SLEEP IN CHILDREN WITH AUTISM SPECTRUM DISORDER
LONGITUDINAL AND CROSS-SECTIONAL INVESTIGATIONS

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BA (Hons) Psychology

A thesis submitted to The University of Western Australia in fulfilment of the requirements for the degree of Doctor of Philosophy in the discipline of Psychology, and in partial completion of the requirements for the Master of Clinical Neuropsychology degree.

School of Psychological Science, The University of Western Australia
Cognition, Autism and Neurodevelopment Laboratory
May 2019
Thesis Declaration

I, Amelia Anne Host, certify that:

This thesis has been substantially accomplished during enrolment in the degree. This thesis does not contain material which has been submitted for the award of any other degree or diploma in my name, in any university or other tertiary institution.

No part of this work will, in the future, be used in a submission in my name, for any other degree or diploma in any university or other tertiary institution without the prior approval of The University of Western Australia and where applicable, any partner institution responsible for the joint-award of this degree.

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The research involving human data reported in this thesis was assessed and approved by The University of Western Australia Human Research Ethics Committee. Approval # RA/4/1/6487; Curtin University Human Research Ethics Office. Approval # HR123/2014. Written consent has been received and archived for the research involving participant data reported in this thesis. Prior to commencing the relevant work described in this thesis, approval was also obtained from the Catholic Education Office of Western Australia.

The work described in this thesis was funded by the School of Psychological Science of The University of Western Australia. Data from the Val Lishman Project at Curtin University (reported in Chapter 2) were funded through the Lishman Health Foundation. This research was supported by an Australian Government Research Training Program (RTP) Scholarship.

This thesis contains published work and/or work prepared for publication, some of which has been co-authored.

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Date: 20/09/2018
Abstract

Sleep is essential for social, emotional, and cognitive well-being, as well as learning and development. Sleep problems are particularly common in children with autism spectrum disorder (ASD). Within this group, poor sleep has been found to be directly associated with core clinical symptoms, as well as cognition and daytime behaviours more broadly. Despite a large volume of research addressing sleep problems in children with ASD, there remain some inconsistencies in findings across methods employed to assess sleep. Furthermore, few studies have examined the cognitive and behavioural sequelae of poor sleep in detail. To build on the existing literature, the current thesis aims to examine the tools used to assess sleep, and then investigate the relationships between sleep and daytime features of ASD using both longitudinal and cross-sectional study designs.

The first empirical study reported in this thesis (Chapter 2) is a psychometric evaluation of the most commonly used sleep questionnaire, the Children’s Sleep Habits Questionnaire (CSHQ). Participants were parents (N = 458) of children with ASD. A revised five-factor solution provided the most appropriate fit for the data (Children’s Sleep Habits Questionnaire-Revised; CSHQ-R). Overall, these five factors show some consistency with the subscales proposed in the original CSHQ. Further, correlations showed that the CSHQ-R subscales corresponded well to the subscales of another well-validated paediatric sleep scale. Overall, these slight revisions may provide a more appropriate measurement structure for assessing sleep in ASD samples.

Using the CSHQ-R developed in Chapter 2, the remaining studies of the thesis aimed to add to the current understanding of sleep in ASD. In Chapter 3,
longitudinal predictive associations between sleep problems and core ASD symptoms were explored. Sleep and ASD symptoms were assessed over a three to five year period in a sample of primary school-aged children with ASD (N = 45). The findings showed that ASD symptom severity and sleep problems were (i) individually stable over time, (ii) positively associated with each other at each time point, and (iii) robustly associated with each other across time points. However, sleep problems at initial assessment did not account for significant additional variance in ASD symptoms at follow-up when controlling for initial ASD symptom; similarly, ASD symptoms at initial assessment did not add to the prediction of sleep problems at follow-up when controlling for initial sleep problems.

Given the stability of sleep problems identified in Chapter 3, Chapter 4 sought to build on the current understanding of the differences in sleep problems in children with and without ASD using a multi-method approach to sleep assessment. Participants were children with ASD (n = 39) and typically developing (TD) children (n = 28) aged 6-12 years and their parents. Consistent with past literature, children with ASD had more severe sleep problems than their TD peers on parent-rated measures; however, few differences were found when reviewing sleep diaries or actigraphy-derived measures of sleep. Further, there was poor agreement between CSHQ-R subscales and sleep diaries/actigraphy recording across both groups, highlighting the complexity of establishing sleep “problems” in paediatric groups.

Finally, Chapter 5 examined relationships between sleep problems, challenging daytime behaviours, and both parent-rated and performance-based executive functioning (EF) in ASD (n = 32) and TD (n = 32) groups. There were consistent associations between parent-reported sleep, EF, and challenging behaviours across both groups. Indeed, parent-rated EF mediated the relationship
between parental perceptions of poor sleep and challenging behaviours in the TD group; however, this relationship fell short of reaching significance in the ASD group. No consistent relationships were found between objective assessments of sleep and EF or behaviour. Again, these results underscore the need for multi-method sleep assessment and raise questions regarding the association between sleep and challenging behaviours.

Together, these findings highlight potential revisions to a commonly used sleep questionnaire within ASD samples, whilst emphasising that sleep is best assessed using both objective and subjective tools. Parental reports of sleep problems are enduring over time. However, when employing multi-method assessment of sleep, few differences emerge between ASD and TD groups in objectively assessed sleep parameters, even in the context of high parent-reported sleep problems. Given that objective sleep parameters are broadly consistent between groups, alternative explanations for the association between parent-rated sleep and daytime behaviours were considered. The theoretical and clinical implications of these findings are discussed in Chapter 6.
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Preamble

This thesis is presented as a series of papers. Chapter 1, the General Introduction, provides the research context, background literature and rationale for the thesis. Chapter 2 has been submitted for publication and revised based on feedback provided. All remaining empirical chapters (Chapters 3-5) are presented as manuscripts that have not yet been submitted for publication. Chapter 6 is a theoretical chapter that discusses the key findings of the thesis. Given that each empirical chapter has been written to stand individually, there is a degree of repetition between the General Introduction and the subsequent introductions for Chapters 2-5. While written to be read in isolation, chapters have been structured to progress logically from one to the next, with a short preamble at the start of each chapter detailing how it is linked to the previous papers and the aims of the thesis more broadly.
Chapter 2:
Submitted for publication, revisions completed, planned for resubmission.

Student contribution to Chapter 2:
The candidate completed the study design, literature review, data analyses and interpretations of the findings, and prepared and revised the manuscript. Claire Mitchell provided advice and guidance during data analysis. Data were collected through the Western Australian Autism Biological Registry (WAABR; Telethon Kids Institute), and the Val Lishman Project (VLP; Curtin University). Co-authors of the manuscript provided guidance during this process as well as substantive comments on draft manuscripts.

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Chapter 3:
Not yet submitted for publication.


_Longitudinal assessment of sleep and autism spectrum disorder symptom severity._ (Unpublished manuscript). University of Western Australia, Australia.

Student contribution to Chapter 3:
The candidate completed the study design, literature review, data analyses and interpretations of the findings, and prepared and revised the manuscript. Data presented for the first time point (T1) were collected by researchers and research assistants in connection with the WAABR (Telethon Kids Institute). Data presented for the second time point (T2) were collected by the candidate with support from research assistants at the Telethon Kids Institute. Co-authors of the manuscript provided guidance during this process as well as substantive comments on draft manuscripts.

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18/9/2018
Chapter 4:
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Chapter 5:
Not yet submitted for publication.

Student contribution to Chapter 4 and 5:
The candidate completed the study design, literature review, data collection and analyses, interpretations of the findings, and prepared and revised both manuscripts. Co-authors of manuscripts provided guidance during this process as well as substantive comments on draft manuscripts.

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## Abbreviations

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<tr>
<td>AD</td>
<td>Autistic Disorder</td>
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<tr>
<td>ADHD</td>
<td>Attention Deficit Hyperactivity Disorder</td>
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<td>ADOS</td>
<td>Autism Diagnostic Observation Schedule</td>
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<td>AS</td>
<td>Asperger Syndrome</td>
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<tr>
<td>ASD</td>
<td>Autism Spectrum Disorder</td>
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<tr>
<td>BEDS</td>
<td>Behavioural Evaluation of Disorders of Sleep Scale</td>
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<td>BRI</td>
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<td>BRIEF</td>
<td>Behavioural Rating Inventory of Executive Functioning</td>
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<td>BT ^</td>
<td>Bedtime</td>
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<tr>
<td>CBCL</td>
<td>Children’s Behaviour Checklist</td>
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<td>CFA</td>
<td>Confirmatory Factor Analysis</td>
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<td>CSHQ</td>
<td>Children’s Sleep Habits Questionnaire</td>
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<tr>
<td>CSHQ-R</td>
<td>The Children’s Sleep Habits Questionnaire - Revised as detailed in Chapter 2</td>
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<tr>
<td>DSM</td>
<td>Diagnostic and Statistical Manual of Mental Disorders</td>
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<tr>
<td>EF</td>
<td>Executive Functioning</td>
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<td>EFA</td>
<td>Exploratory Factor Analysis</td>
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<td>Ext B</td>
<td>Externalising Behaviours Index CBCL</td>
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<td>FSIQ</td>
<td>Full-Scale Intelligent Quotient</td>
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<td>ID</td>
<td>Intellectual Disability</td>
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<td>MI</td>
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<td>NIHEx</td>
<td>National Institute of Health Examiner Battery</td>
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<td>NREM</td>
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<td>PDD-NOS</td>
<td>Pervasive Developmental Disorder Not Otherwise Specified</td>
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<td>Polysomnography</td>
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<tr>
<td>REM</td>
<td>Rapid Eye Movement (sleep)</td>
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<td>RRBI</td>
<td>Restricted and Repetitive Behaviour and Interests</td>
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<td>Abbreviation</td>
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<tr>
<td>SDB</td>
<td>Sleep Disordered Breathing</td>
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<td>Sleep Disturbances Scale for Children</td>
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<td>SOffT</td>
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<td>Sleep Onset Time</td>
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<td>SP ^</td>
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<td>WASO</td>
<td>Wake After Sleep Onset</td>
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<td>WT ^</td>
<td>Wake Time</td>
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^ When prefaced with “D_” refers to the abbreviation for the corresponding variable calculated from the sleep diary, rather than actigraphy.
Chapter 1: General Introduction

1.1 Preamble to Chapter 1

The first chapter presented in this thesis is a general introduction which summarises the existing literature regarding sleep in children with autism spectrum disorder (ASD). For context, reference is also made to sleep in typically developing children and sleep across the lifespan. After establishing sleep patterns across typically developing populations and those with ASD, the literature review shifts to focus on methods used for assessing sleep in childhood, and more specifically, assessing sleep in children with ASD. The chapter then addresses the developmental trajectory of sleep problems in children with ASD, followed by a discussion of some of the possible explanations for these sleep problems. Finally, the review outlines the commonly reported functional sequelae of poor sleep in children with ASD with a focus on cognition and daytime challenging and externalising behaviours. The chapter concludes with a description of the aims of the current research as well as an outline of the empirical studies which follow.
1.2 Defining Sleep

Sleep is essential for social, emotional, and cognitive well-being, and for learning and development. Insufficient or problem sleep throughout childhood can have a significant impact on a child’s well-being, making sleep a vitally important area of developmental research. Sleep is comprised of cycles of various sleep stages broadly categorised as rapid eye movement (REM) and non-rapid eye movement (NREM; Berry & Wagner, 2015b). As may be expected, REM sleep is characterised by rapid eye movements, as well as reduced voluntary muscle tone. NREM is generally divided into three stages: N1, N2 and N3 (previously stages 1-4), with the final stage, often referred to as “slow wave sleep”, characterised by delta waves on electroencephalography (high amplitude, low-frequency waves; Berry & Wagner, 2015a). While important, a full discussion and review of sleep architecture are outside of the scope of the current thesis, which will focus primarily on sleep quantity, indicators of sleep quality, and “sleep problems”.

Regulation of sleep and wake is often considered in the context of both homeostatic “sleep dependent” and circadian “sleep independent” processes (Gregory & Sadeh 2016; Wyatt, 2014). Internal homeostatic processes are, in large part, determined by the duration of time since the last sleep period, as well as past sleep duration. Crudely summarised, the drive for sleep builds across the day, increasing from initial waking and reaching its peak at a time just before the next sleep onset. Conversely, the circadian system, based roughly on a 24-hour schedule, promotes wakefulness during the daylight hours and sleep during the night (Wyatt, 2014).

In discussing sleep problems, it is important to note that the presence of sleep problems does not describe the underlying causal or maintaining factors associated
with the problem. Sleep problems – including difficulties with sleep initiation or maintenance, parasomnias, and daytime somnolence – may be the result of an underlying sleep disorder, or stem from a range of other physiological, psychological, behavioural, or environmental factors. As currently defined by the International Classification of Sleep Disorder (ICSD-3; American Academy of Sleep Medicine, 2014) there are six main categories of sleep disorders. These sleep disorders include insomnia; sleep-related breathing disorders; central disorders of hypersomnolence; circadian rhythm sleep-wake disorders; parasomnias; and sleep-related movement disorders (American Academy of Sleep Medicine, 2014). While these disorders may account for an observed sleep problem, there may also be alternative or coexisting factors which lead to, or exacerbate, these problems.

1.3 Sleep across Childhood and Adolescence

While sleep is critically important, sleep needs and sleep patterns change considerably over the lifespan, leading to changes in what constitutes “problem” sleep. Throughout childhood, for example, distinct sleep periods consolidate into a single overnight block (Galland, Taylor, Elder, & Herbison, 2012) and sleep duration shortens (Galland et al., 2012; Ohayon, Carskadon, Guilleminault, & Vitiello, 2004). As such, a toddler napping twice during the day would not be considered problematic; however, a primary school age student still regularly sleeping for long periods during the day may raise concern. Given these changes, a sound understanding of “typical” sleep across the lifespan is important in understanding and assessing sleep problems.

Current guidelines indicate that children aged 6-13 years should sleep for between 9 and 11 hours per night (Hirshkowitz et al., 2015). However, sleep quantity represents only one domain of sleep health. Recommendations exist also for sleep
quality, with guidelines available for a number of indicators such as sleep onset latency (SOL; for children aged 6-12 years of age “good” sleep is indicated by SOL < 30 mins), the number of overnight awakenings of greater than 5 mins (i.e. “night wakings” with good sleep indicated by < 1), wake after sleep onset (WASO < 20 mins) and sleep efficiency (SE > 85%; Ohayon et al., 2017).

Within typically developing (TD) populations sleep is consolidated into a single overnight period of 7.5 to 9.5 hours per night by school age (Scholle et al., 2011). Throughout primary school, sleep duration continues to decline (Galland et al., 2012) due to a gradual delay in sleep onset times coupled with fairly consistent rise times. As such, sleep periods reduce to an average of between 6.8 to 9 hours by 12 years of age (Galland et al., 2012; Pesonen et al., 2014; Sadeh, Dahl, Shahar, & Rosenblat-Stein, 2009). Across adolescence, bedtimes continue to become progressively later due to shifts in the circadian rhythm (Crowley, Acebo, & Carskadon, 2007). Despite this, rise times, usually determined by the start of the school day, tend to remain constant, truncating the time spent in bed (i.e. opportunity for sleep) and subsequently the sleep period. Total sleep time (TST) therefore, reduces to approximately 6 to 9 hours by 18 years of age (Scholle et al., 2011). Sleep duration continues to shorten into adulthood, while SOL steadily increases (Ohayon et al., 2004). Currently, there is some evidence that healthy TD children are sleeping less than they have done historically (Matricciani, Olds, & Petkov, 2012). Moreover, as may be inferred from the discrepancies between sleep recommendations and normative data detailed above, many children are not meeting sleep duration guidelines (Matricciani, Olds, Blunden, Rigney, & Williams, 2012). While these general patterns are observed in the population, it is critical to highlight that there are also considerable individual differences in sleep duration and sleep need (Jenni,
Molinari, Caflisch, & Largo, 2007), hence fairly wide windows for what is recommended (i.e. 9-11 hours).

As sleep patterns change across the lifespan, so do sleep problems. Sleep initiation and maintenance difficulties are the most common problems throughout childhood, with 10-40% experiencing long SOL and frequent night wakings (Calhoun, Fernandez-Mendoza, Vgontzas, Liao, & Bixler, 2014; Fricke-Oerkermann et al., 2007; Xicheng Liu et al., 2005; Meltzer, Johnson, Crosette, Ramos, & Mindell, 2010; Pesonen et al., 2014; Spruyt, O’Brien, Cluydts, Verleye, & Ferri, 2005). For example, in a large community-based sample of 2 to 14-year-old children (N = 1038) at least one symptom of insomnia was reported in 41% of participants (although, only 18% were reported to experience more than one symptom of insomnia; Archbold, Pituch, Panahi, & Chervin, 2002). In another study of school-age TD children, 31% were reported to experience difficulty with initiating and maintaining sleep (Spruyt et al., 2005). Consistent with other estimates, Calhoun et al. (2014) found that 19% of children from the general population had symptoms of insomnia when defined as a parental report of trouble falling asleep or waking up during the night “often” or “very often” over the past two months. Of note, when compared to objective assessment of sleep, the study found that children with parent-reported insomnia symptoms did indeed have statistically poorer sleep on some sleep parameters, such as an increased SOL, compared to children without those reported symptoms (Calhoun et al., 2014). However, the magnitude of the differences in these parameters was relatively small (e.g. difference in SOL of only 5 minutes) leading to questions regarding the clinical significance of these findings.

Importantly, within TD populations, most sleep problems are transient (Clarkson, Williams, & Pa, 1986) and tend to diminish with age (Barclay, Gehrman,
In a long-running longitudinal study, Gregory and O’Connor (2002) found that parent-reported sleep problems (calculated from the Child Behaviour Checklist; CBCL; Achenbach & Rescorla, 2001) decreased steadily over the course of 11 years, from age 4 years to 15 years of age. Similarly, Fricke-Oerkermann et al. (2007) assessed self- and parent-reported sleep over a three-year period in children who were 9 years old at initial assessment (N = 832). While 30-40% of the children had problems falling asleep at the first assessment, one-third of these children no longer had sleep onset difficulties (self- or parent-report) a year later (Fricke-Oerkermann et al., 2007). Finally, Laberge et al. (2001) reported on sleep data from a large-scale longitudinal study (N =1146) with a parent-reported questionnaire completed annually from the age of 10 through to 13 years. While the study focused on sleep scheduling variables, the percentage of children perceived to have difficulty falling asleep was found to significantly diminish with age, as did the percentage of children who woke frequently during the night (Laberge et al., 2001). Despite this, there is also some evidence that sleep problems, namely difficulty with sleep initiation, may increase again towards adolescence, particularly when reviewing adolescent self-report data (Sivertsen, Harvey, Pallesen, & Hysing, 2017).

As noted, sleep problems are potentially of great consequence to emotional and physical well-being, as well as learning and memory. Inadequate sleep has been associated with mood disorders (e.g. anxiety and depression), poorer health indicators (e.g. obesity), deficits in cognitive skills (e.g. attention) and decreased academic achievement (for reviews see Chaput et al., 2016; Gregory & Sadeh, 2016). Given that sleep is related to a number of health, behavioural and cognitive outcomes, it is imperative that sleep problems are identified and treated when they
emerge. Further, from a public health perspective, it is important to identify groups who are most at risk of sleep problems in order to focus screening and intervention programs.

### 1.4 Sleep in Children with Neurodevelopmental Disorders

While sleep problems are common throughout childhood in general, there is considerable evidence that they are even more common in children with neurodevelopmental disorders (NDD) such as intellectual disability (ID; Robinson & Richdale, 2004), Down syndrome (Breslin et al., 2014; Dyken, Lin-Dyken, Poulton, Zimmerman, & Sedars, 2003), Prader-Willi syndrome (Nixon & Brouillette, 2002), Angelman syndrome (Clayton-Smith, 1993), Smith-Magenis syndrome (e.g. Potocki et al., 2000), attention deficit hyperactivity disorder (ADHD; Sung, Hiscock, Sciberras, & Efron, 2008) and autism spectrum disorder (ASD; Allik, Larsson, & Smedje, 2008; Richdale & Schreck, 2009). Indeed, the rate of sleep problems in groups with NDD has been reported to range from around 65-75% across NDD, with rates extending to over 90% in children with severe global neurological injury (Tietze et al., 2012). Needless to say, these rates are markedly higher than the rate of 10-40% described in the TD paediatric sleep literature (Calhoun et al., 2014; Fricke-Oerkerermann et al., 2007; Xicheng Liu et al., 2005; Meltzer et al., 2010; Pesonen et al., 2014; Spruyt et al., 2005). While children with a range of conditions commonly present with sleep disturbance, the rates and types of sleep problems appear to differ between diagnoses (Stores, 1992). For example, children with Down syndrome tend to present with high prevalence of sleep-disordered breathing (SDB; Breslin et al., 2014; Rosen, 2011), likely related to craniofacial abnormalities inherent to the syndrome (Slaats et al., 2015; Tan, Gozal, & Kheirandish-Gozal, 2013), while children with ADHD tend to present with difficulties in initiating and maintaining
sleep (see Spruyt & Gozal, 2011b for review). Most recently, a study assessing sleep patterns across a range of NDD found syndrome-specific profiles (Trickett, Heald, Oliver, & Richards, 2018). Namely, while all NDD groups had greater sleep disturbance than TD peers, children with Smith-Magenis syndrome experienced more symptoms of SDB and early morning waking, while children with ASD experienced more difficulties with sleep onset (Trickett et al., 2018).

Despite ongoing research regarding the specific sleep profile of children with ASD (Goldman et al., 2009), there is some agreement that these children most commonly present with symptoms of insomnia (Cotton & Richdale, 2010; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Malow, Marzec, et al., 2006), with evidence that the rate and severity of these sleep problems exceed those of several other clinical groups including Down syndrome, Prader-Willi syndrome, non-specific ID, and epilepsy (Cotton & Richdale, 2010; Tsai et al., 2012). In addition to this, there are also reports that children with ASD present with sleep problems characterised by fragmented and irregular sleep-wake patterns (Honomichl, Goodlin-Jones, Burnham, Gaylor, & Anders, 2002; Miano et al., 2007), apnoea (Aathira et al., 2017; Schreck & Mulick, 2000) and parasomnias (Xianchen Liu, Hubbard, Fabes, & Adam, 2006; Polimeni, Richdale, & Francis, 2005; Schreck & Mulick, 2000). Sleep disturbance in children with ASD may also be differentiated from that of children with other NDD by greater bedtime resistance and sleep anxiety (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014). The sleep profiles generally seen in ASD are described more comprehensively below (section 1.5).

### 1.5 Autism Spectrum Disorder

ASD is the term used to describe a developmental condition characterised by difficulties with social communication and interaction (American Psychiatric
According to the current Diagnostic and Statistical Manual of Mental Disorders (DSM), ASD should be diagnosed in the context of persistent deficits in social communication and social interaction and a restricted and repetitive pattern of behaviour, interests, or activities (DSM-5; American Psychiatric Association, 2013). These symptoms must be observed early in the developmental period and not be better explained by another condition such as global developmental delay (American Psychiatric Association, 2013). Current criteria have collapsed the individual diagnoses of Autistic Disorder (AD), Asperger Syndrome (AS) and Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) from the DSM-IV (American Psychiatric Association, 2000) into a single “spectrum” disorder, due to the fact that there is considerable literature questioning both the validity and utility of subcategories of autistic disorders (e.g. see Ozonoff, 2012). The DSM-5 also specifies severity criteria to capture the intensity and duration of symptoms, and the degree of related impairment (American Psychiatric Association, 2013). Regardless of which DSM version is utilised, there is evidence of reasonable concordance in diagnostic practices across criteria (i.e. between the DSM-IV and DSM-5; Mazurek et al., 2017).

Since initial description in the scientific literature (see Kanner, 1943), prevalence estimates for ASD have increased considerably. While early estimates suggest an incidence rate of around 4.5:10,000 (Lotter, 1966), more recent reports suggest rates as high as 1:68 (Baio, 2014). A large volume of research suggests that the rise in prevalence across time likely reflects a combination of a range of factors such as a broader awareness of the condition, clearer diagnostic criteria, and improved access to diagnostic services (for review, see Fombonne, 2003). Consistent with this, changes over time have often been temporally linked with revisions to the
diagnostic criteria (e.g. see Bennett & Goodall, 2016; Hartley-McAndrew, Mertz, Hoffman, & Crawford, 2016).

At present, the aetiology of autism remains unclear. Much of the research in this area has been driven by the observation that the incidence of ASD is considerably higher in males, with a 4:1 diagnostic bias (Werling & Geschwind, 2013). In this context, there is a plethora of research exploring the biological bases for these sex differences in ASD. Findings indicate that increased exposure to certain hormones (e.g. testosterone and cortisol) during critical periods of prenatal development, may contribute to structural and functional changes in the brain (see Lai, Lombardo, Auyeung, Chakrabarti, & Baron-Cohen, 2015, for review). In addition to this, there is strong agreement for a genetic component (Krumm, O’Roak, Shendure, & Eichler, 2014), with a recent meta-analysis of twin studies indicating heritability of 64-91% (Tick, Bolton, Happé, Rutter, & Rijsdijk, 2016). As noted by Krumm et al. (2014), this line of investigation has identified some of the syndromic causes for behavioural presentations consistent with ASD (e.g. Rett syndrome); however, these syndromes account for a relatively small portion of cases (<10%; Krumm et al., 2014). Many other genes have been identified as contributing towards risk for ASD, albeit with inconsistencies in the strength of these findings (e.g. Chaste et al., 2016; Taniai, Nishiyama, Miyachi, Imaeda, & Sumi, 2008). Currently, the specific mechanisms and potential environmental and epigenetic contributions remain elusive (Tordjman et al., 2014). Given the high heritability of ASD, the presence of still unknown genetic aetiologies is presumed. However, ASD will most likely be related to a vast number of genes and gene combinations, with multiple pathways to the behaviours observed (Tordjman et al., 2014).
Individuals diagnosed with ASD commonly present with a range of comorbid medical and psychiatric conditions (Bauman, 2010; Leyfer et al., 2006; Simonoff et al., 2008), of which sleep problems are one of the most frequently reported (Mannion, Leader, & Healy, 2013; Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2008; Patzold, Richdale, & Tonge, 1998; Richdale & Schreck, 2009). Importantly, these are of considerable concern to parents and are an independent predictor of maternal stress (Hoffman et al., 2008). Given the high prevalence sleep problems and the impact on both the child and family systems, sleep problems have been identified in the literature as a high priority for ongoing research (Mindell et al., 2006).

1.6 Sleep in Autism Spectrum Disorder

To date, there is considerable evidence that sleep problems are highly prevalent in ASD across the lifespan, from pre-school years (e.g. Goodlin-Jones, Tang, Liu, & Anders, 2008), to school-aged children (e.g. Giannotti, Cortesi, Cerquiglini, Vagnoni, & Valente, 2011; Xianchen Liu et al., 2006; Souders et al., 2009; Wiggs & Stores, 2004; for review see Cortesi, Giannotti, Ivanenko, & Johnson, 2010), adolescents (e.g. Allik et al., 2008; Baker, Richdale, Short, & Gradisar, 2013) and adults (e.g. Baker & Richdale, 2017; Godbout, Bergeron, Limoges, Stip, & Mottron, 2000; Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005). For example, in a community sample of 167 children with ASD (mean age 8.8) it was found that 86% of children had at least one parent-reported sleep problem (Xianchen Liu et al., 2006). Bedtime resistance (54%), insomnia (56%), and parasomnias (53%) were the most frequently reported concerns; however, difficulty waking in the morning and daytime sleepiness were also common, occurring in more than one-third of those surveyed (Xianchen Liu et al.,
Similarly, in an adolescent population, 46% of participants with ASD reported a sleep problem, a rate which is three times that of their TD peers (Baker et al., 2013).

Although sleep problems are common across a range of NDD, there is some evidence that the rate and severity of these problems may be highest in children with ASD when compared to those with other NDD (Krakowiak et al., 2008). As noted previously, the types of sleep problems across NDD also tend to vary. For example, while many children with NDD present with symptoms of SDB, parents of children with ASD tend to report difficulties with falling and then staying asleep during the night, ultimately impacting upon total sleep duration (Cotton & Richdale, 2010; Krakowiak et al., 2008; Patzold, Richdale, & Tonge, 1998). Studies have also reported difficulty waking in the morning (Richdale & Prior, 1995) or early waking (Taira, Takase, & Sasaki, 1998). Somewhat consistent with subjective reports of sleep, some studies using polysomnography (PSG) and actigraphy data (reviewed below) have also found that children with ASD tend to take a long time to fall asleep, and wake frequently during the night (Goodlin-Jones, Tang et al., 2009; Limoges et al., 2005; Malow, Marzec et al., 2006; Wiggs & Stores, 2004). However, as detailed below, findings from studies employing objective measures of sleep are more varied compared to those utilising parent-report measures.

1.7 Assessment of Sleep Problems in ASD

In order to accurately estimate the prevalence of sleep disturbance, establish the most common types of sleep problems, and gain an understanding of the daytime correlates of sleep, it is essential to obtain a valid and reliable measure of sleep itself. Broadly speaking, tools used to assess various aspects of sleep can be divided into
objective measures such as PSG and actigraphy, and subjective measures such as sleep diaries, sleep questionnaires and single item responses.

1.7.1 Objective measures of sleep

*Polysomnography*

PSG refers to a collection of recordings, including of neurological activity (electroencephalography), heart rhythm (electrocardiography), musculoskeletal activation (electromyography) and eye movements (electrooculography) as well as measures of respiratory effort and blood oxygen saturation (Aurora et al., 2011; Kushida et al., 2005). Together, these electrophysiological data are interpreted to provide precise determinations of sleep onset and offset, sleep architecture (i.e. sleep stages) and other occurrences in sleep such as apnoea and limb movements (Aurora et al., 2011; Kushida et al., 2005). Given that PSG relies on physiological changes during sleep, it is considered the ‘gold standard’ of sleep assessment (Aurora et al., 2011; Kushida et al., 2005) and is often used as the benchmark against which to validate other measures such as actigraphy or sleep diaries (e.g. Gregory et al., 2011; Lichstein et al., 2006; Meltzer, Walsh, Traylor, & Westin, 2012).

While PSG is able to provide clear, objective data, there are limitations associated with this method. The setup, monitoring and interpretation require the presence and skill of a trained technician. Moreover, the use of electrodes and monitoring equipment usually necessitates study participation in a sleep laboratory. Not only are these requirements associated with expense and inconvenience (Khatwa, Ramgopal, Singh, Zarowski, & Kothare, 2013), but sleep in a laboratory setting is also likely to result in a “first-night effect” whereby sleep patterns (e.g. SE, REM sleep, WASO) are atypical due to the unfamiliarity of surroundings as well as restrictions in movement and the discomfort of monitoring equipment (Coble,
Kupfer, Taska, & Kane, 1984; Le Bon et al., 2001). This phenomenon has led to suggestions that PSG recordings, especially for children, should take place over two nights (Verhulst, Schrauwen, De Backer, & Desager, 2006); with the first night to allow for acclimatisation to the setting and equipment, and the second to record sleep parameters.

Within ASD populations specifically, studies have confirmed the presence of the first night effect for at least some PSG variables (e.g. WASO and SE; Buckley et al., 2013). However, others have suggested that within this population, sleep may be so disrupted on the first night of PSG that night-two data may reflect a “catch up effect” whereby sleep is longer and deeper than usual due to accumulated sleep debt from the previous night (Malow, Marzec et al., 2006). As such, the authors suggested that within paediatric ASD populations it may be necessary for PSG recordings to take place over three consecutive nights to observe the child’s typical sleep without either a first night or catch-up effect (Malow, Marzec et al., 2006). Despite this suggestion, there are no published studies to date which have examined sleep in individuals with ASD through three or more nights of PSG recordings. Moreover, due to the need for placement of electrodes, some have noted that children with ASD (and associated sensory sensitivities) may find it difficult to tolerate the procedure at all (Hodge, Parnell, Hoffman, & Sweeney, 2012). In this context, it is likely that samples will be impacted by a selection bias, with only those children who can tolerate PSG (i.e. those who are less severely affected/less sensory) being able to participate in such studies. Given these challenges inherent to PSG studies, ASD sample sizes are often small (e.g. n = 18, Bruni et al., 2007; n = 13, Elia et al., 2000; n = 16, Miano et al., 2007) while those with larger samples (e.g. n = 60, Buckley et al., 2010) have often collected only one night of sleep data, precluding the ability to
account for the first night effect. Overall, while PSG is the most appropriate tool for determining sleep architecture and detecting conditions such as sleep apnoea and periodic leg movement disorder, it can be difficult to record a ‘typical’ night sleep, especially in ASD populations. Given this fact, PSG may be of limited benefit in establishing habitual sleep-wake patterns especially within ASD populations (Hodge et al., 2012).

**Actigraphy**

Given the limitations associated with PSG, actigraphy is a commonly used alternative method for measuring sleep in both TD and ASD populations. A recent review noted that the use of actigraphy in academic literature had risen markedly over the past 20 years (Sadeh, 2011). As noted by Sadeh (2011), while the rate of actigraphy to PSG studies was approximately 1:10 in the early 1990s, as of 2009 the ratio had increased to one actigraphy study for every four PSG studies, highlighting a relative increase in actigraphy when compared to other tools for objective sleep assessment (Sadeh, 2011).

Actigraphy uses an accelerometer, generally in the form of a watch-like device, to record gross motor activity from which sleep-wake cycles are inferred (Ancoli-Israel et al., 2003; Sadeh & Acebo, 2002). Sleep assessment using actigraphy generally requires that the participant wear an actigraph on their non-dominant wrist for one to two weeks (Sadeh & Acebo, 2002). Data regarding movement (and often light exposure) are recorded as epochs (typically 30 sec or 60 sec) and stored on the watch-like device until it is collected by the clinician/researcher and downloaded for analysis (Ancoli-Israel et al., 2003). Given that the device can be worn at home, it is both less invasive and less expensive than PSG. Further, as the actigraphy device is worn for a longer period of time (~1 week)
it can provide more detailed information regarding habitual sleep patterns which cannot be gleaned from PSG (Ancoli-Israel et al., 2003). Sleep variables commonly derived from actigraphy recordings are detailed in Table 1.1 (adapted from Meltzer, Montgomery-Downs, Insana, & Walsh, 2012).

While an actigraph can be mildly irritating (i.e. wearing a watch continuously for >24-hour period), studies have found that there does not appear to be a first night effect with the watch (Arora, Omar, & Taheri, 2016), suggesting that it does not generally cause sufficient discomfort to impact sleep. Further, while some have suggested children with ASD may find the actigraphy device difficult to tolerate (Hering, Epstein, Elroy, Iancu, & Zelnik, 1999; Oyane & Bjorvatn, 2005), in practice, others have found that the majority of their sample were able to wear the device for the full study period (e.g. Wiggs & Stores, 2004).

Despite the fact that there are no set guidelines for which device or scoring algorithm to use, a number of devices and algorithms have been validated against PSG across the lifespan: from infancy (So, Buckley, Adamson, & Horne, 2005); to childhood (Spruyt, Gozal, Dayyat, Roman, & Molfese, 2011); to adulthood (Marino et al., 2013). With regard to school-aged children specifically, a recent study assessed not only intra-device reliability, but also agreement between two of the most commonly used brands of actigraphs, and comparisons of each of these brands to PSG (Meltzer, Walsh et al., 2012). Importantly, intra-device reliability was high. This indicates that the use of multiple devices, provided that they are of the same make and model (as in the current research), would not compromise the reliability of the results. Actigraphy to PSG epoch-by-epoch comparisons indicated good agreement, with sensitivity (i.e. the ability of actigraphy to identify sleep) ranging from 89-97%.
### Table 1.1

*Details of Common Actigraphy Variables and How They Are Calculated.*

<table>
<thead>
<tr>
<th>Variable</th>
<th>Abbreviation</th>
<th>Units</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Diary</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bedtime</td>
<td>D_BT</td>
<td>(HH:MM)</td>
<td>time the child attempted to fall asleep as recorded by event marker or sleep diary</td>
</tr>
<tr>
<td>Wake Time</td>
<td>D_WT</td>
<td>(HH:MM)</td>
<td>time of the child’s final awakening in the morning as recorded by event marker or sleep diary</td>
</tr>
<tr>
<td>Time in Bed</td>
<td>D_TIB</td>
<td>(min)</td>
<td>duration between bedtime and wake time (i.e. sleep opportunity)</td>
</tr>
<tr>
<td><strong>Actigraphy</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep Onset Time</td>
<td>SOT</td>
<td>(HH:MM)</td>
<td>time of the first of a predetermined number of consecutive minutes of sleep (e.g. 1, 5 or 10 mins) following bedtime</td>
</tr>
<tr>
<td>Sleep Offset Time</td>
<td>SOffT</td>
<td>(HH:MM)</td>
<td>time of the last of a predetermined number of consecutive minutes of sleep (e.g. 1, 5 or 10 mins) before wake time</td>
</tr>
<tr>
<td>Sleep Period</td>
<td>SP</td>
<td>(min)</td>
<td>duration between sleep onset and sleep offset times</td>
</tr>
<tr>
<td>Wake After Sleep Onset</td>
<td>WASO</td>
<td>(min)</td>
<td>number of minutes scored as wake during the sleep period</td>
</tr>
<tr>
<td>Total Sleep Time</td>
<td>TST</td>
<td>(min)</td>
<td>duration of sleep during the sleep period (SP - WASO)</td>
</tr>
<tr>
<td>Sleep Efficiency</td>
<td>SE</td>
<td>(%)</td>
<td>percentage of time spent asleep whilst in bed ((TST/D_TIB) x 100)</td>
</tr>
<tr>
<td>Sleep Onset Latency</td>
<td>SOL</td>
<td>(min)</td>
<td>duration between bedtime and sleep onset time</td>
</tr>
</tbody>
</table>

*Note. Table adapted from Table 4 in “Use of actigraphy for assessment in pediatric sleep research” Meltzer et al. (2012)*

Chapter 1: General Introduction
Consistent with previous reports, specificity (i.e. the ability of actigraphy to identify wake) was lower, ranging from 54-77% (Meltzer, Walsh et al., 2012). Despite this, overall accuracy was good, ranging from 87-90% (Meltzer, Walsh et al., 2012). Given that actigraphy infers sleep-wake cycles from movement, and that it is possible to wake during the night without much movement, WASO may remain undetected. Due to this, and the resulting low specificity of actigraphy, some have raised concerns regarding the validity of the sleep quality variables produced by actigraphy (i.e. WASO, SE etc.; Ancoli-Israel et al., 2003; Sadeh, 2011; Sadeh & Acebo, 2002; So et al., 2005). However, and of importance for the current thesis, within school-aged children, the Philips Respironics MiniMitter Actiwatch-2 (which is used in the following studies) was found to have some of the highest agreement with PSG, recording sensitivity (i.e. detecting sleep) of 93% and specificity (i.e. detecting wake) of 71%, leading to overall accuracy of 90% (Meltzer, Walsh et al., 2012).

1.7.2 Subjective measures of sleep

Sleep diaries

Sleep diaries (or sleep logs) require that the child or their parent/guardian record sleep information such as bed and wake time, time of sleep onset and periods of wakefulness during the night (Hodge et al., 2012). This information is generally recorded each morning with regard to the previous night. During the day, note can also be made of any daytime napping, or periods when the watch has been removed. While sleep diaries can be used alone, they are often used as a complementary data source to actigraphy (Acebo et al., 1999; Kushida et al., 2005; Sadeh & Acebo, 2002). In general, within the paediatric sleep literature, there is evidence that diary and actigraphy recordings have strong concordance for sleep schedule variables such
as sleep onset and TST; however, studies have found less consistency across measures of sleep quality such as NW, WASO or SE (Ancoli-Israel et al., 2003; Werner, Molinari, Guyer, & Jenni, 2008).

Questionnaires

Structured sleep questionnaires offer an alternative (or adjunct) in sleep assessment. Compared to PSG and actigraphy, questionnaires offer the benefit of being inexpensive, time effective, and easy to administer. Commonly used measures include the Sleep Disturbances Scale for Children (SDSC; Bruni et al., 1996), Behaviour Evaluation of Disorders of Sleep (BEDS; Schreck, Mulick, & Rojahn, 2003) and the Children’s Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000). While these questionnaires may not provide clear information regarding sleep schedule variables (e.g. daily sleep or wake times) they do provide valuable information regarding the parent perception of sleep quality and a range of sleep behaviours which are not readily captured by objective measures. This includes scales assessing bedtime resistance, sleep-related anxiety, and daytime somnolence. Within the ASD sleep literature the CSHQ is, by far, the most widely used questionnaire (Aathira et al., 2017; Adams, Matson, & Jang, 2014; Doo & Wing, 2006; Goldman et al., 2009; Goodlin-Jones, Tang et al., 2008; Hodge et al., 2014; Hoffman et al., 2008; Hoffman, Sweeney, Gilliam, & Lopez-Wagner, 2006; Honomichl et al., 2002; Lambert et al., 2016; Mannion & Leader, 2016; Meltzer, 2011; Souders et al., 2017). Despite being the most commonly used scale, a recent review of paediatric sleep questionnaires noted that the CSHQ met only 5 of the 11 steps necessary for sound instrument development (Spruyt & Gozal, 2011a). Moreover, despite the frequent use of the CSHQ for children with ASD for more than a decade, until this year, only one study had assessed the psychometric
properties and factor structure of the CSHQ in this population (see Johnson et al., 2016).

Outside of limitations of the CSHQ specifically, it must be noted that when using questionnaires in general, parents are generally reporting on behaviours which they have likely not witnessed directly (e.g. waking during the night). Due to this, it is possible that they may over- or under-estimated the occurrence of these behaviours. This is reflected in the fact that questionnaire subscales – which are intended to assess sleep parameters such as SOL, NW, and sleep duration (SD) – tend to have small or non-significant correlations with corresponding PSG and actigraphy variables. For example, in a study of 91 TD children aged 6-11 years, Holley, Hill, and Stevenson (2010) found expected correlations between the parent-rated Sleep Onset Delay subscale of the CSHQ and actigraphy SOL (r = .35) and Sleep Duration (CSHQ) and actigraphy sleep duration (r = -.28); however, while statistically significant, the magnitude of these relationships was small. Further, no statistically significant correlations were found between parent-reported night wakings and actigraphy WASO (Holley et al., 2010). Conversely, in a study conducted by Markovich, Gendron, & Corkum (2015), comparing the CSHQ, actigraphy, and PSG, the authors reported that only one subscale of the CSHQ (NW) correlated with any of the actigraphy variables (CSHQ night wakings r = -.42 with TST and r = .47 with WASO). No relationships were noted between any of the CSHQ subscale scores and PSG variables. Of note, however, the study also failed to find any relationship between actigraphy (collected for one week) and PSG (collected over a single night), indicating that PSG recorded sleep may have been impacted by the first night effect (Markovich et al., 2015). While there is poor concordance between parent report and objective measures in TD samples, it is
possible that there may be different patterns of concordance in samples with NDDs due to differences in wake behaviour, such as children with NDDs being more inclined to draw the attention of their parents when awake.

**Single item response**

Finally, a number of studies include or rely upon parental response to a single item with either a Likert or binary scale (e.g. “Is your child a good sleeper?” or “Does your child have a sleep problem?”). There is some evidence that parent-response on a single-item measure of sleep may be consistent with findings from PSG (Malow, Marzec et al., 2006).

1.7.3 **Concordance between measurement tools in ASD**

Importantly, the discrepancies in the rate and severity of sleep problems in ASD may be, at least in part, attributable to differences in the tools used to assess sleep. That is, while the vast majority of questionnaire data indicates that parents report high levels of enduring sleep problems in children with ASD, the findings from objective sleep assessment are far less consistent. Specifically, studies using parent-report responses on the CSHQ (e.g. Couturier et al., 2005; Giannotti et al., 2011; May et al., 2015; Souders et al., 2009), BEDS (e.g. Schreck & Mulick, 2000; Polimeni et al. 2005) and the SDSC (e.g. Miano et al., 2007; Paavonen et al., 2008) often indicate that children with ASD had higher scores than their TD peers on at least one subscale of each questionnaire. However, while some studies using objective measures have confirmed findings of sleep disturbance (Allik, Larsson, & Smedje, 2006; Giannotti et al., 2011), many others have found either mixed results or no group differences between children with ASD and their TD peers (Bruni et al., 2007; Souders et al., 2009; Tani et al., 2004, 2005). While use of different measures
may account for some of the discrepancies, the inclusion of often large heterogenous samples make cross-study comparisons difficult.

Relatively few have directly examined cross-method concordance within a single sample with ASD to better understand these discrepancies. Further, of those that have examined agreement between measures, many have only reported on general, broad findings, such as general agreement between the percentage of those defined as having “good” or “poor” sleep (Souders et al., 2009), rather than specific agreement between comparable subscales (e.g. SOL subscale of the CSHQ and SOL variable derived from actigraphy).

Goldman et al. (2009) examined group differences between PSG and actigraphy derived sleep parameters across three groups of children: those who were TD (n = 16); those with ASD and parent-rated good sleep (n = 15); and those with ASD and parent-rated poor sleep (n = 27). Significant differences between “good” and “poor” sleepers with ASD emerged across a number of actigraphy variables (SOL, WASO and SE), however, when examining PSG data, group differences were found only for SOL. Taken together, this highlights inconsistencies across measures. While the authors concluded that parental reports of sleep problems were corroborated by the objective measures given, it is pertinent to note that the actigraphy and PSG data were based on an average across two nights, without allowing for an initial night to acclimatise to the laboratory/equipment (Goldman et al., 2009). As such, results may have been impacted by a first night effect. This is particularly notable given that a prior study had found differences between parent-reported “good” and “poor” sleepers with ASD based on recordings from their first night of PSG; however, no differences based on the second night of PSG recordings (i.e. after an opportunity to acclimatise to the setting; Malow, Marzec et al., 2006).
Consistent with this, another PSG study noted that the types of sleep problems reported by parents of children with ASD were only partially confirmed by the objective data (Miano et al., 2007), while studies employing actigraphy to assess sleep have also noted poor agreement between parent-reported sleep and objective sleep parameters (Hering et al., 1999; Wiggs & Stores, 2004).

Only two studies directly comparing the agreement between objective and subjective reports of sleep patterns within an ASD sample have been identified. The first study reviewed agreement between sleep diary and actigraphy recorded sleep parameters (using an Actiwatch) in a clinical sample of 32 school-aged children (M=10.8 years) with Asperger syndrome and high-functioning autism. Findings noted strong agreement between the two measures for both TST (r = .73) and SOL (r = .80; Allik et al., 2006). Interestingly, for both of these variables, concordance was actually stronger in the ASD group compared to the TD group (TST r = .64 and SOL r = .55). The second study comparing the same two measures (i.e. sleep diary and actigraphy: Mini Mitter Actiwatch), in a primary-school aged sample (M =44.4 months), found only moderate agreement for TST (r = .58) and SOL (r = .43). While there were strong correlations for SOT (r = .81) and SOffT (r = .91), it is notable that agreement between measures was considerably lower for WASO duration (r = .24) and WASO number (r = .27; Goodlin-Jones, Tang et al., 2008). While the two aforementioned studies examined agreement between parent-reported sleep based on the sleep diary and actigraphy recordings, no studies were identified which directly compared comparable subscales of a sleep questionnaire with actigraphy recordings in the ASD population, as has been done in the TD paediatric sleep literature (e.g. Markovich, Gendron, & Corkum, 2015). However, one study, comprised of pre-school-aged children with a range of NDD including ASD, did report good
agreement for sleep scheduling variable between CSHQ, sleep diaries and actigraphy (Mini Mitter Actiwatch; Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008).

1.8 Developmental Trajectory of Sleep Problems in ASD

While the developmental trajectory of sleep is well defined in TD populations (detailed above), there is a paucity of literature regarding the development of sleep problems in children with ASD. Historically, few have explored sleep patterns in ASD longitudinally and developmental comparisons have been hindered in cross-sectional studies due to study designs which often group children across wide age brackets. In recent years, however, there have been a few studies published which have sought to fill this gap (Allik et al., 2008; Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2011; Hodge et al., 2014; Humphreys et al., 2014; Mannion & Leader, 2016; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012).

Using a cross-sectional approach, Hodge et al. (2014) examined parent-reported sleep problems in children with and without ASD across three age groups; 3-5 years, 6-9 years, and 10-17 years. The study found that within the ASD sample there were no differences in the rate of sleep problems across age groups. This was in contrast to the TD control group where age-related differences in sleep emerged, with significantly fewer children categorised as having sleep problems in the older, compared to the younger, groups. Of note, due to the fact that children with ASD experienced persistent sleep problems across all ages, group differences between the ASD and TD samples only emerged for the 6-9 and 10-17 year-olds (Hodge et al., 2014).

With regard to longitudinal analysis, Sivertsen et al. (2012) reported on data from a large (N = 3700) population-based study. Using parental responses on a single
sleep question, children aged 7-9 years with ‘autism spectrum problems’ (i.e. above the cut-off of 17 on the Autism Spectrum Screening Questionnaire) were 10 times more likely to experience a sleep problem at initial assessment compared to the rest of the sample. Importantly, at follow-up two years later, these children had a significantly reduced rate of improvement in sleep compared to their peers (8.2% compared to 52.4%; Sivertsen et al., 2012). Similarly, Humphreys et al. (2014) examined parental reports of bed and wake time across 8 time points in children aged 6 months to 11 years in a large population-based cohort study (N ~ 11000). The results indicated that from 30 months of age until the end of the study period (at 11 years), children with ASD slept less each night than those without ASD. This difference was attributed to both later bedtimes and earlier waking times. Despite this, the trajectories of sleep problems were fairly consistent across groups (Humphreys et al., 2014). In a smaller scale study (N = 56) of children aged 5-19 years with a clinical diagnosis of ASD, 91.5% of those with sleep problems at initial assessment continued to experience sleep problems two years later (Mannion & Leader, 2016). Conversely, May et al. (2015) examined parent-reported sleep over a one-year period, finding that while, when compared to a TD control group, children with ASD (n = 46) had higher levels of sleep problems (CSHQ) at baseline, this decreased over the course of the study period.

Compared to reports of sleep problems, objectively measured sleep patterns over time, appear quite consistent between TD and ASD groups (Allik et al., 2008; Anders et al., 2011; Fletcher et al., 2017). In a six-month follow-up study using actigraphy, Anders et al. (2011) reported that the trajectory of sleep parameters was similar across community-based samples of pre-school aged children with and without ASD, noting comparable reductions in sleep duration and night wakings.
from the first assessment to the second (Anders et al., 2011). However, it was also noted that at follow-up, children with ASD appeared to present with more night-to-night variability in their sleep (Anders et al., 2011). Similarly, in an older sample of children with ASD (N = 16; M = 11.1 years) Allik et al. (2008) reported that across a 2-3 year period, the trajectory of actigraphy assessed sleep patterns was similar across ASD and TD groups with gradually later sleep onset times and shorter sleep durations (Allik et al., 2008). Similarly, a more recent study which examined both parent-report and actigraphy-derived sleep profiles over a 1-year period found that the course of sleep was comparable between groups (actigraphy). However, at both time points, children with ASD presented with notable night-to-night variability in sleep quality (Fletcher et al., 2017).

In summary, based on the current literature, it appears that while parents of TD children report a reduction in sleep problems over time, parents of children with ASD report higher levels of enduring sleep problems at all ages. Objectively assessed sleep parameters appear to show a similar developmental trajectory for TD and ASD groups; however, there are some indications that while the trajectory is similar, sleep quality is poorer across time points in those with ASD.

1.9 Aetiology of Sleep Problems in ASD

At present, there remains conjecture regarding the causes for sleep problems in ASD. To some extent, this may be attributable to shifting descriptions within the ASD literature of what constitutes a sleep problem. Often the term “sleep problems” refers to any report (generally a parent report) of difficulties with sleep onset and maintenance, including behavioural difficulties surrounding bedtime. The term also encapsulates excessive somnolence throughout the day.
The causes of sleep problems are likely varied. The literature does not specify the existence of specific sleep phenotypes which are observable in all individuals with ASD. Instead, sleep difficulties are better understood by way of a combination of predisposing, precipitating or perpetuating factors. Specifically, sleep problems may be the result of physiological, psychological, behavioural, or environmental factors.

There is evidence that children with ASD may be predisposed to sleep disturbance due to neurobiological or genetic abnormalities which underlie ASD (Richdale & Schreck, 2009). Other likely causes detailed in the literature include the presence of comorbid medical (e.g. gastrointestinal) or neurological (e.g. epilepsy) or primary sleep disorders (Accardo & Malow, 2015; Klukowski, Wasilewska, & Lebensztejn, 2015; Mannion, Leader & Healy, 2013). Due to the prevalence of medical comorbidities and the behavioural features of ASD, the use of stimulant, antidepressant, antipsychotic and antiepileptic medication is common (Murray et al., 2014), which may also precipitate sleep problems. In addition, the presence of a coexisting mood disturbance (i.e. depression or anxiety) or challenging behaviours may contribute (Gabriels et al., 2005; Hollway et al., 2013; Mayes & Calhoun, 2009; Paavonen et al., 2003; Sikora, Johnson, Clemons, & Katz, 2012). Finally, environmental or family factors (e.g. routine, limit setting, etc.; Cotton & Richdale, 2010) can also precipitate and perpetuate sleep problems. These interrelated factors are discussed in more detail below.

1.9.1 Predisposing factors

There is consistent evidence that sleep problems are strongly associated with both social and communication deficits, as well as highly restricted and repetitive behaviours or interests (RRBI; Hollway & Aman, 2011). For example, Tudor,
Hoffman, and Sweeney, (2012) found a relationship between parent-reported communication problems and parent-reported sleep problems; while Hollway et al. (2013) found that clinician-rated reciprocal social interactions were able to predict the severity of parent-reported sleep problems. Parent-reported RRBI have also been found to be associated with increased parent-reported sleep problems (Park et al., 2012; Tudor et al., 2012). While many studies have discussed the relationship in terms of sleep problems exacerbating the presentation of ASD symptoms, others have contested that these links may reflect the possibility that individuals with ASD may be at heightened risk for sleep problems due to underlying neurophysiological and neurochemical features of the condition itself (Cohen et al., 2014).

A number of studies have suggested a link between specific neurobiological profiles in children with ASD and predisposition to poor sleep. Within ASD populations there is growing evidence that some sleep problems may, at least in part, be accounted for by circadian disturbances. A range of physiological processes are entrained to a 24-hour cycle known as the circadian rhythm (Van der Heijden, Stoffelsen, Popma, & Swaab, 2018). While these processes, including sleep-wake cycles, changes in body temperature, and release of various hormones (e.g. melatonin and cortisol) are endogenous, they can also be modulated by exogenous cues such as sunlight and temperature which are often referred to as “zeitgebers” (Guénolé et al, 2011; Van der Heijden, Stoffelsen, Popma, & Swaab, 2018).

With regard to core features of ASD, there are suggestions that children with ASD, who have impairments in social interaction, may misinterpret or not notice social cues which are important zeitgebers for human circadian rhythms, leading to disruption in the entrainment of the sleep-wake cycle (Glickman, 2010). Further,
there is evidence of abnormalities in the production and synthesis of certain circadian relevant hormones (Nicholas et al., 2007; Yang et al., 2015).

Melatonin, a neurohormone produced from serotonin in the pineal gland, is critical in regulating sleep-wake cycles (Bauman, 2010). Within ASD populations, there are findings of mutations in circadian-relevant genes (Nicholas et al., 2007; Yang et al., 2015). Further, urinary 6-sulphatoxymelatonin, the major metabolite of melatonin, has been found to be lower in children and adolescents with ASD when compared to TD controls (Tordjman et al., 2012; Tordjman, Anderson, Pichard, Charbuy, & Touitou, 2005). Interestingly, while reduced 6-sulphatoxymelatonin has been found to be related to increased daytime sleepiness in a small sample of children with ASD (n = 24), it was not associated with changes in PSG measured sleep parameters (Leu et al., 2011). Moreover, a recent study documented normal overnight serum (rather than urinary metabolite) melatonin levels in children with ASD (Goldman et al., 2014). Despite this, and supporting the notion that differences in melatonin pathways in ASD may play a role in sleep disturbance, exogenous melatonin has been found to be efficacious in treating sleep problems within this population (for systematic review, see Guénolé et al., 2011; for meta-analysis, see Rossignol & Frye, 2011).

Another explanation is found in common physical medical comorbidities. As noted previously, children with ASD often present with a number of medical comorbidities such as epilepsy and gastrointestinal issues (Bauman, 2010; Doshi-Velez, Ge, & Kohane, 2014; Tuchman & Rapin, 2002), which have been found to be related to sleep problems both within TD children and other clinical populations (Batista & Nunes, 2007; Noronha et al., 2009). This has led to the suggestion that it may be these comorbidities which predispose children with ASD to sleep problems
(Xianchen Liu et al., 2006). Within this population a diagnosis of epilepsy has been found to be associated with sleep disturbance (Accardo & Malow, 2014) which is of importance given that approximately 30% of children with ASD experience seizures at some time (Tuchman & Rapin, 2002). In addition to this, children with ASD have a higher relative risk of gastrointestinal symptoms (Bauman, 2010; Doshi-Velez, Ge, & Kohane, 2014) which also have been found to be associated with sleep problems (Aldinger, Lane, Veenstra-VanderWeele, & Levitt, 2015). More generally, pain as a result of medical comorbidities is predictive of overall sleep disturbance as well as sleep duration, parasomnias, and SDB in ASD (Tudor, Walsh, Mulder, & Lerner, 2014).

With regard to underling sleep architecture, there is some evidence of differences in sleep architecture, based on PSG studies. That is, in addition to PSG findings regarding sleep onset and offset (which are broadly consistent with actigraphy findings; Elia et al., 2000; Lambert et al., 2016; Limoges et al., 2005; Miano et al., 2007), there are mixed findings of possible disruptions in both NREM and REM sleep. Early studies in this field reported a reduction in the overall percentage of REM sleep, which was fragmented by NREM sleep (Diomedi et al., 1999) as well as evidence for REM sleep behaviour disorder in some children with ASD (Thirumalai, Shubin, & Robinson, 2002). However, these findings have often not been replicated. A number of more recent studies have reported that there were no group difference in REM duration or timing (Bruni et al., 2007; Elia et al., 2000; Lambert et al., 2016; Limoges et al., 2005; Miano et al., 2007; Tani et al., 2004), between those with and without ASD, with only one study recording a lower number of rapid eye movements during REM sleep (i.e. decreased REM density; Limoges et al., 2005). Some of these studies have, however, found discrepancies in NREM
sleep, with an increased duration of stage 1 sleep (Limoges et al., 2005), and reductions in slow-wave sleep (stages 3 and 4; Lambert et al., 2016; Limoges et al., 2005); although these were often only subtle. One research group has also examined cyclic alternating pattern during NREM, describing some differences in sleep stability between children with ASD and their TD peers (Bruni et al., 2007; Miano et al., 2007). While these differences were noted, PSG studies varied in their consideration of protentional confounders, such as accounting for the first-night effect, or ensuring adequate duration of sleep.

1.9.2 Precipitating factors

Outside of physical comorbidities, psychiatric conditions including both internalising and externalising disorders, are commonly linked to sleep problems in the wider TD population and are likely to impact upon sleep in ASD (DeVincent, Gadow, Delosh, & Geller, 2007; Hollway & Aman, 2011; Hollway, Aman, & Butter, 2013; Rzepecka, Mckenzie, McClure, & Murphy, 2011). Studies have found that children with ASD and poor sleep experience more depressive symptoms than those with good sleep (Malow, Marzec et al., 2006), while others have noted that both symptoms of depression and anxiety are related to sleep disturbance in ASD (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; DeVincent et al., 2007; Mayes & Calhoun, 2009). Further, there is evidence that a number of sleep problems are related to both hyperactivity (Chervin et al., 2002; DeVincent et al., 2007; Golan, Shahar, Ravid, & Pillar, 2004; Goldman et al., 2011; Mayes & Calhoun, 2009; Park et al., 2012) and other challenging behaviours (Gillott, Furniss, & Walter, 2001; Goldman et al., 2011; Rzepecka et al., 2011) within ASD populations. Of note, the relationship between internalising and externalising disorders and sleep is a
complicated one, with evidence that the relationship is likely bidirectional (see Gregory & Sadeh, 2016, for review).

Given the common medical and psychiatric comorbidities seen in ASD there are high rates of the use of medications such as stimulants, antidepressants, antipsychotics and antiepileptics in this population (Murray et al., 2014). In particular, within paediatric groups, stimulant medications have been found to disrupt sleep (Sangal et al., 2006). Within ASD samples specifically, there is evidence that use of medications, including antipsychotics (Liu et al. 2006; Krakowiak et al., 2008), and stimulants (Mayes, Calhoun, Bixler, Vgontzas et al., 2009) may be associated with sleep problems. However, as noted by Hollway and Aman (2011) use of these medications are indicators of other behavioural and psychiatric features which may impact upon sleep directly.

1.9.3 Perpetuating factors

As noted by Cortesi et al. (2010), given the types of sleep problems observed in ASD (e.g. increase SOL), poor sleep may be best accounted for by behavioural issues and environmental factors, such as parental limit-setting or bedtime routines (Cortesi et al., 2010); indeed, it was an earlier finding of Wiggs and Stores (2004) who had established that sleep problems in a group of children with ASD were best accounted for by behavioural and sleep-onset association sleep disorders. In general, sleep quality is facilitated by sound sleep hygiene practices such as regular bedtimes, reduction in stimulation in the period before sleep, reduced screen time and consistent routines around bedtime (Mindell, Meltzer, Carskadon, & Chervin, 2009). For example, within TD populations, shorter sleep duration and difficulties with morning waking have been found to be associated with increased screen time (> 2 hours per day; Garmy, Nyberg, & Jakobsson, 2012). Importantly, while screen use in
the bedroom is associated with reduced sleep duration across all children (i.e. both ASD and TD populations), the association has been found to be strongest for children with ASD (Engelhardt, Mazurek, & Sohl, 2013). Similarly, within ASD populations, consistent routines such as adherence to a set bedtime and consistent sleep location, have been found to be associated with fewer parent-reported sleep problems (Henderson, Barry, Bader, & Jordan, 2011). Together, these findings indicate that sound sleep hygiene practices may be of even greater importance for children with ASD.

1.9.4 Other considerations

Given that sleep problems are common across children with a range of NDD, some have suggested that sleep problems in ASD may be best accounted for by common features across NDD, such as the presence of a comorbid ID. While some studies have identified an association between intelligence quotient (IQ) and the severity of sleep problems in ASD (Giannotti et al., 2008; Taylor, Schreck, & Mulick, 2012), others have not replicated this finding (Krakowiak et al., 2008), with sleep problems commonly observed even in the absence of an ID (Richdale & Schreck, 2009).

Overall, given the heterogeneity of ASD, sleep disturbances within this population likely occur as a result of a complex interaction between a number of biological, psychological, and environmental factors. This complex aetiology makes the design of interventions challenging, with the suitability of different interventions, or the combination of various interventions, depending on the specific underlying causes of sleep problems. As interventions for sleep disorders in ASD are not a focus of this thesis, they will not be reviewed here in detail; however, for a comprehensive overview, see recent meta-synthesis by Cuomo et al. (2017).
1.10 Functional impact of poor sleep in ASD

As noted previously, the interest in sleep problems in the literature is largely due to the fact that poor sleep is understood to impact upon daytime functioning in a number of domains including cognition (e.g. Gruber et al., 2010; Kopasz et al., 2010; Paavonen et al., 2010), mood (for reviews see Gregory & Sadeh, 2012, 2016), and behavioural regulation (e.g. Aronen, Paavonen, Fjällberg, Soininen, & Törrönen, 2000; Becker, Langberg, & Evans, 2015; Lavigne et al., 1999; Touchette et al., 2007).

Historically, much of the literature in this area has centred on neurotypical adult populations (e.g. see Waters & Bucks, 2011, for review). Within adult populations, the most significant cognitive deficits are seen in attentional vigilance and processing speed, with some evidence of deficits in the domains of memory and executive functioning (EF). A meta-analysis exploring the effects of short-term total sleep deprivation on cognition found performance decrements in both simple and complex attention as well as working memory, with a smaller impact on short-term memory (Lim & Dinges, 2010).

A number of theories have been put forward to explain the impact of poor sleep or sleep deprivation on daytime cognition. One popular explanation is that the observed deficits are the result of “state instability” (Doran, Van Dongen, & Dinges, 2001) caused by competition between the desire for sleep and circadian factors promoting daytime wakefulness. This results in fluctuations in vigilance, producing a pattern of variable performance over time. According to this framework, cognitive decrements could be explained by the central function that vigilance (i.e. attention) plays a large role in many higher-order tasks, such as working memory and planning. An alternative group of theories is that sleep loss has a direct impact on
neurobiological functions. It is proposed that sleep may impact directly on the prefrontal cortex (Beebe & Gozal, 2002) and the hippocampus (Walker & Stickgold, 2006) producing temporary changes in cerebral metabolism. These changes are thought to impact on the efficiency of the brain regions, resulting in deficits in EF and memory performance. Due to the principal role of the prefrontal cortex in modulating emotions and behaviours, chronic sleep loss is also linked to increased negative mood and behavioural disturbances.

While historically much of the literature has centred on adult sleep and daytime correlates, the past few decades have seen an increased focus of research for paediatric populations as well. Meta-analytic findings from studies examining cognition in otherwise healthy TD children show that reduced sleep duration is associated with problems in cognition and internalising/externalising behaviour (Astill, Van der Heijden, Van Ijzendoorn, & Van Someren, 2012) while sleep variables such as sleep quality, sleep quantity, and daytime alertness are all positively related to school outcomes (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010).

As noted, within ASD populations specifically, many studies have focused on the link between sleep and the core features of ASD. For example, there is evidence that habitually shorter sleep duration and poor sleep quality are related to reduced verbal skills (Taylor, Schreck, & Mulick, 2012), communication deficits (Schreck, Mulick, & Smith, 2004), and increased RRBI (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Goldman et al., 2011; Malow, McGrew, Harvey, Henderson, & Stone, 2006; Schreck, Mulick, & Smith, 2004; Taylor et al., 2012). However, these aforementioned findings are based on correlational research and as such, causal associations cannot be established.
1.10.1 Cognition

Consistent with the TD literature (e.g. Buckhalt, El-Sheikh, Keller, & Kelly, 2009; El-Sheikh, Buckhalt, Keller, Cummings, & Acebo, 2007) poor sleep in ASD has been found to be associated with lower non-verbal (Elia et al., 2000; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005) and full-scale (Taylor et al., 2012) intelligence, as well as poorer school outcomes (Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt, 2003). Again, these findings are based on correlational research. Furthermore, little work has focused on understanding these general impairments at a more detailed level, with few studies exploring the relationships between sleep and specific cognitive domains (such as attention or EF) which may drive these associations.

Of the few studies that have examined the relationship between sleep and specific cognitive domains in individuals with ASD (Limoges, Bolduc, Berthiaume, Mottron, & Godbout, 2013; Maski et al., 2015), findings have been mixed. Limoges et al. (2013) assessed sleep and cognition in a small sample of young adults with ASD. They found that, while PSG-determined signs of poor sleep (e.g. increased SOL, increased stage 1 sleep and increased WASO) were associated with impaired selective attention and procedural memory, no associations were found with working memory. Maski et al. (2015) assessed sleep-dependent memory consolidation in children with ASD. While children with ASD were found to have reduced SE when compared to controls, both groups demonstrated better memory consolidation after a period of sleep; albeit, to a lesser degree than TD controls. This highlights the similar role that sleep plays in stabilising memory for children across groups, regardless of diagnosis (Maski et al., 2015).
1.10.2 Behavioural regulation

With regard to mood and emotional regulation, again consistent with findings from the TD literature (e.g. Baum et al., 2014; Dagys et al., 2012; Gregory et al., 2005; Gregory & O’Connor, 2002; Roberts & Duong, 2013; Tamura & Tanaka, 2014), sleep problems in ASD have been found to be associated with anxiety and depression (Hollway et al., 2013; Mayes & Calhoun, 2009; Mazurek & Petroski, 2015; Rzepecka et al., 2011). Again, while the results reported are based on correlational research, more recent longitudinal findings suggest that in school-aged children, parent-reported sleep problems indeed predict symptoms of anxiety one year later (May et al., 2015), while others have found that reductions in parent-reported sleep problems over time were also associated with reduced symptoms of anxiety 12–15 months later (Fletcher et al., 2017).

Problem sleep in ASD has been similarly linked to daytime behaviour regulation issues (Gabriels et al., 2005; Paavonen et al., 2003; Mayes & Calhoun, 2009; Sikora et al., 2012; Taylor et al., 2012), with a number of studies finding a link between sleep problems and challenging behaviours (Gabriels et al., 2005; Mayes & Calhoun, 2009; Sikora et al., 2012; Taylor et al., 2012). For example, many studies have found that children with ASD and parent-reported “good sleep” have significantly fewer externalising behaviours than those children with ASD and parent-reported “poor sleep” (Adams, Matson, & Jang, 2014; Goldman et al., 2009, 2011; Sikora et al., 2012). Others have reported that sleep problems can account for up to 32% of the variance in challenging daytime behaviours (Mazurek & Sohl, 2016) such as increased tantrums and aggression (Henderson et al., 2011; Goldman et al., 2011). Furthermore, behavioural problems have been found to improve after a sleep remediation program involving melatonin (Paavonen et al., 2003).
Importantly, while the findings of a link between behavioural problems and sleep are fairly consistent in the literature, the majority of studies have relied on parent-reported measures of both sleep and behaviour. For example, Sikora et al. (2012) found significantly higher parent-reported behavioural difficulties in those children with both ASD and parent-reported sleep problems when compared to those without sleep problems. Goldman et al. (2009) assessed sleep in children with ASD using both parent-report (single item) and actigraphy, concluding that children with parent-reported “poor sleep” had more inattention and hyperactivity than those with parent-reported “good sleep”. However, while Goldman et al. (2009) employed actigraphy to assess sleep, no comparisons were made between actigraphy-derived sleep problems and daytime behaviours.

1.11 Overview of the Current Research and Summary of Aims

Given the considerable impact of poor sleep on health, cognition, mood and behaviour, an understanding of sleep profiles, especially within vulnerable populations, is imperative. Children with ASD have been identified in the literature as one such vulnerable population, leading to a marked recent increase in the number of studies focusing on sleep problems and their correlates within this group. Despite the focus on sleep in this population, a number of gaps in knowledge remain.

As elaborated upon below, the current thesis aims to address a number of these gaps with a focus on, firstly, exploring the measurement of sleep and sleep problems in ASD; secondly, establishing the trajectory of sleep problems and their relation to ASD symptom severity over time; and thirdly, examining the broader behavioural impact of sleep problems in ASD, with a focus on the potential underlying cognitive mechanisms.
Firstly, to gain a clear understating of sleep, valid and reliable assessment of this behaviour is essential. Despite extensive use of the CSHQ, assessment of the psychometric properties of the scale within the ASD population is limited. To this end, Chapter 2 presents a study exploring the use of the CSHQ in a large, well-characterised sample of school-aged children with ASD. This chapter reports an analysis of the factor structure of the scale as well as preliminary concurrent reliability through comparisons with the SDSC.

Following this, Chapter 3 explores the stability of sleep problems over time in children with ASD and examines the association between sleep problems and core symptoms of the disorder. Specifically, given suggestions that sleep problems may be explained by symptom severity in ASD, the study investigates the longitudinal predictive relationship between parent-reported sleep problems and core ASD symptoms over a period of three to five years.

The following two chapters investigate differences in sleep profiles between ASD and TD children of school age, as well as how sleep variables relate to challenging daytime behaviours. Chapter 4 focuses on differences in sleep patterns between ASD and TD groups as assessed by both parent-report and actigraphy. This chapter also examines how parent-reported sleep problems compare to objectively measured sleep parameters across ASD and TD groups. Chapter 5 explores how these sleep profiles (both objective and subjective) relate to challenging externalising behaviour. Finally, this study considers potential cognitive factors – namely, EF skills – which may account for the relationship between sleep and challenging behaviours.

Finally, the results of the four empirical chapters are summarised in Chapter 6, the general discussion. Here, the main findings and limitations of the studies are
discussed and interpreted in the context of the current sleep literature. Suggestions for future research are described along with clinical implications of the findings. Given that the growing focus on sleep ASD populations are due, in large part, to consistent parental reports of sleep problems, it is important for research to ensure that these concerns are fully explored. In refining assessment of sleep problems and examining their daytime correlates, it is hoped that these findings may contribute to guiding intervention and mitigate or reduce the potential daytime sequelae of poor sleep.
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Chapter 2: Psychometric Properties of the Children’s Sleep Habits Questionnaire in Children with Autism Spectrum Disorder

2.1 Preamble to Chapter 2

As the first of the empirical chapters presented in this thesis, Chapter 2 focuses on the most commonly employed questionnaire measure for sleep in children with autism spectrum disorder (ASD): the Children’s Sleep Habits Questionnaire (CSHQ). Review of the literature highlights some of the gaps in past explorations of the psychometric properties of the scale in ASD populations. To address these gaps, the study then details factor analyses leading to a revised version of the CSHQ (CSHQ-R), as well as a preliminary assessment of convergent validity of the CSHQ-R. This chapter creates a foundation for subsequent papers as the CSHQ-R is employed in the three remaining empirical chapters to examine sleep in children with ASD.
Psychometric Properties of the Children’s Sleep Habits Questionnaire in Children with Autism Spectrum Disorder

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Abstract

Sleep problems are common in children with autism spectrum disorder (ASD). Despite limited psychometric evaluation, the Children’s Sleep Habits Questionnaire (CSHQ) is among the most widely used measure of sleep in ASD. The purpose of this study was to explore the psychometric properties of the CSHQ in a large community-based sample of children with ASD (N = 458). Confirmatory factor analyses were conducted to replicate previous CSHQ subscales; however, models demonstrated poor fit. Follow-up exploratory factor analyses showed that a five-factor solution yielded the most appropriate fit for the current data. Overall, these five factors show some consistency with the subscales proposed in the original CSHQ. The suggested five subscales may provide a more appropriate measurement structure for assessing sleep in ASD.

Keywords: Autism. Autism Spectrum Disorder. Children’s Sleep Habits Questionnaire. Sleep. Sleep disturbance. Sleep questionnaire.
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2.2 Introduction

Fifty to eighty percent of children with autism spectrum disorder (ASD) have been reported to experience problems with sleep (Couturier et al., 2005; Fadini et al., 2015; Paavonen et al., 2008; Rzepecka, Mckenzie, McClure, & Murphy, 2011; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012). Commonly reported sleep problems include delays in sleep onset and early waking (Patzold, Richdale, & Tonge, 1998; Richdale & Prior, 1995), as well as fragmented sleep, with an increase in both the number and duration of episodes of wake after sleep onset (Wiggs & Stores, 2004). Sleep problems in ASD not only impact the child’s cognitive functioning (Elia et al., 2000; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt, 2003; Schreck, Mulick, & Smith, 2004; M. A. Taylor, Schreck, & Mulick, 2012) and behaviour (Gabriels et al., 2005; Paavonen et al., 2003; Sikora, Johnson, Clemons, & Katz, 2012; M. A. Taylor et al., 2012), but also parental stress and well-being (Doo & Wing, 2006). Given the rising incidence of ASD (currently 1:68; Baio, 2014) and the associations between sleep and functioning for both the child and family system, this area of study has gained increasing focus.

In order to adequately detect sleep problems in ASD, studies have relied heavily on parent-rated questionnaires. While tools, such as polysomnography (PSG) or actigraphy, exist to capture objective sleep parameters, these are both costly and may be poorly tolerated by children with ASD who have sensory sensitivities (Hodge, Parnell, Hoffman, & Sweeney, 2012). Further, although such tools have been found to reliably record sleep parameters (i.e. sleep and wake time), they do not capture other bedtime behaviours such as bedtime resistance or co-sleeping. Due to this, a valid and reliable questionnaire-based measure of sleep and sleep-related
behaviours in children with ASD is important in ongoing sleep research and in clinical practice.

One such questionnaire is the Children’s Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000) which has frequently been used to measure sleep problems in ASD. While the CSHQ was grounded in a sound theoretical framework (detailed below), there are several limitations to its current use. Firstly, although theoretically sound, the original scale was subject to only limited psychometric analysis during the scale development stage. This was highlighted in a recent review of paediatric sleep scales, which found that the CSHQ met only 5 of the 11 steps necessary for sound instrument development (see Spruyt & Gozal, 2011). Specifically, neither exploratory nor confirmatory factor analyses (EFA and CFA respectively) were conducted in the original CSHQ study (Owens et al., 2000), and have not been performed for an English-speaking, typically developing (TD), school-aged population since. This is in contrast to the Sleep Disturbances Scale for Children (SDSC; Bruni et al., 1996), which met all 11 steps of scale development (Spruyt & Gozal, 2011). Secondly, despite the frequent use of the CSHQ for children with ASD for more than a decade, at the time of completing the current research, only one other study had assessed the psychometric properties and factor structure of the CSHQ in this population, albeit with notable limitations as will be discussed below (see Johnson et al., 2016). Again, this is in contrast to the SDSC which, while not validated in ASD population, has been validated in a sample of children with a range of clinical and neurodevelopmental disorders (NDD; Marriner, Pestell, Bayliss, McCann, & Bucks, 2017). Further, while the CSHQ was originally intended for primary school aged children (4-10 years of age), many have used the scale outside of this age range without evidence that the scale’s interpretation is consistent across age groups (e.g. Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014). Only one
study has explored the factor structure of the CSHQ in a TD, pre-school aged sample (see Sneddon, Peacock, & Crowley, 2013), finding a four-factor solution with just 24 items, which differed substantially from the structure originally proposed for the CSHQ. There have also been inconsistencies in methods to adapt the scale for use in pre-school aged children. For example, previous studies have removed or adjusted scoring on certain items which were thought to be developmentally inappropriate, without thorough psychometric analysis (e.g. Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008; Sneddon et al., 2013). Two of the primary concerns detailed above, relating to sound scale development and lack of psychometric evaluation in ASD, will be further discussed below.

2.2.1 Original construction and validation of the CSHQ

The CSHQ was developed as a screening tool, to measure sleep problems in children aged between 4-10 years (Owens et al., 2000). Items were based on the International Classification of Sleep Disorders (ICSD) manual. As described by the authors, they were written to address the major presenting complaints in primary school-aged children: bedtime behaviour (including sleep onset); sleep duration; anxiety around sleep; behaviours during sleep (i.e. waking during the night); sleep-disordered breathing (SDB) and parasomnias. Questions were also developed to target morning waking and somnolence throughout the day (Owens et al., 2000). An initial 48 items were reduced to a 33-item scale (two items of which were repeated in different categories) based on qualitative analysis (i.e. 15 items were deemed redundant or ambiguous based on author consensus; Owens et al., 2000). This ‘original’ CSHQ (hereafter referred to as CSHQ-original) was assessed for internal consistency in a large sample of children both with (n =154) and without (n = 469) sleep disturbance (Owens et al., 2000). However, the scale was never subject to item-
level or structure analysis (such as EFA or CFA). Rather, items were “group[ed] conceptually” based on the eight domains outlined above (Owens et al., 2000, pp. 8) with subscales containing one to eight items each. Internal consistency values of the subscales ranged from $\alpha = 0.36$ (unacceptable) to $\alpha = 0.93$ (good); with fewer than half of the subscales demonstrating internal consistency above the minimally acceptable level of $\alpha = 0.70$ (Nunnally & Bernstein, 1994).

While there has yet to be any structural validation of the subscales of the CSHQ-original in an English speaking TD school-aged sample, there have been attempts to assess psychometric properties in different language versions of the CSHQ through both EFA and CFA (Chinese: Liu, Wang, Tang, Wen, & Li, 2014; Spanish: Lucas-de la Cruz et al., 2016; German: Schlarb, Schwerdtle, & Hautzinger, 2010; and Dutch: Waumans et al., 2010). Of the factor analyses using non-English versions of the scale, none have confirmed the original subscales suggested by Owens et al. (2000). Rather, they have all found different factor solutions (ranging from four to eight factors) with different numbers of items (27 to 29). While these inconsistencies may reflect true cross-cultural differences in sleep behaviours, it is also possible that the limited psychometric analysis in the initial stages of scale development has played an ongoing role in these inconsistencies. These discrepancies highlight the need for further, systematic scale development.

2.2.2 The CSHQ in ASD

Up to the time of completion of the present study, only one other study had examined the psychometric properties of the CSHQ in children with ASD. Johnson et al. (2016) completed a principal components analysis with data from 310 English-speaking children with ASD, aged 2-10 years. The analyses yielded a five-component solution, based on retention of 27 of the 33 original items (Johnson et al.,
2016); hereafter their final model will be referred to as the “CSHQ-ASD” model. However, as noted by Johnson et al. (2016), their study was limited by the fact that over 50% of the sample had marked disruptive behaviours as they were recruited through a study focusing on parental training. A further 11% of the sample were recruited specifically due to known behavioural sleep disturbance, and other potential participants were excluded based on medical sleep conditions such as sleep apnoea. Given these limitations, and considering the widespread use of the CSHQ-original in ASD populations, it is critical to explore the factor structure in a representative sample of children with ASD without these same biases.

2.2.3 The current study
In summary, the main concerns regarding the current use of the CSHQ are that the original CSHQ lacked thorough psychometric evaluation; and that apart from one recent study (Johnson et al., 2016), this scale has often been employed in ASD groups without any attempts at exploring the factor structure in this population. Therefore, the aims of the current study were to: (i) attempt to replicate both the CSHQ-original and CSHQ-ASD, using CFA in a sample of children with ASD, to establish whether either of the two previously proposed scales demonstrates good fit for the data; (ii) if the items did not demonstrate good fit with the existing scales, explore the component structure of the CSHQ in a sample of children with ASD to create a revised scale; and (iii) examine the concurrent reliability of the revised scale.

2.3 Method
2.3.1 Participants
The current study included 458 children with ASD, aged between 2 and 18 years. Cases were drawn from the Western Australian community through two previously conducted studies: the Western Australian Autism Biological Registry
Chapter 2: Psychometric Properties of the Children’s Sleep Habits Questionnaire in Children with Autism Spectrum Disorder

(WAABR; 266, 58%; Telethon Kids Institute, see L. J. Taylor et al., 2013 for more detail); and the Val Lishman Project (VLP; 192, 42%; Curtin University). Initial recruitment for both studies was directly from the Western Australian community through advertising in diagnostic clinics, intervention centres, community noticeboards, and university/institute open days. The only criterion for study inclusion was a clinical diagnosis of ASD. Ethics approvals were granted by the Human Ethics Committee at Princess Margaret Hospital for Children (1845/EP) and Curtin University Human Research Ethics Office (HR123/2014) respectively. Participants from these databases were included in the current study if their parent or caregiver had completed the CSHQ (minimum of 85% items answered).

Caregivers reported a clinical diagnosis of Autistic Disorder (AD; 74.2%), Asperger Syndrome/High Functioning Autism (AS; 18.1%) or Pervasive Developmental Disorder–Not Otherwise Specified (PDD-NOS; 7.6%) for their child (Table 2.1). In Western Australia, this diagnosis, based on the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2000; 2013) criteria, is established through clinical team consensus involving a paediatrician, psychologist, and speech-language pathologist (Glasson et al., 2008).

2.3.2 Measures

Children’s Sleep Habits Questionnaire

The CSHQ (Owens et al., 2000) is available in 48-item and 33-item versions, with scoring for the core 33-items common across both. While 42% of the current sample completed the 48-item version (i.e. all VLP participants), analyses were conducted only on the 33 items common between the two subsamples. Items were rated on a three-point scale. Items 1-31 are rated on a scale 1 = “Rarely” (0-1 night/week), 2 = “Sometimes” (2-4 nights/week) and 3 = “Usually” (5-7 nights/week).
### Table 2.1
Demographic Information for the Current Sample

<table>
<thead>
<tr>
<th>Characteristics</th>
<th>WAABR</th>
<th>VLP</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n = 266</td>
<td>n = 192</td>
</tr>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Child's age category</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Pre-school (2-5 years)</td>
<td>114 (42.9)</td>
<td>51 (26.6)</td>
</tr>
<tr>
<td>Primary (elementary) school (6-11 years)</td>
<td>108 (40.6)</td>
<td>86 (44.8)</td>
</tr>
<tr>
<td>High school (12-18 years)</td>
<td>44 (16.5)</td>
<td>55 (28.6)</td>
</tr>
<tr>
<td>Child's sex</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Male</td>
<td>213 (80.1)</td>
<td>156 (81.3)</td>
</tr>
<tr>
<td>Female</td>
<td>53 (19.9)</td>
<td>36 (18.8)</td>
</tr>
<tr>
<td>ASD Diagnosis (clinician diagnosed)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Autistic Disorder</td>
<td>227 (85.3)</td>
<td>113 (58.9)</td>
</tr>
<tr>
<td>Asperger Syndrome</td>
<td>17 (6.4)</td>
<td>66 (34.4)</td>
</tr>
<tr>
<td>PDD-NOS</td>
<td>22 (8.3)</td>
<td>13 (6.8)</td>
</tr>
<tr>
<td>Comorbidities (clinician diagnosed)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Seizure a</td>
<td>44 (16.5)</td>
<td>25 (13)</td>
</tr>
<tr>
<td>ID or GDD</td>
<td>73 (27.4)</td>
<td>38 (19.8)</td>
</tr>
<tr>
<td>Any Physical Condition</td>
<td>66 (24.8)</td>
<td>40 (20.8)</td>
</tr>
<tr>
<td>Any Mental Health Diagnosis</td>
<td>19 (7.1)</td>
<td>36 (18.8)</td>
</tr>
<tr>
<td>Child’s age at clinical diagnosis</td>
<td></td>
<td></td>
</tr>
<tr>
<td>&lt; 3</td>
<td>60 (22.6)</td>
<td>36 (18.7)</td>
</tr>
<tr>
<td>3 years</td>
<td>63 (23.7)</td>
<td>46 (24.0)</td>
</tr>
<tr>
<td>4-6 years</td>
<td>79 (29.7)</td>
<td>66 (34.4)</td>
</tr>
<tr>
<td>7-10 years</td>
<td>33 (12.4)</td>
<td>29 (15.1)</td>
</tr>
<tr>
<td>&gt;10</td>
<td>17 (6.4)</td>
<td>15 (7.8)</td>
</tr>
<tr>
<td>Not reported</td>
<td>14 (5.3)</td>
<td>0</td>
</tr>
<tr>
<td>Caregiver completing the questionnaire</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mother</td>
<td>175 (65.8)</td>
<td>177 (92.2)</td>
</tr>
<tr>
<td>Father</td>
<td>8 (3.0)</td>
<td>12 (6.3)</td>
</tr>
<tr>
<td>Both parents together</td>
<td>69 (25.9)</td>
<td>0</td>
</tr>
<tr>
<td>Other/Not reported</td>
<td>14 (5.3)</td>
<td>3 (1.6)</td>
</tr>
<tr>
<td>Has taken melatonin at any time</td>
<td>24 (9.0)</td>
<td>54 (28.1)</td>
</tr>
<tr>
<td>Child's age (months) at time of CSHQ</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Mean</td>
<td>94.24</td>
<td>111.50</td>
</tr>
<tr>
<td>SD</td>
<td>46.50</td>
<td>49.63</td>
</tr>
<tr>
<td>Range</td>
<td>27-215</td>
<td>24-215</td>
</tr>
</tbody>
</table>

Note. WAABR = Western Australian Autism Biological Registry; VLP = the Val Lishman Project; PDD-NOS = Pervasive Developmental Disorder–Not Otherwise Specified; ID = Intellectual Disability; GDD = Global Developmental Delay. a Has had at least one seizure at any time.
For items 32 and 33, 1 = “Not Sleepy”, 2 = “Very Sleepy”, and 3 = “Falls Asleep”. The scale has a scoring range of 33-99, with higher scores indicating greater levels of sleep disturbance. To this end, five items are reverse scored.

*Sleep Disturbances Scale for Children*

The SDSC (Bruni et al., 1996) is a 26-item parent-report questionnaire examining a child’s sleep habits over the previous 6 months. Statements regarding sleep behaviours are rated on a five-point scale indicating how often the behaviour is observed (e.g. 1 = “Never”, to 5 = “Always”). The questionnaire includes six subscales: difficulty in initiating and maintaining sleep (DIMS); disorders of excessive somnolence (DOES); sleep hyperhidrosis (SH; i.e. night sweats); sleep breathing disorders (SBD); disorders of arousal (DOA) and sleep-wake transition disorders (SWTD), which are then summed to provide a total sleep score. The scale developers reported good internal consistency (Cronbach’s $\alpha = 0.71$ to 0.79), good test-retest reliability ($r = 0.71$), as well as acceptable sensitivity (0.89) and specificity (0.74) in detecting children with sleep disorders (Bruni et al., 1996). In the current study, both the total and subscales scores (excluding SH) were used to examine the concurrent validity of the revised CSHQ in a random subsample of children with ASD in the current sample (n = 36), for whom data from both the SDSC and CSHQ were available.

*Demographic information*

Parents also provided information on their child’s clinical diagnosis of ASD, clinician-diagnosed comorbidities, and use of medication to support sleep (i.e. melatonin supplements; see Table 2.1).
2.3.3 Statistical analyses

All general data analyses, including data screening, were performed using SPSS Statistics Version 22.0 for Windows. CSHQ data were screened for missing values using Little’s MCAR test. Item data were missing completely at random (MCAR), $\chi^2 (1285) = 1361.41, p > 0.05$, and fewer than 5% (0.66%) of total values were missing, which can be considered inconsequential (Schafer, 1999). Missing values were therefore imputed using Expectation Maximization procedures.

![Figure 2.1. Model A Confirmatory factor analysis model for the CSHQ-original. Note. Residual errors are not shown in the model for the sake of simplicity. BR = Bedtime Resistance; SD = Sleep Duration; SIAnx = Sleep Anxiety; NW = Night-time Wakings; Para = Parasomnias; SDB = Sleep Disordered Breathing; DS = Daytime Sleepiness.](image)

As the currently suggested subscales of the CSHQ (-original and -ASD) had not previously been confirmed through CFA, initial analysis was conducted to determine the fit for these models. CFA was conducted using PASW Statistics AMOS, Version 22. As two models were identified in the literature, each was tested on the current data: the CSHQ-original (Owens et al., 2000; Figure 1) and the CSHQ-ASD (Johnson et al., 2016; Figure 2). As noted previously, one of the subscales (Sleep Onset Delay) on the CSHQ-original had only one item (see Table 2.2, Item 7). As CFA in AMOS cannot be performed with single-item factors, this
item (“Child falls asleep within 20 minutes after going to bed”) was grouped with conceptually similar items on the Sleep Duration subscale. This revision also maintained good internal consistency for the subscale with Cronbach’s $\alpha = 0.78$.

Acceptable model fit was evaluated using an absolute close-fit index, Root Mean Square Error of Approximation (RMSEA), and two incremental close fit indices, the Comparative Fit Index (CFI), and Tucker-Lewis Index (TLI). Guidelines provided by Hu and Bentler (1999) state that values greater than 0.95 for the CFI and TLI, and less than 0.05 for RMSEA, indicate “relatively good” fit between the observed data and the hypothesised model. Given the sensitivity of chi-square to sample size (Hooper, Coughlan, & Mullen, 2008), chi-square adjusted for degrees of freedom was calculated, where reasonable fit is indicated when the value is less than three (Hu & Bentler, 1999).

Figure 2.2. Model B Confirmatory factor analysis model for the CSHQ-ASD. 
Note. Residual errors are not shown in the model for the sake of simplicity. SRP = Sleep Routine Problems; IS = Insufficient Sleep; SOAP = Sleep Onset Association Problems; Para/SDB = Parasomnias/Sleep Disordered Breathing; SlAnx = Sleep Anxiety.

As the use of EFA was dependent on whether or not the current data fit the already proposed CSHQ factor structures, the results of the CFA analyses will be presented first below.
### Table 2.2

**Internal Consistency, Means, Standard Deviations and Item-Total Correlations for the Original 33-Item, 8-Subscale Children’s Sleep Habits Questionnaire**

<table>
<thead>
<tr>
<th>Item #</th>
<th>Item wording</th>
<th>Rarely Freq</th>
<th>Rarely %</th>
<th>Sometimes Freq</th>
<th>Sometimes %</th>
<th>Usually Freq</th>
<th>Usually %</th>
<th>M</th>
<th>SD</th>
<th>r</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td><strong>Bedtime Resistance (BR)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>1</td>
<td>Child goes to bed at the same time at night R</td>
<td>36</td>
<td>7.86</td>
<td>66</td>
<td>14.41</td>
<td>356</td>
<td>77.73</td>
<td>1.30</td>
<td>0.61</td>
<td>0.40</td>
</tr>
<tr>
<td>2</td>
<td>Child falls asleep alone in own bed R</td>
<td>58</td>
<td>12.66</td>
<td>28</td>
<td>6.11</td>
<td>372</td>
<td>81.22</td>
<td>1.31</td>
<td>0.69</td>
<td>0.32</td>
</tr>
<tr>
<td>3</td>
<td>Child falls asleep in parent’s or sibling’s bed</td>
<td>357</td>
<td>77.95</td>
<td>56</td>
<td>12.23</td>
<td>45</td>
<td>9.83</td>
<td>1.32</td>
<td>0.64</td>
<td>0.34</td>
</tr>
<tr>
<td>4</td>
<td>Child needs parent in the room to fall asleep</td>
<td>314</td>
<td>68.56</td>
<td>51</td>
<td>11.14</td>
<td>93</td>
<td>20.31</td>
<td>1.52</td>
<td>0.81</td>
<td>0.36</td>
</tr>
<tr>
<td>5</td>
<td>Child struggles at bedtime</td>
<td>265</td>
<td>57.86</td>
<td>111</td>
<td>24.24</td>
<td>82</td>
<td>17.90</td>
<td>1.60</td>
<td>0.77</td>
<td>0.55</td>
</tr>
<tr>
<td>6</td>
<td>Child is afraid of sleeping alone</td>
<td>305</td>
<td>66.59</td>
<td>79</td>
<td>17.25</td>
<td>74</td>
<td>16.16</td>
<td>1.49</td>
<td>0.76</td>
<td>0.46</td>
</tr>
<tr>
<td>2.</td>
<td><strong>Sleep Onset Delay (SOD)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>1.85</td>
<td>0.79</td>
<td></td>
</tr>
<tr>
<td>7</td>
<td>Child falls asleep within 20 minutes after going to bed R</td>
<td>115</td>
<td>25.11</td>
<td>161</td>
<td>35.15</td>
<td>182</td>
<td>39.74</td>
<td>1.85</td>
<td>0.79</td>
<td>0.35</td>
</tr>
<tr>
<td>3.</td>
<td><strong>Sleep Duration (SD)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>5.01</td>
<td>2.01</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>Child sleeps too little</td>
<td>223</td>
<td>48.69</td>
<td>126</td>
<td>27.51</td>
<td>109</td>
<td>23.80</td>
<td>1.75</td>
<td>0.82</td>
<td>0.55</td>
</tr>
<tr>
<td>9</td>
<td>Child sleeps the right amount R</td>
<td>116</td>
<td>25.33</td>
<td>111</td>
<td>24.24</td>
<td>231</td>
<td>50.44</td>
<td>1.75</td>
<td>0.83</td>
<td>0.55</td>
</tr>
<tr>
<td>10</td>
<td>Child sleeps about the same amount each day R</td>
<td>62</td>
<td>13.54</td>
<td>109</td>
<td>23.80</td>
<td>287</td>
<td>62.66</td>
<td>1.51</td>
<td>0.72</td>
<td>0.49</td>
</tr>
<tr>
<td>4.</td>
<td><strong>Sleep Anxiety (SlA)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>6.34</td>
<td>2.21</td>
<td></td>
</tr>
<tr>
<td>(rep) 4</td>
<td>Child needs parent in the room to fall asleep</td>
<td>314</td>
<td>68.56</td>
<td>51</td>
<td>11.14</td>
<td>93</td>
<td>20.31</td>
<td>1.52</td>
<td>0.81</td>
<td>0.36</td>
</tr>
<tr>
<td>11</td>
<td>Child is afraid of sleeping in the dark</td>
<td>274</td>
<td>59.83</td>
<td>63</td>
<td>13.76</td>
<td>121</td>
<td>26.42</td>
<td>1.67</td>
<td>0.87</td>
<td>0.30</td>
</tr>
<tr>
<td>(rep) 6</td>
<td>Child is afraid of sleeping alone</td>
<td>305</td>
<td>66.59</td>
<td>79</td>
<td>17.25</td>
<td>74</td>
<td>16.16</td>
<td>1.49</td>
<td>0.76</td>
<td>0.46</td>
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<tr>
<td>5.</td>
<td><strong>Night Wakings (NW)</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>4.79</td>
<td>1.78</td>
<td></td>
</tr>
<tr>
<td>13</td>
<td>Child moves to someone else’s bed during the night</td>
<td>313</td>
<td>68.34</td>
<td>84</td>
<td>18.34</td>
<td>61</td>
<td>13.32</td>
<td>1.45</td>
<td>0.72</td>
<td>0.42</td>
</tr>
<tr>
<td>14</td>
<td>Child awakens once during the night</td>
<td>183</td>
<td>39.96</td>
<td>163</td>
<td>35.59</td>
<td>112</td>
<td>24.45</td>
<td>1.84</td>
<td>0.79</td>
<td>0.46</td>
</tr>
<tr>
<td>15</td>
<td>Child awakens more than once during the night</td>
<td>291</td>
<td>63.54</td>
<td>106</td>
<td>23.14</td>
<td>61</td>
<td>13.32</td>
<td>1.50</td>
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<td>0.57</td>
</tr>
<tr>
<td></td>
<td>Description</td>
<td>Count</td>
<td>%</td>
<td>R1</td>
<td>R2</td>
<td>R3</td>
<td>R4</td>
<td>R5</td>
<td>R6</td>
<td></td>
</tr>
<tr>
<td>---</td>
<td>-----------------------------------------------------------------------------</td>
<td>-------</td>
<td>------</td>
<td>-----</td>
<td>-----</td>
<td>-----</td>
<td>-----</td>
<td>-----</td>
<td>-----</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>Parasomnias (Para)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>16</td>
<td>Child wets the bed at night</td>
<td>329</td>
<td>71.83</td>
<td>55</td>
<td>12.01</td>
<td>74</td>
<td>16.16</td>
<td>1.44</td>
<td>0.75</td>
<td>0.19</td>
</tr>
<tr>
<td>17</td>
<td>Child talks during sleep</td>
<td>276</td>
<td>60.26</td>
<td>137</td>
<td>29.91</td>
<td>45</td>
<td>9.83</td>
<td>1.50</td>
<td>0.67</td>
<td>0.36</td>
</tr>
<tr>
<td>18</td>
<td>Child is restless and moves a lot during sleep</td>
<td>161</td>
<td>35.15</td>
<td>148</td>
<td>32.31</td>
<td>149</td>
<td>32.53</td>
<td>1.97</td>
<td>0.82</td>
<td>0.50</td>
</tr>
<tr>
<td>19</td>
<td>Child sleepwalks during the night</td>
<td>406</td>
<td>88.65</td>
<td>39</td>
<td>8.52</td>
<td>13</td>
<td>2.84</td>
<td>1.14</td>
<td>0.42</td>
<td>0.33</td>
</tr>
<tr>
<td>20</td>
<td>Child grind teeth during sleep</td>
<td>279</td>
<td>60.92</td>
<td>117</td>
<td>25.55</td>
<td>62</td>
<td>13.54</td>
<td>1.53</td>
<td>0.72</td>
<td>0.28</td>
</tr>
<tr>
<td>21</td>
<td>Child awakens alarmed by a frightening dream</td>
<td>322</td>
<td>70.31</td>
<td>122</td>
<td>26.64</td>
<td>14</td>
<td>3.06</td>
<td>1.33</td>
<td>0.53</td>
<td>0.40</td>
</tr>
<tr>
<td>22</td>
<td>Child awakens during night screaming, sweating etc.</td>
<td>358</td>
<td>78.17</td>
<td>83</td>
<td>18.12</td>
<td>17</td>
<td>3.71</td>
<td>1.26</td>
<td>0.51</td>
<td>0.45</td>
</tr>
<tr>
<td>7</td>
<td>Sleep Disordered Breathing (SDB)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>23</td>
<td>Child snores loudly</td>
<td>309</td>
<td>67.47</td>
<td>116</td>
<td>25.33</td>
<td>33</td>
<td>7.21</td>
<td>1.40</td>
<td>0.62</td>
<td>0.27</td>
</tr>
<tr>
<td>24</td>
<td>Child seems to stop breathing during sleep</td>
<td>419</td>
<td>91.48</td>
<td>30</td>
<td>6.55</td>
<td>9</td>
<td>1.97</td>
<td>1.10</td>
<td>0.37</td>
<td>0.30</td>
</tr>
<tr>
<td>25</td>
<td>Child snorts and/or gasps during sleep</td>
<td>381</td>
<td>83.19</td>
<td>62</td>
<td>13.54</td>
<td>15</td>
<td>3.28</td>
<td>1.20</td>
<td>0.48</td>
<td>0.37</td>
</tr>
<tr>
<td>8</td>
<td>Daytime Sleepiness (DS)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>26</td>
<td>Child wakes up by him/herself</td>
<td>61</td>
<td>13.32</td>
<td>91</td>
<td>19.87</td>
<td>306</td>
<td>66.81</td>
<td>1.46</td>
<td>0.72</td>
<td>0.06</td>
</tr>
<tr>
<td>27</td>
<td>Child wakes up in a negative mood</td>
<td>208</td>
<td>45.41</td>
<td>202</td>
<td>44.10</td>
<td>48</td>
<td>10.48</td>
<td>1.65</td>
<td>0.66</td>
<td>0.43</td>
</tr>
<tr>
<td>28</td>
<td>Adults or siblings wake up child</td>
<td>234</td>
<td>51.09</td>
<td>179</td>
<td>39.08</td>
<td>45</td>
<td>9.83</td>
<td>1.59</td>
<td>0.66</td>
<td>0.23</td>
</tr>
<tr>
<td>29</td>
<td>Child has difficulty getting out of bed in the morning</td>
<td>287</td>
<td>62.66</td>
<td>115</td>
<td>25.11</td>
<td>56</td>
<td>12.23</td>
<td>1.50</td>
<td>0.70</td>
<td>0.35</td>
</tr>
<tr>
<td>30</td>
<td>Child takes a long time to become alert in the morning</td>
<td>282</td>
<td>61.57</td>
<td>117</td>
<td>25.55</td>
<td>59</td>
<td>12.88</td>
<td>1.51</td>
<td>0.71</td>
<td>0.38</td>
</tr>
<tr>
<td>31</td>
<td>Child seems tired in the morning</td>
<td>175</td>
<td>38.21</td>
<td>202</td>
<td>44.10</td>
<td>81</td>
<td>17.69</td>
<td>1.79</td>
<td>0.72</td>
<td>0.43</td>
</tr>
<tr>
<td>32</td>
<td>Watching TV *</td>
<td>349</td>
<td>76.20</td>
<td>84</td>
<td>18.34</td>
<td>25</td>
<td>5.46</td>
<td>1.29</td>
<td>0.56</td>
<td>0.34</td>
</tr>
<tr>
<td>33</td>
<td>Riding in a car*</td>
<td>318</td>
<td>69.43</td>
<td>99</td>
<td>21.62</td>
<td>41</td>
<td>8.95</td>
<td>1.40</td>
<td>0.65</td>
<td>0.22</td>
</tr>
<tr>
<td></td>
<td>Total CSHQ</td>
<td>49.60</td>
<td>9.90</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note. Italicised/bold items were removed prior to exploratory factor analysis due to low corrected item-total correlation (>0.2). r = corrected item-total correlations.

R Denotes items which have been reverse scored. * Denotes item scored on a different scale (1 = Not sleepy, 2 = Very Sleepy, 3 = Falls asleep).
2.4 Results

2.4.1 Confirmatory analyses

As can be seen in Table 2.3, the models derived from the CSHQ-original (Owens et al. 2000) and the CSHQ-ASD (Johnson et al. 2016) were tested using the full data set (i.e. aged 2-18 years; N = 458) and using subsets of the current data set reflecting the age of the sample used by each author respectively (Owens et al. 2000, 4-10 years; current n = 284; Johnson et al. 2016, 2-10 years; current n = 339). Poor fit was recorded for each of the previously suggested CSHQ factor structures, regardless of the age of the sample (Table 2.3).

Table 2.3

<table>
<thead>
<tr>
<th>Sample</th>
<th>Model</th>
<th>( \chi^2 ) (df)</th>
<th>( \chi^2/df )^a</th>
<th>RMSEA (^b)</th>
<th>CFI (^c)</th>
<th>TLI (^c)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Full sample (2-18 yrs)</td>
<td>Model A</td>
<td>3051.295 (495)*</td>
<td>6.164</td>
<td>0.106 (0.103-0.110)</td>
<td>0.446</td>
<td>0.409</td>
</tr>
<tr>
<td></td>
<td>Model B</td>
<td>2234.212 (324)*</td>
<td>6.896</td>
<td>0.113 (0.109-0.118)</td>
<td>0.476</td>
<td>0.433</td>
</tr>
<tr>
<td>4-10 yrs</td>
<td>Model A</td>
<td>2143.740 (495)*</td>
<td>4.331</td>
<td>0.109 (0.104-0.113)</td>
<td>0.439</td>
<td>0.402</td>
</tr>
<tr>
<td>2-10 yrs</td>
<td>Model B</td>
<td>1695.846 (324)*</td>
<td>5.234</td>
<td>0.112 (0.107-0.117)</td>
<td>0.516</td>
<td>0.476</td>
</tr>
</tbody>
</table>

Note. Each model was tested on the full data set (i.e. aged 2-18 years) and on a subset of the current data set reflecting the original sample (i.e. 4-10 years olds for CSHQ-original, n = 284; and 2-10 year olds for CSHQ-ASD, n = 339). RMSEA = Root Mean Square Error of Approximation with 90% confidence intervals; CFI = Comparative Fit Index; TLI = Tucker/Lewis Index.

*a Ideal value <3; \(^b\) Ideal value, <0.05; \(^c\) Ideal value, >0.95
*Denotes p<0.001
2.4.2 Exploratory analyses

Given that fit was poor across both previously suggested factor structures for the CSHQ, EFA was performed to examine potential revisions to subscales within the CSHQ. Prior to EFA, item analysis was conducted for all 33-items. Items with corrected item-total correlations lower than 0.2 were inspected in the context of the overall internal consistency of the scale and ultimately deleted before EFA. Parallel analysis (Horn, 1965) and scree test (Cattell, 1966) criteria were used to determine the number of factors extracted. Items which did not load above |0.30| or with cross-loadings of greater than |0.30| were removed from the final solution. Internal consistencies were recalculated once the final subscales were identified.

**Item analysis**

Corrected item-total correlations, descriptive statistics and endorsement frequencies for each item are presented in Table 2.2. All items demonstrated reasonable means and variances (item means ranged from 1.1 to 1.97 as can be seen in Table 2.2; mean item M = 1.5, item mean SD = 0.21). However, consistent with expectations, some of the item-total correlations were low (below 0.2), indicating that these items were performing poorly in the context of the full-scale. Item 16, “Child wets the bed at night-time” and item 26 “Child wakes up by him/herself” both correlated below 0.2 with the total. Further, no satisfactory factor solution was found when these items were included in the analysis, as such, items 16 and 26 were removed from further analysis.

**Factor structure**

A maximum likelihood procedure with direct oblimin rotation was conducted on the remaining 31 items. While the scree test suggested that four to six factors be extracted from the data set, parallel analysis indicated that a four-factor solution was most appropriate. Upon review of the factor loadings, a further 4 items were removed.
sequentially as they did not load above |0.30| on any of the four factors (items 12, 33, and 32; removed based on lowest loading first) or due to cross-loadings above |0.30| on multiple factors (Item 15). The four factors accounted for 38.54% of the variance. However, items from the SDB and Parasomnias subscales were grouped into a single factor. Given the impact this grouping would have on the clinical utility of the test (i.e. it is less useful to have SDB symptoms and Parasomnia symptoms grouped together), a five-factor solution was explored with the remaining 27 items. This five-factor model was found to be more consistent with the subscales in the CSHQ-original, with the items on the SDB subscale and the Parasomnias subscale neatly splitting into two separate factors. All other factors remained unchanged from the four-factor solution, with the exception of a single cross-loading item between Factor 3 and Factor 5 (Item 6). This revised five-factor solution also accounted for more of the variance in the data (42.39% as opposed to 38.54%). Final item loadings are presented in Table 2.4.

Review of the items grouped into each factor appeared consistent with the reporting of sleep problems in the ASD literature. The subscales that emerged were “Sleep Initiation and Maintenance (Sleep I & M)”, which was comprised of all three items on the original Sleep Duration subscale, as well as similar items from the Bedtime Resistance and Night-time Awakening subscales. Factor 2 was comprised solely of items from the original Daytime Sleepiness subscale, excluding 32 and 33 which were removed. However, review of the item wording indicated that all items were most reflective of morning waking behaviour, rather than the child’s level of alertness or somnolence during the rest of the day. As such, the factor was re-named “Morning Lethargy”. 
Table 2.4  
**Final Factor Loadings for the Revised Five-Factor CSHQ-R**

<table>
<thead>
<tr>
<th>Orig. subscale</th>
<th>Abbreviated question</th>
<th>Factor 1 Sleep I &amp; M</th>
<th>Factor 2 Morning Lethargy</th>
<th>Factor 3 Co-sleeping</th>
<th>Factor 4 SDB</th>
<th>Factor 5 Para</th>
</tr>
</thead>
<tbody>
<tr>
<td>SD</td>
<td>9 Sleeps the right amount</td>
<td>0.77</td>
<td>0.89</td>
<td>0.01</td>
<td>0.09</td>
<td>-0.02</td>
</tr>
<tr>
<td>SD</td>
<td>8 Sleeps too little</td>
<td>0.63</td>
<td>0.75</td>
<td>0.02</td>
<td>0.12</td>
<td>0.02</td>
</tr>
<tr>
<td>SD</td>
<td>10 Sleeps the same amount</td>
<td>0.43</td>
<td>0.58</td>
<td>-0.11</td>
<td>-0.10</td>
<td>0.04</td>
</tr>
<tr>
<td>SD</td>
<td>7 Asleep within 20 minutes</td>
<td>0.30</td>
<td>0.45</td>
<td>-0.22</td>
<td>-0.01</td>
<td>-0.02</td>
</tr>
<tr>
<td>BR</td>
<td>1 Bed at the same time at night</td>
<td>0.32</td>
<td>0.40</td>
<td>-0.10</td>
<td>-0.29</td>
<td>0.04</td>
</tr>
<tr>
<td>NW</td>
<td>14 Awakens once during the night</td>
<td>0.28</td>
<td>0.35</td>
<td>0.07</td>
<td>-0.12</td>
<td>-0.06</td>
</tr>
<tr>
<td>BR</td>
<td>5 Struggles at bedtime</td>
<td>0.35</td>
<td>0.34</td>
<td>-0.16</td>
<td>-0.22</td>
<td>0.08</td>
</tr>
<tr>
<td>DS</td>
<td>29 Difficulty getting out of bed</td>
<td>0.70</td>
<td>0.01</td>
<td><strong>-0.84</strong></td>
<td>0.04</td>
<td>0.01</td>
</tr>
<tr>
<td>DS</td>
<td>30 Takes a long time to be alert</td>
<td>0.66</td>
<td>0.00</td>
<td><strong>-0.80</strong></td>
<td>-0.01</td>
<td>0.06</td>
</tr>
<tr>
<td>DS</td>
<td>28 Adults or siblings wake up</td>
<td>0.31</td>
<td>-0.02</td>
<td><strong>-0.57</strong></td>
<td>-0.01</td>
<td>-0.08</td>
</tr>
<tr>
<td>DS</td>
<td>31 Seems tired</td>
<td>0.34</td>
<td>0.14</td>
<td><strong>-0.44</strong></td>
<td>0.09</td>
<td>0.09</td>
</tr>
<tr>
<td>DS</td>
<td>27 Wakes up in a negative mood</td>
<td>0.27</td>
<td>0.14</td>
<td><strong>-0.39</strong></td>
<td>0.02</td>
<td>0.09</td>
</tr>
<tr>
<td>BR</td>
<td>2 Falls asleep alone in own bed</td>
<td>0.63</td>
<td>0.08</td>
<td>0.05</td>
<td><strong>-0.79</strong></td>
<td>0.12</td>
</tr>
<tr>
<td>BR</td>
<td>3 Falls asleep in others’ bed</td>
<td>0.55</td>
<td>-0.09</td>
<td>-0.03</td>
<td><strong>-0.75</strong></td>
<td>0.09</td>
</tr>
<tr>
<td>BR/SIA</td>
<td>4 Needs parent in the room</td>
<td>0.38</td>
<td>-0.03</td>
<td>0.02</td>
<td><strong>-0.55</strong></td>
<td>-0.03</td>
</tr>
<tr>
<td>BR/SIA</td>
<td>6 Afraid of sleeping alone</td>
<td>0.47</td>
<td>-0.03</td>
<td>-0.06</td>
<td><strong>-0.53</strong></td>
<td>-0.05</td>
</tr>
<tr>
<td>NW</td>
<td>13 Moves bed during the night</td>
<td>0.42</td>
<td>0.16</td>
<td>0.11</td>
<td><strong>-0.52</strong></td>
<td>-0.08</td>
</tr>
<tr>
<td>SDB</td>
<td>25 Snorts and/or gasps</td>
<td>0.60</td>
<td>-0.01</td>
<td>0.00</td>
<td>-0.08</td>
<td><strong>0.75</strong></td>
</tr>
<tr>
<td>SDB</td>
<td>23 Snores loudly</td>
<td>0.45</td>
<td>-0.07</td>
<td>-0.05</td>
<td>0.01</td>
<td><strong>0.67</strong></td>
</tr>
<tr>
<td>SDB</td>
<td>24 Seems to stop breathing</td>
<td>0.42</td>
<td>0.01</td>
<td>0.02</td>
<td>-0.02</td>
<td><strong>0.63</strong></td>
</tr>
<tr>
<td>Para</td>
<td>21 Awakens alarmed by dream</td>
<td>0.40</td>
<td>-0.08</td>
<td>-0.09</td>
<td>-0.05</td>
<td>0.05</td>
</tr>
<tr>
<td>Para</td>
<td>22 Awakens screaming etc.</td>
<td>0.39</td>
<td>0.08</td>
<td>0.05</td>
<td>-0.09</td>
<td>0.09</td>
</tr>
<tr>
<td>Para</td>
<td>17 Talks during sleep</td>
<td>0.34</td>
<td>0.05</td>
<td>0.03</td>
<td>0.03</td>
<td>0.19</td>
</tr>
<tr>
<td>Para</td>
<td>18 Restless during sleep</td>
<td>0.37</td>
<td>0.19</td>
<td>-0.06</td>
<td>0.02</td>
<td>0.10</td>
</tr>
<tr>
<td>SIA</td>
<td>11 Afraid of sleeping in the dark</td>
<td>0.22</td>
<td>-0.06</td>
<td>-0.12</td>
<td>-0.12</td>
<td>-0.07</td>
</tr>
<tr>
<td>Para</td>
<td>19 Sleepwalks</td>
<td>0.24</td>
<td>0.05</td>
<td>0.01</td>
<td>0.01</td>
<td>0.15</td>
</tr>
<tr>
<td>Para</td>
<td>20 Grinds teeth during sleep</td>
<td>0.21</td>
<td>0.11</td>
<td>0.07</td>
<td>0.05</td>
<td>0.14</td>
</tr>
</tbody>
</table>

Note. Italics indicate items that load onto different factor from their original subscale. Factor loadings greater than |0.30| are in bold. \( h \) = communality; \( \lambda \) = factor loading. SD = Sleep Duration; BR = Bedtime Resistance; NW = Night-time Wakings; DS = Daytime Sleepiness; SIA = Sleep Anxiety; SDB = Sleep Disordered Breathing; Para = Parasomnias; Sleep I & M = Sleep Initiation and Maintenance.
Factor 3 was comprised mostly of items from the original Bedtime Resistance subscale, as well as one item from the Night-time Awakening Subscale, all of which pertained to co-sleeping or requiring company while falling asleep, rather than other signs of bedtime anxiety (e.g. afraid of the dark). As such, this factor was renamed “Co-sleeping”. The remaining two factors (4 and 5) retained their original names (SDB and Parasomnias) as can be seen in Table 2.4.

While the current sample size was sufficient for exploratory analysis (subject-to-item ratio of 14), it was insufficient to split into training and validation data sets. As such, CFA of the newly defined factor structure was not performed.

2.4.3 Reliability

Internal consistencies, as well as subscale means and standard deviations for the revised five-factor model, are presented in Table 2.4. The internal consistencies ($\alpha = 0.71 - 0.80$) were all above acceptable minimal levels (Nunnally & Bernstein, 1994) and improved compared to those for the CSHQ-original subscales reported by Owens et al. (2000; $\alpha = 0.36 - 0.93$). The revised 27-item scale had good overall internal consistency ($\alpha = 0.85$). No further improvements in internal consistency could be gained, for either the subscales or the total scale, through the removal of additional items.

2.4.4 Validity

To examine the convergent validity of the current scale, scores on the revised CSHQ subscales were compared to comparable subscales of the SDSC in a smaller subset of children ($n = 36$). As can be seen in Table 2.5, bivariate correlation analysis revealed that comparable subscales and total scores of the two questionnaires were all strongly positively correlated ($r = .58$ to $.80$).
Table 2.5

Convergent Validity of the CSHQ-Revised; Correlations with the SDSC

<table>
<thead>
<tr>
<th>CSHQ subscales</th>
<th>SDSC subscales</th>
<th>DIMS</th>
<th>DOES</th>
<th>SH</th>
<th>SBD</th>
<th>DOA</th>
<th>SWTD</th>
<th>SDSC total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sleep I &amp; M</td>
<td>.716**</td>
<td>.516**</td>
<td>.286</td>
<td>.567**</td>
<td>.318</td>
<td>.343*</td>
<td>.687**</td>
<td></td>
</tr>
<tr>
<td>Morning Lethargy</td>
<td>.272</td>
<td>.584**</td>
<td>.049</td>
<td>-.027</td>
<td>.276</td>
<td>.313</td>
<td>.377*</td>
<td></td>
</tr>
<tr>
<td>Co-sleeping</td>
<td>.491**</td>
<td>.051</td>
<td>.328</td>
<td>.247</td>
<td>.228</td>
<td>.198</td>
<td>.404*</td>
<td></td>
</tr>
<tr>
<td>SDB</td>
<td>.275</td>
<td>.381*</td>
<td>.353*</td>
<td>.755**</td>
<td>.025</td>
<td>.183</td>
<td>.438**</td>
<td></td>
</tr>
<tr>
<td>Parasomnias</td>
<td>.474**</td>
<td>.306</td>
<td>.338*</td>
<td>-.018</td>
<td>.762**</td>
<td>.686**</td>
<td>.661**</td>
<td></td>
</tr>
<tr>
<td>CSHQ-R total</td>
<td>.711**</td>
<td>.559**</td>
<td>.384*</td>
<td>.388*</td>
<td>.549**</td>
<td>.556**</td>
<td>.796**</td>
<td></td>
</tr>
</tbody>
</table>

Note. n = 36; Values in bold represent correlations between subtests which are most conceptually similar. CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing. SDSC = The Sleep Disturbances Scale For Children; DIMS = Difficulty in Initiating and Maintaining Sleep; DOES = Disorders of Excessive Somnolence; SH = Sleep Hyperhidrosis; SBD = Sleep Breathing Disorders; DOA = Disorders of Arousal; SWTD = Sleep-Wake Transition Disorders.

* p < .05; ** p < .01.

In addition to positive associations between CSHQ-R and SDSC subscales, validity of the CSHQ-R may be inferred from the fact that, when compared to those who had not taken medication for sleep, children who had either current or past use of melatonin supplements (indicative of sleeping difficulties) also had significantly higher scores on the newly defined scales (Table 2.5).

<table>
<thead>
<tr>
<th></th>
<th>Melatonin M (SD)</th>
<th>No Melatonin M (SD)</th>
<th>t</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sleep I &amp; M</td>
<td>13.74 (3.70)</td>
<td>11.17 (3.43)</td>
<td>5.96</td>
<td>&lt; .001**</td>
</tr>
<tr>
<td>Morning Lethargy</td>
<td>8.90 (2.63)</td>
<td>7.86 (2.44)</td>
<td>3.37</td>
<td>.001**</td>
</tr>
<tr>
<td>Co-sleeping</td>
<td>7.71 (2.92)</td>
<td>6.97 (2.61)</td>
<td>2.23</td>
<td>.026*</td>
</tr>
<tr>
<td>SDB</td>
<td>3.98 (1.32)</td>
<td>3.63 (1.08)</td>
<td>2.45</td>
<td>.013*</td>
</tr>
<tr>
<td>Parasomnias</td>
<td>11.20 (3.02)</td>
<td>10.21 (2.75)</td>
<td>2.84</td>
<td>.005**</td>
</tr>
<tr>
<td>Total</td>
<td>45.55 (8.69)</td>
<td>39.87 (8.20)</td>
<td>5.52</td>
<td>&lt; .001**</td>
</tr>
</tbody>
</table>

CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing

* p < .05; ** p < .01.
2.5 Discussion

This study aimed to replicate previous CSHQ-original and CSHQ-ASD models, evaluate the psychometric properties of the CSHQ in a large, representative, community-based sample of children with ASD. While neither the CSHQ-original subscales nor the CSHQ-ASD factor structure fit the current data, a revised scale could be derived from EFA. The revised 27-item, five-factor scale required relatively minor changes from the CSHQ-original and demonstrated promising signs of external validity.

2.5.1 Confirmatory analyses

To our knowledge, ours is the first paper to attempt to confirm both the revised factor structure (CSHQ-ASD) presented by Johnson et al. (2016) or the CSHQ-original subscales in an ASD sample until this year. While no one has examined the proposed factor structure of the CSHQ-original in an English speaking primary school-aged sample (either TD or ASD), a number of past confirmatory analyses have been conducted on other language versions of the CSHQ (Chinese: Liu et al., 2014; Spanish: Lucas-de la Cruz et al., 2016; German: Schlarb et al., 2010; and Dutch: Waumans et al., 2010) as well as a pre-school sample (Sneddon et al., 2013), all of which failed to confirm the original subscales proposed by Owens et al. (2000). Consistent with these past findings, the current study was unable to find a good fit for the CSHQ-original subscales based on the current data. Together, these studies, along with our findings, highlight the need to exercise caution with using these original subscales.

With regard to the CSHQ-ASD (Johnson et al., 2016), poor fit may be explained by differences between sample characteristics. While both the current paper and Johnson et al. (2016) included only children with a diagnosis of ASD, the
current sample is a broad community-based sample. In contrast, Johnson et al. (2016) included a large number of children with known clinically significant behavioural disturbance (57%) and a small proportion (11%) with known behavioural sleep disturbance, but without any medical sleep condition (e.g. sleep apnoea, restless leg syndrome). Conversely, the current sample was a well characterised and representative community-based sample not selected for elevated behavioural disturbances or sleep problems.

2.5.2 Exploratory analyses

Given poor fit of the two competing confirmatory models (CSHQ-original and CSHQ-ASD) in the current sample, exploratory analyses were performed. After the removal of six items, based on item-level and factor analysis, a five-factor solution was considered the best fit for the data. This model was easily interpretable and maintained similar conceptual groupings as the original CSHQ based on the ICSD categories.

Two items (16 and 26) were removed during item-level analysis. Item 26, “Child wakes up by him/herself”, has commonly been recorded as a problematic item (Liu et al., 2014; Waumans et al., 2010) due to ambiguity inherent in the wording. This item could be an indication of either good sleep (i.e. wakes in own time, indicating that the child is refreshed) or poor sleep (i.e. wakes early due to poor sleep maintenance). Qualitatively, issues were also identified with parental responses to Item 16 regarding bedwetting. Many parents wrote that their child was still wearing nappies but then continued to respond to the question by either selecting, with similar frequencies, that this was “usually” a problem (3) or that it was “rarely” a problem (1). This again highlights ambiguity in the item, with differing interpretations of whether bed-wetting is interpreted as a “problem” if the child is still in nappies. This
item has not been reported as problematic in other studies conducting either item-level analysis or EFA and CFA; although it has been removed prior to formal analysis in some of the studies with younger participants (Goodlin-Jones et al., 2008; Sneddon et al., 2013). It is likely that the current issues may be specific to younger populations or those with developmental delay. While item removal was both theoretically and statistically most appropriate for this setting, if retained in other versions of the scale, it may be useful to provide more guidance on how to respond if the child is in nappies (e.g. “If your child is still wearing nappies please tick 3”).

EFA led to the removal of a further 4 items (items 12, 15, 32 and 33; based on low loadings or high cross-loadings). Items 32 and 33 have been removed from the final factor solution of many of the published models (e.g. Johnson et al., 2016 - deleted item 33 only; Liu et al., 2014; Sneddon et al., 2013). This may be explained by the fact that these items are rated on a scale with different labels (“Not Sleepy”, “Sleepy”, “Falls Asleep” v. “Rarely”, “Sometimes”, “Usually”), which seems to have the effect of artificially inflating shared variance between these two items and reducing shared variance with the remaining 31 items (this same pattern was noted by Schlarb et al., 2010). Item 12, “Child has trouble sleeping away from home”, may again be less relevant in younger populations and those with developmental delays as they may be less likely to sleep away from home.

The four-factor solution was revised to a final five-factor solution due to improvements in clinical utility, whilst retaining sound psychometric properties. In particular, it was deemed useful to have separate SDB and Parasomnia subscales, rather than having these scales combined. Despite this, it is noted that the overall explained variance remained quite low. Overall, the final five subscales (Sleep I & M, Morning Lethargy, Co-sleeping, SDB, Parasomnias) shared some similarities to
the original conceptually-based subscales presented by Owens et al. (2000; see Table 2.5).

Similarities were also noted between current subscales and those recorded in other factor analyses. For example, the current Sleep I & M was similar to the subscales Sleep Routine Problems (items 1, 5, 7, 8, 9, 10 in common; Johnson et al., 2016) and Sleep Initiation (items 1, 5, 7, 14 in common; Sneddon et al., 2013), while Morning Lethargy was similar to Insufficient Sleep reported by Johnson et al. (2016; items 27, 28 29 and 30 in common). Hence, while neither the CSHQ-original (Owens et al., 2000) nor CSHQ-ASD (Johnson et al., 2016) models could be confirmed, and other models could not be compared due to a lack of reporting factor loadings (e.g. Liu et al., 2014; Waumans et al., 2010), there are promising consistencies between current and past analyses. This highlights the sound theoretical basis for the questionnaire, despite limited psychometric analysis during scale development. The internal consistencies for revised subscales were all above acceptable minimal levels and improved compared to those for the CSHQ-original subscales reported by Owens et al. (2000). Further, the total CSHQ-R had good overall internal consistency.

Of note, the revised scale no longer has a Sleep Anxiety scale, despite the retention of three out of the four original Sleep Anxiety items. It would be useful for future research to assess the relationship between daytime anxiety and items/subscales on the CSHQ-R. It may be that indications of anxiety are captured through individual items on the scale, or a subscale (such as Co-sleeping), despite the absence of a specific anxiety subscale.

The revised Co-sleeping subscale is also of interest. It is unclear from the current data whether bed-sharing behaviour should be considered problematic or not. While bed-sharing behaviour was conceptualised as an indication of either anxiety or bedtime resistance in the CSHQ-original, in the current EFA, these bed-sharing items
group together, rather than align with anxiety or bedtime-resistance items. It would be worthwhile for further research to explore whether bed sharing is different in ASD and TD groups, and whether or not this behaviour is perceived as problematic by parents of children with ASD.

Retention of the SDB subscale in the current solution is notable given that it has often been lost in other revisions to the questionnaire (e.g. Johnson et al., 2016; Sneddon et al., 2013). While low in frequency (1-5% in paediatric groups, Ehsan et al., 2017), SDB is an important medical condition for which to screen and is certainly no less relevant in ASD populations, as evidenced by scores on the SDB subscale in the current sample (M = 3.70, SD = 1.19), and for past TD samples such as Owens et al. (2000; M = 3.24, SD = 0.63) and Schlarb et al. (2010; M = 3.27, SD = 0.56).

Of note, a very recent study reported on the factor structure of the CSHQ in children with ASD (Katz et al., 2018). Their findings were comparable to those presented here, especially for the Sleep I & M, Morning Lethargy, and Co-sleeping subscales (Katz et al., 2018)

2.5.3 Convergent validity

Encouragingly, the current study showed good agreement with another well developed and validated questionnaire used to assess sleep problems in children, the SDSC. While the subscales in the two questionnaires do not map perfectly, agreement between subscales was highest for conceptually comparable scales. Further, the finding that those who reported use of melatonin supplements had significantly higher scores across scales provides support for the scale’s validity, as melatonin is commonly used to treat sleep problems in ASD (Cuomo et al., 2017; Rossignol & Frye, 2011), with use of this medication therefore being a strong indication that sleep problems have been previously identified. While these initial
findings support the validity of the CSHQ-R, it would be beneficial to compare the revised scale against other sleep questionnaires in a larger sample and to assess for the scale’s sensitivity in distinguishing those children with clinically identified sleep problems, from those without.

2.5.4 Conclusion

In summary, the inability to confirm the factor structure for either the CSHQ-original or the CSHQ-ASD suggests that these subscales and scoring procedures should be avoided in children with ASD. However, moderate-to-high internal consistencies for all five revised subscales, as well as clear theoretical grounds for item groupings in the CSHQ-R, provides strong support for the suggested revisions to the original scale, which could improve the reliability and utility of the CSHQ for children with ASD.

While this modified version of the CSHQ shows promise for use in children with ASD, the current sample size was insufficient to split into training and validation data sets. As such, the five-factor solution presented has not been confirmed through CFA. As a CFA was not performed on the final model, formal invariance testing for age was not possible. Given that the scale is often employed in paediatric samples of all ages, it will be worthwhile for future studies to assess the factor structure across age groups. Further work is required to confirm the reliability and validity of the instrument.

The present study presents data from a large well-characterised and representative community sample of children with ASD. Results indicate that revisions to the original CSHQ subscales and scoring may be more appropriate for assessing sleep in children with ASD. Internal consistency was acceptable for all revised subscales and high for the total score. Initial measures of convergent validity
seem promising. However, before further use, it would be preferable for the revised structure to be explored in a larger data set through CFA and compared to other measures of sleep.
2.6 References


Elia, M., Ferri, R., Musumeci, S. A., Del Gracco, S., Bottitta, M., Scuderi, C., …


Chapter 3: Longitudinal Assessment of Sleep and Autism Spectrum Disorder Symptom Severity

3.1 Preamble to Chapter 3

Using the Children’s Sleep Habits Questionnaire-Revised (CSHQ-R) developed in Chapter 2, the following paper addresses the associations between sleep problems in autism spectrum disorder (ASD) and the core behavioural features of the condition. As noted in Chapter 1, while many studies have detailed associations between sleep problems and core symptoms of ASD, few have examined whether predictive relationships exist between these features. Further, the direction of these relationships remains unclear. Employing a longitudinal design in a sample of children with ASD (N = 45), the current study builds on past literature by exploring longitudinal predictive associations between these symptoms. Across the three to five year follow-up period, the results also highlight the stability and persistence of sleep problems within this population, highlighting the need for further examination of the extent and consequences of poor sleep for children with ASD.
Longitudinal Assessment of Sleep and Autism Spectrum Disorder Symptom Severity

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\textsuperscript{2} Telethon Kids Institute, The University of Western Australia, West Perth, Australia

\textsuperscript{3} Clinical Research Centre, Graylands Hospital, North Metropolitan Health Service Mental Health, Mount Claremont, WA, Australia
Abstract

Poor sleep has been found to be directly associated with the core symptoms of autism spectrum disorder (ASD), including repetitive behaviours and interests and social and communication difficulties. However, it remains unclear whether early sleep problems exacerbate the expression of ASD symptoms, or whether more severe early ASD symptoms lead to the development or maintenance of sleep problems. Accordingly, the current study sought to examine associations between sleep and ASD symptoms across time. Sleep and ASD symptoms were assessed over a three to five year period in a sample of 45 children with ASD who were aged 7-12 years at follow-up. The findings showed that ASD symptom severity and sleep problems were: (i) individually stable over time; (ii) positively associated with each other at each time point; and (iii) robustly associated with each other across time points. However, after controlling for initial sleep problems, initial ASD symptom severity did not add significantly to the prediction of sleep problems at follow-up, and similarly, after controlling for initial symptom severity, initial sleep problems did not add significantly to the prediction of ASD symptom severity at follow-up.

3.2 Introduction

Autism spectrum disorder (ASD) refers to a neurodevelopmental spectrum condition, characterised by difficulties with social communication as well as restricted and repetitive behaviours and interests (American Psychiatric Association, 2013). In addition to these core behavioural features, comorbid medical and psychiatric conditions are frequently noted (Bauman, 2010; Leyfer et al., 2006; Simonoff et al., 2008) including difficulties with initiating and maintaining sleep. Current estimates indicate that as many as 80% of children with ASD will experience sleep problems (Aathira et al., 2017; Couturier et al., 2005; Goldman et al., 2009; Polimeni, Richdale, & Francis, 2005; Rzepecka et al., 2011; Souders et al., 2009; Wang et al., 2016).

Poor sleep is known to have a significant impact on daytime emotional and behavioural regulation (for a review, see Gregory & Sadeh, 2012) and cognitive performance (Gruber et al., 2010; Paavonen et al., 2010) in children who are typically developing (TD). Similar associations have been reported in ASD, with poorer sleep associated with lower cognitive functioning (e.g. academic performance, general intelligence quotient; Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005; Paavonen, Niemenen-von Wendt, Vanhala, Aronen, & von Wendt, 2003; Richdale & Schreck, 2009; Sikora, Johnson, Clemons, & Katz, 2012; M. A. Taylor, Schreck, & Mulick, 2012), and behaviour regulation, with associated challenging behaviours (Adams, Matson, & Jang, 2014; Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Masurek & Sohl, 2016)). Furthermore, sleep has been linked to the frequency and severity of the core symptoms of ASD (Adams, Matson, Cervantes, & Goldin, 2014; Hoffman et al., 2005; Hundley, Shui, & Malow, 2016; Park et al., 2012;
Schreck, Mulick, & Smith, 2004; Tudor, Hoffman, & Sweeney, 2012). That is, in addition to the common correlates of poor sleep seen in the wider TD paediatric population, there is evidence that poor sleep in ASD is also directly associated with more severe overall ASD symptoms, including restricted interests and repetitive behaviours (RRBI) and limitations in social and communication skills.

Past research has linked a range of sleep problems (e.g. bedtime resistance, limited total sleep time, delayed sleep onset latency, poor sleep quality and symptoms of SDB) with ASD symptom severity (Gabriels et al., 2005; Hoffman et al., 2005; Park et al., 2012; Schreck et al., 2004; Tudor et al., 2012). For example, fewer hours of sleep per night has been found to be predictive of ASD symptom severity, including both communication deficits and RRBI (Schreck et al., 2004; Tudor et al., 2012), as well as being associated with lower clinician-rated verbal skills, socialisation and communication skills (M. A. Taylor et al., 2012). Increased parent-rated sleep problems on various different sleep questionnaires have been found to be associated with poorer social interaction (Hollway, Aman, & Butter, 2013), increased RRBI (Goldman et al., 2009, 2011) and increased severity of ASD symptoms in general (Mayes & Calhoun, 2009). Finally, within those with ASD, communication abnormalities and RRBI have been found to be associated with an increased risk of sleep problems (Park et al., 2012). While the majority of these studies have relied on parent- or clinician-rated measures of sleep and ASD symptoms, two studies have looked at the link between objective sleep parameters and ASD symptom severity. Commensurate with parental reports, shorter total sleep time as assessed by polysomnography (PSG), was reported to be associated with poorer parent-rated non-verbal communication skills (Elia et al., 2000); while
actigraphy recorded sleep fragmentation has been found to be associated with RRBI (Goldman et al., 2009).

As indicated by these data, there is consistent evidence that sleep quality and quantity is related to the severity of core symptoms of ASD. However, as noted by Adams et al. (2014) the direction of this relationship remains unclear. While many studies have discussed the relationship in terms of sleep problems exacerbating the presentation of ASD symptoms, others have contested that individuals with ASD may be at heightened risk for sleep problems due to underlying genetic factors potentially common to both sleep disturbance and ASD (Veatch et al., 2014). For example, there is evidence to suggest reduced nocturnal secretion of melatonin (a neurohormone known to be involved in the regulation of sleep-wake cycles) in children and adolescents with ASD when compared to TD controls (Tordjman et al., 2012; Tordjman, Anderson, Pichard, Charbuy, & Touitou, 2005). These differences are particularly pronounced for those with more severe ASD symptoms (Tordjman et al., 2012). Other researchers speculate that the core features of ASD, such as reduced social awareness or engagement, may lead to decreased attention to social cues that are important zeitgebers for human circadian rhythms. In turn, this may lead to interruptions to the entrainment of sleep-wake cycles (Glickman, 2010).

These theories are not mutually exclusive, and as is the case in much of the sleep-health literature (see Gregory & Sadeh, 2012), a bidirectional relationship is likely (Adams, Matson, Cervantes et al., 2014). However, most previous investigations have employed cross-sectional designs, and have thus been unable to detect how sleep and ASD symptoms may influence each other in a temporal way. While a handful of longitudinal studies have been conducted to track sleep in children with ASD (Allik, Larsson, & Smedje, 2008; May et al., 2015; Fletcher et al.,
2017; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012), none have looked at the relationship between changes in sleep patterns and symptom severity over time. As such, it remains unclear whether early sleep problems may exacerbate the expression of ASD symptoms for children with this diagnosis, or whether more severe ASD symptoms may lead to the development or maintenance of sleep problems throughout childhood.

To address this gap in the literature, the current study sought to re-examine the association between sleep and ASD symptoms by assessing relationships over time in a sample of children with ASD who had been followed up over a three to five year period. Specifically, the three aims were as follows: first, to examine the relationship between sleep and core ASD symptoms cross-sectionally at two separate time points; second, to understand changes in ASD symptoms and sleep patterns over time in children with ASD; and third, to investigate longitudinal predictive associations between sleep disturbance and symptom severity in ASD.

We hypothesised that: (i) greater ASD symptom severity would be linked to greater sleep difficulties at each time point; (ii) both ASD symptoms and sleep disturbance would be relatively stable over time; and (iii) ASD symptoms and sleep disturbance would be associated across time points (i.e. T1 sleep associated with T2 ASD symptoms and vice versa). As there are a range of differing (and not mutually exclusive) theoretical positions aiming to explain the link between sleep and ASD symptoms, no specific hypotheses were made regarding longitudinal predictive associations. Rather, models will examine both whether initial sleep characteristics predict ASD symptom severity at a later time point (after controlling for initial ASD symptoms), and whether initial ASD symptom severity predicts sleep problems at follow-up (after controlling for initial sleep problems).
3.3  Method

3.3.1  Participants

Participants were recruited from the Western Australian Autism Biological Registry (WAABR) at the Telethon Kids Institute in Perth, Western Australia (see L. J. Taylor et al., 2013 for more detail). All children recruited to the WAABR study had a clinical ASD diagnosis (i.e. either Autistic Disorder, AD; Asperger Syndrome, AS; or Pervasive Developmental Disorder Not Otherwise Specified, PDD-NOS) based on the Diagnostic and Statistical Manual of Mental Disorders criteria (4th ed. DSM–IV; American Psychiatric Association, 2000). In Western Australia, this diagnosis is established by multidisciplinary team consensus (paediatrician, psychologist, and speech-language pathologist; Glasson et al., 2008). Diagnosis was confirmed at initial enrolment by the study team using the Autism Diagnostic Observation Schedule-Generic (ADOS-G; Lord et al. 2000).

The current study included children who met the following criteria: (a) initial enrolment between February 2011 and September 2013; (b) were administered the ADOS and had both Children’s Sleep Habits Questionnaire (CSHQ) and Social Responsiveness Scale (SRS) data available from enrolment; (c) were aged 7-12 years\(^1\) at follow-up; (d) had consented to being contacted for future research.

Of the families in the WAABR registry, 143 families met criteria for the current study. Forty-five participants (31.47% of those contacted) returned the questionnaire packs. Of these children, 38 had a clinical diagnosis of AD, one child had a diagnosis of AS, and six children had a diagnosis of PDD-NOS.

\(^1\) While children were aged 7-12 years when the follow-up packs were posted, some families were delayed in their reply and as such, three children in the sample were aged 13 years at the time of reply.
Ethics approval for this study was granted by the Human Research Ethics Committee at The University of Western Australia in Perth, Western Australia (RA/4/1/6487).

3.3.2 Procedure

The current study involved repeat testing at a second time point (T2) using the same sleep and ASD symptoms questionnaires from T1, with a view to measuring change over time. In October 2016 (T2), 143 families were contacted via post seeking interest to participate in the current study. Information posted included an information sheet, consent form, and symptom questionnaires, along with a reply paid envelope. A summary of the measures completed at the two time points can be found in Table 3.1.

<table>
<thead>
<tr>
<th>Time point completed</th>
<th>T1</th>
<th>T2</th>
</tr>
</thead>
<tbody>
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</tr>
<tr>
<td>Clinical Diagnosis</td>
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<td>-</td>
</tr>
<tr>
<td>Comorbidities</td>
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<td>✓</td>
</tr>
<tr>
<td>Current use of prescribed medication †</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>Current use of CAM †</td>
<td>✓</td>
<td>✓</td>
</tr>
<tr>
<td>Any use of melatonin since T1</td>
<td>-</td>
<td>✓</td>
</tr>
<tr>
<td>Behavioural sleep intervention since T1</td>
<td>-</td>
<td>✓</td>
</tr>
<tr>
<td>ASD symptom measures</td>
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<tr>
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<td>-</td>
</tr>
<tr>
<td>SRS-2</td>
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<td>✓</td>
</tr>
<tr>
<td>ASD sleep measures</td>
<td></td>
<td></td>
</tr>
<tr>
<td>CSHQ</td>
<td>✓</td>
<td>✓</td>
</tr>
</tbody>
</table>

*Note. CAM = Complementary or alternative medication; ADOS-G=Autism Observation Schedule-Generic; SRS-2 = Social Responsiveness Scale-2; CSHQ = Children’s Sleep Habits Questionnaire. † Responses were screened for use of melatonin and entered as a single variable - 'Use of melatonin' from birth to T2.*
Of the 45 responders, a majority were males (n = 41, 91%). Age at T1 ranged from 31 to 106 months (M = 63.22; SD = 20.11) while age at T2 ranged from 84 to 163 months (M = 119.93; SD = 21.56). Sleep and ASD symptom profiles at T1 did not differ between T2 responders and T2 non-responders on CSHQ total scores; \( t(117) = 1.16, p = .401 \) (M = 47.36, SD = 10.43 and M = 49.94, SD = 11.41 respectively) or initial (T1) ADOS-2 comparison scores; \( t(141) = 0.018, p = .718 \) (M = 5.91, SD = 2.08 and M = 5.92, SD = 2.26 respectively).

### 3.3.3 Measures used at T1

**Autism Observation Schedule-Generic**

To check clinical diagnosis of ASD, participants were assessed on the ADOS-G (Lord et al., 2000). The ADOS is a semi-structured assessment tool which employs play-like activities to create opportunities for the assessor to observe behaviours and communication skills relevant to the diagnosis of ASD. Diagnostic categories are derived from raw scores, accounting for age and verbal ability. While participants completed the ADOS-G, scoring was completed using the ADOS-2 procedures which offer the benefit of comparison scores across different modules (Lord et al., 2012).

**Demographic information**

Parents completed a clinical history form detailing their child’s clinical diagnosis and any comorbidities such as intellectual disability (ID), global developmental delay (GDD), attention deficit hyperactivity disorder (ADHD) as well as use of prescribed medication or complementary/alternative medication. Parents were advised that the primary caregiver should complete the questionnaires, with all completed by the same caregiver.
3.3.4 Measures used at T1 and T2

**The Social Responsiveness Scale-2 (SRS-2)**

The SRS-2 (Constantino & Gruber, 2012) is a 65-item parent-rated questionnaire used to examine a range of ASD-related social and communication behaviours in children. Responses are made on a four-point scale ranging from 0 = “Not True” to 3 = “Almost Always True”. The SRS-2 yields a total score (maximum score of 195) and four social subscale scores - Awareness, Cognition, Communication and Motivation, as well as one focusing on RRBI. Across each subscale higher scores indicate greater symptom severity. For those with fewer than 20% missing values, scores were prorated using the item mean score across the sample.

**Children’s Sleep Habits Questionnaire-Revised (CSHQ-R)**

The CSHQ (Owens, Spirito, & McGuinn, 2000) is a 33-item parent-report measure used to examine sleep behaviour over a “typical week” period. Items are rated on a three-point scale where 1 = “Rarely” (0-1 night per week), 2 = “Sometimes” (2-4 nights per week) and 3 = “Usually” (5-7 nights per week). This scale has been revised for use in ASD populations (CSHQ-R; Host et al. 2018), with the total score based on 27 of the 45 items (i.e. maximum total score of 81). Within ASD populations, Host et al. (2018) recommended the use of five subscale scores: sleep initiation and maintenance (Sleep I & M), morning lethargy, co-sleeping, sleep disordered breathing (SDB) and parasomnias. Again, higher scores indicate greater sleep problems; as such, four positively phrased items are reverse scored. For those

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N.B. While a stringent 15% criterion was used in Chapter 2, for which the sample size was substantial, it was necessary to use a less stringent criterion in the subsequent studies because of the desire to maintain reasonable levels of power with the smaller samples.
with fewer than 20% missing values, scores were prorated using the item mean score across the sample.

**Demographic information**

At T1 and T2 parents again provided information regarding their child’s ASD diagnosis, comorbidities and use of sleep medication, including complementary or alternative therapies. At T2 parents also reported on any behavioural sleep intervention between T1 and T2.

**3.3.5 Statistical analyses**

Data were screened for outliers using a criterion of 3.29 SD from the mean for each variable (Field, 2013). Based on this criterion, three univariate outliers were identified on three of the sleep subscales (Morning Lethargy, SDB and Parasomnias). These were dealt with using a Winsorizing approach. Preliminary analyses were completed to examine data distributions and correlations amongst the study variables. Paired sample t-tests were used to examine the change (or stability) in sleep and ASD symptoms over time. To determine which variables to include in the regression models, correlational analyses were conducted to examine the relationships between demographic characteristics (using point-biserial correlations for dichotomous variables), sleep problems, and ASD symptoms. Correlational analyses were also employed to determine whether sleep problems and ASD symptoms were correlated with each other at the baseline (T1) and at follow-up three to five years later (T2). Where questionnaire data was missing, pairwise deletion was employed to maximise available sample size.

Following correlational analysis, two hierarchical multiple regression analyses were conducted to examine whether:
1. Sleep problems (CSHQ-R total) at T1 predicted ASD symptoms (SRS-2 total) at T2, above and beyond the effects of demographic variables and ASD symptoms (SRS-2 total) at T1.

2. ASD symptoms (SRS-2 total) at T1 predicted sleep problems (CSHQ-R total) at T2, above and beyond the effects of demographic variables and sleep problems (CSHQ-R total) at T1.

While the SRS-2 and CSHQ-R both include a number of subscales, due to the small sample size and the need to maintain power while restricting the Type 1 error rate, only a single total score for each was included in the regression models. For all analyses, statistical significance was tested using an alpha of 0.05.

3.4 Results

The study follow-up period ranged from 36-71 months (M = 56.18, SD = 9.11). Length of follow-up in months was not correlated with ASD symptom severity or sleep problems at T1 or T2 (all \( r \) values < |.23|, all \( p \) values >.153). Overall, both sleep and ASD symptoms were stable over time with no significant differences found between scores at T1 and T2 on either the SRS-2 total, \( t(44) = 1.45, p = .157, \) or the CSHQ-R total, \( t(44) = 0.60, p = .550. \) While there were no significant differences in sleep across T1 and T2 for any of the CSHQ-R subscales, as can be seen in Table 3.2, both Social Awareness and Social Motivation improved with age.

3.4.1 Correlational analyses

As presented in Table 3.3, parental reports of total sleep problems and ASD symptoms were moderate to strongly associated at both T1 (total CSHQ-R and SRS-2, \( r = .449 \)) and T2 (total CSHQ-R and SRS-2, \( r = .538 \)). T1 sleep was also associated with T2 sleep, with strong correlations between the T1 and T2 scores for each of the subscales (Table 3.4). Similarly, T1 ASD symptoms were associated with
T2 ASD symptoms, again with moderate to strong correlations between scores for the two time points for all subscales (Table 3.4). Finally, T1 sleep problems were associated with T2 ASD symptoms ($r = .499$), and T1 ASD symptoms were associated with T2 sleep problems ($r = .440$; Table 3.3).

Point-biserial or Pearson correlations were employed to determine which variables to include in the regression models. As can be seen in Table 3.3, neither sleep problems nor ASD symptom severity were associated with sex (T1) or age (T1 or T2). Similarly, having a clinical diagnosis of GDD at T1 was not associated with sleep or ASD symptoms at T1 or T2. As such, neither sex, nor age, nor GDD was included as a control variable in the regression models detailed below.

Table 3.2
Means (SD) and Paired Sample T-Tests Comparing SRS-2 and CSHQ-R Variables from T1 to T2

<table>
<thead>
<tr>
<th></th>
<th>T1 M</th>
<th>T1 SD</th>
<th>T2 M</th>
<th>T2 SD</th>
<th>t</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>SRS-2</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social Awareness</td>
<td>14.20</td>
<td>3.19</td>
<td>12.14</td>
<td>3.72</td>
<td>3.08</td>
<td>.004**</td>
</tr>
<tr>
<td>Social Cognition</td>
<td>19.58</td>
<td>6.43</td>
<td>19.54</td>
<td>6.93</td>
<td>0.04</td>
<td>.966</td>
</tr>
<tr>
<td>Social Communication</td>
<td>33.29</td>
<td>8.81</td>
<td>30.91</td>
<td>11.88</td>
<td>1.36</td>
<td>.184</td>
</tr>
<tr>
<td>Social Motivation</td>
<td>15.13</td>
<td>6.51</td>
<td>13.20</td>
<td>6.22</td>
<td>2.23</td>
<td>.032*</td>
</tr>
<tr>
<td>RRBI</td>
<td>19.01</td>
<td>6.77</td>
<td>19.38</td>
<td>6.94</td>
<td>-0.36</td>
<td>.725</td>
</tr>
<tr>
<td>CSHQ-R</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep I &amp; M</td>
<td>10.86</td>
<td>3.73</td>
<td>10.98</td>
<td>3.00</td>
<td>-0.24</td>
<td>.812</td>
</tr>
<tr>
<td>Morning Lethargy</td>
<td>7.23</td>
<td>2.10</td>
<td>7.49</td>
<td>2.27</td>
<td>-0.93</td>
<td>.358</td>
</tr>
<tr>
<td>Co-sleeping</td>
<td>7.02</td>
<td>2.72</td>
<td>6.43</td>
<td>2.09</td>
<td>1.68</td>
<td>.101</td>
</tr>
<tr>
<td>SDB</td>
<td>3.64</td>
<td>0.95</td>
<td>3.60</td>
<td>0.94</td>
<td>0.28</td>
<td>.782</td>
</tr>
<tr>
<td>Parasomnia</td>
<td>10.04</td>
<td>2.94</td>
<td>9.66</td>
<td>2.60</td>
<td>1.01</td>
<td>.318</td>
</tr>
</tbody>
</table>

Note. SRS = Social Responsiveness Scale; RRBI = Restricted and Repetitive Behaviours and Interests; CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing

* $p < .05$; ** $p < .01$. 

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### Table 3.3

Demographics and Correlations between Participant Characteristics, Sleep Problems and ASD Symptoms at T1 and T2

<table>
<thead>
<tr>
<th>T1 Variables</th>
<th>Mean (SD)</th>
<th>n (%)</th>
<th>SRS-2 Total</th>
<th>CSHQ-R Total</th>
<th>SRS-2 Total</th>
<th>CSHQ-R Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>T1 Age</td>
<td>63.22 (20.11)</td>
<td>- .023</td>
<td>.109</td>
<td>.048</td>
<td>- .012</td>
<td></td>
</tr>
<tr>
<td>Sex (male) †</td>
<td>41 (91.1)</td>
<td>- .113</td>
<td>-.191</td>
<td>- .100</td>
<td>-.263</td>
<td></td>
</tr>
<tr>
<td>Comorbidities</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ID</td>
<td>2 (4.4)</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>GDD †</td>
<td>13 (28.9)</td>
<td>.271</td>
<td>-.106</td>
<td>.248</td>
<td>.213</td>
<td></td>
</tr>
<tr>
<td>ASD Symptoms</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SRS-2 Total</td>
<td>102.53 (28.69)</td>
<td>.449**</td>
<td></td>
<td>.654**</td>
<td>.440**</td>
<td></td>
</tr>
<tr>
<td>Sleep Problems</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CSHQ-R Total</td>
<td>38.80 (8.76)</td>
<td></td>
<td>.499**</td>
<td></td>
<td>.714**</td>
<td></td>
</tr>
<tr>
<td>T2 Variables</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>T2 Age</td>
<td>119.93 (21.56)</td>
<td></td>
<td>.046</td>
<td>.000</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Use of melatonin (birth to T2) †</td>
<td></td>
<td></td>
<td>.254</td>
<td>.387**</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Comorbidities</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ID †</td>
<td>10 (22.2)</td>
<td></td>
<td>.385*</td>
<td>.073</td>
<td></td>
<td></td>
</tr>
<tr>
<td>ADHD †</td>
<td>12 (26.7)</td>
<td></td>
<td>.367*</td>
<td>.177</td>
<td></td>
<td></td>
</tr>
<tr>
<td>ASD Symptoms</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SRS-2 Total</td>
<td>94.89 (29.64)</td>
<td></td>
<td></td>
<td>.538**</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep Problems</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CSHQ-R Total</td>
<td>38.24 (7.50)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Note. T1 age range = 31 - 106 months. T2 age range 84 - 163 months. 8 children were diagnosed with ID between T1 and T2. No one had a diagnosis of ADHD at T1. No one had a diagnosis of GDD at T2. Point-biserial correlations used for dichotomous variables where Male = 1 and Female = 0; Use of melatonin = 1, No melatonin = 0; GDD/ID/ADHD = 1, Not diagnosed =0. ID at T1 was not included as only two participants reported this diagnosis. † Indicates point-biserial correlations used to examine the relationship between continuous and dichotomous variables to determine which (if any) demographic variables were included in the regression analyses. All other correlations are Pearson’s r coefficients. * p < .05; ** p < .01.
While not associated with sleep, having a clinical diagnosis of either ID or ADHD at T2 was associated with having a higher total score on the SRS-2 (i.e. more severe ASD symptoms) at T2 ($r = .385$ and $r = .367$ respectively). Accordingly, these two comorbidities were included as control variables in regression Model 1, predicting ASD symptoms at T2. A history of having used melatonin for sleep problems (any time from birth to T2; measured at T2) was associated with sleep problems at T2 ($r = .387$); as such, this variable was included as a control variable in the regression analyses predicting sleep at T2 (Model 2).

### 3.4.2 Regression analyses

In Model 1, hierarchical regression analyses were conducted to examine whether T1 sleep problems (T1 CSHQ-R total; entered at step 3) predicted ASD symptoms at T2 (T2 SRS-2 total), after controlling for comorbid ID and ADHD (from T2; entered at step 1), and baseline ASD symptoms (T1 SRS-2 total; entered at step 2). Results are summarised in Table 3.5. After controlling for comorbidities, $F(2, 32) = 5.82$, $p < .01$, $R^2 = .267$, ASD symptoms at T1 were a strong predictor of ASD symptoms at T2, $F(1, 31) = 19.27$, $p < .001$, $\Delta R^2 = .281$. Sleep problems at T1 did not add any predictive value over and above this in predicting ASD symptoms at T2, $F(1, 30) = 2.14$, $p = .153$, $\Delta R^2 = .030$. 

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Table 3.4

Correlations between T1 and T2 Scores for Subscales of the SRS-2 and CSHQ-R

<table>
<thead>
<tr>
<th>SRS-2</th>
<th></th>
<th>CSHQ-R</th>
<th></th>
</tr>
</thead>
<tbody>
<tr>
<td>Social Awareness</td>
<td>.353*</td>
<td>Sleep I &amp; M</td>
<td>.566**</td>
</tr>
<tr>
<td>Social Cognition</td>
<td>.666**</td>
<td>Morning Lethargy</td>
<td>.651**</td>
</tr>
<tr>
<td>Social Communication</td>
<td>.530**</td>
<td>Co-sleeping</td>
<td>.542**</td>
</tr>
<tr>
<td>Social Motivation</td>
<td>.676**</td>
<td>SDB</td>
<td>.530**</td>
</tr>
<tr>
<td>RRBI</td>
<td>.587**</td>
<td>Parasomnia</td>
<td>.575**</td>
</tr>
<tr>
<td>Total</td>
<td>.654**</td>
<td>Total</td>
<td>.714**</td>
</tr>
</tbody>
</table>

*Note. SRS = Social Responsiveness Scale; RRBI = Restricted and Repetitive Behaviours and Interests; CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing
* p < .05; ** p < .01.

In Model 2, analyses examined whether T1 ASD symptoms (T1 SRS-2 total; entered at step 3) predicted sleep problems at T2 (T2 CSHQ-R total), after controlling for use of melatonin (from T2; entered at step 1), and baseline sleep problems (T1 CSHQ-R total; entered at step 2). Outcomes are summarised in Table 3.5. After controlling for melatonin use, $F(1, 37) = 5.72, p < .01, R^2 = .134$, sleep at T1 was a strong predictor of sleep at T2, $F(1, 36) = 38.37, p < .001, \Delta R^2 = .447$; however, ASD symptoms at T1 did not add any predictive value over and above that of sleep and melatonin use entered at earlier steps, $F(1, 35) = 1.08, p = .306, \Delta R^2 = .013$. 

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Table 3.5
Hierarchical Multiple Regression Analyses Predicting ASD Symptoms at T2 (Model 1), and Sleep at T2 (Model 2)

<table>
<thead>
<tr>
<th></th>
<th>R^2</th>
<th>ΔR^2</th>
<th>Beta</th>
</tr>
</thead>
<tbody>
<tr>
<td>Model 1 predicting ASD symptoms at T2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>n = 35</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Step 1</td>
<td>.267</td>
<td>.267**</td>
<td>.358*</td>
</tr>
<tr>
<td>ID</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ADHD</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Step 2</td>
<td>.548</td>
<td>.281***</td>
<td></td>
</tr>
<tr>
<td>ID</td>
<td></td>
<td></td>
<td>.266*</td>
</tr>
<tr>
<td>ADHD</td>
<td></td>
<td>.218</td>
<td></td>
</tr>
<tr>
<td>ASD Symptoms at T1</td>
<td></td>
<td>.553***</td>
<td></td>
</tr>
<tr>
<td>Step 3</td>
<td>.578</td>
<td>.030</td>
<td></td>
</tr>
<tr>
<td>ID</td>
<td></td>
<td></td>
<td>.292*</td>
</tr>
<tr>
<td>ADHD</td>
<td></td>
<td>.205</td>
<td></td>
</tr>
<tr>
<td>ASD Symptoms at T1</td>
<td></td>
<td>.432**</td>
<td></td>
</tr>
<tr>
<td>Sleep at T1</td>
<td></td>
<td>.213</td>
<td></td>
</tr>
<tr>
<td>Model 2 predicting sleep at T2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>n = 39</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Step 1</td>
<td>.134</td>
<td>.134*</td>
<td></td>
</tr>
<tr>
<td>Use of melatonin</td>
<td></td>
<td></td>
<td>.366*</td>
</tr>
<tr>
<td>Step 2</td>
<td>.581</td>
<td>.447**</td>
<td></td>
</tr>
<tr>
<td>Use of melatonin</td>
<td></td>
<td></td>
<td>.192</td>
</tr>
<tr>
<td>Sleep at T1</td>
<td></td>
<td>.691***</td>
<td></td>
</tr>
<tr>
<td>Step 3</td>
<td>.593</td>
<td>.013</td>
<td></td>
</tr>
<tr>
<td>Use of melatonin</td>
<td></td>
<td></td>
<td>.186</td>
</tr>
<tr>
<td>Sleep at T1</td>
<td></td>
<td>.636***</td>
<td></td>
</tr>
<tr>
<td>ASD Symptoms at T1</td>
<td></td>
<td>.126</td>
<td></td>
</tr>
</tbody>
</table>

*Note.* The control variables for Model 1 are diagnosis of comorbid intellectual disability (ID) and comorbid attention deficit hyperactivity disorder (ADHD); the control variable for Model 2 is use of melatonin to aid sleep.

* p < .05; ** p < .01; *** p < .001.

3.5 Discussion

This study sought to examine the associations between sleep and ASD symptom severity over a three to five year follow-up period. The findings showed
that: (i) ASD symptom severity and sleep problems were positively associated at each time point; (ii) both ASD symptom severity and sleep disturbance severity were stable across time in ASD children; and (iii) ASD symptoms and sleep disturbance were robustly associated across time points (i.e. T1 sleep was associated with T2 ASD symptoms, and vice versa). However, after controlling for baseline sleep problems, initial ASD symptoms were not predictive of follow-up sleep problems. Similarly, after controlling for baseline ASD symptoms, initial sleep problems were not predictive of follow-up ASD symptom severity.

Consistent with previous literature (Gabriels et al., 2005; Goldman et al., 2009, 2011; Hoffman et al., 2005; Hollway et al., 2013; Mayes & Calhoun, 2009; Park et al., 2012; Schreck et al., 2004; M. A. Taylor et al., 2012; Tudor et al., 2012), sleep problems and ASD symptoms were strongly and positively associated at both T1 and T2. In addition to this, both ASD symptoms and sleep problems were remarkably stable over time, with few group differences between T1 and T2. While this is, to our knowledge, the longest follow-up period for assessing sleep in ASD (up to five years), the findings are consistent with literature that has used cross-sectional designs. Past studies have found that sleep problems in ASD (when present) are similar across age, with no significant differences in sleep parameters across age groups (Goldman et al., 2012; Mayes & Calhoun, 2009; Patzold, Richdale & Tonge, 1998; Schreck & Mulick, 2000; Williams, Sears, & Allard, 2004). While sleep problems decline across childhood in TD children, children with ASD tend to experience more persistent sleep disturbance (Allik et al., 2008; Richdale & Prior, 1995; Sivertsen et al. 2012). This difference in the developmental trajectory of sleep problems in children with and without ASD is also reflected in the fact that between-group differences in sleep between ASD and TD children are most consistently
found at late primary school age. For example, while Goodlin-Jones et al. (2008) found no differences between sleep parameters in children aged 2-6 years with and without ASD, most studies assessing sleep in children aged between 5 and 16 years have found significant group differences (Couturier et al., 2005; Hoffman et al., 2006; May et al., 2015; Souders et al., 2009).

While both sleep and ASD symptoms were inter-related at T1 and T2, no temporal association was found between them after accounting for stability in ASD symptoms and sleep over time. This suggests that sleep problems at T1 did not impact significantly on ASD symptom severity at T2, and that symptom severity at T1 did not impact on sleep problems at T2. Given that these domains correlated with each other both at and across time points, this does not mean that the two domains are independent. Rather, this might suggest that there was insufficient variability over time in sleep or ASD symptoms that could then be explained by the alternate construct. Indeed, initial (T1) sleep accounted for such a large portion of the variance (58.1%) in sleep problems at T2, that there was little variance left over to explain. Similarly, ASD symptoms at T1 accounted for a substantial portion of the variance (54.8%) in symptoms at T2.

Despite the interesting findings yielded by this study, the small sample size (n = 35-39) must be acknowledged. In particular, we were unable to explore differences across sleep subscales in how they predict ASD symptom subscale scores (and vice versa). It may be that if assessed at the subscale level, different predictive associations between sleep and ASD symptoms may emerge. Indeed, Hundley et al. (2016) found that while repetitive motor movements were related to sleep problems, another component of RRBI, insistence on sameness, was not.
To our knowledge, this is the first study to explore developmental changes in sleep in ASD over such a long period (3-5 years) and as such, it provides an insight into the remarkable stability of sleep problems over time in ASD. While we cannot make any assertions about differences between sleep in ASD and TD based on the current data, the results are commensurate with a growing body of evidence that children with ASD do not appear to outgrow sleep problems in the way that TD children are known to. This is an important finding. If children with ASD do not outgrow sleep problems, there is even greater need to establish effective sleep interventions for this population to ensure that sleep problems are remedied.

In addition to examining developmental changes in sleep over longer time periods, it would be beneficial for future research to explore changes in sleep problems, ASD symptoms (including subclinical autistic traits) and their relationship over time, in both ASD and TD groups.
3.6 References


Hoffman, C. D., Sweeney, D. P., Gilliam, J. E., Apodaca, D. D., Lopez-Wagner, M.


Mayes, S. D., & Calhoun, S. L. (2009). Variables related to sleep problems in


Chapter 4: Parent-Reported and Actigraphy-Derived Sleep Parameters in Children With and Without Autism Spectrum Disorder

4.1 Preamble to Chapter 4

Given the persistent nature of sleep problems in autism spectrum disorder (ASD) as outlined in Chapter 3, Chapter 4 examines these sleep problems in more detail using a multi-method approach to sleep assessment. As noted in Chapter 1, there is some indication in the literature that while sleep problems are persistently reported by parents, findings from objective assessment of sleep are less consistent. However, given inter-sample discrepancies, the divergent findings are difficult to reconcile. While a number of studies have used multi-method assessment, few have directly examined the relationships between measures of sleep. The current study sought to build on the existing literature by exploring both parent-reported and actigraphy-derived sleep assessment in a single sample of children both with and without ASD.
Parent-Reported and Actigraphy-Derived Sleep Parameters in Children With and Without Autism Spectrum Disorder

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Abstract

The current study explored group differences in parent-reported and actigraphy-derived sleep parameters in primary school-age children with and without autism spectrum disorder (ASD). The study also sought to explore agreement between these methods of sleep assessment, examining the concordance between actigraphy, sleep diaries and the Children’s Sleep Habits Questionnaire-Revised (CSHQ-R). The sample consisted of 67 children aged 6-12 years, 39 of whom had a diagnosis of ASD. Consistent with hypotheses, while children with ASD had more severe sleep problems on parent-rated measures, few differences emerged when reviewing sleep diaries or actigraphy-derived measures of sleep parameters. Overall, there was poor agreement between CSHQ-R subscales and sleep diaries/actigraphy recording, though moderate to strong agreement on comparable diary and actigraphy variables. These findings emphasise the complexity of establishing “sleep problems” in children with ASD and strengthen support for use of multi-method sleep assessment.

4.2 Introduction

Autism spectrum disorder (ASD) refers to a spectrum condition characterised by difficulty with social communication as well as restricted and repetitive behaviours and interests (RRBI; American Psychiatric Association, 2013). The rates of both medical and psychiatric comorbidities are high in those with diagnosed with ASD (Bauman, 2010; Leyfer et al., 2006; Simonoff et al., 2008). Sleep problems are one of the most frequently reported comorbidities, with rates ranging from 30-80%, compared to only 10-40% for their typically developing (TD) peers (Aathira et al., 2017; Couturier et al., 2005; Elrod et al., 2016; Fadini et al., 2015; Goldman et al., 2009; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Paavonen et al., 2008; Polimeni, Richdale, & Francis, 2005; Rzepecka, Mckenzie, McClure, & Murphy, 2011; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012; Souders et al., 2009; Wang et al., 2016). Rates were lowest when restricted to clinician-diagnosed sleep disorders (e.g. Elrod et al., 2016) as opposed to more broadly defined “sleep problems” (i.e. above a cut-off on a questionnaire: Aathira et al., 2017; Couturier et al., 2005; Rzepecka et al., 2011).

While high rates of sleep problems are noted in the literature, there are inconsistencies in results, likely as a function of the tools used to measure sleep. The most widely used measure of sleep in ASD is the parent-rated Children’s Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000; see Hodge, Parnell, Hoffman, & Sweeney, 2012). Studies employing this scale persistently report higher rates of sleep problems in children with ASD compared to those without ASD. Indeed, children with ASD differ from TD peers on most subscales of the CSHQ, including those measuring bedtime resistance, sleep onset delay, sleep duration, night waking, and parasomnias (Couturier et al., 2005; Giannotti et al.,
2008; Giannotti, Cortesi, Cerquiglini, Vagnoni, & Valente, 2011; Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008; Goodlin-Jones, Tang, Liu, & Anders, 2008; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Hoffman et al., 2005; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Souders et al., 2009). Similar findings have been reported using other parent-rated sleep questionnaires such as the Behaviour Evaluation of Disorders of Sleep (BEDS; Schreck, Mulick, & Rojahn, 2003; e.g. Polimeni et al. 2005), the Simonds and Parraga Sleep Questionnaire (Simonds and Parraga 1982; e.g. Wiggs & Stores, 2004) the Sleep Disturbances Scale for Children (SDSC; Bruni et al., 1996; e.g. Miano et al., 2007; Paavonen et al., 2008) or other single-item measures (e.g. Malow et al., 2006).

Compared to the myriad of studies utilising questionnaire data, relatively few have assessed sleep in ASD using objective measures such as actigraphy or polysomnography (PSG). Actigraphy is a method of sleep assessment which uses an accelerometer to record gross motor activity, from which sleep-wake cycles are inferred (Ancoli-Israel et al., 2003; Sadeh & Acebo, 2002). Of the studies which have employed objective measures in ASD samples, the extent of sleep problems appear considerably less clear. While some actigraphy studies agree with findings from questionnaire studies regarding delayed sleep onset latency (SOL) and reduced sleep efficiency (SE; Allik, Larsson, & Smedje, 2006; Baker, Richdale, Short, & Gradisar, 2013; Souders et al., 2009), others found no difference between community derived ASD and TD samples on these measures (Goodlin-Jones, Sitnick et al., 2008; Tani et al., 2005; Goldman et al., 2009; Hering, Epstein, Elroy, Iancu, & Zelnik, 1999). Findings are similarly discrepant across other measures of sleep quality including wake after sleep onset (WASO), and total sleep time (TST; Allik et al., 2006; Baker et al., 2013; Goldman et al., 2009; Goodlin-Jones et al., 2008; Hare, Jones, & Evershed, 2006; Souders et al., 2009; Tani et al., 2005). Consistent with
this, there are conflicting results with PSG data, with some reporting group
differences between community derived ASD and TD samples across sleep
parameters such as TST, SOL and SE (Giannotti et al., 2011; Lázár et al., 2010;
Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005), while others found no
differences (Bruni et al., 2007; Elia et al., 2000; Goldman et al., 2009; Miano et al.,
2007), except for TST (Elia et al., 2000; Miano et al., 2007). Overall, while a recent
meta-analysis examining data from PSG and actigraphy studies did conclude that
there were significant ASD and TD group differences in TST, SOL and SE, it was
also noted that these were mostly small effects (Elrod & Hood, 2015). Collectively,
the literature suggests that while children with ASD are reported by their parents to
experience a number of sleep-related problems, the results are less consistent in
studies which employ objective measures of sleep.

It is possible that conflicting results between studies may be explained by the
heterogeneity of ASD samples across studies (e.g. age, symptom severity, level of
disability, intellectual ability etc.). Given that ASD is an umbrella term which covers
a wide range of symptom presentation, other factors such as symptom severity or
intellectual disability (ID) may account for some of the differences in sleep patterns
between studies. Another explanation is that findings are influenced by the
assessment methods employed. Previous studies have identified some discrepancies
between assessment methods (i.e. objective vs subjective) within ASD samples
(Goodlin-Jones, Tang et al., 2008; Hering et al., 1999; Wiggs & Stores, 2004). These
discrepancies have led to the suggestion that parents of children with ASD may be
hypervigilant to sleep problems, leading to misperceptions about sleep (Hering et al.,
1999). Alternatively, questionnaires may be capturing parental concerns about
psychological factors and behavioural issues which are perceived as sleep problems
(such as bedtime anxiety or co-sleeping) but which do not impact sleep itself (i.e. duration, SE) and which are therefore not captured by actigraphy.

As alluded to, one way to explore whether or not these differences in findings are due to sample characteristics is to examine agreement between objective and subjective measures within samples. Within the TD paediatric sleep literature, a number of studies have examined how subscales on the CSHQ compare to actigraphy and PSG derived sleep parameters (Alfano, Patriquin & De Los Reyes, 2015; Holley, Hill & Stevenson, 2010; Markovich, Gendron, & Corkum, 2015) with mixed results. However, of the many studies exploring sleep in ASD, relatively few have included both objective and subjective measures of sleep. Of the studies with both objective (actigraphy or PSG) and subjective (sleep diary or questionnaire) measures of sleep, many omit direct comparisons between the measures (Bruni et al., 2007; Goodlin-Jones, Tang et al., 2008; Miano et al., 2007). Where direct comparisons are made, there are conflicting results with some showing agreement between measures (e.g. Goldman et al., 2009; Malow et al., 2006), and others finding poor concordance between parent-rated questionnaire data and objective sleep assessment (Hering et al., 1999; Wiggs and Stores, 2004; Goodlin-Jones et al., 2008). Interestingly, when comparing parental-responses on sleep diaries with actigraphy, studies have found good agreement between sleep parameters (e.g. Allik et al., 2006).

As noted, sleep questionnaires do not necessarily assess the same sleep behaviours as subjective sleep diaries, or objective measures such as actigraphy. In response to the suggestion that parents of children with ASD may misperceive sleep parameters, leading to an over-reporting of sleep problems on subjective measures (Hering et al., 1999), it would be useful to compare parents’ responses on both sleep diaries (i.e. parents’ impression of sleep parameters – e.g. onset/offset of sleep etc.)
and on a questionnaire measure. Agreement between the sleep diary and actigraphy variables, in the context of poor agreement between comparable questionnaire and actigraphy variables, would suggest that while parents have knowledge of sleep patterns, this does not necessarily correspond to their perception of whether or not the sleep behaviour is a problem.

Given that the majority of questionnaire studies report high levels of sleep disturbance, in the context of mixed results from studies employing objective assessment of sleep, the primary aim of the current study was to explore group differences between primary school-age children with and without ASD, using both parent-reported and actigraphy sleep data in a single sample. Specifically, it was hypothesised that children with ASD would differ from their TD peers across subscales of the CSHQ, but that differences would be less consistent across sleep diary or actigraphy-derived variables. Secondly, as few studies have reported on direct comparisons between objective and subjective assessment of sleep, the current study sought to explore agreement between these methods of sleep assessment, assessing the agreement between actigraphy, sleep diaries and the CSHQ. It was anticipated that while the relationship between comparable CSHQ subscales and actigraphy variables would be weak, there would be stronger agreement between comparable actigraphy and sleep diary variables.

4.3 Method

4.3.1 Participants

A total of 82 primary school aged children (6-12 years), both with (n = 45) and without (n = 37) diagnosis of ASD were recruited for participation in the current study. The majority of children with ASD were recruited primarily from the Western Australian Autism Biological Registry (WAABR) at the Telethon Kids Institute.
(TKI) in Perth, Western Australia (for details see Taylor et al., 2013). Additional participants \( n = 7 \) were recruited via incidental contact with the Autism Research Team members at TKI on institute “open days”, following involvement in other studies, or via the Autism Research Team’s Facebook page. All children had a clinical ASD diagnosis (i.e. either Autistic Disorder, AD; Asperger Syndrome/High Functioning Autism, AS; or Pervasive Developmental Disorder Not Otherwise Specified, PDD-NOS; based on the Diagnostic and Statistical Manual of Mental Disorders criteria 4th ed. DSM–IV; American Psychiatric Association, 2000; or ASD based on DSM-5; American Psychiatric Association, 2013). In Western Australia, this diagnosis is determined by consensus amongst all members of a multidisciplinary team comprised of a paediatrician, psychologist, and speech-language pathologist (Glasson et al., 2008). Diagnosis was confirmed in all WAABR participants using the Autism Diagnostic Observation Schedule-Generic (84% of ASD sample\(^3\); ADOS-G; Lord et al. 2000). Two participants with ASD were subsequently excluded due to insufficient questionnaire data (i.e. greater than 20% missing values as detailed below).

TD children were recruited through local primary schools (e.g. school newsletters). For this group, children were excluded if they had a parent-reported diagnosis of any pervasive developmental disorder or developmental delay \( n = 1 \). An additional three participants were excluded due to insufficient questionnaire data. TD children were also screened for autistic-like traits using the Children’s version of the Autism Spectrum Quotient (AQ-Child; Auyeung, Baron-Cohen, Wheelwright, &

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\(^3\) While the majority of participants with ASD in Chapters 4 and 5 had their diagnosis confirmed by ADOS, the decision was made to include the small number of participants for whom these data were not available.
None of the children in the TD group scored above the cut-off for clinically elevated symptoms of ASD (i.e. none above 76; M = 51.46, SD = 12.49)

4.3.2 Procedure

The study procedure was approved by The University of Western Australia Human Research Ethics Committee (RA/4/1/6487). All parents received an information sheet about the study and provided written consent. Following enrolment, parents and the participating children were met during a school or home visit and provided with the parent questionnaires, sleep diary and Actiwatch. Both parents and children were given a detailed verbal explanation of how to use the Actiwatch and complete the sleep diary. Children were requested to wear the watch on their non-dominant wrist for seven days and nights (i.e. five weeknights and two weekend nights). At the end of the seven days, the questionnaires and Actiwatch were collected. All data collection took place during the school semester. The CSHQ was completed by parents at the end of the week, with reference to the seven-day period during which the child was wearing the Actiwatch. Parents were advised that questionnaires be completed by the primary caregiver, with all completed by the same caregiver. As greater than 90% of informants were the child’s mother, there was too little data from fathers to systematically examine possible differences between informants.

4.3.3 Measures

Demographic Information

Parents provided information on their child’s clinical diagnoses such as ID, global developmental delay (GDD), attention deficit hyperactivity disorder (ADHD), prescribed medication, and use of complementary or alternative medication. Within the ASD group parents also provided details of the ASD diagnosis, including
information regarding who provided the diagnosis. Included in the demographic questionnaires was a single item to assess parents’ perception of sleep, where parents rated their child’s sleep overall on a five-point scale (1 “Very Good” to 5 “Very Poor/ Difficulties almost every night”).

**Parent-Reported Sleep Problems**

*Children’s Sleep Habits Questionnaire-Revised (CSHQ-R).*

The CSHQ-R (Host et al., 2018) is a sleep questionnaire validated for use in ASD populations. It is derived from the 33-item parent-report CSHQ developed by Owens et al (2000) to examine sleep behaviour over the preceding week. Items are rated on a three-point scale (from 1 = “Rarely”, to 3 = “Usually”). Five subscales as summed from the 27 items, including sleep initiation and maintenance (Sleep I & M), morning lethargy, co-sleeping, sleep disordered breathing (SDB) and parasomnias. A total CSHQ-R score is derived by summing scores for these five scales. Across all subscales, a higher score is indicative of greater sleep problems (four positively phrased items are reverse scored). As noted above, five participants had insufficient CSHQ-R data (> 20% missing data) for inclusion in the analysis. Where participants had fewer than 20% missing values, missing item scores were prorated using the item mean across the ASD and TD samples separately (<1% of total items).

**Sleep diaries**

Sleep diaries (or sleep logs) were used both as an adjunct to the actigraphy (detailed below) and also as a separate measure of parent-reported sleep parameters. Sleep diaries were completed during the same week as actigraphy recording. Each morning, parents – with help from their participating child – were asked to record their child’s bedtime (D_BT), sleep onset time (D_SOT), and wake time (D_WT). At the end of the day, it was requested that they record both naps and instances where
the watch was not worn. Reports of D_BT, D_SOT and D_WT were used to calculate the time spent in bed (D_TIB), sleep period time (D_SP) and sleep onset latency (D_SOL); these measures are detailed in Table 4.1.

**Objective Measure of Sleep Problems**

To gain an objective measure of sleep patterns, participants wore an Actiwatch-2 (Philips Respironics MiniMitter Actiwatch). Use of the Actiwatch was informed by methods set out by Fawkes et al. (2015). Each watch was configured to record in 60-second epochs. Data were scored in line with guidelines for the use of actigraphy in paediatric groups (Fawkes et al., 2015; Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). Descriptions of the actigraphy variables, including how they were calculated are detailed in Table 4.1. These were scored using the Respironics software (detailed below). All actigraphy data were checked and scored by the same researcher (A. Host) with data for 20% of the participants confirmed by a second researcher (F. Waters).

For inclusion in the analysis, participants required a minimum of 5 nights of valid actigraphy recordings (Acebo et al., 1999), with each of the final variables calculated from averaging across the available nights. Due to Actiwatch malfunction during data collection (n = 6), and participants being unwilling or unable to tolerate wearing the Actiwatch (n = 3), valid actigraphy data were available for only 67 participants (ASD = 39 [90.69%]; TD = 28 [84.84%]). The mean number of nights for actigraphy recordings was 6.82 (SD = .49).

**4.3.4 Statistical analyses**

Respironics Actiware Software (Version 6.0.2) was used to analyse the Actigraphy data. As indicated by Meltzer, Walsh, Traylor and Westin (2012) the default scoring setting of medium sensitivity (i.e. 40 activity counts per epoch) was
used. Remaining analysis was completed using SPSS Version 22.0 for Windows. Data for each group (ASD and TD) were screened for outliers using a criterion of 3.29 SD from the mean for each variable. Outliers were dealt with using a winsorizing approach, reducing the score to be one unit higher than the next highest value (Field, 2013).

To address the first hypothesis regarding group differences across sleep measures, independent sample t-tests were computed to test group differences for the CSHQ-R subscales, as well as the sleep diary and actigraphy parameters. Corrections to the alpha level for multiple tests were not applied since more conservative testing would have compromised power in the studies where sample sizes were modest. Subsequent to this, chi-squared analyses were conducted to compare the TD and ASD samples on two overall measures of the rate of sleep problems. For the first measure, based on parent perceptions of sleep problems, responses from the single sleep item were re-coded to create two groups: “good” and “problem” sleepers. Good sleepers were those whose parents indicated they were “Very Good” or “Generally Good” sleepers (problems < 2 nights per week). Poor sleepers were those whose parents indicated they had problems more than two nights per week. For the second measure, based on objective assessment of sleep, participants were separated into “good” and “problem” sleepers based on actigraphy, using normative data reported by Hirshkowitz et al. (2015) and Ohayon et al. (2017). Actigraphy cut-offs for poor sleep quality were derived from guidelines for school-aged children presented by Ohayon et al. (2017): SOL of greater than 45 mins, WASO greater than 40 mins and SE less than 74%. A participant was classified as having a “sleep quality” problem if they met cut-off for at least two of these three variables.
Table 4.1

Details of the Actigraphy Variables Used and How They Were Calculated

<table>
<thead>
<tr>
<th>Variable</th>
<th>Abbreviation</th>
<th>Units</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Diary</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Bedtime</td>
<td>D_BT</td>
<td>(HH:MM)</td>
<td>time recorded in sleep diary for when the child attempted to fall asleep</td>
</tr>
<tr>
<td>Wake Time</td>
<td>D_WT</td>
<td>(HH:MM)</td>
<td>time recorded in sleep diary for when the child woke up</td>
</tr>
<tr>
<td>Time In Bed</td>
<td>D_TIB</td>
<td>(min)</td>
<td>duration between bedtime and wake time</td>
</tr>
<tr>
<td>Sleep Onset Time</td>
<td>D_SOT</td>
<td>(HH:MM)</td>
<td>time recorded in sleep diary for when the child fell asleep</td>
</tr>
<tr>
<td>Sleep Period</td>
<td>D_SP</td>
<td>(min)</td>
<td>duration between diary sleep onset and diary wake time</td>
</tr>
<tr>
<td>Sleep Onset Latency</td>
<td>D_SOL</td>
<td>(min)</td>
<td>duration between bedtime and diary sleep onset time</td>
</tr>
<tr>
<td><strong>Actigraphy</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep Onset Time</td>
<td>SOT</td>
<td>(HH:MM)</td>
<td>time of the first of 10 consecutive minutes scored as sleep after bedtime</td>
</tr>
<tr>
<td>Sleep Offset Time</td>
<td>SOffT</td>
<td>(HH:MM)</td>
<td>time of the last of 10 consecutive minutes scored as sleep before wake time</td>
</tr>
<tr>
<td>Sleep Period</td>
<td>SP</td>
<td>(min)</td>
<td>duration between sleep onset and sleep offset times</td>
</tr>
<tr>
<td>Wake After Sleep Onset</td>
<td>WASO</td>
<td>(min)</td>
<td>number of minutes scored as wake between sleep onset and sleep offset time</td>
</tr>
<tr>
<td>Total Sleep Time</td>
<td>TST</td>
<td>(min)</td>
<td>sleep period time (mins) minus wake after sleep onset (mins)</td>
</tr>
<tr>
<td>Sleep Efficiency</td>
<td>SE</td>
<td>(%)</td>
<td>percentage of time spent asleep whilst in bed ((TST/TIB)x100)</td>
</tr>
<tr>
<td>Sleep Onset Latency</td>
<td>SOL</td>
<td>(min)</td>
<td>duration between bedtime and sleep onset time</td>
</tr>
</tbody>
</table>

*Note.* Table adapted from Table 4 in “Use of actigraphy for assessment in pediatric sleep research” Meltzer et al. (2012)
Actigraphy cut-off for a “sleep quantity” problem was derived from Hirshkowitz et al. (2015); defined as TST of less than 7 hours. It is noted that the cut-offs chosen are unequivocally conservative definitions of sleep problems.

To address the second hypothesis (i.e. agreement between subjective and objective measures), CSHQ-R subscale scores (excluding the SDB subscale) were compared to sleep diary and actigraphy variables using Pearson’s correlation analyses. The SDB subscale of the CSHQ-R was not included in this analysis as symptoms of SDB are not assessed by actigraphy or sleep diaries (Markovich et al., 2015).

4.4 Results

4.4.1 Participant characteristics

Demographic variables are detailed in Table 4.2. The final sample consisted of 39 children with ASD and 28 TD children. The average age of the ASD group was 117.38 months (SD = 20.56), while the average age for the TD group was 119.32 months (SD = 16.76); with no significant difference between the groups, \( t (65) = 0.41, p = .683 \). Moreover, there were no significant differences in ethnicity between the groups \( \chi^2(2) = 4.84, p = .089 \); however, there were significantly more males in the ASD sample \( \chi^2(1) = 4.92, p = .027 \).

As can be seen in Table 4.2, twenty-five (66.7%) of the children with ASD had a diagnosis of AD, seven (17.9%) had a diagnosis of AS and five (12.8%) had a diagnosis of PDD-NOS. With regard to comorbidities, developmental disorders reported were ADHD, ID, Specific Language Impairment and Specific Learning Disability (e.g. dyslexia; see Table 4.2). The most frequently reported medical comorbidities included allergies, asthma, and gastrointestinal problems.
A number of children with ASD were taking prescription medications (33.3%; mostly stimulants or mood stabilisers and those used to treat allergies and asthma or gastrointestinal symptoms). Three TD children (10.7%) were also taking medications. For the majority of participants, medications had been stable for a minimum of six months prior to enrolment in the study, excluding two children whose medications had been stable for six and four weeks respectively.

With regard to reporting, the vast majority of informants were the child’s mother (greater than 90%), as such, there was too little data from fathers to systematically examine possible differences between informants.

Specifically regarding sleep, three children (TD = 1; ASD = 2) had previously received a diagnosis of sleep apnoea; however, all three reported subsequent tonsillectomy or adenotonsillectomy. In addition to this, eleven children with ASD (28.2%) were currently taking medication prescribed for sleep-related problems. Medications reported were melatonin (n = 10; dose 2-10mg) and clonidine (n = 3). All were reported to have been stable on these medications for at least six months prior to enrolment in the study. While none of the TD children were taking any medication prescribed for sleep, one child was taking over the counter melatonin (1mg) “when required”.
Table 4.2
Demographic Information for the Current Sample

<table>
<thead>
<tr>
<th></th>
<th>TD</th>
<th>ASD</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>n = 28</td>
<td>n = 39</td>
</tr>
<tr>
<td></td>
<td>n (%)</td>
<td>n (%)</td>
</tr>
<tr>
<td>Sex (male)</td>
<td>17 (60.7)</td>
<td>33 (84.6)</td>
</tr>
<tr>
<td>First language</td>
<td></td>
<td></td>
</tr>
<tr>
<td>English</td>
<td>25 (89.3)</td>
<td>36 (92.3)</td>
</tr>
<tr>
<td>Other</td>
<td>2 (7.1)</td>
<td>1 (2.6)</td>
</tr>
<tr>
<td>Not specified</td>
<td>1 (3.6)</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Ethnicity</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>25 (89.3)</td>
<td>26 (66.7)</td>
</tr>
<tr>
<td>Other</td>
<td>2 (7.1)</td>
<td>6 (15.4)</td>
</tr>
<tr>
<td>Not specified</td>
<td>1 (3.6)</td>
<td>7 (17.9)</td>
</tr>
<tr>
<td>ASD diagnosis (clinician diagnosed)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Autistic Disorder</td>
<td>Nil.</td>
<td>25 (64.1)</td>
</tr>
<tr>
<td>Asperger Syndrome</td>
<td>Nil.</td>
<td>7 (17.9)</td>
</tr>
<tr>
<td>PDD-NOS</td>
<td>Nil.</td>
<td>5 (12.8)</td>
</tr>
<tr>
<td>Not specified</td>
<td>Nil.</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Comorbidities (clinician diagnosed)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Epilepsy</td>
<td>Nil.</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Gastrointestinal problems</td>
<td>Nil.</td>
<td>4 (10.3)</td>
</tr>
<tr>
<td>Asthma</td>
<td>Nil.</td>
<td>3 (7.7)</td>
</tr>
<tr>
<td>Allergies</td>
<td>4 (14.3)</td>
<td>10 (25.6)</td>
</tr>
<tr>
<td>Other medical diagnoses</td>
<td>2 (7.1)</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Intellectual Disability</td>
<td>Nil.</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Attention Deficit Hyperactivity Disorder</td>
<td>Nil.</td>
<td>12 (30.8)</td>
</tr>
<tr>
<td>Specific Language Impairment</td>
<td>Nil.</td>
<td>1 (2.6)</td>
</tr>
<tr>
<td>Specific Learning Disorder</td>
<td>Nil.</td>
<td>1 (2.6)</td>
</tr>
<tr>
<td>Past diagnosis of sleep disorder</td>
<td>1 (3.6)</td>
<td>5 (12.8)</td>
</tr>
<tr>
<td>Prescribed medications</td>
<td></td>
<td></td>
</tr>
<tr>
<td>For sleep problem</td>
<td>Nil.</td>
<td>11 (28.2)</td>
</tr>
<tr>
<td>For non-sleep related diagnosis</td>
<td>3 (10.7)</td>
<td>13 (33.3)</td>
</tr>
<tr>
<td>Maternal education</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Did not complete Year 12 Cert</td>
<td>Nil.</td>
<td>3 (8.0)</td>
</tr>
<tr>
<td>Completed Year 12 Cert</td>
<td>7 (25.0)</td>
<td>9 (23.1)</td>
</tr>
<tr>
<td>Undergraduate degree</td>
<td>11 (39.3)</td>
<td>15 (38.5)</td>
</tr>
<tr>
<td>Postgraduate degree</td>
<td>9 (32.1)</td>
<td>2 (5.1)</td>
</tr>
<tr>
<td>Not specified</td>
<td>1 (3.6)</td>
<td>10 (26)</td>
</tr>
</tbody>
</table>

Note. PDD-NOS = Pervasive Developmental Disorder-Not Otherwise Specified
4.4.2 Group differences across sleep patterns

Subjective sleep problems

The mean total CSHQ-R score across all participants was 36.55 (SD = 6.74). Table 4.3 shows that children with ASD had significantly higher total scores than the TD children. In addition to this, those with ASD also had significantly higher scores on subscales measuring Sleep I & M, SDB and Parasomnias; however, the two groups did not differ on the Morning Lethargy or the Co-sleeping subscales.

Contrary to CSHQ-R data, few differences emerged between ASD and TD groups on sleep diary parameters. As can be seen in Table 4.3, the children with ASD only differed from TD children in that they had an earlier D_WT (getting up 22 mins earlier on average), compared to TD children.

Objective sleep patterns

Few differences emerged between groups on objective sleep parameters. As can be seen in Table 4.3, the children with ASD had an earlier SOffT (on average waking 27 mins earlier) than the TD children.

Rate of sleep problems

Based on parental response to the single item measure of sleep, the percentage of participants who were perceived as having a sleep problem differed by group; \( \chi^2(1) = .718, p = .007 \). Of the children in the TD group, 19% (n = 7) were reported to have a sleep problem, while 51.4% (n = 19) of those with ASD were reported to have a sleep problem.

As detailed above, participants were also grouped into those with or without sleep problems based on actigraphy assessed sleep duration or sleep quality markers.

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For reference, total CSHQ score using the full 33-item version of the questionnaire yielded means (SD) of 40.79 (5.70) for the TD group and 46.45 (7.95) for the ASD group which are comparable to those reported in the paediatric and ASD sleep literature.
Based on these criteria, 25% (n = 7) of TD children and 36% (n = 14) of children with ASD had a sleep quality problem; however, the difference between groups was not statistically significant $\chi^2(1) = .899, p = .343$. Similarly, while 11% (n = 3) of TD children and 15% (n = 6) of children with ASD had a sleep duration problem, this difference was not statistically significant, $\chi^2(1) = .306, p = .580$.

Table 4.3
Means (SD) and Independent Sample T-Tests Comparing Sleep Diary, Actigraphy and CSHQ-R between Children with (N = 39) and without (N = 28) ASD

<table>
<thead>
<tr>
<th></th>
<th>ASD M (SD)</th>
<th>TD M (SD)</th>
<th>t</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Sleep Diary</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>D_BT (HH:MM)</td>
<td>20:42 0:51</td>
<td>20:50 0:37</td>
<td>-0.72</td>
<td>.471</td>
</tr>
<tr>
<td>D_SOT (HH:MM)</td>
<td>21:10 0:56</td>
<td>21:15 0:37</td>
<td>-0.45</td>
<td>.652</td>
</tr>
<tr>
<td>D_WT (HH:MM)</td>
<td>6:38 0:50</td>
<td>7:00 0:22</td>
<td>-2.16</td>
<td>.034*</td>
</tr>
<tr>
<td>D_TIB (min)</td>
<td>595.90 38.79</td>
<td>609.74 34.18</td>
<td>-1.51</td>
<td>.135</td>
</tr>
<tr>
<td>D_SP (min)</td>
<td>565.65 40.79</td>
<td>583.98 32.65</td>
<td>-1.92</td>
<td>.060</td>
</tr>
<tr>
<td>D_SOL (min)</td>
<td>27.07 15.89</td>
<td>23.08 13.57</td>
<td>1.05</td>
<td>.296</td>
</tr>
<tr>
<td><strong>Actigraphy</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOT (HH:MM)</td>
<td>21:18 0:59</td>
<td>21:28 0:48</td>
<td>-0.73</td>
<td>.466</td>
</tr>
<tr>
<td>SOFFT (HH:MM)</td>
<td>6:12 0:49</td>
<td>6:39 0:31</td>
<td>-2.57</td>
<td>.013*</td>
</tr>
<tr>
<td>SP (min)</td>
<td>533.95 39.93</td>
<td>551.22 42.41</td>
<td>-1.70</td>
<td>.094</td>
</tr>
<tr>
<td>TST (min)</td>
<td>464.73 42.67</td>
<td>486.63 46.88</td>
<td>-1.99</td>
<td>.051</td>
</tr>
<tr>
<td>WASO (min)</td>
<td>69.00 23.35</td>
<td>64.59 16.86</td>
<td>0.85</td>
<td>.397</td>
</tr>
<tr>
<td>SE (%)</td>
<td>78.27 6.81</td>
<td>79.85 5.77</td>
<td>-1.00</td>
<td>.322</td>
</tr>
<tr>
<td>SOL (min)</td>
<td>34.58 18.87</td>
<td>36.86 27.48</td>
<td>-0.40</td>
<td>.689</td>
</tr>
<tr>
<td><strong>CSHQ-R</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep I &amp; M</td>
<td>10.85 3.33</td>
<td>9.32 2.51</td>
<td>2.04</td>
<td>.045*</td>
</tr>
<tr>
<td>Morning Lethargy</td>
<td>7.49 2.04</td>
<td>7.64 2.20</td>
<td>-0.30</td>
<td>.766</td>
</tr>
<tr>
<td>Co-sleeping</td>
<td>6.44 1.85</td>
<td>5.86 1.96</td>
<td>1.23</td>
<td>.222</td>
</tr>
<tr>
<td>SDB</td>
<td>3.65 0.99</td>
<td>3.21 0.42</td>
<td>2.20</td>
<td>.031*</td>
</tr>
<tr>
<td>Parasomnias</td>
<td>10.01 2.26</td>
<td>8.18 1.36</td>
<td>3.83</td>
<td>.000***</td>
</tr>
<tr>
<td>Total</td>
<td>38.50 7.13</td>
<td>33.86 5.14</td>
<td>2.94</td>
<td>.005**</td>
</tr>
</tbody>
</table>

Note. D_BT = diary bedtime; D_WT = diary wake time; D_TIB = diary time in bed; SOT = sleep onset time; SOFFT = sleep offset time; SP = sleep period; WASO = wake after sleep onset; TST = total sleep time; SE = sleep efficiency; SOL = sleep onset latency; CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing.
* p < .05; ** p < .01; *** p < .001.
4.4.3 Agreement between objective and subjective assessment

Across measures of sleep, a number of variables were associated with age in both the ASD and TD groups (e.g. CSHQ-R SDB and Co-sleeping; sleep diary D_BT, D_SOT, D_TIB and SP; and actigraphy SOT and TST). Given the number of variables associated with age, all further analysis employed age as a covariate. Conversely, none of the sleep variables were related to the diagnosis of comorbid developmental or medical conditions, or the use of medication (point-biserial correlations for dichotomous variables), except for sleep medications (related to SOT, SOFFT and SOL within the ASD group). As such, only use of sleep medication was included as a covariate for the ASD group.

As can be seen in Table 4.4 there were no associations between actigraphy and CSHQ-R subscale scores for the TD group. Within the ASD group, two relationships were noted. Firstly, later morning waking was associated with higher scores on the Parasomnias subscales. Secondly, an inverse relationship was found between SOL (actigraphy) and Co-sleeping behaviours (on CSHQ-R), indicating that increased co-sleeping was associated with a shorter time between going to bed and falling asleep.

With regard to the sleep diary variables, again, there were very few associated with any of the CSHQ-R subscale scores. Within the TD group, Table 4.5 shows a moderate positive correlation between time taken to fall asleep, that is D_SOL (sleep diary), and Sleep I & M subscale score (CSHQ-R), indicating that those TD children who recorded longer times between bedtime and sleep onset also scored higher on the subscale assessing difficulty with initiating and maintaining sleep.
Table 4.4
Partial Correlations Controlling for Age and Use of Sleep Medication between the Actigraphy and CSHQ-R for Both the ASD and TD Groups

<table>
<thead>
<tr>
<th></th>
<th>Sleep I &amp; M</th>
<th>Morning Lethargy</th>
<th>Co-Sleeping</th>
<th>Parasomnias</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>TD Group</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOT</td>
<td>.095</td>
<td>.186</td>
<td>.248</td>
<td>.097</td>
<td>.197</td>
</tr>
<tr>
<td>SOffT</td>
<td>.201</td>
<td>.189</td>
<td>.278</td>
<td>.203</td>
<td>.289</td>
</tr>
<tr>
<td>SP</td>
<td>.058</td>
<td>-.062</td>
<td>-.058</td>
<td>.058</td>
<td>.012</td>
</tr>
<tr>
<td>TST</td>
<td>.055</td>
<td>.046</td>
<td>-.070</td>
<td>.091</td>
<td>.049</td>
</tr>
<tr>
<td>WASO</td>
<td>-.007</td>
<td>-.247</td>
<td>.045</td>
<td>-.097</td>
<td>-.094</td>
</tr>
<tr>
<td>SE</td>
<td>.071</td>
<td>.218</td>
<td>-.036</td>
<td>.229</td>
<td>.165</td>
</tr>
<tr>
<td>SOL</td>
<td>.057</td>
<td>.054</td>
<td>.126</td>
<td>-.142</td>
<td>.017</td>
</tr>
<tr>
<td><strong>ASD Group</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOT</td>
<td>.189</td>
<td>.218</td>
<td>-.074</td>
<td>.311</td>
<td>.263</td>
</tr>
<tr>
<td>SOffT</td>
<td>-.078</td>
<td>.270</td>
<td>-.137</td>
<td>.340*</td>
<td>.151</td>
</tr>
<tr>
<td>SP</td>
<td>-.323</td>
<td>.049</td>
<td>-.070</td>
<td>.018</td>
<td>-.147</td>
</tr>
<tr>
<td>TST</td>
<td>-.228</td>
<td>.043</td>
<td>-.066</td>
<td>.010</td>
<td>-.104</td>
</tr>
<tr>
<td>WASO</td>
<td>-.130</td>
<td>-.019</td>
<td>-.011</td>
<td>-.013</td>
<td>-.076</td>
</tr>
<tr>
<td>SE</td>
<td>-.092</td>
<td>.028</td>
<td>.232</td>
<td>.032</td>
<td>.023</td>
</tr>
<tr>
<td>SOL</td>
<td>-.139</td>
<td>-.134</td>
<td>-.463*</td>
<td>-.064</td>
<td>-.246</td>
</tr>
</tbody>
</table>

*Note.* Use of sleep medication was controlled only in the ASD group as none of the TD participants were taking prescription medication for sleep. SOT = sleep onset time; SOffT = sleep offset time; SP = sleep period; WASO = wake after sleep onset; TST = total sleep time; SE = sleep efficiency; SOL = sleep onset latency; CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing. *p < .05

Within the ASD group, a moderate positive correlation was found between D_SOT (sleep diary) and the Parasomnias subscale (CSHQ-R), indicating that those whose diaries recorded earlier sleep times also scored higher on the subscale assessing night-time parasomnias.
Table 4.5
Partial Correlations Controlling for Age and Use of Sleep Medication between the Sleep Diary and CSHQ-R for Both the ASD and TD Groups

<table>
<thead>
<tr>
<th></th>
<th>Sleep I &amp; M</th>
<th>Morning Lethargy</th>
<th>Co-Sleeping</th>
<th>Parasomnias</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD Group</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>D_SOT</td>
<td>.156</td>
<td>.179</td>
<td>.160</td>
<td>.259</td>
<td>.271</td>
</tr>
<tr>
<td>D_WT</td>
<td>.101</td>
<td>.025</td>
<td>.237</td>
<td>.169</td>
<td>.187</td>
</tr>
<tr>
<td>D_SP</td>
<td>-1.18</td>
<td>-2.04</td>
<td>-2.017</td>
<td>-1.92</td>
<td>-1.94</td>
</tr>
<tr>
<td>D_SOL</td>
<td>.388*</td>
<td>-2.052</td>
<td>.035</td>
<td>.124</td>
<td>.196</td>
</tr>
<tr>
<td>ASD Group</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>D_SOT</td>
<td>.229</td>
<td>.220</td>
<td>.024</td>
<td>.369**</td>
<td>.330</td>
</tr>
<tr>
<td>D_WT</td>
<td>.124</td>
<td>.327</td>
<td>-2.217</td>
<td>.314</td>
<td>.251</td>
</tr>
<tr>
<td>D_SP</td>
<td>-2.037</td>
<td>.192</td>
<td>-2.285</td>
<td>.064</td>
<td>.025</td>
</tr>
<tr>
<td>D_SOL</td>
<td>-2.054</td>
<td>-2.237</td>
<td>-2.207</td>
<td>.104</td>
<td>-1.126</td>
</tr>
</tbody>
</table>

Note. Use of sleep medication was controlled only in the ASD group as none of the TD participants were taking prescription medication for sleep. D_BT = diary bedtime; D_WT = diary wake time; D_SP = diary sleep period; D_SOL = diary sleep onset latency; CSHQ-R = Children’s Sleep Habits Questionnaire-Revised; Sleep I & M = Sleep Initiation and Maintenance; SDB = Sleep Disordered Breathing.

* p < .05; ** p < .01.

The sleep diary variables showed better correspondence with actigraphy measures (Table 4.6) with a number of relationships emerging for both the ASD and TD groups. Strong positive correlations were found between comparable measures such as SOT (actigraphy) and D_SOT (diary), as well as SOffT (actigraphy) and D_WT (diary). Similarly, moderate positive relationships were found between SP/TST (actigraphy) and D_SP (diary). However, while there was a moderate positive relationship between SOL (actigraphy) and D_SOL (diary) for the ASD group, there was no relationship between these variables for the TD group, indicating that while parents of children with ASD had reasonable knowledge of the duration of time taken for their child to fall asleep, the parents of TD children did not.
Table 4.6
Partial Correlations Controlling for Age and Use of Sleep Medication between the Actigraphy (rows) and Sleep Diary (columns) for both the ASD and TD Groups

<table>
<thead>
<tr>
<th></th>
<th>D_SOT</th>
<th>D_WT</th>
<th>D_SP</th>
<th>D_SOL</th>
</tr>
</thead>
<tbody>
<tr>
<td>TD Group</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOT</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOffT</td>
<td>.753**</td>
<td>.654**</td>
<td>-.431*</td>
<td>.020</td>
</tr>
<tr>
<td>SP</td>
<td>.478*</td>
<td>.781**</td>
<td>.009</td>
<td>.362</td>
</tr>
<tr>
<td>TST</td>
<td>-.491*</td>
<td>-.111</td>
<td>.526**</td>
<td>.290</td>
</tr>
<tr>
<td>WASO</td>
<td>-.324</td>
<td>.038</td>
<td>.432*</td>
<td>.274</td>
</tr>
<tr>
<td>SE</td>
<td>-.273</td>
<td>-.332</td>
<td>.083</td>
<td>-.043</td>
</tr>
<tr>
<td>SOL</td>
<td>.041</td>
<td>.007</td>
<td>-.046</td>
<td>.219</td>
</tr>
<tr>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>ASD Group</td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOT</td>
<td>.912**</td>
<td>.722**</td>
<td>-.014</td>
<td>.084</td>
</tr>
<tr>
<td>SOffT</td>
<td>.548**</td>
<td>.863**</td>
<td>.494*</td>
<td>-.154</td>
</tr>
<tr>
<td>SP</td>
<td>-.486**</td>
<td>.125</td>
<td>.662**</td>
<td>-.310</td>
</tr>
<tr>
<td>TST</td>
<td>-.462**</td>
<td>-.030</td>
<td>.447*</td>
<td>-.329</td>
</tr>
<tr>
<td>WASO</td>
<td>.079</td>
<td>.229</td>
<td>.175</td>
<td>.097</td>
</tr>
<tr>
<td>SE</td>
<td>-.300</td>
<td>-.394*</td>
<td>-.174</td>
<td>-.375*</td>
</tr>
<tr>
<td>SOL</td>
<td>.243</td>
<td>.380*</td>
<td>.251</td>
<td>.400*</td>
</tr>
</tbody>
</table>

Note. Correlations for theoretically corresponding variables are in bold italics. Use of sleep medication was controlled only in the ASD group as none of the TD participants were taking prescription medication for sleep. D_BT = diary bedtime; D_WT = diary wake time; D_TIB = diary time in bed; SOT = sleep onset time; SOffT = sleep offset time; SP = sleep period; WASO = wake after sleep onset; TST = total sleep time; SE = sleep efficiency; SOL = sleep onset latency.

* p < .05; ** p < .01.

4.5 Discussion

The two aims of the current study were to explore group differences between primary school-age children with and without ASD (using both parent-reported and actigraphy sleep data) and to assess cross-method concordance between actigraphy, sleep diaries, and the CSHQ-R. In brief, while children with ASD had both higher rates and higher levels of sleep problems on parent-rated measures, few differences emerged on actigraphy measures of sleep.

Interestingly, as with actigraphy, few differences emerged when assessing parent reports of sleep scheduling using sleep diaries. Overall, there was poor
agreement between CSHQ-R subscales and sleep diaries or actigraphy recording, but moderate to strong agreement on comparable diary and actigraphy variables.

When looking at the rates of children having either “poor” or “good” sleep, significantly more children with ASD than TD children were considered by their parents to have a sleep problem. However, despite parent-reported sleep problems on this single-item measure, when groupings were based on actigraphy recordings, there were no significant differences between the ASD and TD samples in the rates of children meeting predetermined cut-offs for “poor” sleep (quality or quantity).

Consistent with the literature, children with ASD were reported by their parents to have higher levels of sleep problems than their TD peers across most subscales of the CSHQ (e.g. Couturier et al., 2005; Giannotti et al., 2008; Giannotti, Cortesi, Cerquiglini, Vagnoni, & Valente, 2011; Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008; Goodlin-Jones, Tang, Liu, & Anders, 2008; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Hoffman et al., 2005; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Souders et al., 2009). As the current study used a revised version of the CSHQ, direct comparisons between subscales are not possible; however, in the current study TD and ASD children did not differ on Co-sleeping behaviour (e.g. “falls asleep in others’ bed” or “needs parent in the room to fall asleep”), or Morning Lethargy (e.g. “difficulty getting out of bed in the morning”, “takes a long time to be alert”).

Despite parent reports of sleep problems on the CSHQ-R, analysis of parent recordings in sleep diaries, and review of actigraphy, revealed few group differences for sleep parameters. Indeed, group differences for the diaries and actigraphy emerged only on the D_WT (diary) and SOffT (actigraphy) variables, where children with ASD woke and got up approximately 25 mins earlier than their TD peers.
Despite differences in rise time, this did not translate into significant group differences in the total amount of time spent in bed or sleeping (albeit with the later approaching significance).

Few have reported group differences in SOffT (Allik, Larsson, & Smedje, 2006; Hering et al 1999) while a number found no such difference (e.g. Baker, Richdale, Short, & Gradisar, 2013; Goodlin-Jones, Tang, Liu, & Anders, 2008; Souders et al., 2009; Tani et al., 2005). Further, the most consistently reported group differences based on actigraphy data – increased SOL and decreased SE (e.g. Allik et al., 2006; Baker et al., 2013; Hare, Jones, & Evershed, 2006; Souders et al., 2009) – were not replicated in the current study. This further highlights the inconsistencies across studies when employing objective assessment of sleep.

In this context, it is pertinent to note that a recent meta-analysis examining objectively measured sleep parameters in ASD identified a number of moderator variables impacting on the findings of group differences between ASD and TD samples (Elrod & Hood 2015). Namely, studies which included children with ID were more likely to identify group differences in TST while those which included children on medication were less likely to report differences in SE (Elrod & Hood 2015). The fact that only two children in the current sample had been diagnosed with an ID may have contributed to the finding of comparable sleep times between the groups. Further, many children in the current sample, especially those with ASD, were taking medications including those prescribed to aid sleep. As such, these children may not have differed from their TD peers on SE due to the benefit from medication.

Importantly, however, parent-ratings of sleep were collected over the same week-long period as sleep assessment. Therefore, despite comparable sleep
parameters over this week period, parents still reported significantly higher levels of sleep disturbance. Given that approximately one-third of the participants with ASD were taking sleep medication, it is possible that parents experience of their child’s past sleep problems (which presumably preceded use of sleep medication) continued to impact their current perception despite improvements in actual sleep parameters (i.e. no difference from peers). Alternatively, it may be that even if sleep parameters improve, the child continues to present with a range of disruptive sleep-related behaviours (i.e. bedtime resistance). It would be beneficial for future intervention studies to assess differences in both parent-report and objective assessment of sleep to determine if various treatments (i.e. medication, behavioural intervention etc.) have comparable outcomes across measures.

With regard to cross-method concordance, overall there was little relationship between the CSHQ-R subscales and diary or actigraphy measures for either group. Notably, subscales on the CSHQ-R which were conceptually most similar to variables obtained from actigraphy/sleep diary (e.g. Sleep I & M) were not related to these measures for either group. Rather, within the ASD group, higher scores on the Parasomnias subscale were associated with later wake times (SOffT; actigraphy) and Co-sleeping behaviour was associated with reduced time taken to fall asleep (SOL; actigraphy). With regard to the latter, in general (and for the purpose of scoring the CSHQ), co-sleeping by school-age is considered indicative of increased sleep problems. This highlights a clear discrepancy between the information gleaned from different measures, where children are still falling asleep within a reasonable time (actigraphy) but in an environment (parent’s bed) that is not ideal, which is captured only by questionnaire responses. Overall, higher scores on the CSHQ from parents of children with ASD may be reflecting negative experiences around bedtime such as a
rigid ritual of bedtime routine, which ultimately do not impact upon sleep onset or offset times. Future reach may benefit from further exploring parental perceptions of bedtime behaviours such as routines and co-sleeping, to establish whether these are perceived as a problematic, or acceptable practices to reduce their child’s distress and aid sleep.

When reviewing sleep diaries, within the TD group a relationship was found between comparable measures from the sleep diary and CSHQ-R, with an association between the Sleep I & M subscale and time taken to fall asleep (SOL). Within the ASD group, those whose parents recorded in their diaries that they fell asleep earlier in the evening also had higher scores on the CSHQ-R Parasomnias subscale. While these relationships are logical, other actigraphy and sleep diary variables that may have been expected to correspond to CSHQ-R subscales (e.g. such as Sleep I & M with WASO or SE) were not related.

Despite this, when comparing the sleep diary variables to actigraphy-derived sleep parameters, a number of relationships emerged amongst comparable variables, suggesting good agreement between actigraphy and parent reporting of their children’s sleep parameters. Interestingly, agreement between comparable measures were consistently (though only slightly) higher for the ASD group when compared to the TD group. This is important as it indicates that parents (especially those with children with ASD) have good knowledge of their children’s sleep patterns. In this context it seems less likely that elevated scores on questionnaire measures represent a misperception and reduced awareness of sleep patterns as suggested by Hering et al. (1999), but rather that sleep questionnaires are capturing features of sleep behaviours which are of concern, even if they do not relate to changes in underlying sleep parameters, such as less time spent asleep.
It is acknowledged that within the current study, it is possible that the strong agreement between actigraphy and diary may have been influenced by the fact that participating children were requested to assist their parents in completing the sleep diaries, while the CSHQ-R was completed by parents independently. However, it is also noted that the CSHQ-R was completed at the end of the seven-day actigraphy period, and as such, parents would have had knowledge of their responses in the sleep diary at the time of completing the CSHQ-R (i.e. they did not fill out the CSHQ-R, and then subsequently find out that their child was going to sleep later than they had previously thought).

Another factor that must be considered is the limitations of actigraphy as a tool to assess sleep. The device used in the current thesis (Philips Respironics MiniMitter Actiwatch-2) has been found to have some of the highest agreement with PSG, recording sensitivity (to sleep) of 93% and specificity (to wake) of 71% (Meltzer, Walsh et al., 2012). However, it is widely acknowledged the best application of actigraphy is in detecting sleep timing (SOT, SOffT, TST) with lower agreement for sleep quality variables such as WASO and SE (Ancoli-Israel et al., 2003; Sadeh, 2011; Sadeh & Acebo, 2002; So et al., 2005). Of relevance to the current research, one study examining actigraphy in groups with neurodevelopmental disorders, found specificity to be as low as 24% (Sitnick, Goodlin-Jones, & Anders, 2008). As such, findings regarding sleep quality variables must be interpreted with caution.

A further limitation of the current study is that a number of children were taking medication, including those which may have inadvertently impacted upon sleep; however, as noted above, none of the sleep variables assessed were associated with the use of medication. Additionally, while the current study did include children...
with a range of clinical comorbidities (e.g. ID, epilepsy) it was noted that the presence of these conditions was not related to sleep within the current sample.

With regard to sampling, it is acknowledged that the ASD and TD samples were not matched for sex. Finally, while efforts were made to reduce recruitment bias it is also possible that the families most inclined to engage with the current research are those who have children with sleep problems. Importantly, however, the TD children’s scores on the CSHQ were comparable to those reported in other studies.

Overall, children with ASD have higher parent-rated sleep problems than their TD peers; however, few differences emerge when analysing sleep parameters, either by parent diary reports or actigraphy. Contrary to suggestions that parents of children with ASD may over-report sleep problems, agreement between actigraphy and sleep diaries indicates that parents seem to have good knowledge of sleep parameters (i.e. accurate sleep diary reports), though continue to express concerns around sleep behaviours which may not directly impact objective sleep parameters.
4.6 References


Chapter 4: Parent-Reported and Actigraphy-Derived Sleep Parameters in Children With and Without Autism Spectrum Disorder


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Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?

5.1 Preamble to Chapter 5

Given the persistent nature of sleep problems in autism spectrum disorder (ASD), as outlined in Chapter 3, and the high levels of parent-reported sleep problems detailed in Chapter 4, this final empirical chapter sought to examine the relationship between these sleep problems and daytime behaviour and executive functioning. As noted in Chapter 1, poor sleep in ASD has been found to be associated with a range of cognitive and behavioural outcomes. However, there is a paucity of literature examining these associations at a more detailed level. The current study sought to build on the existing understanding of the impact of sleep on daytime executive functioning and challenging externalising behaviour in ASD, with reference to similar findings in the broader TD paediatric sleep literature.
Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours In Children With and Without Autism Spectrum Disorder?

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Abstract
The current study explored the relationship between sleep (using objective and subjective assessment) and daytime challenging behaviours (parent-rated) and executive functioning (EF; parent-rated and performance-based measures) in primary school-age children with (n = 32) and without (n = 32) autism spectrum disorder ASD. The results showed that, within each group, parental reports of sleep problems, challenging behaviours and EF skills were all related; however, objectively assessed sleep (actigraphy) was not related to behaviour or EF. Further, performance-based measures of EF were not related to sleep (objective or subjective assessment) or reports of challenging behaviours. Mediation analysis indicated that parent-reported poor behavioural EF explained the relationship between parent-reported sleep and challenging behaviours in the TD group; however, results fell short of reaching statistical significance in the ASD group. Given that objectively assessed sleep was not related to any measure of cognition or behaviour, alternative explanations for the association between parent-rated sleep and behaviours were considered.

5.2 Introduction

Sleep is essential for social, emotional, and cognitive well-being. There is considerable evidence that poor sleep has an adverse impact on daytime cognition (e.g. Gruber et al., 2010; Kopasz et al., 2010; Paavonen et al., 2010), academic achievement (e.g. Gozal, 1998), mood (for reviews see Gregory & Sadeh, 2012, 2016) and behavioural regulation (e.g. Aronen, Paavonen, Fjällberg, Soininen, & Törrönen, 2000; Becker, Langberg, & Evans, 2015; Lavigne et al., 1999; Touchette et al., 2007). For example, experimentally restricting sleep duration has been found to result in lower mood and increased difficulty with regulating behaviour (Baum et al., 2014). Further, there are meta-analytic findings indicating that sleep variables such as sleep quality, sleep duration, and daytime alertness are all positively related to school outcomes (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010). Across the lifespan, the domains of attentional control and executive functioning are thought to be particularly sensitive to poor sleep (e.g. Bell-McGinty et al., 2004; Bucks, Olaithe, & Eastwood, 2013; Chee, 2004; Chee & Chuah, 2008; Gozal & Kheirandish-Gozal, 2007; Jackson et al., 2013; Jones & Harrison, 2001; Nilsson et al., 2005).

Given the link between sleep and daytime functioning, it is of great concern that as many as 40% of typically developing (TD) children experience sleep problems (Calhoun, Fernandez-Mendoza, Vgontzas, Liao, & Bixler, 2014; Hochadel, Frölich, Wiater, Lehmkuhl, & Fricke-Oerkermann, 2014). However, there is evidence that sleep problems are even more common in groups with neurodevelopmental disorders (NDD) such as intellectual disability (ID; Wiggs & Stores, 1996), attention deficit hyperactivity disorder (ADHD; Cassoff, Wiebe, & Gruber, 2012) and autism spectrum disorder (ASD). Sleep problems are a particular
issue for children with ASD, with prevalence estimates based on parental-reports indicating that up to almost 80% of children with ASD experience sleep problems throughout childhood (Aathira et al., 2017; Couturier et al., 2005; Goldman et al., 2009; Polimeni, Richdale, & Francis, 2005; Rzepecka et al., 2011; Souders et al., 2009; Wang et al., 2016). Indeed, direct comparisons suggest that children with ASD have more severe parent-reported sleep problems than both neurotypical children (e.g. Couturier et al., 2005; Giannotti et al., 2008; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Hoffman, Sweeney, Gilliam, & Lopez-Wagner, 2006; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Souders et al., 2009), and children with other NDD (e.g. Krakowiak et al., 2008).

ASD refers to a spectrum condition which is neurodevelopmental in nature and characterised by difficulty with social communication as well as restricted and repetitive behaviours and interests (RRBI; American Psychiatric Association, 2013). While the past decade has seen considerable improvements in early detection and intervention, children with ASD still present with heightened risk for many medical and psychiatric comorbidities (Bauman, 2010; Mattila et al., 2010; Salazar et al., 2015). As noted by Bauman (2010) these comorbidities, such as sleep disturbance, have a negative impact on the child’s development and behaviour. While, of course, they do not fully account for the core symptoms of ASD, there is evidence that comorbidities such as sleep problems are associated with increased RRBI (Gabriels, Cuccaro, Hill, Ivers, & Goldson, 2005), reduced verbal skills (Elia et al., 2000; Gabriels et al., 2005; Taylor, Schreck, & Mulick, 2012) and communication deficits (Schreck, Mulick, & Smith, 2004).

In addition to these links with the core features of ASD, others have focused on the link between sleep problems and daytime challenging, or externalising,
behaviours (Gabriels et al., 2005; Sikora, Johnson, Clemons, & Katz, 2012; Taylor et al., 2012). Externalising behaviours, such as aggression, hyperactivity and rule-breaking, provide one of the biggest challenges for families of children with ASD. The majority of children with ASD present with at least one challenging behaviour (Matson & Nebel-Schwalm, 2007), with some studies suggesting that the rate is as high as 94% (Jang, Dixon, Tarbox, & Granpeesheh, 2011; Matson, Wilkins, & Macken, 2009). These behaviours can lead to difficulty engaging with education (Lecavalier, Leone, & Wiltz, 2006) and are associated with significant stress within the family environment (Lecavalier et al., 2006; McStay, Dissanayake, Scheeren, Koot, & Begeer, 2014). Indeed, challenging behaviours are the strongest predictor of parental stress over and above other child and parent factors (Lecavalier et al., 2006). Moreover, these challenging behaviours do not tend to abate over time (Matson, Mahan, Hess, Fodstad, & Neal, 2010).

With regard to the link between externalising behaviours and sleep, a number of studies have found that children with ASD who were “good sleepers” (as reported by their parents) had significantly fewer externalising behaviours than those children with ASD who were “poor sleepers” (Adams, Matson, & Jang, 2014; Goldman et al., 2009, 2011; Sikora et al., 2012). Others have reported that sleep problems are associated with these behaviour problems (Fadini et al., 2015; Mazurek & Sohl, 2016), and can account for up to 32% of the variance in challenging daytime behaviours (Mazurek & Sohl, 2016). However, much of the past literature has relied heavily on parent-reported measures of both sleep and behaviour. For example, Goldman et al. (2009) assessed both sleep and behaviour in children with ASD using both parent-report and actigraphy, concluding that children with parent-reported poor sleep had more inattention and hyperactivity than those without parent-reported sleep
problems. However, while Goldman et al. (2009) employed actigraphy to assess sleep, no direct comparisons were made between actigraphy derived sleep problems and daytime behaviours.

Further evidence for a link between sleep and behaviour comes from clinical literature which has shown that externalising behaviours improve slightly following the successful treatment of sleep problems using exogenous melatonin (Paavonen et al., 2003). Further, and most recently, Cohen et al. (2018) found a strong temporal relationship between sleep and future externalising behaviours using real-time data in young people with low-functioning ASD who were living in residential facilities. The 24-hour care provided within these facilities allowed for regular (15 min - 30 min) carer recordings of sleep (i.e. observation of sleep/wake) and challenging daytime behaviours (i.e. aggression, self-injury, tantrums, etc). Here, behaviour was most accurately predicted with the use of approximately one week of prior sleep data, rather than just the previous night’s sleep (Cohen et al., 2018). Collectively these studies provide emerging evidence for a causal relationship between sleep and behaviour, indicating that poor sleep, in particular, habitual poor sleep, may lead to an increase in challenging daytime behaviours.

While the association between externalising behaviours and sleep in ASD is documented, there has been little focus on the reasons why poor sleep may lead to behavioural problems within this population. This is a vital question as it may help to inform interventions for both sleep and for managing challenging behaviours. As such, it is important to explore potential cognitive factors which may underlie the relationship between sleep and externalising behaviours in ASD. One such cognitive factor is executive control. Executive functions (EF) encompass a range of higher-order cognitive processes which control lower-order cognitive processes for the
purpose of goal-directed behaviour (Espy, 2004; Miller & Cohen, 2001; Miyake et al., 2000; Miyake & Friedman, 2012). The link between sleep and EF is well established in the paediatric sleep literature (Anderson, Storfer-Isser, Taylor, Rosen, Carol, & Redline, 2015; Bernier, Beauchamp, Bouvette-Turcot, Carlson, & Carrier, 2013; Edmed, Rossa, Kenardy, Anderson, & Smith, 2017; Moreau, Rouleau, & Morin, 2013). Furthermore, previous studies have also described a link between deficits in EF and challenging behaviours both in TD children and in those with ASD (Visser, Berger, Van Schrojenstein Lantman-De Valk, Prins, & Teunisse, 2015). However, despite considerable evidence of reduced executive control in ASD (Demetriou et al., 2017) beyond that explained by general intellectual functioning (Narzisi, Muratori, Calderoni, Fabbro, & Urgesi, 2013), to our knowledge, there are no published studies examining executive deficits in the context of poor sleep within this population.

While the relationships between sleep, executive dysfunction and challenging behaviours have not been considered together within populations with ASD, there is evidence of EF mediating the relationship between sleep and daytime outcomes in a number of TD paediatric populations. For example, in a large sample of healthy, TD pre-school aged children, one study found that the longitudinal relationship between sleep problems at three years of age and symptoms of hyperactivity at nine years of age was moderated by deficits in EF at four years of age (Kidwell, Hankey, Nelson, Espy, & Nelson, 2017). Similarly, Warren, Riggs and Pentz (2017) reported that the longitudinal relationship between poor sleep and future rule-breaking externalising behaviours (i.e. use of cigarettes and alcohol) was mediated through deficits with cognitive inhibitory functions (i.e. a component of EF).
Therefore, given that children with ASD are persistently found to exhibit high levels of executive dysfunction (Demetriou et al., 2017), and that deficits in EF appear to be related to challenging behaviours in ASD (Visser et al., 2015), it seems likely that the association between sleep and externalising, challenging behaviours in ASD may, at least in part, be accounted for by the impact that poor sleep has on EF. As such, the primary aim of the current study was twofold.

Firstly, the study sought to explore whether there was an association between sleep and externalising behaviours in ASD. It was hypothesised that children with ASD would have more challenging externalising behaviours than TD children and that these challenging behaviours would be related to measures of sleep.

Secondly, the study sought to determine whether the relationship between sleep and challenging behaviours (should one exist) would be mediated by differences in either parent-rated or performance-based measures of EF. Specifically, it was hypothesised that children with ASD would have poorer performance on tasks of EF and more problems noted on a parent-rated measure of EF. Within each group (ASD and TD), those with more sleep problems were anticipated to have poorer performance on tasks of EF and more problems noted on a parent-rated measure of EF. Finally, it was expected that executive deficits across parent-report and performance-based measures would mediate the relationship between sleep problems and externalising behaviours in both children with and without ASD.
5.3  Method

5.3.1 Participants

The current sample consisted of 72 children aged 6-12 years, either with (n = 35) or without (n = 37) a diagnosis of ASD. These participants were a subset of those detailed in Host, Maybery, Waters, & Whitehouse (2018). The majority of children with ASD were recruited through the Western Australian Autism Biological Registry (WAABR) at the Telethon Kids Institute (TKI) in Perth, Western Australia. All children had a clinical ASD diagnosis based on either the fourth or fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (American Psychiatric Association, 2000; 2013), which in Western Australia, is determined by consensus between a Paediatrician, Psychologist, and Speech-Language Pathologist (Glasson et al., 2008). WAABR participants had their diagnosis confirmed using the Autism Diagnostic Observation Schedule (ADOS-G; Lord et al. 2000; ADOS-2; Lord et al., 2012). Three participants with ASD were subsequently excluded due to insufficient questionnaire data (i.e. greater than 20% missing values as detailed below).

Recruitment of TD children was completed through local primary schools. One child with a parent-reported diagnosis of ASD was excluded from analysis. TD children were also screened for autistic-like traits using the children’s version of the Autism Spectrum Quotient (AQ-Child; Auyeung, Baron-Cohen, Wheelwright, & Allison, 2008). None of the children in the TD group scored above the cut-off for clinically elevated symptoms of ASD (i.e. none above 76; M = 49.80, SD = 12.63). Four TD participants were excluded due to insufficient questionnaire data.

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3 While children were aged 6-12 years when consented for the study and completion of the questionnaires, one child turned 13 just before completing participation, being aged 13 years 0 months at the time of wearing the actigraphy device. Given that questionnaire data was collected at 12 year, this age has been included for purpose of analysis.
5.3.2 Procedure

University of Western Australia Human Research Ethics Committee approved the current study (RA/4/1/6487). All parents were provided with a written information sheet regarding the study and signed consent to participate. Parent questionnaires, sleep diary and an actigraphy device were provided to families either at a school or a home visit. Use of the Actiwatch and a description of how to complete the sleep diary was provided to all parents and their participating child. Children were requested to wear the Actiwatch on their non-dominant wrist for seven days and nights (i.e. five weeknights and two weekend nights). At the end of the seven days, children completed computer-based EF tasks and the questionnaires and Actiwatches were collected. All data collection took place during the school semester. Parents were advised that the primary caregiver should complete the questionnaires, with all completed by the same caregiver.

5.3.3 Measures

Demographic Information

A clinical history form was used to collect information regarding ASD diagnosis, clinical comorbidities (e.g. ID and ADHD) and use of medication (prescription and complementary/alternative). Children completed the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler 1999) to gain an estimate of their full-scale intelligence quotient (FSIQ; missing for n = 2 of the ASD sample).

Sleep Problems

Subjective measure of sleep

The Children’s Sleep Habits Questionnaire-Revised (CSHQ-R; Host et al., 2017; derived from CSHQ; Owens et al., 2000) was employed to examine sleep. Items are rated on a three-point scale, noting the extent to which behaviours were present over the course of the preceding week (i.e. while the child was wearing the
Actiwatch). The CSHQ-R produces a total score through summing all 27 items, as well as five subscales, sleep initiation and maintenance (Sleep I and M), morning lethargy, co-sleeping, sleep-disordered breathing (SDB) and parasomnias. Across all scales, a higher score denotes greater sleep disturbance (four positively phrased items are reverse scored). The CSHQ-R scores could not be calculated for three participants (4%; TD = 2; ASD = 1) missing more than 20% of data. In instances where participants had fewer than 20% missing values, item scores were prorated using the item mean (<1% of item scores).

Objective measure of sleep

An Actiwatch-2 (Philips Respironics MiniMitter Actiwatch) was used to obtain an objective measure of sleep patterns. Consistent with guidelines (Fawkes et al., 2015; Meltzer, Montgomery-Downs, Insana, & Walsh, 2012) parents and their participating child completed a sleep diary over the study period. Bedtime and wake time were scored manually using data from the sleep diary (Fawkes et al., 2015; Meltzer et al., 2012). Each Actiwatch was configured to record in 60-second epochs. Actigraphy variables were scored using the Respironics software (detailed below). Actigraphy-derived sleep onset (SOT) was the clock time of the first of ten consecutive minutes of sleep following bedtime (as recorded in the sleep diary; D_BT) while sleep offset (SOffT) was the last of ten consecutive minutes of sleep prior to wake time (as recorded in the sleep diary; D_WT; Meltzer et al., 2012). Additional variables were: total sleep time (TST); wake after sleep onset (WASO); sleep efficiency (SE; TST/D_TIB); and sleep onset latency (SOL). All actigraphy data were scored according to strict protocol by the same researcher (A. Host), with scoring cross-checked for 20% of the participants by a second researcher (F. Waters).
Participants were required to have a minimum of five night’s valid actigraphy recordings to be included in the analysis (Acebo et al., 1999) with each of the final variables calculated from averaging across the available nights. Based on this criterion, valid actigraphy data were available for only 57 participants (ASD = 30; TD = 27). The mean number of nights for actigraphy recordings was 6.81 (SD = .48). Missing data were attributable to participants being unwilling or unable to tolerate wearing the device (n = 2) and Actiwatch malfunction (n = 5).

**Externalising Behaviours**

The Children’s Behaviour Checklist (CBCL; Achenbach & Rescorla, 2001) is a 113-item parent-rated questionnaire used to examine problem behaviours in children. Parents rate the presence of each problem behaviour over the past 6 months on a three-point scale (0 = “Not True” to 2 = “Very Often True”). Responses are summed to create eight subscales, as well as two composites reflecting total internalising problems and total externalising problems; these are then standardised and presented as T scores. The CBCL has strong evidence for both reliability and validity (Achenbach & Rescorla, 2001), and has been employed previously to assess behavioural problems in children with ASD (Hanratty et al., 2015; also see Patzold, Richdale, & Tonge, 1998; Richdale & Baglin, 2015). In the current study, the externalising behaviours composite (Ext B) was used as a measure of daytime challenging behaviours. Seven participants (9%) had at least one missing item score, so for these items scores were prorated using the item mean score across ASD and TD samples separately (<1% of item scores).

**Executive Functioning**

Two methods were employed to assess EF: 1) The Behavioural Rating Inventory of Executive Functioning (BRIEF; Gioia, Isquith, Guy, & Kenworthy,
selected tasks from the NIH Examiner Battery (NIHEx; Kramer et al., 2014).

**Parent-rated executive functioning**

The BRIEF is an 86-item parent-reported questionnaire designed to assess EF in children between five and 18 years of age. Parents rate the extent to which each behaviour was a problem for their child over the preceding six-month period. Responses are scored on a three-point scale (1 = “Never” to 3 = “Often”). Items are summed to create eight subscales as well as two composite index scores reflecting behavioural regulation (Behavioural Regulation Index, BRI; comprised of inhibit, shift and emotional control subscales) and metacognitive skills (Metacognition Index, MI; comprised of initiate, working memory, plan/organise, organisation of materials and monitor subscales). Raw scores are converted to age and sex-adjusted T scores with higher scores indicating poorer EF. The BRIEF has sound psychometric properties (internal consistency ranging from 0.80 to 0.98; Gioia, Isquith, Guy & Kenworthy, 2000) and has been found to have utility in a range of clinical populations, including children with ASD (Gioia et al. 2000, 2002). Of note, it has been identified as the most common measures of EF used in the ASD and EF literature (Demetriou et al., 2017). In the current study, BRI and MI index scores were used as parent-rated measures of EF.

**Performance-based executive functioning**

Three subtests from the NIHEx battery were employed as performance-based measures of EF: the Flanker task; the NBack task (1-Back); and the Continuous Performance Test (CPT).

The Flanker task is a measure of response inhibition. Participants were requested to look at a fixation point in the centre of the screen before being presented
with a row of five fish in which the target (centre) fish was either pointing in the same direction (congruent) or in a different direction (incongruent) to the other fish. Participants were requested to indicate whether the target fish was facing left or right by pressing the corresponding arrow key. The task included up to three practice blocks; the participant would advance to the test trials if at least 75% of the trials in one practice block were answered correctly. The test block consisted of 48 trials, with 50% congruent trials and 50% incongruent trials. The primary score from the Flanker task is calculated from the median reaction time on incongruent trials and the median accuracy on incongruent trials.

The 1-Back is a test of spatial working memory. Participants were shown a series of white squares in various locations on a black screen. They were then requested to indicate whether each square was presented in the same or different locations as the one previous. The tasks consisted of two practice blocks; the child advanced to the testing block if at least 7 of the 10 trials in one practice block were answered correctly. The testing blocks consisted of 30 trials: 10 “yes” and 20 “no” trials. The programme provides an adjusted overall 1-Back score (using d-prime) which accounts for both hits and false alarms.

The CPT is a measure of attentional control and response inhibition. Participants were presented with different shapes (all white) in the centre of a black screen and were requested to press a key (left arrow) when they saw the target image (e.g. a white five-pointed star). The task included three practice blocks; the child advanced to the testing block if at least 16 trials (out of 20) in one practice block were answered correctly. The test block consisted of 100 experimental trials, 80% of which displayed the target image. The measure used was the total number of trials...
for both target and non-target trials where the child’s response or non-response was correct (i.e. overall accuracy).

All tasks include practice items. A task was automatically discontinued if the participant was unable to complete the practice trials successfully. As non-completion was a reflection of inability to meet task demands, those who attempted, but were unable to complete the task were scored one unit below the next lowest score. Each of the tasks had a moderate to strong positive correlation with the others (Flanker and CPT: \( r (62) = 0.52, p < .001 \); Flanker and 1-Back: \( r (60) = 0.58, p < .001 \); CPT and 1-Back: \( r (60) = 0.46, p < .001 \)). Given these relationships, before data analysis, variables for the three tasks were converted to z-scores and summed to create an EF composite score (NIHExComp; Cronbach’s \( \alpha = 0.77 \)).

5.3.4 Statistical analyses

Actigraphy data were analysed using the Respironics Actiware Software (Version 6.0.2). As indicated by Meltzer, Walsh, Traylor and Westin (2012), the default scoring setting was used (medium sensitivity; 40 activity counts per epoch). Remaining analysis was completed using SPSS Version 22.0 for Windows. Data for each group (ASD and TD) were screened for outliers using a criterion of 3.29 SD from the mean for each variable. Outliers were dealt with using a Winsorizing approach, reducing the score to be one unit higher than the next highest value (Field, 2013).

Independent sample t-tests were computed to determine group differences across measures of sleep, challenging behaviour and EF. Subsequent to this, Pearson’s correlation analyses were employed to explore trends in the relationships

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6 A review of the strengths and limitations of this approach is included in the discussion.
between sleep, behaviour and EF. Where age was found to correlate with either behaviour or EF measures it was introduced as a covariate in testing relationships between sleep problems and the relevant day-time variable. Given that consistent relationships were found only between parent-reported variables, regression analyses were employed to examine whether perceptions of sleep problems (CSHQ-R total) predicted externalising behaviours (CBCL Ext B subscale) within either the TD or ASD groups. Finally, a mediation model was conducted to examine the role of executive behavioural regulation (i.e. the BRIEF BRI) in the relationship between poor sleep and challenging externalising behaviours. Mediation was computed in SPSS with a ‘PROCESS’ macro using the Preacher and Hayes (2004) indirect regression method. In light of the relatively small sample size, corrections to the alpha level for multiple tests were not applied since more conservative testing would have compromised power.

5.4 Results

5.4.1 Participant characteristics

Subsequent to exclusions detailed above, the final sample consisted of 32 children with ASD and 32 TD children. Demographic information for these participants (representing a subset of those detailed in Host, Maybery, Waters, & Whitehouse, 2018) are detailed in Table 5.1. There was no significant difference between the ASD and TD groups for age \( t(62) = 0.37, p = .714 \) (\( M = 119.31, SD = 17.29 \); \( M = 120.84, SD = 15.89 \) respectively). The groups did not differ on their estimated FSIQ, \( t(60) = 1.35, p = .182 \); with a mean score of 104.67 (SD = 17.82) for the ASD group and 109.88 (SD = 12.18) for the TD group. There were, however, significantly more males in the ASD sample \( \chi^2(1) = 8.33, p = .004 \). Given that over
90% of informants were the child’s mother, there was too little data from fathers to systematically examine possible differences between informants.

Twenty (63%) of the children with ASD had a diagnosis of AD, eight (25%) had a diagnosis of AS, and four (12%) had a diagnosis of PDD-NOS. Common comorbid medical and developmental disorders are also detailed in Table 5.1. No child in either group had been diagnosed with an ID and none had an estimated FSIQ less than 70 (range: 74-137 for ASD; 79-133 for TD).

The most frequently reported medical comorbidities included allergies, asthma, and gastrointestinal problems. As can be seen in Table 5.1, eleven (34%) children with ASD and two TD children were taking prescribed medications. For the majority of these participants, medication had been stable for a minimum of 6 months prior to enrolment in the study, excluding two children whose medications had been stable for six and four weeks respectively.

With regard to sleep, one TD child and two children with ASD had previously received a diagnosis of sleep apnoea and with subsequent tonsillectomy/adenotonsillectomy. Nine children with ASD (28%) were prescribed medication for sleep-related problems including melatonin (n = 8; dose 2-10mg), clonidine (n = 3), and amitriptyline (n = 1). Use of these medications had been stable on for at least six months prior to enrolment in the study. One TD child was taking over the counter melatonin (1mg) “when required”, though none were prescribed medication for sleep.
Table 5.1  
**Demographic Information for the Current Sample**

<table>
<thead>
<tr>
<th></th>
<th>TD</th>
<th>ASD</th>
</tr>
</thead>
<tbody>
<tr>
<td>n= 32</td>
<td>n= 32</td>
<td></td>
</tr>
<tr>
<td><strong>Sex (Male)</strong></td>
<td>19 (59.4)</td>
<td>29 (90.6)</td>
</tr>
<tr>
<td>First language</td>
<td></td>
<td></td>
</tr>
<tr>
<td>English</td>
<td>30 (93.8)</td>
<td>31 (96.9)</td>
</tr>
<tr>
<td>Other</td>
<td>2 (6.3)</td>
<td>1 (3.1)</td>
</tr>
<tr>
<td><strong>Ethnicity</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Caucasian</td>
<td>29 (90.6)</td>
<td>22 (68.8)</td>
</tr>
<tr>
<td>Other</td>
<td>3 (9.4)</td>
<td>6 (18.75)</td>
</tr>
<tr>
<td>Not specified</td>
<td>Nil</td>
<td>4 (12.5)</td>
</tr>
<tr>
<td><strong>ASD diagnosis (clinician diagnosed)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Autistic Disorder</td>
<td>Nil</td>
<td>20 (62.5)</td>
</tr>
<tr>
<td>Asperger Syndrome</td>
<td>Nil</td>
<td>8 (25.0)</td>
</tr>
<tr>
<td>PDD-NOS</td>
<td>Nil</td>
<td>4 (12.5)</td>
</tr>
<tr>
<td><strong>Comorbidities (clinician diagnosed)</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Intellectual Disability</td>
<td>Nil</td>
<td>Nil</td>
</tr>
<tr>
<td>Attention Deficit Hyperactivity Disorder</td>
<td>Nil</td>
<td>11 (34.4)</td>
</tr>
<tr>
<td>Specific Language Impairment</td>
<td>Nil</td>
<td>1 (3.1)</td>
</tr>
<tr>
<td>Specific Learning Disability</td>
<td>Nil</td>
<td>1 (3.1)</td>
</tr>
<tr>
<td>Intellectual Disability</td>
<td>Nil</td>
<td>Nil</td>
</tr>
<tr>
<td>Epilepsy</td>
<td>Nil</td>
<td>Nil</td>
</tr>
<tr>
<td>Gastrointestinal problems</td>
<td>Nil</td>
<td>3 (9.4)</td>
</tr>
<tr>
<td>Asthma</td>
<td>Nil</td>
<td>3 (9.4)</td>
</tr>
<tr>
<td>Allergies</td>
<td>6 (18.8)</td>
<td>7 (21.9)</td>
</tr>
<tr>
<td>Other medical diagnosis</td>
<td>2 (6.3)</td>
<td>3 (9.4)</td>
</tr>
<tr>
<td><strong>Past diagnosis of sleep problem</strong></td>
<td>1 (3.1)</td>
<td>2 (6.3)</td>
</tr>
<tr>
<td>Prescribed medications</td>
<td></td>
<td></td>
</tr>
<tr>
<td>For sleep problem</td>
<td>Nil</td>
<td>9 (28.1)</td>
</tr>
<tr>
<td>For non-sleep related diagnosis</td>
<td>3 (9.4)</td>
<td>11 (34.4)</td>
</tr>
<tr>
<td><strong>Maternal education</strong></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Completed Year 10 Cert</td>
<td>Nil</td>
<td>2 (6.2)</td>
</tr>
<tr>
<td>Completed Year 12 Cert</td>
<td>9 (28.1)</td>
<td>7 (21.9)</td>
</tr>
<tr>
<td>Undergraduate degree</td>
<td>14 (43.8)</td>
<td>12 (37.5)</td>
</tr>
<tr>
<td>Postgraduate degree</td>
<td>9 (28.1)</td>
<td>3 (9.4)</td>
</tr>
<tr>
<td>Not specified</td>
<td>Nil</td>
<td>8 (25)</td>
</tr>
<tr>
<td><strong>Abbreviated FSIQ</strong></td>
<td>109.88 (12.18)</td>
<td>104.67 (17.82)</td>
</tr>
</tbody>
</table>

*Note.* PDD-NOS = Pervasive Developmental Disorder-Not Otherwise Specified; FSIQ = Full-Scale Intelligent Quotient; FSIQ data unavailable for 2 participants.

Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder? 182
5.4.2 Effects of age and sex and comorbidities

As can be seen in Table 5.2, increasing age was associated with fewer parent-reported sleep problems for the TD group, but not the ASD group. Increasing age was also associated with later sleep onset time and reduced total sleep for both the ASD and TD groups. Finally, older age was also associated with stronger performance on performance-based EF tasks for both the TD and ASD groups. No other variables were related to age in either group, however, given the number of variables associated with age, further analysis employed age as a covariate.

Within the TD group, none of the sleep or behaviour variables were related to the diagnosis of co-morbid developmental or medical conditions or the use of medication (point-biserial correlations for dichotomous variables). Within the ASD sample, use of sleep medication was related to both SOT \( r (29) = -.51, p = .004 \) and SOL and \( r (29) = -.39, p = .038 \); while the presences of a comorbid developmental disorder was related to SOffT \( r (29) = -.54, p = .003 \). As such, use of sleep medication was controlled for in further analysis examining SOT and SOL variables. Analysis did not control for presence of a comorbid developmental condition given known high concordance between sleep and these diagnoses (e.g. ADHD problems) with likely bidirectional relationships.

There were no significant sex differences on any of the sleep, behaviour, or EF measures (parent-rated or performance-based) except for sleep onset in the TD group, and sleep offset for both groups (Table 5.3). Within the TD group, females fell asleep and woke earlier; while within the ASD group, females woke later than males. Given that groups were not matched for sex and given that a select number of sex differences in sleep patterns emerged in both the ASD and TD samples (see Table 5.3), all observed differences between ASD and TD groups were also
Table 5.2
Summary of Correlations Between Measures of Sleep, Externalising Behaviours and Executive Functioning for the ASD Group (above the diagonal) and TD Group (below the diagonal)

<table>
<thead>
<tr>
<th></th>
<th>Age</th>
<th>CSHQ</th>
<th>SOT</th>
<th>SOffT</th>
<th>TST</th>
<th>WASO</th>
<th>SE</th>
<th>SOL</th>
<th>Ext B</th>
<th>BRI</th>
<th>MI</th>
<th>NIHEx</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>-</td>
<td>-0.049</td>
<td>0.498**</td>
<td>0.253</td>
<td>-0.511**</td>
<td>0.116</td>
<td>-0.352</td>
<td>0.136</td>
<td>-0.160</td>
<td>-0.004</td>
<td>-0.075</td>
<td>0.375*</td>
</tr>
<tr>
<td>2</td>
<td>CSHQ-R</td>
<td>-0.353*</td>
<td>-</td>
<td>0.217</td>
<td>0.294</td>
<td>0.041</td>
<td>-0.103</td>
<td>0.136</td>
<td>-0.212</td>
<td>0.446*</td>
<td>0.396*</td>
<td>0.139</td>
</tr>
<tr>
<td>3</td>
<td>SOT</td>
<td>0.394*</td>
<td>0.021</td>
<td>-</td>
<td>0.734**</td>
<td>-0.639**</td>
<td>-0.048</td>
<td>-0.488**</td>
<td>0.608**</td>
<td>0.151</td>
<td>0.126</td>
<td>0.109</td>
</tr>
<tr>
<td>4</td>
<td>SOffT</td>
<td>-0.018</td>
<td>0.268</td>
<td>0.522**</td>
<td>-</td>
<td>-0.087</td>
<td>0.153</td>
<td>-0.301</td>
<td>0.482**</td>
<td>0.316</td>
<td>0.187</td>
<td>0.207</td>
</tr>
<tr>
<td>5</td>
<td>TST</td>
<td>-0.453*</td>
<td>0.210</td>
<td>-0.571**</td>
<td>0.351</td>
<td>-</td>
<td>-0.315</td>
<td>0.751**</td>
<td>-0.347</td>
<td>0.014</td>
<td>-0.023</td>
<td>-0.037</td>
</tr>
<tr>
<td>6</td>
<td>WASO</td>
<td>0.091</td>
<td>-0.133</td>
<td>-0.287</td>
<td>-0.567**</td>
<td>-0.461*</td>
<td>-</td>
<td>-0.675**</td>
<td>-0.025</td>
<td>0.170</td>
<td>0.052</td>
<td>0.172</td>
</tr>
<tr>
<td>7</td>
<td>SE</td>
<td>-0.228</td>
<td>0.215</td>
<td>-0.335</td>
<td>0.383*</td>
<td>0.840**</td>
<td>-0.625**</td>
<td>-</td>
<td>-0.559**</td>
<td>-0.162</td>
<td>-0.079</td>
<td>-0.238</td>
</tr>
<tr>
<td>8</td>
<td>SOL</td>
<td>0.275</td>
<td>-0.047</td>
<td>0.632**</td>
<td>0.473*</td>
<td>-0.242</td>
<td>-0.240</td>
<td>-0.379</td>
<td>-</td>
<td>0.135</td>
<td>0.065</td>
<td>0.174</td>
</tr>
<tr>
<td>9</td>
<td>Ext B (CBCL)</td>
<td>0.015</td>
<td>0.413*</td>
<td>-0.053</td>
<td>0.202</td>
<td>0.312</td>
<td>-0.323</td>
<td>0.242</td>
<td>0.274</td>
<td>-</td>
<td>0.810**</td>
<td>0.452*</td>
</tr>
<tr>
<td>10</td>
<td>BRI (BRIEF)</td>
<td>-0.033</td>
<td>0.494**</td>
<td>-0.150</td>
<td>0.100</td>
<td>0.217</td>
<td>0.019</td>
<td>0.122</td>
<td>0.143</td>
<td>0.699**</td>
<td>-</td>
<td>0.483**</td>
</tr>
<tr>
<td>11</td>
<td>MI (BRIEF)</td>
<td>-0.084</td>
<td>0.355*</td>
<td>-0.116</td>
<td>0.059</td>
<td>0.073</td>
<td>0.235</td>
<td>0.054</td>
<td>0.010</td>
<td>0.479**</td>
<td>0.684**</td>
<td>-</td>
</tr>
<tr>
<td>12</td>
<td>NIHExComp</td>
<td>0.427*</td>
<td>0.043</td>
<td>0.158</td>
<td>0.060</td>
<td>-0.071</td>
<td>-0.142</td>
<td>0.136</td>
<td>0.014</td>
<td>0.195</td>
<td>0.036</td>
<td>-0.081</td>
</tr>
</tbody>
</table>

Note. CSHQ = Total score on the Children’s Sleep Habits Questionnaire-Revised; Actigraphy variables: SOT = Sleep onset Time; SOffT = Sleep Offset Time; TST = Total Sleep Time; WASO = Wake After Sleep Onset; SE= Sleep Efficiency; SOL= Sleep Onset Latency. Ext B (CBCL) = Externalising Behaviours index from the Child Behaviour Checklist; BRI (BRIEF) = Behavioural Regulation Index from the Behavioural Rating Inventory of Executive Functioning; MI = Metacognition Index from the BRIEF; NIHExComp = Composite score calculated from the NIH Examiner Battery

*p < .05; **p < .01.
## Table 5.3

*Means (SD) and Independent Sample T-Tests Comparing Scores on Measures of Sleep, Externalising Behaviours and Executive Functioning Across Males and Females for Both the ASD and TD Groups*

<table>
<thead>
<tr>
<th></th>
<th>Male</th>
<th>Female</th>
<th>t</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>TD group</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CSHQ-R</td>
<td>33.21</td>
<td>33.56</td>
<td>-.19</td>
<td>.850</td>
</tr>
<tr>
<td>SOT</td>
<td>21:47:30</td>
<td>21:06:31</td>
<td>2.40</td>
<td>.024*</td>
</tr>
<tr>
<td>SOffT</td>
<td>6:53:20</td>
<td>6:19:37</td>
<td>3.20</td>
<td>.004**</td>
</tr>
<tr>
<td>TST</td>
<td>484.43</td>
<td>481.92</td>
<td>.14</td>
<td>.892</td>
</tr>
<tr>
<td>WASO</td>
<td>61.39</td>
<td>71.18</td>
<td>-1.50</td>
<td>.145</td>
</tr>
<tr>
<td>SE</td>
<td>80.46</td>
<td>78.94</td>
<td>.65</td>
<td>.525</td>
</tr>
<tr>
<td>SOL</td>
<td>40.75</td>
<td>28.82</td>
<td>1.13</td>
<td>.267</td>
</tr>
<tr>
<td>Ext B (CBCL)</td>
<td>51.84</td>
<td>48.77</td>
<td>.85</td>
<td>.403</td>
</tr>
<tr>
<td>BRI (BRIEF)</td>
<td>50.42</td>
<td>48.77</td>
<td>.44</td>
<td>.662</td>
</tr>
<tr>
<td>MI (BRIEF)</td>
<td>49.79</td>
<td>53.69</td>
<td>-8.9</td>
<td>.380</td>
</tr>
<tr>
<td>NIHExComp</td>
<td>1.58</td>
<td>.87</td>
<td>1.49</td>
<td>.146</td>
</tr>
<tr>
<td><strong>ASD group</strong></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>CSHQ-R</td>
<td>37.86</td>
<td>43.00</td>
<td>-1.11</td>
<td>.274</td>
</tr>
<tr>
<td>SOffT</td>
<td>6:07:05</td>
<td>7:15:59</td>
<td>-2.58</td>
<td>.015*</td>
</tr>
<tr>
<td>TST</td>
<td>465.26</td>
<td>493.80</td>
<td>-1.05</td>
<td>.304</td>
</tr>
<tr>
<td>WASO</td>
<td>66.04</td>
<td>80.08</td>
<td>-.94</td>
<td>.356</td>
</tr>
<tr>
<td>SE</td>
<td>78.59</td>
<td>78.17</td>
<td>.10</td>
<td>.924</td>
</tr>
<tr>
<td>SOL</td>
<td>35.95</td>
<td>41.34</td>
<td>-.44</td>
<td>.664</td>
</tr>
<tr>
<td>Ext B (CBCL)</td>
<td>57.38</td>
<td>59.33</td>
<td>-.24</td>
<td>.815</td>
</tr>
<tr>
<td>BRI (BRIEF)</td>
<td>69.03</td>
<td>69.33</td>
<td>-.05</td>
<td>.964</td>
</tr>
<tr>
<td>MI (BRIEF)</td>
<td>69.14</td>
<td>69.33</td>
<td>.03</td>
<td>.974</td>
</tr>
<tr>
<td>NIHExComp</td>
<td>-1.19</td>
<td>-1.81</td>
<td>.41</td>
<td>.686</td>
</tr>
</tbody>
</table>

*Note.* CSHQ = Total score on the Children’s Sleep Habits Questionnaire-Revised; Actigraphy variables: SOT = Sleep onset Time; SOffT = Sleep Offset Time; TST = Total Sleep Time; WASO = Wake After Sleep Onset; SE = Sleep Efficiency; SOL = Sleep Onset Latency. Ext B (CBCL) = Externalising Behaviours index from the Child Behaviour Checklist; BRI (BRIEF) = Behavioural Regulation Index from the Behavioural Rating Inventory of Executive Functioning; MI = Metacognition Index from the BRIEF; NIHExComp = Composite score calculated from the NIH Examiner Battery

* $p < .05$; ** $p < .01$. 

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Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?
examined using a subsample comprised of only male participants (ASD = 29; TD = 19).

5.4.3 ASD and TD Group Differences

Compared to TD participants, children with ASD had significantly higher scores on the CSHQ (indicating more sleep problems), as well as earlier SOFFT (i.e. wake up time as recorded by actigraphy) in the morning (Table 5.4). Children with ASD were also rated as having more externalising behaviours on the CBCL and more difficulty applying executive skills in day-to-day life (BRI and MI of the BRIEF; Table 5.4). Consistent with parental reports, compared to TD participants, children with ASD also had significantly poorer performance on the EF tasks (NIHExComp; Table 5.4).

When assessing group differences across male participants only, similar patterns emerged, except that there was no longer a significant group difference for CBCL Ext B \( t (43) = 1.52, p = .135 \) (ASD: M = 57.38, SD = 13.44 and TD: M = 51.84, SD = 9.80).

5.4.4 Sleep and externalising behaviours within groups

As can be seen in Table 5.2, there were significant, moderate, positive correlations between the CSHQ and Ext B scores for both the ASD and TD groups, which were unchanged after controlling for age; ASD: \( r (26) = .44, p = .018 \) and TD: \( r (29) = .45, p = .012 \). This indicates that across both groups, those children who were reported by their parent to have more sleep problems were also reported by their parents to exhibit more challenging, externalising behaviours, even after controlling for the relationship that each of these variables had with age. With regard to objective sleep data, as can be seen in Table 5.2, no associations were found between any of the actigraphy variables and Ext B for either group. However, after
controlling for both age and sleep medication in the ASD group, Ext B scores were related to SOT scores, \( r(23) = .50, p = .012 \).

Table 5.4

Means (SD) and Independent Sample T-Tests Comparing Scores for Sleep, Externalising Behaviours and Executive Functioning between Children with and without ASD

<table>
<thead>
<tr>
<th></th>
<th>ASD M (SD)</th>
<th></th>
<th>TD M (SD)</th>
<th></th>
<th>( t )</th>
<th>( p )</th>
</tr>
</thead>
<tbody>
<tr>
<td>CSHQ-R</td>
<td>38.34 (7.64)</td>
<td>SOT</td>
<td>21:18:08 (1:04:38)</td>
<td>21:31:24 (0:47:53)</td>
<td>-0.88</td>
<td>.381</td>
</tr>
<tr>
<td>SOT</td>
<td>21:18:08 (1:04:38)</td>
<td>SOffT</td>
<td>6:13:59 (0:47:55)</td>
<td>6:40:05 (0:31:30)</td>
<td>-0.43</td>
<td>.695</td>
</tr>
<tr>
<td>TST</td>
<td>468.12 (44.82)</td>
<td>WASO</td>
<td>67.44 (24.55)</td>
<td>65.24 (17.24)</td>
<td>.39</td>
<td>.786</td>
</tr>
<tr>
<td>SE</td>
<td>78.55 (7.09)</td>
<td></td>
<td>79.90 (5.88)</td>
<td>7.78</td>
<td>.441</td>
<td></td>
</tr>
<tr>
<td>SOL</td>
<td>36.49 (19.90)</td>
<td></td>
<td>36.06 (27.32)</td>
<td>.07</td>
<td>.946</td>
<td></td>
</tr>
<tr>
<td>Ext B (CBCL)</td>
<td>57.59 (13.29)</td>
<td>Ext B (CBCL)</td>
<td>50.59 (10.03)</td>
<td>2.33</td>
<td>.023</td>
<td></td>
</tr>
<tr>
<td>BRI (BRIEF)</td>
<td>69.06 (10.73)</td>
<td>MI (BRIEF)</td>
<td>49.75 (12.26)</td>
<td>7.36</td>
<td>&lt;.001 ***</td>
<td></td>
</tr>
<tr>
<td>NIHExComp</td>
<td>-1.25 (2.45)</td>
<td>NIHExComp</td>
<td>1.31 (3.33)</td>
<td>-5.08</td>
<td>&lt;.001 ***</td>
<td></td>
</tr>
</tbody>
</table>

Note. CSHQ = Total score on the Children’s Sleep Habits Questionnaire-Revised; Actigraphy variables: SOT = Sleep onset Time; SOffT = Sleep Offset Time; TST = Total Sleep Time; WASO = Wake After Sleep Onset; SE = Sleep Efficiency; SOL = Sleep Onset Latency. Ext B (CBCL) = Externalising Behaviours index from the Child Behaviour Checklist; BRI (BRIEF) = Behavioural Regulation Index from the Behavioural Rating Inventory of Executive Functioning; MI = Metacognition Index from the BRIEF; NIHExComp = Composite score calculated from the NIH Examiner Battery
* \( p < .05 \); ** \( p < .01 \).

5.4.5 Sleep and executive functioning within groups

Within the TD group, higher scores on the CSHQ were significantly correlated with higher scores on both the BRI and the MI (Table 5.2); however, after controlling for age, the relationship between sleep and MI no longer reached significance; \( r(29) = .35, p = .054 \). This suggest that there was a trend towards higher levels of parent-rated sleep problems being associated with greater difficulty in their
children applying executive skills in day-to-day life; especially behavioural regulation skills.

Within the ASD group, there was a significant, moderate, positive correlation between sleep problems and the BRI subscale of the BRIEF, which remained after controlling for age, $r (29) = .40, p = .027$; however, there was no relationship between CSHQ and MI subscale scores (Table 5.2). Thus, within the ASD group, parent-rated sleep was associated with parent-rated executive difficulties in regulating behaviour, but not associated with parent-rated difficulties with higher-order metacognitive skills.

With regard to EF, there was no relationship between parent-reported sleep problems and performance-based measures of EF (NIHExComp) for children with or without ASD (Table 5.2). Further, there were no significant relationships between actigraphy-based sleep variables and any of the EF measures (either parent-rated or performance-based).

### 5.4.6 Executive functioning and externalising behaviours

As can be seen in Table 5.2, there were significant, moderate to strong, positive correlations between Ext B and both of the BRIEF index scales (BRI and MI) for the ASD and TD groups which remained significant after controlling for age; for the BRI, ASD: $r (26) = .82, p < .001$ and TD: $r (29) = .70, p < .001$ and for the MI, ASD: $r (26) = .45, p = .017$ and TD: $r (29) = .48, p = .006$. That is, those children who were reported by their parent to have difficulty applying metacognitive skills (MI) or regulating behaviour (BRI) in day-to-day life also exhibited more externalising behaviours (Ext B). However, as with sleep, there was no relationship between Ext B and the performance-based measure of EF for either group (Table 5.2).
5.4.7 Relationship between measures of executive functioning

Within both the ASD and TD groups there were significant, strong, positive correlations between the BRI and MI scales of the BRIEF which remained significant after controlling for age, ASD: \( r (29) = .48, p = .006 \) and TD: \( r (29) = .68, p < .001 \). However, these index scores were not correlated with performance-based EF (NIHExComp; Table 5.2). That is, while parent ratings of the child’s ability to apply metacognitive and behavioural regulation skills in day-to-day life were associated, these skills were not associated with the child’s performance on computer-based executive tasks\(^7\).

5.4.8 Sleep, executive functioning, and externalising behaviours

With regard to the relationship between parent-reported sleep and externalising behaviours, regression analyses were employed to examine whether sleep problems (CSHQ-R total) predicted externalising behaviours (Ext B subscale) within either the TD or ASD groups. As age was associated with CSHQ-R total for the TD group, it was included as a control in the first step of the regression model. The model did not control for co-morbid developmental or medical conditions, or the use of medication as these were not related to scores on the CSHQ-R or Ext B.

Within the TD group, after controlling for age, sleep still predicted externalising behaviours, independently explaining a significant proportion of the variance; \( \Delta R^2 = .20, F (2, 29) = 7.25, p = .012 \). Within the ASD group, after controlling for age, sleep still predicted externalising behaviours, \( \Delta R^2 = .192, F (1, 26) = 6.36, p = .018 \).

\(^7\) Given that the measurement of EF is not the focus of the current chapter, or thesis more broadly, inconsistency across results from questionnaire-based and task-based measures of EF not discussed in detail. However, it is noted that this is a common finding across the literature, with questionnaire-based measures generally considered to have greater sensitivity to other variables. For example, see Albein-Urios et al (2018).
Given that the BRI was the only measure of EF in the current study to correlate with both sleep (CSHQ-R) and externalising behaviours (Ext B), a mediation model was conducted to examine the role of executive behavioural regulation (i.e. the BRI) in the relationship between poor sleep and challenging externalising behaviours (Table 5.5). Again, age, though not comorbidities nor medications, was included as a control variable.

Table 5.5
Mediation Model and Standardised Beta Coefficients for Predicted Relationships Between Sleep, Executive Functioning and Externalising Behaviours in ASD and TD Groups After Controlling for Age

<table>
<thead>
<tr>
<th></th>
<th>c</th>
<th>a</th>
<th>b</th>
<th>c'</th>
<th>a'b</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>TD</strong></td>
<td>0.48 *</td>
<td>0.55 **</td>
<td>0.64 **</td>
<td>0.13 ns</td>
<td>0.35 *</td>
</tr>
<tr>
<td><strong>ASD</strong></td>
<td>0.44 *</td>
<td>0.38 *</td>
<td>0.75 **</td>
<td>0.16 ns</td>
<td>0.28 ns</td>
</tr>
</tbody>
</table>

Note. a = path from sleep problems (predictor) to executive functioning (EF; mediator); b = path from EF (mediator) to externalising behaviour (dependent variable), controlling for sleep problems; c = direct path from sleep problems to externalising behaviour; ab = indirect path (via EF) from sleep problems to externalising behaviour; c' = path from sleep problems to externalising behaviour, controlling for EF

* p < .05; ** p < .01.; ns = non-significant.

Within the TD group, after controlling for age, the model explained 50% of the variance in externalising behaviours, $F (3, 28) = 9.32, p < .001$. Importantly, the
indirect pathway was significant (path ab; bootstrapped indirect path = .35; CI 0.07 to 0.64). That is, in TD children, reduced behavioural regulation skills explained the relationship between sleep and challenging, externalising behaviours.

Within the ASD group, the model explained 69% of the variance in externalising behaviours, \( F(2, 26) = 27.43, p < .001 \). That is, as with the TD group, sleep was a significant predictor of externalising behaviours. However, the mediated path did not meet the conventional criterion for significance (path ab; bootstrapped indirect path = .28; CI -0.04 to 0.47). That is, analysis for the ASD group yielded similar results to the TD group, except for the confidence interval for the indirect path which failed to exclude zero.

5.5 Discussion

The primary aim of the current study was to explore the relationships between sleep and externalising behaviours in ASD and to determine whether or not the relationship was mediated by differences in either parent-rated or performance-based measures of EF. The findings showed that: (i) compared to TD children, those with ASD had higher parent-rated sleep problems (CSHQ) and challenging behaviours (CBCL Ext B), as well as poorer performance on parent-rated and performance-based measures of EF (BRIEF MI and BRI, NIHExComp); (ii) within each group, parental reports of sleep problems, challenging behaviours, and behavioural regulation EF skills (BRI) were all related; however, after controlling for age, neither meta-cognitive EF skills (MI) nor performance-based EF (NIHExComp) were related to parent-rated sleep; (iii) sleep assessed using actigraphy was not related to any EF measure (parent-rated and performance-based) nor to challenging behaviours; (iv) reduced behavioural EF (BRI) explained the relationship between
parent-reported sleep and externalising behaviours in the TD group; however, the comparable effect fell short of reaching significance in the ASD group.

Firstly, as anticipated based on substantial previous research (Couturier et al., 2005; Giannotti et al., 2008; Hodge et al., 2014; Hoffman et al., 2006; May et al., 2015; Souders et al., 2009), those with ASD had higher parent-rated sleep problems compared to TD children. Despite this, when measured objectively using actigraphy, the groups differed only on waking time (SOffT), with the ASD group waking approximately 27 mins earlier than the TD group. This is consistent with data (from an overlapping sample) presented in Host et al. (2018) where findings are discussed in more detail. Broadly, it is noteworthy that even in a sample with clear parent-reported sleep difficulties, objective assessment of sleep found little evidence of differences in sleep parameters.

Also consistent with past literature, relative to the TD children, the current ASD sample presented with poorer performance on both parent-rated and performance-based measures of EF (Demetriou et al., 2017) and more parent-rated challenging behaviours (Jang et al., 2011; Matson & Nebel-Schwalm, 2007; Matson et al., 2009). However, when comparing only males within the samples, this difference for challenging behaviours was no longer significant.

With regard to the relationships between these measures, as anticipated, increased challenging behaviours were related to poorer parent-rated EF (metacognition and behavioural regulation). Further, parent-rated sleep problems were associated with both challenging externalising behaviours and executive dysfunction (behavioural regulation). However, sleep was not associated with parent-related meta-cognitive EF skills (after controlling for age in the TD group) or performance-based EF. Moreover, there were no significant relationships between
objective measures of sleep and externalising behaviour, or any measure of EF. For reasons enumerated below, these findings of interest in the context of existing literature suggesting relationships between both sleep and challenging behaviours in ASD groups (Adams et al., 2014; Fadini et al., 2015; Goldman et al., 2009, 2011; Mazurek & Sohl, 2016; Sikora et al., 2012), and sleep and EF in TD samples (Anderson et al., 2015; Bernier et al., 2013; Edmed et al., 2017; Moreau et al., 2013).

First, with regard to the relationship between sleep and challenging externalising behaviours, as noted previously, the current study differed from past literature in that direct comparisons were made between actigraphy-assessed sleep and daytime behaviours, rather than parent reports of both (e.g. Fadini et al., 2015; Goldman et al., 2011; Mazurek & Sohl, 2016; Sikora et al., 2012). Interestingly, within the ASD sample, after controlling for both age and sleep medication, challenging behaviours were related to sleep onset, with children with more challenging behaviours falling asleep later than those with lower levels of parent-reported challenging behaviours. Challenging behaviours were, however, not related to differences in other actigraphy-based sleep parameters (i.e. TST, SE, WASO, etc.).

Second, the study provided the first examination of EF in the context of sleep in ASD sample. While the current finding of a relationship between parent-reported sleep and EF is consistent with the TD literature in this area (e.g. Anderson et al., 2015; Bernier et al., 2013; Edmed et al., 2017; Moreau et al., 2013), again the lack of relationship between objective assessment of sleep and either parent-rated or performance-based EF is unexpected. As noted, past studies in TD groups have found associations between objectively assessed poor sleep and performance-based measures of complex attention and EF (e.g. Vriend, Davidson, Shaffner, Corkum, &...
Further, there are clear theoretical reasons as to why reduced sleep may impact upon EF (e.g. Beebe & Gozal, 2002; Doran, Van Dongen, & Dinges, 2001).

One consideration which may impact results in the current study was that very few children in either group had markedly reduced sleep duration as defined by Hirshkowitz et al. (2015; <7 hours). Given that deficits in neurocognitive functioning are generally reported in the context of sleep deprivation or restriction (de Bruin, van Run, Staaks, & Meijer, 2017; Lowe, Safati, & Hall, 2017), the fact that the majority of children slept for approximately 7.8 (ASD) to 8 hours (TD) would suggest that sleep parameters may not have been sufficiently impaired in either group to contribute to executive dysfunction. It may be that over a certain threshold of sleep (i.e. 7 hours), other factors (i.e. other cognitive skills) are more important in executive control. However, while Hirshkowitz et al. (2015) detailed that less than 7 hours was problematic for children aged 6-12 years, guidelines recommend ideal sleep duration of 9-11 hours, with 7 hours recorded as the lower limit of what “may be appropriate”. This indicates that while few in the current samples were averaging less sleep that the absolute minimum duration, most were sleeping less than recommended. Consistent with this suggestion, in a sample of 8-12 year old TD children (N = 32), Vriend et al. (2013) found that sleep restriction of just one hour, leading to average sleep duration of approximately 8 hours, was associated with performance decrements on aspects of EF (e.g. working memory, complex attention).

Alternatively, it could be that the current measure of EF, being composite measures across a number of EF domains, was not sensitive enough to detect performance decrements. While the decision was made to collapse performance across EF tasks into a single composite (to avoid increased type 1 error associated...
with multiple comparisons in a small sample), it may be prudent for future studies to examine performance on these EF tasks separately in larger samples. Moreover, while none of the children in either group (ASD or TD) presented with an ID, future studies may consider more rigorous control of other cognitive variables such as IQ.

Given that relationships did exist between parent-rated sleep, challenging behaviours and executive behavioural regulation, mediation was computed with these variables only. As anticipated, reduced behavioural regulation skills explained the relationship between sleep and externalising behaviours in the TD group; however, results fell short of reaching significance in the ASD group.

As noted previously, the relationship between sleep, behaviour and EF was anticipated due to findings of EF mediating the relationship between sleep and daytime behaviour across TD paediatric populations (e.g. Kidwell et al., 2017; Warren, Riggs, & Pentz, 2017). While the mediated effect did not reach significance in the ASD group, there was a similar pattern of relationships between sleep, behavioural regulation and externalising behaviour across the two groups. At the outset, this was conceptualised in the context of known pathways through which poor sleep is likely to impact upon cognition, and in turn, behavioural control. For example, changes in neurological activity in the prefrontal cortex (Horne 1993; Yoo et al. 2007), which is implicated in executive control, may lead to increased challenging behaviours. Interestingly, however, relationships were mainly noted for parent-rated measures rather than objective measures of sleep. Given that changes in most sleep parameters (i.e. TST, SE, WASO, etc.) were not related to daytime behaviours or EF, alternative explanations for the association between parent-rated sleep and behaviours must be considered.
In this context, it is pertinent to note that the relationships between sleep and daytime behaviour in childhood are complex and likely bidirectional (see for review Gregory & Sadeh 2012). The bidirectional nature of these relationships has also been noted within the field of ASD specifically. In a comprehensive review, Hollway and Aman (2011) suggested a theoretical model accounting for sleep problems in ASD. The framework detailed a bidirectional relationship between sleep and externalising behaviours, which both impact on, and are impacted by, over-arousal and insomnia (Hollway & Aman, 2011). In light of this, it is possible that increased challenging behaviours may lead to behavioural sleep problems which are captured by the CSHQ-R, such as bedtime refusal or bed-sharing. This is consistent with the current finding of a relationship between challenging behaviours and sleep onset time within the ASD group after controlling for age. This may indicate that challenging behaviours throughout the day lead to difficulty settling at night (i.e. increased arousal, longer SOL). However, it is also noted that past research directly examining the relationships between sub-types of sleep problems and behaviour found that parent-reported bedtime resistance was not significantly related to other daytime behaviour problems (Mazurek & Sohl, 2016). Rather, the authors found that poor sleep maintenance (characterised by parent-reported night-time awakenings) was most consistently associated with daytime behaviour problems (Mazurek & Sohl, 2016), adding support to the initial suggestion that poor sleep may contribute to challenging behaviours, rather than the reverse.

With regard to the current sample, it is acknowledged that the data described are cross-sectional in nature, and as such, causality cannot be determined. It will be pertinent for future research to explore the longitudinal predictive associations between sleep, behaviour and EF in more detail to delineate the predictors and
outcomes of both sleep parameters and parental perceptions of sleep. Further, while there is some evidence that externalising behaviours may improve slightly following successful treatment of sleep problems (Paavonen et al., 2003), additional treatment research, examining the potential cognitive and behavioural benefits of improved sleep parameters, may also help to shed light on these complex relationships.

Overall, the current study provides preliminary evidence that while parental reports of sleep problems, challenging behaviours, and behavioural regulation EF skills were related, these variables do not appear to be related when measured objectively. When examining parent report in more detail, there is evidence that reduced behavioural regulation skills can explain the relationship between sleep and externalising behaviours for the TD children; however, results fell short of reaching significance in the ASD group.
Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?

5.6 References


Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?


Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?

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Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?


Chapter 5: Do Executive Functioning Skills Mediate the Relationship Between Sleep Problems and Daytime Externalising Behaviours in Children With and Without Autism Spectrum Disorder?

Pediatrics, 102(3), 616–620. doi:10.1542/peds.102.3.616


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Nilsson, J. P., Soderstrom, M., Karlsson, A. U., Lekander, M., Akerstedt, T.,


Chapter 6: General Discussion

Across the paediatric sleep literature, there are consistent reports of sleep problems in children with autism spectrum disorder (ASD). Within this group, poor sleep has previously been identified as being associated with both the core symptoms of ASD (Allik, Larsson, & Smedje, 2006; Gabriels, Cucaro, Hill, Ivers, & Goldson, 2005; Goldman et al., 2011; Schreck & Mulick, 2000; Taylor, Schreck, & Mulick, 2012), as well as other daytime behaviours (Adams, Matson, & Jang, 2014; Goldman et al., 2011; Mazurek & Sohl, 2016; Sikora, Johnson, Clemons, & Katz, 2012; Taylor et al., 2012). However, despite considerable research literature addressing sleep problems in children with ASD, there are inconsistencies regarding the rate and severity of these problems. This is likely, at least in part, to be a function of the methods used to assess sleep. Further, few studies have examined the potential cognitive mechanisms which underlie behavioural sequelae of poor sleep. To build on the existing literature, the overarching aims of the current thesis were to examine the tools used to assess sleep, to investigate group differences in sleep across ASD and typically developing (TD) samples, and to investigate the relationships between sleep, daytime behaviour and cognition in greater detail.
6.1 Summary of the Main Findings

To address the aims detailed above, the current thesis included: psychometric examination of a widely used sleep scale, the Children’s Sleep Habits Questionnaire (CSHQ; Chapter 2); a longitudinal assessment of the relationships between sleep problems and core ASD features over time (Chapter 3); an investigation of the differences in sleep between ASD and TD groups using a multi-method sleep assessment approach (Chapter 4); and exploration of the relationship between sleep and challenging behaviours including identification of potential underlying cognitive mechanisms (Chapter 5). In addressing these questions, Chapter 2 detailed revisions to the CSHQ based on psychometric evaluation. The revised subscales (CSHQ-R) showed good agreement with another, well validated, sleep questionnaire, the Sleep Disturbances Scale for Children (SDSC; Bruni et al., 1996). Overall, the chapter provided support for use of the CSHQ-R as an accurate measure of sleep in children with ASD. Chapter 3, assessed the stability of sleep problems and ASD symptoms over time in the ASD population, which were closely related at both time points. Both ASD symptom severity and sleep disturbance severity were notably stable across the study period. However, despite robust associations between sleep and ASD symptoms at each time point, after controlling for baseline symptoms at presentation, no longitudinal predictive associations were identified between sleep and core ASD symptoms. Given the enduring nature of sleep problems identified in Chapter 3, Chapter 4 sought to build on the current understanding of sleep problems in children with and without ASD using a multi-method approach to sleep assessment. Consistent with past literature, children with ASD had more severe parent-rated sleep problems (i.e. CSHQ-R) than their TD peers; however, this was not reflected in objective assessment of sleep (i.e. actigraphy). Moreover, few
differences emerged on parent-rated assessment of sleep parameters (sleep diary). These findings highlight the complexity of establishing “sleep problems” in children with ASD and reinforce the need for broad assessment of sleep, capturing both sleep parameters from actigraphy and sleep diaries, as well as perceptions of sleep from caregiver reporting. Finally, Chapter 5 examined the relationship between sleep problems (assessed using parent-report and actigraphy), challenging daytime behaviours (parent-report) and executive functioning (EF; assessed using parent-report and cognitive testing). While there were consistent associations between parent-reported sleep and parent-reports of daytime behaviour and EF, the study did not identify relationships between objective actigraphy assessments of sleep and challenging behaviour or any measure of EF, except that within the ASD group, sleep onset time (SOT) was associated with challenging behaviours after controlling for use of sleep medication. No associations were found between performance-based measures of EF and parent-reports of sleep or behaviour.

These findings, along with their theoretical and clinical implications, are discussed in more detail below. Further, the limitations of the current findings and suggestions for future research are outlined. Finally, the broader implications of these findings are described.

### 6.2 Measuring Parental Perceptions of Sleep in ASD

To allow for further exploration of sleep in children with ASD, the first study sought to examine the most widely used measure of sleep in ASD, the CSHQ (Owens, Spirito, & McGuinn, 2000). Although polysomnography (PSG) is the gold standard tool for assessing sleep parameters, it is costly and potentially difficult to use with children who have sensory sensitivities (Hodge, Parnell, Hoffman, & Sweeney, 2012). Actigraphy, an alternative objective measure of sleep, is widely...
used, though it is still relatively costly (when compared to questionnaires) and requires training to ensure data are collected and interpreted appropriately (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). As such, much of the research literature focusing on sleep in ASD has relied solely on parent-reported questionnaires such as the CSHQ. Questionnaires offer the benefit of being inexpensive, quick and easily administered, as well as being able to capture information about broader sleep-related behaviours, over a longer time frame, which are not reflected in objective assessment of sleep-wake parameters.

While the CSHQ is the most widely used questionnaire in research examining sleep in ASD samples, it was not developed for use in this population. Further, the original CSHQ was subject to only limited psychometric analysis during the scale development stage, as highlighted in a review of paediatric sleep scales (see: Spruyt & Gozal, 2011). Moreover, despite the frequent use of the CSHQ in ASD populations, at the time of completing the current research, only one other study had assessed the psychometric properties and factor structure of this instrument in an ASD sample (Johnson et al., 2016). Johnson et al. (2016) completed a principal components analysis and found that a five-component solution was the best fit for their data. However, the authors noted that their study was limited by sampling factors (e.g. over 50% of the sample had marked disruptive behaviours; 11% recruited specifically due to known behavioural sleep disturbance; Johnson et al., 2016).

The study described in Chapter 2 found that neither the original CSHQ structure (SCHQ-Original) nor the “CSHQ-ASD” structure proposed by Johnson et al. (2016) provided a good fit for the data in the current, large, representative, community-based sample of children with ASD. This was consistent with a number
of past studies in various TD paediatric populations which had been unable to confirm the CSHQ-original factor structure (Liu, Wang, Tang, Wen, & Li, 2014; Lucas-de la Cruz et al., 2016; Schlarb, Schwerdtle, & Hautzinger, 2010; Sneddon, Peacock, & Crowley, 2013; Waumans et al., 2010). As neither model fit the current data, exploratory analyses were conducted to revise the scale. This analysis yielded a five-factor solution (the CSHQ-R model) based on 27 of the original items. This model was easily interpretable and required relatively minor changes from the CSHQ-original model. While demonstrably different from the CSHQ-original model, a number of consistencies emerged in item groupings which were commensurate with the original scale based on the International Classification of Sleep Disorders (ICSD). Similarly, there were some overlapping features between the CSHQ-ASD and current CSHQ-R models. Of note, a very recent study reported on the factor structure of the CSHQ in a larger sample of children with ASD (Katz et al., 2018). In a large (N = 4979) sample of children aged 4 to 10 years, Katz et al. (2018) explored the factor structure of the items in the CSHQ (n = 1437) leading to a revised scale which was then confirmed through confirmatory analysis (n = 1435). A number of items were removed during their initial item analysis and exploratory factor analysis (EFA) after which, the best fit for the data was a four-factor solution. While all items on the SDB subscale were removed, leading to the absence of this scale, the remaining four factors were remarkably comparable to those presented in Chapter 2. Specifically, the Sleep anxiety/Co-sleeping subscale described by Katz et al. (2018) was exactly the same as the current Co-sleeping scale. The Sleep Initiation and Duration and Daytime Alertness subscales defined by Katz et al (2018) were comparable to the current Sleep I & M and Morning Lethargy subscales in Chapter 2, with the exception of one question on each. These minor discrepancies were due to
differences in the items removed before or during EFA. (Katz et al., 2018). Finally, while a number of the items on the Katz et al (2018) Night-waking/Parasomnias subscales were comparable to those in the current Parasomnia scale, more differences emerged, again due to differences in the items removed before or during EFA. Namely, Katz et al (2018) included items regarding night-time waking which were removed during our analyses. Taken together, these consistencies were considered promising signs of factorial validity, with subscales mapping appropriately to the underlying and separable constructs which drive poor sleep (i.e. difficulties with sleep initiation/maintenance, parasomnias, sleep-disordered breathing, etc.). Consistencies between the revised factor structure and theoretical conceptualisation of poor sleep provide further support for the proposed scale. Moreover, the revised scale had strong internal consistency with values all above acceptable minimal levels and improved compared to those for the CSHQ-original subscales reported by Owens et al. (2000). The total scale also had good overall internal consistency.

Assessed in a smaller sub-sample of the participants, the revised scales derived from the CSHQ-R demonstrated good agreement with another well developed and validated questionnaire used to assess sleep problems in children, the SDSC (Bruni et al., 1996). Agreement between subscales was highest for those measuring conceptually similar constructs. Further, support for the scale’s validity was reflected in the finding that those who reported use of melatonin supplements had significantly higher scores across subscales. Use of this medication is a strong indication that sleep problems had previously been identified (Cuomo et al., 2017; Rossignol & Frye, 2011).
Taken together, the current findings suggest caution in using the original CSHQ or CSHQ-ASD factor structures in children with ASD. Instead, the current study presents strong evidence that the revised scale is a psychometrically sound method for assessing sleep in this population. However, it is also noted that while the CSHQ-R structure shows promise for use in children with ASD, the current sample size was insufficient to split into training and validation data sets, and as such, the scale has not been confirmed through confirmatory factor analysis. Nonetheless, the analysis suggests that the revised scale is an improvement to the previously heavily-employed original CSHQ.

6.3 Examining Temporal Relationships between Sleep Problems and ASD Symptoms

Within the literature to date, there are a number of potential explanations for the high rates of reported sleep problems in ASD. The most commonly cited explanations in the literature include the presence of a common comorbid medical or neurological disorder, behavioural problems, mood disturbance, use of medications and environmental or family factors (Cotton & Richdale, 2010). Others have suggested that sleep disturbance may be related to either the neurobiological or genetic abnormalities inherent in ASD (including irregularities in endogenous melatonin synthesis leading to disruptions to the circadian rhythm) or the core behavioural features associated with ASD – that is, increased restricted and repetitive behaviours (RRBI) or decreased social and communication skills (Richdale & Schreck, 2009).

Specifically with regard to the core symptoms of ASD, a considerable body of research has identified links between the presence and severity of sleep problems and both social and communication deficits as well as RRBI (Adams, Matson,
Cervantes, & Goldin, 2014; Hollway & Aman, 2011; Hollway, Aman, & Butter, 2013; Kozlowski, Matson, Belva, & Rieske, 2012; Park et al., 2012; Schreck, Mulick, & Smith, 2004; Tudor, Hoffman, & Sweeney, 2012). As noted by Adams et al. (2014), although this association has been well investigated in past literature, the majority of research has conceptualised this link as a unidirectional relationship, examining only whether sleep problems may lead to increased ASD symptomatology (e.g. Kozlowski et al., 2012; Schreck et al., 2004). However, as proposed by Adams et al. (2014) this is likely a bidirectional relationship with core symptoms of ASD potentially exacerbating, and being exacerbated by, problem sleep.

In light of this, the study outlined in Chapter 3 examined the associations between sleep and ASD symptom severity over time, examining both the stability of sleep and ASD symptoms, as well as the longitudinal predictive relationships between these variables. The study found that ASD symptom severity and sleep problems were positively associated at each time point and across time points (i.e. sleep problems at initial assessment were associated with ASD symptoms at follow-up and vice versa). However, after controlling for baseline sleep problems, initial ASD symptoms were not predictive of follow-up sleep problems; and similarly, after controlling for baseline ASD symptoms, initial sleep problems were not predictive of follow-up ASD symptom severity. The finding of strong associations at and across time points is consistent with previous literature (Gabriels et al., 2005; Goldman et al., 2009, 2011; Hoffman et al., 2005; Hollway et al., 2013; Mayes & Calhoun, 2009; Park et al., 2012; Schreck et al., 2004; Taylor et al., 2012; Tudor et al., 2012).

Contrary to the hypothesis that poor sleep and ASD symptoms may be mutually causal (Adams et al., 2014), no temporal relationships were found between sleep and symptoms after accounting for stability in ASD symptoms and sleep over time.
However, given the relatively small sample size in the current study (n = 35 - 39), it was not valid to explore relationships across sleep and ASD symptom subscales to determine whether there were particular features of sleep which may predict certain ASD symptoms (and vice versa). It remains possible that if assessed at the subscale level, different predictive associations between sleep and ASD symptoms emerge. Further, in light of findings in subsequent chapters of the current thesis, regarding discrepancies between sleep problems identified by objective assessment (e.g. actigraphy) when compared to subjective assessment, conclusions drawn from questionnaires alone must be interpreted with caution.

6.4 Assessing the Longitudinal Stability of Sleep Problems in ASD

The study presented in Chapter 3 also revealed that reports of the severity of sleep disturbance were remarkably stable across time in children with ASD. To our knowledge, this study represents the longest follow-up period for assessing sleep in ASD (i.e. up to 5 years post initial assessment). The findings were consistent with those of the small number of other longitudinal studies (with shorter follow-up periods) which describe ongoing reports of sleep problems over time (Allik, Larsson, & Smedje, 2008; Fletcher et al., 2017; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Richdale & Prior, 1995; Sivertsen et al., 2012). While there are few longitudinal studies compared to the plethora of cross-sectional data, the current findings are also consistent with evidence from cross-sectional studies which have found that older children with ASD are reported to have similar rates of sleep disturbance as their younger counterparts (Goldman et al., 2012; Mayes & Calhoun, 2009; Patzold, Richdale & Tonge, 1998; Schreck & Mulick, 2000; Wiggs & Stores, 2004; Williams, Sears, & Allard, 2004). While no conclusions can be drawn
regarding potential differences between sleep trajectories across ASD and TD groups based on the current data, the results are commensurate with a growing body of evidence that children with ASD do not appear to outgrow sleep problems in the way that TD children have been found to. That is, group differences between children with ASD and their TD peers tend to emerge more consistently for older children (i.e. late primary school age) when sleep problems start to abate in TD children (Couturier et al., 2005; Hoffman, Sweeney, Gilliam, & Lopez-Wagner, 2006; May et al., 2015; Souders et al., 2009). These findings indicate that children with ASD do not outgrow sleep problems, and so it is critical that sleep problems are identified and addressed as early as possible.

6.5 Comparison of Sleep Patterns in Children with and without ASD

Given the persistent nature of reported sleep problems in ASD identified in Chapter 3, the study outlined in Chapter 4 examined sleep problems in more detail using a multi-method approach to sleep assessment. As described in the General Introduction (section 1.6.3), there is some indication in the literature that while sleep problems are persistently reported by parents, findings from objective assessment of sleep are less consistent. The two aims of the study were, therefore, to explore group differences between primary school-age children with and without ASD using both parent-reported and actigraphy sleep data, and to assess the concordance between these measures.

As hypothesised based on findings from past literature (e.g. Couturier et al., 2005; Goodlin-Jones, Sitnick, Tang, Liu, & Anders, 2008; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Hoffman et al., 2005; May et al., 2015; Souders et al., 2009), children with ASD were reported by their parents to have higher levels of
sleep problems than their TD peers across most subscales of the CSHQ. However, when sleep parameters were assessed using either sleep diary or actigraphy variables, the two groups appeared quite similar. Indeed, group differences were found only for two comparable actigraphy/diary variables capturing the time the child woke in the morning (DWT, diary; and SOffT, actigraphy). This earlier wake time did not, however, translate into significant group differences in the total amount of time spent in bed or sleeping. As identified in the paper, while a number of previous studies that used actigraphy found group differences across a range of sleep parameters, only two other published studies (Allik et al., 2006; Hering, Epstein, Elroy, Iancu, & Zelnik, 1999) reported group differences for SOffT. This highlights the inconsistencies across studies when using objective assessment of sleep, possibly attributable to differences in sampling (i.e. age, comorbidities, mediation, etc.).

When looking at the rates of children who met the predefined definitions for “poor” or “good” sleep, significantly more children with ASD than TD children were considered by their parents to have a sleep problem. Despite this, ASD and TD groups did not differ in the number of children identified as having “poor” or “good” sleep based on actigraphy recordings using predetermined cut-offs for “poor” sleep quality and quantity.

Importantly, in employing both objective and subjective assessment of sleep, the study presented in Chapter 4 demonstrated that even in the context of clear parent-reported sleep difficulties for the children with ASD, objective sleep parameters were comparable between the ASD and TD samples. To further investigate these discrepancies, this study also examined agreement between the objective and subjective assessment of sleep, comparing the CSHQ-R subscale scores to variables from both sleep diaries and actigraphy. Few associations were
found between the CSHQ-R subscale scores and diary or actigraphy measures for either group, including those variables which were intended to measure similar constructs (e.g. CSHQ-R Sleep I & M with actigraphy SOL or WASO). Consistent with a small number of studies which have reviewed the agreement between sleep measures in ASD (e.g. Hering et al., 1999; Wiggs & Stores, 2004), these findings highlight the discrepancy between the information obtained from different tools. Hering et al., (1999) found that children with ASD and reported sleep problems (n = 8) only differed from TD children without reported sleep problems (n = 8) on actigraphy assessed SOffT, but no other objective sleep parameter. The authors concluded that this may reflect a bias in parental-reports. Similarly, in a much larger sample of children aged 5-16 years (N = 69) with ASD, children who were reported to experience increased “sleeplessness” did not differ from those who were not reported to be “sleepless” on objectively assessed parameters (Wiggs & Stores, 2004).

While researchers have hypothesised that these discrepancies may be a reflection of parents of children with ASD misperceiving sleep (Hering et al., 1999), the current research found strong agreement between a number of comparable sleep diary and actigraphy variables. This agreement indicates that parents generally have good knowledge of their children’s sleep patterns regardless of whether their child is TD or has a diagnosis of ASD. In this context, elevated scores on questionnaire measures in the ASD group were interpreted as a reflection of ongoing parental concerns regarding sleep behaviours which were separate to, and independent of, underlying sleep parameters (i.e. hours spent sleeping). For example, a child may have increased bedtime refusal, but nevertheless still fall asleep at approximately the same time as others their age. A complementary explanation proposed by Wiggs and
Stores (2004) is that children with ASD may be less able to self-sooth, leading to increased parental involvement in their bedtime routine and therefore increased perception (or knowledge) of sleep problems.

One potential contributing factor not explored in the current study was whether or not parental perceptions of poor sleep may represent a “lag” in perception from past poor sleep patterns. A number of children in the ASD sample were taking prescription medication for sleep (e.g. melatonin). This treatment suggests that sleep problems had been identified in the past. It may be that use of medication had improved sleep patterns (as measured by actigraphy) but that past experience of poor sleep continued to influence parents’ perception of sleep. However, given there were similar discrepancies between objective and subjective measures for ASD and TD groups, this may also reflect that fact that these tools are simply capturing different aspects of sleep. Therefore, it is possible that while sleep parameters had improved subsequent to treatment, the child maintained a number of broader bedtime related behaviours which were problematic.

6.6 Examining the Relationships between Sleep Problems and Challenging Behaviours

Given the persistent nature of reported sleep problems as outlined in Chapter 3 and the severity of these problems detailed in Chapter 4, the final empirical chapter sought to examine the relationship between sleep problems and daytime challenging behaviour in children with and without ASD. As noted in Chapter 1, reports of poor sleep in ASD have been found to be associated with a range of behavioural outcomes. However, much of this literature has focused on subjective assessment of both sleep and behaviour. The initial aim of the final study was to explore the
relationships of both objective and subjective assessments of sleep with parent-reports of challenging behaviours.

It was found that, compared to TD children, those with ASD had higher levels of parent-reported sleep problems, as well as increased levels of parent-reported challenging behaviours. Within each group, parental-reports of sleep problems were related to their reports of challenging behaviours. However, objective assessment of sleep using actigraphy was mostly unrelated to the parent-reported challenging behaviours, except SOT within the ASD group. This finding is interesting given that the existing literature in ASD samples has relied solely on parent-reports (e.g. Adams, Matson, & Jang, 2014; Fadini et al., 2015; Goldman et al., 2009, 2011; Mazurek & Sohl, 2016; Sikora et al., 2012). To our knowledge, the study presented in Chapter 5 is the first to examine challenging behaviours in the context of objectively-assessed sleep within an ASD population. It is pertinent to note that this is not dissimilar to findings from Fletcher et al., (2017) who found that anxiety was related to parental responses on the CSHQ but not actigraphy-derived sleep profiles. As noted by the authors, this may be attributed to shared method variance (i.e. parent report for both); however, it was also suggested that anxiety may have particular associations with sleep behaviours, though not sleep timing (Fletcher et al., 2017). These two explanations were also considered in Chapter 5, with differences possibly indicating that while challenging behaviours are related to the perception of sleep or bedtime related behaviours, they are not necessarily related to objective sleep parameters.
6.7 Examining the Relationships between Sleep Problems and Executive Functioning

In addition to exploring the relationship between sleep and challenging behaviours, the study presented in Chapter 5 also examined how both sleep and challenging behaviours related to EF skills. While challenging behaviours and reduced cognitive skill have often been discussed in the ASD literature, to our knowledge, no studies have examined EF in the context of sleep in ASD. This is surprising given the well-established link between sleep and EF in the paediatric sleep literature more broadly (Anderson, Storfer-Isser, Taylor, Rosen, Carol, & Redline, 2015; Bernier, Beauchamp, Bouvette-Turcot, Carlson, & Carrier, 2013; Edmed, Rossa, Kenardy, Anderson, & Smith, 2017; Moreau, Rouleau, & Morin, 2013) and that reduced executive control is widely acknowledged in populations with ASD (Demetriou et al., 2017).

Consistent with the existing literature, the findings in Chapter 5 indicate that the children with ASD had poorer performance on both parent-rated and performance-based measures of EF, relative to the TD children. However, within each group (ASD and TD), though parent-reported sleep was related to parent-reported EF (behavioural regulation, not meta-cognitive skill), there was no relationship between sleep and a performance-based EF. Further, there was no relationship between objective assessment of sleep and either parent-rated or performance-based EF. Again, these results were unexpected given that studies of TD children have found associations between objectively assessed poor sleep and performance-based measures of EF (i.e. measures of complex attention, working memory, and inhibition; e.g. Sadeh & Acebo, 2002; Vriend, Davidson, Shaffner, Corkum, & Rusak, 2013).
In light of this, a number of hypotheses regarding differences in the current sample were considered. Firstly, very few children had “problem” sleep of fewer than 7 hours per night (as defined by Hirshkowitz et al., 2015). Given that deficits in neurocognitive functioning are generally reported in the context of sleep deprivation or restriction (de Bruin, van Run, Staaks, & Meijer, 2017; Lowe, Safati, & Hall, 2017), it may be that sleep was too limited in variance in either group; that is, there were few children with deficits in sleep sufficiently pronounced to contribute directly to executive dysfunction. However, while Hirshkowitz et al., (2015) describe less than 7 hours as problematic for children aged 6-12 years, their guidelines recommend that children within the age group ideally sleep for 9-11 hours, with 7 hours recorded as the lower limit of what “may be appropriate”. Therefore, while few in the current samples were averaging less sleep than the absolute minimum duration, most were sleeping less than recommended.

6.7.1 Sleep, Challenging Behaviours and Executive Functioning

With regard to sleep, behaviour, and EF, given that children with ASD are consistently found to exhibit higher levels of executive dysfunction than TD children (Demetriou et al., 2017), and that deficits in EF appear to be related to challenging behaviours in ASD (Visser, Berger, Van Schrojenstein Lantman-De Valk, Prins, & Teunisse, 2015), it was considered likely that the association between sleep and externalising behaviours in ASD may, at least in part, be accounted for by the impact that poor sleep has on EF. Overall, the findings from this analysis can be broadly summarised as: parental perceptions of sleep, challenging behaviours and EF (behavioural regulation) were strongly inter-related; however, no relationships were found between these variables when objective measures of either sleep (i.e. actigraphy) or EF (performance-based measures) were used.
Leaving aside the null findings for objective assessment of sleep and EF, as there were relationships between parent-rated sleep, challenging behaviours and executive behavioural regulation, a pre-defined mediation model was computed with these variables. As hypothesised, reduced behavioural regulation skills explained the relationship between sleep and externalising behaviours in the TD group; however, the mediated effect did not reach significance in the ASD group. Despite this, there was a similar pattern of relationships between sleep, behavioural regulation and externalising behaviour across the two groups.

As noted, these relationships were initially considered in the context of known pathways through which reduced sleep may impact upon cognition, and in turn, behavioural control. That is, for example, changes in neurological activity in the prefrontal cortex (Horne 1993; Yoo et al. 2007) which is implicated in executive control, may lead to decreased behavioural inhibition and, therefore, increased challenging behaviours. However, given that objective sleep parameters were not related to challenging behaviours or EF, alternative explanations for the association between parent-rated sleep and challenging behaviours must also be considered. In this context, it is noted that relationships between sleep and daytime behaviour are likely bidirectional (see for review Gregory & Sadeh, 2012). The bidirectional nature of these relationships has also been noted within the field of ASD specifically (Hollway & Aman, 2011). That is, poor sleep may lead to increased externalising behaviours; while externalising behaviours, associated with over-arousal may lead to difficulties settling at bedtime. It is possible that increased challenging behaviours throughout the day may lead to behavioural sleep problems which are captured by the CSHQ-R, but not actigraphy, such as bedtime refusal.
It is again noted that the current study employed a composite measure of EF, collapsing performance across EF tasks into a single variable to avoid an inflated type 1 error risk associated with multiple comparisons in a small sample. However, it is possible that this combined variable was not sensitive enough to detect performance decrements. As such it may be beneficial for future research to examine these relationships across individual EF tasks in larger samples.

### 6.8 Implications for Research and Clinical Practice

Taken together, the results from this thesis have a number of clinical implications. Firstly, with regard to sleep measurement; in the context of the existing literature regarding the CSHQ, the results of Chapter 2 again highlights concerns regarding the factorial validity of the original scale and its use in ASD. The CSHQ-R appears to provide an improved subscale structure for assessing sleep in this population. While further confirmation of this revised structure and validity of the scale is warranted, this scale shows promising signs of improvement from the CSHQ original and was broadly consistent with that reported by Katz et al (2018).

Moreover, in the context of findings from Chapters 4 and 5 it is pertinent to note that while cost effective and quick to administer, the CSHQ does not provide a good indication of sleep parameters. This conclusion is consistent with previous findings which have found little agreement between measures (Goodlin-Jones, Tang, Liu, & Anders, 2008; Veatch et al., 2016). While Goodlin-Jones et al., (2008) found that parent-reported sleep diaries correlated well with actigraphy, there was little concordance with the CHSQ. Similarly, in a large sample of children with ASD (N = 80), Veatch et al (2016) found that while reports of sleep and wake times were consistent with actigraphy recording, subscales of the CHSQ were not. However, the
authors did find that agreement between measures improved after parental sleep education.

Overall, while questionnaire data are useful indicators of problems related to bedtime routine, sleep behaviours and lethargy in the morning, the subscales do not provide an assessment of actual sleep parameters. The suggestion is not that data from questionnaires are not helpful; however, caution must be taken in interpreting these data, with clear reference made to the perception of sleep and sleep behaviours, rather than sleep duration or quality. Where possible, combined use of questionnaires and objective sleep assessment are imperative in gaining a clear picture of sleep in ASD. While this guideline has been echoed throughout the paediatric sleep literature for the past 5-10 years (e.g. for review see Gregory & Sadeh, 2012), many studies of sleep in ASD continue to focus exclusively on questionnaire data (e.g. Couturier et al., 2005; Giannotti et al., 2008; Hoffman et al., 2005). Where the use of objective assessment is not possible (in either research or clinical settings) due to constraints regarding time, expense, or poor toleration of the equipment, use of sleep diaries may be considered as a reasonable adjunct to questionnaires, with data presented in Chapter 4 suggesting good agreement between diary and actigraphy variables.

Parental reports of problem sleep were broadly incongruent with objective assessment, and the ASD sample sleep parameters were broadly consistent with the TD comparison groups. However, it is also noted that many of the children in the sample were using medication for sleep. Given the current data, it is possible that even with medical intervention (e.g. melatonin) children continue to present with behavioural sleep problems. Alternatively, it may be that even when medication been used to successfully treat sleep patterns (as measured by actigraphy), parents prior experiences of their child’s poor sleep continue to influence their perception of sleep.
beaviours. Clinically, when planning and monitoring interventions, it will be important for those treating children with sleep disturbance to track both changes in sleep patterns (i.e. actigraphy), and changes surrounding bedtime behaviours (i.e. CSHQ-R), and closely consider both medical and behavioural interventions. Another consideration is ongoing parental sleeplessness, secondary to a child’s sleep problems. Studies may also benefit from further exploring parental sleep and associations with both past and current child sleep patterns. Regardless, the findings presented in Chapter 3 highlight the enduring nature of perceived sleep problems in the ASD population. While future research will be required to determine whether or not sustained difficulties are reflected in objective sleep patterns, the enduring nature of sleep concerns, in and of itself, is important. Parental perceptions of sleep problems do not abate with time, highlighting the need for appropriate intervention.

Finally, there are a number of clinical considerations related to the findings detailed in Chapters 3 and 5, regarding the relationships between sleep and daytime features of ASD (i.e. symptom severity, challenging behaviours and EF). While sleep and ASD symptom severity were not found to be predictive when assessed longitudinally, they were closely associated at both assessments. The relationship between symptom severity and sleep suggests that of those with ASD, children who present with greater symptom severity should be particularly closely monitored for sleep disturbance.

While the nature of the relationships between sleep, challenging behaviours and EF remains somewhat unclear, the data presented in Chapter 5 corroborate past findings that parent perceptions of sleep and challenging behaviours are related. Given that no relationships were found between objective measures, it is difficult to determine the reasons for these relationships; however, this does highlight that in a
clinical setting, those presenting with challenging behaviours should also be closely monitored for problematic behaviour around bedtime. As noted by Adams, Matson, and Jang (2014), given the relationship between poor sleep and challenging behaviours, it is imperative that sleep problems are treated so as to mitigate any potential flow-on impact to other areas of development (e.g., cognitive or behavioural regulation).

6.9 Limitations and Direction for Future Research

While this thesis presents one of the longest follow-up periods for longitudinal assessment of sleep in ASD, only subjective parent-report data were available for analysis in this study. Results were consistent with questionnaire data from other longitudinal studies (Fletcher et al., 2017; Richdale & Prior, 1995; Sivertsen et al., 2012), which identified parent-reports of sleep problems persisting over time in people with ASD. Interestingly, of the two studies which have objectively assessed sleep patterns over time in TD and ASD groups, both reported that the trajectory of sleep patterns was similar across groups (Allik et al., 2008; Fletcher et al., 2017). That is, both studies found that while there were group differences on some sleep parameters (actigraphy) at both time points, children with and without ASD had a similar course of sleep patterns, with a reduction in the duration of sleep and delay in sleep timing (i.e. later sleep onset) as they progressed towards adolescence (Allik et al., 2008; Fletcher et al., 2017). However, while sleep trajectories were similar, the majority of children with ASD (76%) were reported to have a sleep problem (on CSHQ) at both initial and follow-up assessment, while only a one-third of TD children had the same persistence in parent-reported sleep problems (Fletcher et al., 2017). It will be important for future longitudinal studies to
use multi-method assessment of sleep over an extended time period to further delineate the time-course of different types of sleep problems across the lifespan.

Across the two studies which included multi-method assessment of sleep (Chapters 4 and 5), parental-reports of problem sleep were broadly not reflected in objective assessment. While this is an important find, consistent with findings from others examining concordance across objective and subjective measurement of sleep using both the CSHQ (Goodlin-Jones, Tang et al., 2008; Veatch et al., 2016) and other sleep questionnaires (e.g. Hering et al., 1999; Wiggs & Stores, 2004), it is noted that there was no examination of night-to-night variability in sleep in the current studies. As outlined, there is emerging evidence that within ASD groups, sleep problems may present as more dramatic changes from one night to the next (i.e. very short sleep one night followed by long sleep the next night; Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2011; Cohen et al., 2018; Fletcher et al., 2017; Malow et al., 2006). These discrepancies may not be apparent when averaging across multiple days of sleep. It is possible that compared to actigraphy-derived averages, actigraphy-derived night-to-night variability in sleep may be more consistent with data obtained from the CSHQ. Compared to averages, measures of sleep variability may also better distinguish the sleep patterns of autistic individuals and their TD peers. However, given that night-to-night variability cannot be reliably assessed with only seven nights of data (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012), it was not possible to explore variability in the current study. It would be of considerable importance for this to be addressed in future research.

Further, it is important to note that while a number of other studies have found poor concordance between actigraphy and parent report, some of these studies still concluded that children with ASD were presenting with more sleep problems
compared to TD comparison groups or normative data. For example, Wiggs & Stores, (2004) noted that while actigraphy parameters did not differ between those with ASD and parent-reported “good sleep” when compared to those with ASD and parent-reported “poor sleep”, both groups appeared to have more disrupted sleep (actigraphy) than normative data derived from a TD sample.

Another consideration when interpreting the (lack of) group difference between those with and without ASD is that the TD group may not have been representative of the broader community, and the null finding may be attributable to a TD population with particularly poor sleep. Though attempts were made to recruit a representative community-based sample, it is possible that families most inclined to reply to sleep research are those who have children with sleep problems (and thus have an increased interest in the subject). Efforts were made to reduce this bias by explicitly stating that the research was to examine all sleep patterns, rather than study sleep problems per se. Importantly, the TD children’s scores on the CSHQ were comparable to those reported in other studies. Further, while sleep duration and quality were on average lower than recommendations (Hirshkowitz et al., 2015; Ohayon et al., 2017), they did not appear to differ markedly from normative data for the same age groups (i.e. 7.5 to 9.5 hours; Scholle et al., 2011). Similarly, it is possible that in choosing an ASD sample who were able to tolerate an Actiwatch, the sample may not have been representative of those with the most severe sleep difficulties. However, as with the TD group it is highlighted that ASD sample’s scores on the CSHQ were comparable to those reported in other studies.

With regard to recruitment, inclusion of a community-based, rather than clinic-based, sample of children with ASD was aimed at reducing possible bias from a clinical sample possibly presenting with elevated levels of comorbid medical and
psychiatric conditions (Mattila et al., 2010). Despite this, it is pertinent to note that many of the children in the sample still presented with a range of clinical comorbidities (e.g. epilepsy, ADHD) and were using medications, including those for sleep. As rates of comorbidities are high in ASD (Mannion & Leader, 2016; Park et al., 2012), inclusion of these participants was decided to ensure that the samples were representative of the broader population. While attempts were made to control for this statistically, it will be important for future studies to employ multi-method assessment of sleep in the context of treatment (e.g. melatonin), to determine the impact of these treatments both on sleep parameters and parental perceptions.

Further, future studies with larger samples are required to enable separation of any possible effects associated with comorbidities such as ADHD. Importantly, this type of research may now be more feasible with the use of specifiers in the DSM-5 which identify comorbid conditions (American Psychiatric Association, 2013).

In the context of use of parent report measures, it may also be of benefit for future studies to explore parent factors, such as parental mental health, parental sleep and parental stress as potential mediating variables in the relationship between sleep and daytime behaviours.

Finally, the limitations of actigraphy and objective tools for assessing sleep must also be considered. As noted at the outset, the most valuable application of actigraphy is in assessing sleep schedule variables due to high sensitivity in detecting sleep (89% to 97%; with lower specificity in detecting wake (54% to 77%; Meltzer, Walsh et al., 2012). Given this lower specificity, some concerns exist regarding the wake-after-sleep variables derived from actigraphy (Ancoli-Israel et al., 2003; Sadeh, 2011; Sadeh & Acebo, 2002; So et al., 2005). Indeed, one study comparing actigraphy and videosomnography (video recording of the sleep period) in children
with a range of neurodevelopmental disorders including ASD, found particularly low specificity (24%; Sitnick, Goodlin-Jones, & Anders, 2008). However, most recently, the device used in the current thesis (Phillips Respironics MiniMitter Actiwatch-2) has been found to have some of the highest agreement with PSG, recording sensitivity of 93% and specificity of 71%, leading to overall accuracy of 90% (Meltzer, Walsh et al., 2012).

6.10 Conclusion

Over the past decade, there has been a marked increase in the focus on sleep in the ASD literature. Much of the literature in this area has focused exclusively on data collected via parent-report questionnaires, in particular, the CSHQ. Given heavy reliance on this questionnaire, the current thesis further developed the CSHQ to ensure valid and reliable assessment of sleep behaviours. While improvements were made to the CSHQ, multimethod assessment of sleep in subsequent chapters reinforced the notion that parental perceptions of sleep behaviours often differ from objective sleep parameters. While children with ASD were found to have high levels of enduring sleep problems based on parental-reports, there were few differences between those with and without ASD when examining objective sleep parameters. This finding reinforces the need for multi-method assessment of sleep, or at least to aim to capture both subjective measures of both sleep behaviours (questionnaires) and sleep parameters (sleep diary). While reports of sleep problems are clearly of concern, highlighting a need for further research into intervention, discord between the measurement tools reinforces the importance of carefully defining and identifying specific sleep problems and their underlying disorders when intervening. Moreover, results demonstrated that while reports of sleep were indeed related to a number of daytime behaviours, objective sleep parameters were not. Applying this
understanding of the differences in sleep problems and sleep parameters will be important in moving forward with ASD sleep research, and in clinical practice when identifying the type of sleep problem and appropriate interventions.
6.11 References


Appendix

CHILD’S SLEEP HABITS QUESTIONNAIRE
(Owens et al., 2000)

All 48 items from the original CSHQ questionnaire are presented below. The 33 items included in scoring for the original CSHQ (Owens et al., 2000) are in italics. The 27 items included in scoring for the CSHQ-R (Host et al., 2017) are in bold.

The following statements are about your child’s sleep habits and possible difficulties with sleep. When answering these questions, think about think about the past week in your child’s life (i.e. while they were wearing the Actiwatch).

Answer USUALLY if something occurs 5 or more times in a week.
Answer SOMETIMES if it occurs 2-4 times in a week.
Answer RARELY if something occurs never or 1 time during a week.

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<th>Table</th>
<th>Usually</th>
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<td>(5-7)</td>
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1. Child goes to bed at the same time at night
2. Child falls asleep within 20 minutes after going to bed
3. Child falls asleep alone in own bed
4. Child falls asleep in parents or siblings bed
5. Child falls asleep with rocking or rhythmic movements
6. Child needs special object to fall asleep (doll, special blanket, etc.)
7. Child needs parent in the room to fall asleep
8. Child is ready to go to bed at bed time
9. Child resists going to bed at bedtime
10. Child struggles at bedtime (cries, refuses to stay in bed, etc.)
11. Child is afraid of sleeping in the dark
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<tr>
<td>12. Child is afraid of sleeping alone</td>
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<td>13. Child sleeps too little</td>
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<td>14. Child sleeps too much</td>
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<td>15. Child sleeps the right amount</td>
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<td>16. Child sleeps about the same amount each day</td>
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<td>17. Child wets bed at night</td>
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<td>18. Child talks during sleep</td>
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<td>19. Child is restless and moves a lot during sleep</td>
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<td>20. Child sleepwalks during the night</td>
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<td>21. Child moves to someone else’s bed during the night (parent, brother, sister, etc.)</td>
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<td>22. Child reports body pains during sleep.</td>
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<td>23. Child grinds teeth during sleep (your dentist may have told you this)</td>
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<td>24. Child snores loudly</td>
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<td>25. Child seem to stops breathing during sleep</td>
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<tr>
<td>26. Child snorts and/or grasp during sleep</td>
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<tr>
<td>27. Child have trouble sleeping away from home (visiting relatives, vacation, etc.)</td>
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<tr>
<td>28. Child complains about problems sleeping</td>
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<td>29. Child awakens during the night screaming, sweating and inconsolable</td>
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<tr>
<td>30. Child awakens alarmed by frightening dream</td>
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<td>31. Child awakens once during the night</td>
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<tr>
<td>32. Child awakes more than once during the night</td>
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<td>33. Child returns to sleep without help after waking</td>
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<td>(5-7)</td>
<td>(2-4)</td>
<td>(0-1)</td>
</tr>
<tr>
<td>35. Child wakes by him/her self</td>
<td></td>
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<tr>
<td>35. Child wakes with an alarm clock</td>
<td></td>
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<tr>
<td>36. Child wakes up in a negative mood</td>
<td></td>
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<tr>
<td>37. Adults or siblings wake up child</td>
<td></td>
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<tr>
<td>38. Child has difficulty getting out of bed in the morning</td>
<td></td>
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<tr>
<td>39. Child takes a long time to become alert in the morning</td>
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<tr>
<td>40. Child wakes up very early in the morning</td>
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<tr>
<td>41. Child has a good appetite in the morning</td>
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<tr>
<td>42. Child naps during the day</td>
<td></td>
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<tr>
<td>43. Child suddenly falls asleep in the middle of active behaviour</td>
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<tr>
<td>44. Child seems tired in the morning</td>
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</tr>
</tbody>
</table>

During the past week, has your child appeared sleepy or fallen asleep during the following:

<table>
<thead>
<tr>
<th></th>
<th>Not sleepy</th>
<th>Very sleepy</th>
<th>Falls Asleep</th>
</tr>
</thead>
<tbody>
<tr>
<td>45. Play alone</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>46. Watching TV</td>
<td></td>
<td></td>
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<tr>
<td>47. Riding in car</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>48. Eating meals</td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>