *Sleep problems and daytime functioning in adolescents with an autism spectrum disorder diagnosis: A systematic review*. PsyArXiv. <https://doi.org/10.31234/osf.io/hjy6p>

IS, MM, HG, CvB and JPT developed the design for the study. IS collected and analyzed the data and wrote the paper. MM, HG, CvB, SP and JPT supervised the analysis and revised the paper.

Chapter 3

Schouwenaars, I. M. G., Magnée, M. J. C. M., Geurts, H. M., van Bennekom, C. A. M., Pillen, S., Waldorp, L.J., Blanken, T.F., & Teunisse, J. P. *A Psychological Network Approach to Sleep and Daytime Functioning in Autistic Teenagers*. [Manuscript submitted for publication]. Research Centre for Social Support and Community Care, HAN University of Applied Sciences

We preregistered the analysis plan for this study at AsPredicted (#34594; https://aspredicted.org/BRG_VMD).

IS, MM, HG, CvB and JPT developed the design for the study. IS, LW and TB deliberated on the statistical analyses. IS collected and analyzed the data and wrote the article. LW and TB supervised the statistical analysis and revised the paper. MM, HG, CvB, SP and JPT supervised the interpretation of the data and revised the paper.

Chapter 4

Schouwenaars, I., Magnée, M., Geurts, H., van Bennekom, C., Pillen, S., & Teunisse, J. (2022). *Using the CeHReS roadmap to develop a sleep intervention for autistic students*. PsyArXiv. <https://doi.org/10.31234/osf.io/h3a4b>

IS, MM, HG, CvB, SP and JPT developed the design for the study. IS collected and analyzed the data and wrote the paper. MM, HG, CvB, SP and JPT supervised the analysis and revised the paper.

Chapter 5

Schouwenaars, I.M.G., Magnée, M.J.C.M., Geurts, H.M., van Bennekom, C.A.M., Pillen, S., Deen, M., & Teunisse, J. (2023). *A School-Based Sleep Intervention for Autistic Teenagers: Effects on Sleep Quality and Daytime Functioning*. PsyArXiv. <https://doi.org/10.31234/osf.io/dz5uf>

We preregistered the analysis plan for this study at AsPredicted (#66893; https://aspredicted.org/XFK_HHG)
IS, MM, HG, CvB, SP and JPT developed the design for the study and deliberated on the statistical analyses. IS collected and analyzed the data and wrote the paper. MD supervised the statistical analysis and revised the paper. MM, HG, CvB, SP and JPT revised the paper.

