SLEEP PROBLEMS IN CHILDREN WITH DEVELOPMENTAL DISORDERS: VOLUME I

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Thesis Overview

This thesis comprises two volumes and is submitted by Andrew Surtees for the Clinical Psychology Doctorate. Volume one includes three chapters. The first chapter contains a meta-analysis of studies that have compared sleep duration and/or sleep quality in people with and without intellectual disabilities. The key finding, after analysis of 26 studies, was that people with intellectual disabilities slept for on average 23 minutes less each night and experienced significantly poorer quality sleep than people without intellectual disabilities. The second chapter details an empirical study of sleep in children with Autism Spectrum Disorders (ASD) and a parent-identified sleep problem. Actigraphy and diary measures found no significant differences between the children with ASD and a comparison group of typically developing children. Questionnaires identified numerous sleep problems (including parasomnias, sleep onset delay and day-time sleepiness) that were more commonly reported in the children with ASD. Chapter three is a summary of the previous two chapters, aimed at a general audience.

Volume Two comprises four Case Practice Reports. Chapter One details the formulation of a 58-year-old man with Obsessive Compulsive Disorder, from Cognitive-Behavioural (CBT) and Systemic perspectives. Chapter Two is a service evaluation of client experience of a group intervention for people with Long Term Conditions. Chapter Three is a Single Case Experimental Design to evaluate the effectiveness of a CBT intervention for sleep and mood problems in a 14-year-old girl. Chapter Four is a case study of a behavioural intervention for challenging behaviour with a 26-year-old man with a severe intellectual disability.

For Ben, who couldn’t wait for this to be finished and will hopefully keep giving us sleepless nights for many years to come.

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CHAPTER I: SHORTER DURATION AND POORER QUALITY SLEEP IN PEOPLE WITH INTELLECTUAL DISABILITIES: A META-ANALYSIS

Abstract

Background. It has long been proposed that people with intellectual disabilities experience more problems with their sleep than do their typically developing peers. A sufficient number of studies have now compared sleep between these groups for the purpose of systematic review and meta-analysis.

Method. Electronic databases were searched systematically, with further papers added from reviews. Search terms were based on variants of terms for intellectual disability, combined with terms for sleep and sleep disorders. The quality of papers reviewed was evaluated using an independent quality framework.

Results. The search returned 21 papers that compared sleep time and/or quality in people with and without intellectual disabilities. The meta-analysis of sleep time revealed that people with an intellectual disability slept for 23 minutes less, on average, than people without an intellectual disability. The analysis of quality also concluded that people with intellectual disabilities experienced poorer sleep: In 93% of comparisons between groups, sleep was found to be of poorer quality in the group of people with intellectual disabilities.

Conclusions. Sleep time was less and of poorer quality in people with intellectual disabilities. Notably, the majority of samples were drawn from populations of people with specified genetic syndromes or developmental disorders, rather than intellectual disability of heterogeneous origin. Similarly, the majority of studies investigated sleep in children, although there was no evidence that problems subsided during adulthood. Most studies used highly-regarded direct measures of sleep, such as polysomnography or actigraphy, although methodological flaws were evident in the identification of samples and the measurement of intellectual disability.

Introduction

Background

Reports of experienced clinicians and a growing body of empirical evidence indicate that children with intellectual disabilities experience problems with sleep more regularly than do typically developing children (Bartlett et al., 1985; Berkman, Didden & Sigafoos, 2001; Quine, 1992; Richdale, Francis, Gavidia-Payne, & Cotton, 2000; Richdale & Baker, 2014; Tietze et al., 2012). The smaller body of evidence on sleep in adults with intellectual disabilities provides no reason to believe that these problems are limited to childhood (Boyle et al., 2010; van de Wouw, Evenhuis, & Echteld, 2012; van de Wouw-van Dijk, 2013). In spite of the growing number of studies conducted with these populations, reviews have concluded that there are significant limitations to knowledge of the scope and severity of sleep problems in different groups of people with intellectual disabilities (Richdale & Baker, 2014). Differences in methodology, characterisation of sleep problems/ intellectual disabilities and sample selection contribute to wide variation in the reported prevalence of sleep problems in populations of people with intellectual disabilities; estimates range from 13% to 86% (Didden & Sigafoos, 2001). Such wide variability in estimates makes it difficult to quantify the scale of the problem with confidence and direct resources to those in most need. The current systematic review includes the first meta-analysis of studies comparing sleep in people with and without intellectual disabilities. It also compares the evidence from parent reports and direct measurement.

Understanding and Defining Sleep

One reason for the variability in findings across studies on sleep in people with intellectual disabilities has likely been the limitations to our understanding of sleep and sleep disorders more broadly. The science of sleep has been developing rapidly over the past 30 years (Randall, 2013) and large variability exists in people with seemingly healthy sleep patterns (Blair et al., 2012). Sleep disorders have been categorised into two types. *Dysomnias* relate directly to difficulties in initiating or maintaining sleep (such as Insomnia, ICD-10, World Health Organisation, 1992; American Sleep Disorders Association, 1997). *Parasomnias* are additional processes that impact on sleep, such as nightmares, sleep apnoea or enuresis. There is some evidence that both types of disorder are more prevalent in people with intellectual disabilities (Didden & Sigafoos, 2001), but variability in quality of sleep can extend beyond the presence of a specified disorder. The likely impact on the individual and their family, both overnight and the following day, is often inferred on the basis of either total sleep time or some measure of sleep quality (Diden & Sigafoos, 2001). Thus, in this review, *Sleep Time* and *Sleep Quality* are treated as distinct dependent variables. Shorter sleep time has been associated with more sleepiness the following day and increased challenging behaviour in people with intellectual disabilities (Johns, 1991; Adams, 2014). Sleep quality remains harder to define and it is broadly accepted that sleep quality is not equivalent to sleep time, although the two are related (Didden & Sigafoos, 2001; Pilcher, Ginter, & Sadowsky, 1997). For instance, fractured sleep time and regularly waking may compromise sleep quality beyond simply reducing the overall duration of sleep (Didden & Sigafoos, 2001).

Sleep in people with intellectual disabilities

Individual differences and potential confounds. The majority of parents of children with intellectual disabilities report that they believe sleep problems in their children are an inevitable result of intellectual disability or brain damage (Quine, 1992). However, people with intellectual disabilities are by no means a homogenous group, so consequently sleep problems are likely of varied aetiology. Factors such as severity of intellectual disability (Didden et al., 2002; van de Wouw et al., 2012) and poor social and communication skills (Quine,1992) have been linked to poor sleep. This suggests that people with intellectual disabilities may be more or less likely to have problems with sleep depending on the degree of their impairment. Similarly, as well as these functional impairments, people with intellectual disabilities are more likely to have physical health conditions, such as epilepsy, posited to have independent mechanisms that would predict poor sleep (Derry, Duncan, & Berkovic, 2006; Doran, Harvey, & Horner, 2006). In addition to physical health conditions, people with intellectual disabilities are thought to be more likely to experience chronic pain, that in many cases is unrecognised and untreated (McGuire, Daly, & Smyth, 2010), which greatly increases the likelihood of sleep problems (Smith, Perlis, Smith, Giles, & Carmody, 2000).

People with intellectual disabilities are also more likely to have comorbid genetic and developmental disorders, many of which have been associated with sleep problems. People with Smith Magenis Syndrome experience inverted melatonin cycles (De Leersnyder, 2006), predictive of difficulties with sleep at night and difficulties maintaining wake and attention in the day time. There is evidence of a significantly increased prevalence of sleep problems in people with Down syndrome (Stores & Stores, 2013), perhaps associated with the physical

differences linked to the condition predisposing people to sleep disordered breathing and sleep apnoea (Chen, Spanò, & Edgin, 2013). Additionally, poorer sleep is reported in children with Angelman syndrome (Miano et al., 2004), Williams Syndrome (Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2013), fragile-X Syndrome (Elia et al., 2000), Prader- Willi Syndrome (Cassidy, Mckillop, & Morgan, 1990), Rett Syndrome (Young et al., 2007), Sanfillippo Syndrome (Colville, Watters, Yule, & Bax, 1996) and Jacobsen Syndrome (Maas, Didden, Korzilius, & Curfs, 2012). Sleep in children with Autism Spectrum Disorders (ASD) has received more attention than other developmental disorders (Richdale & Baker, 2014). Interestingly, increased Autistic symptomology predicts an increased likelihood of a sleep problem (Hoffman et al., 2005; Schreck, 2004). There is also some evidence that intellectual disability is further predictive of sleep problems in people with ASD (Richdale et al., 2000). Given these reports, evidence for sleep problems in people with intellectual disability of heterogeneous origin is examined separately from that in people with genetic syndromes/ developmental disorders as well as together.

Methodological differences in studies of sleep in intellectual disability. Evidence for poor quality sleep in people with intellectual disabilities is often drawn from parent reports (Didden & Sigafoos, 2001), rather than direct measurement. This clearly reduces methodological load on experimenters, individuals and their families, often allowing for larger sample sizes. However, there is conflicting, yet significant, evidence showing parents overestimate their children's sleep difficulties (Hering, Epstein, Elroy, Iancu, & Zelnik, 1999). Additionally, questionnaire measures, the most common tool for such studies, are rarely validated for populations of people with intellectual disabilities (Richdale & Baker, 2014).

Similarly, the majority of samples have been, at least to some degree, self-selecting (although systematic cohort samples have been collected; Boyle et al., 2010; Quine, 1991). In the current review, the impact of measuring sleep directly or through parent report is compared. Furthermore, a set of independent quality criteria against which to weight studies based on the strength of methodology is implemented. Reviewing papers using a quality framework allows for a more objective assessment of different studies based on their methodological strengths and weaknesses (Doi, Barendregt, Khan, Thalib, & Williams, 2015). It can allow for the removal from further analysis of low-quality studies or the statistical tempering of the weight allocated to such studies in the analysis.

Early studies of sleep problems in people with intellectual disabilities rarely included a comparison group of typically developing children (Didden & Sigafoos, 2001), but the last 10-15 years has seen significant development in this area (Tietze et al., 2012). Large cross-sectional cohort studies using random sampling (for instance, Boyle, 2010; Quine, 1991) may provide the most accurate estimates of prevalence of families for whom a child's sleep problem is significant, but because of methodological constraints they typically rely on parent report and provide less in-depth information on the person's sleep. Case-control or group studies, in which a group of children or adults with intellectual disabilities are matched against a typically developing comparison group, are unlikely to provide as accurate an estimate of overall prevalence of sleep problems, but afford several advantages. Firstly, they allow more opportunity to control for potential confounding variables (or at least to systematically report them), such as autism, epilepsy or physical disability. Secondly, they allow for more rigorous examination of sleep, either through more detailed parental questionnaires/ diaries or direct measurement through polysomnography or actigraphy.

Finally, they provide an active comparison group against which to understand the severity of any reported sleep difficulties.

Clinical Impact of Poor Sleep

Poor sleep has a significant biological, psychological, functional and social impact on people. Disturbances to sleep have been associated with physical health conditions (Roth, 2007) and even elevated risk of mortality (Tamakoshi & Ohno, 2004). The psychological impact of poor sleep includes associations between poor sleep and mental health conditions, such as anxiety and depression (Tani et al., 2003). This relationship is also thought to be particularly complicated in somatic disorders, in which sleep interacts with poor physical and mental well-being (Sutton, Moldofsky, & Badley, 2001). Functional impairments in children, as a result of poor sleep are likely, as sleep has been shown to have an impact on attention levels (Lufi, 2014).

Much of the research on the impact of poor sleep in children with intellectual disabilities, has focussed on the increased prevalence of challenging behaviour in children who sleep poorly (Brylewski & Wiggs, 1999; Wiggs & Stores, 1996). Challenging behaviour may be evidence of discomfort for the person themselves and also may impact people around them, such as parents and carers (Emerson, 2001). Sleep problems in children have a negative clinical impact on parents of children with intellectual disabilities, with poorer sleep being associated with increased parent stress (Richdale, Francis, Gavidia-Payne, & Cotton, 2000). It is also worth noting that all of these factors may be self-perpetuating by acting to reinforce difficulties in sleep itself (Tietze et al., 2012).

Rationale

Over the past 25 years, there have been a significant number of studies reporting comparisons of sleep in people with and without intellectual disabilities. However, these studies typically have comparatively few participants and often have to make methodological compromises, such as offsetting the benefits of sample size against depth of data gathering. Additionally, groups of people with intellectual disabilities are not homogenous entities. With this in mind, a meta-analysis of these studies, to examine the evidence for whether people with intellectual disabilities do have a shorter duration and poorer quality of sleep than people without intellectual disabilities, would be timely. This meta-analysis not only evaluates the evidence for the assertion that sleep is worse in people with intellectual disabilities, but also quantifies the effect size of that purported difference. Furthermore, it allows for the examination of the relative effect on any identified difference of independent variable factors (such as inclusion of people with ASD/ genetic disorders), dependent variable factors (such as hours slept vs. sleep quality) and experimental design factors (such as parent report vs. diary vs. direct observation). The introduction of independent quality criteria allows for the impact of studies to be weighted based on their reliability and validity.

Method

Search Strategy

A systematic literature search was conducted using the databases Medline, Embase and Psychinfo in June 2015. All search terms were adapted from van der Wouw et al.'s (2012)

recent systematic review of sleep in adults with intellectual disabilities. Terms for intellectual disability included: intellectual disability, intellectual disturbance, learning disability, mental retardation, mental handicap, mental deficiency, mental disorder, mental incapacity, idiocy, down syndrome, oligophrenia and variants thereof. Terms for sleep time and sleep quality included: sleep, insomnia, dyssomnia, parasomnia, somnolence, hyposomnia and variants thereof. Search terms were required to be included in the abstract, title or keywords of articles. Only empirical, peer-reviewed papers in English were included. For the full search strategy, see Appendix 1.1.1. The final search returned 1590 results. The reference lists of three recent systematic reviews on similar topics: Tietze et al. (2012), van der Wouw (2012) and Richdale and Baker (2014), were also screened for papers that were not returned by the original search.

Paper Selection

Paper selection was completed by the author. Figure 1.1.1 describes the search results and the application of inclusion and exclusion criteria. Studies were excluded on reviewing titles and abstracts if they actively met any of the exclusion criteria, or failed to report the inclusion of participants with intellectual disabilities/ a related disorder (see table 1.1.1) or a measure of sleep time/ quality. If this was not the case, the full paper was retrieved and included/ excluded based on the same criteria.

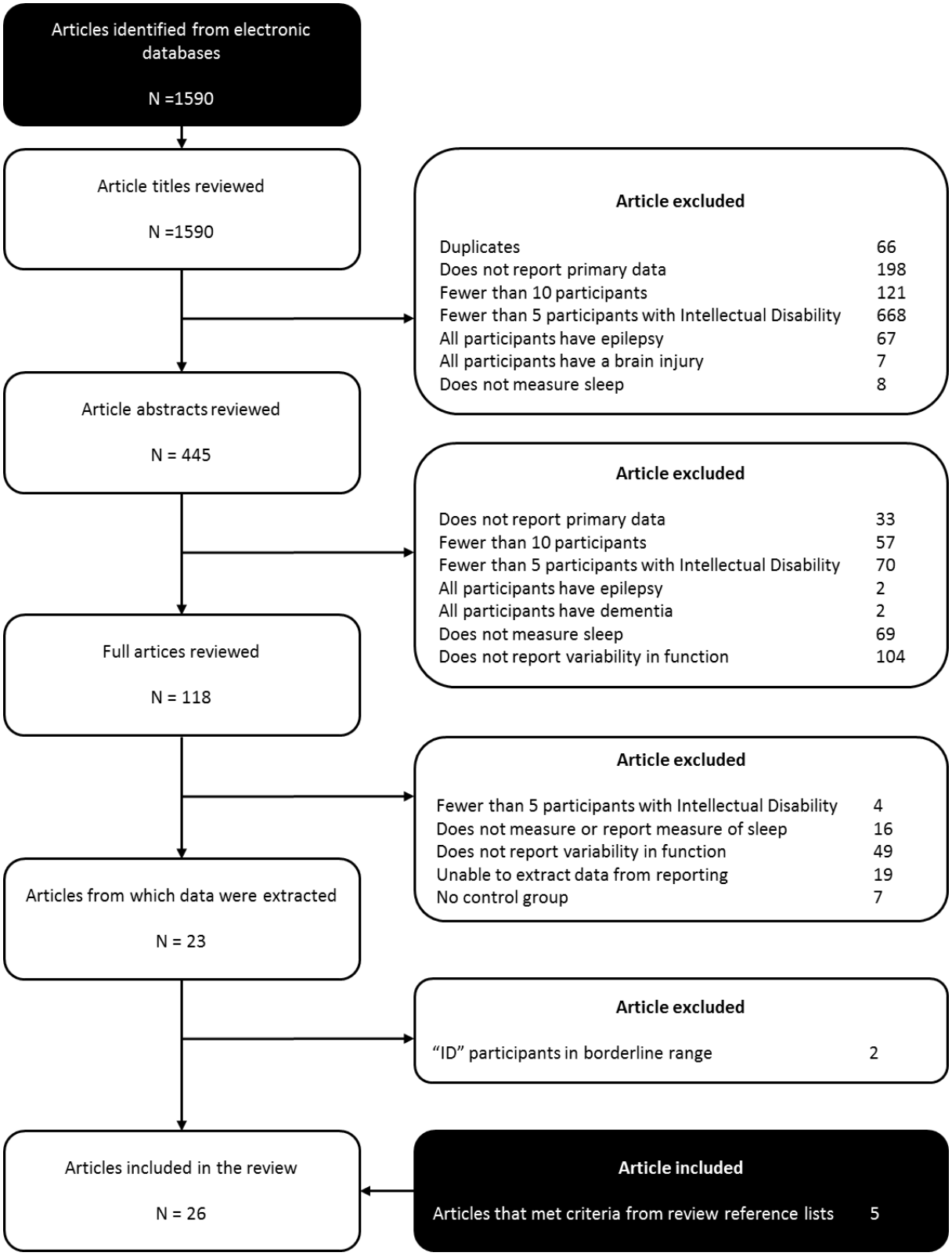


Figure 1.1.1 Process of screening of papers from initial search to final inclusion

Table. 1.1.1 Syndrome groups included with reported IQs and references. Note other syndromes may have been included, but were not returned by the search

Syndrome or Disorder Name	Estimated IQ/ Range of IQs (Reference)
Angelman Syndrome	Mental age 0-2 (Duker, Driel, & Bercken, 2002)
Down Syndrome	Approximately 50, with wide variability (Roizen & Patterson, 2003)
Fragile-X Syndrome	96% have Intellectual Disabilities or Developmental Delay (Bailey, Raspa, Olmsted, & Holiday, 2008).
Prader-Willi Syndrome	Mean IQ approximately 60 (Whittington et al., 2004)
Sanfilippo Syndrome	Majority have mental age 0-2, modal group < 3 months (Valstar, Marchal, Grootenhuis, Colland, & Wijburg, 2011)
Williams Syndrome	IQ of approximately 56 (range: 50–70) (Bellugi, Wang, & Jernigan, 1994; Mervis & Klein-Tasman, 2000)

Exclusion Criteria

Criteria for *participants tested* required that the study included at least five participants with an intellectual disability and at least five without an intellectual disability. For the purposes of this review, participants/ groups of participants were considered to meet the criteria for intellectual disability if reported as such by authors or reported to have a condition/ disorder associated with intellectual disabilities (see table 1.1.1 for included disorders and mean IQs/ mental ages drawn from the literature). Thus, for example, groups of participants with Down Syndrome were included, as Down syndrome is associated with mild to moderate intellectual disability (Roizen & Patterson, 2003), unless evidence of higher intellectual ability was reported for the cohort in the study. However, groups of participants with ASD were not included unless further evidence of intellectual disabilities was reported, as only approximately 55% of people with ASD are thought to have an intellectual disability (Charman et al., 2011). Where standardized IQ tests were reported, papers were considered

to meet criteria if the group of people with intellectual disabilities obtained scores ranged 0 < 85 and the group average was < 70 (one of these criteria was considered sufficient if the other was not reported). Similarly, typically developing comparison groups were required to obtain IQ scores with a range ≥ 70, and an average ≥ 85. Identification of intellectual disability is broadly considered to require the presence of an impairment to functioning, as well as cognitive performance (American Psychiatric Association, 2013; World Health Organisation, 1992). A measure of functioning was not required for participants to be included due to the lack of studies that reported this, but this variable was included within the quality framework for assessing papers. Papers were excluded if all participants with intellectual disabilities had a brain injury, dementia, or epilepsy (due to the widely reported impact on sleep, Doran et al., 2006), but excluding participants with any of these conditions was not a part of the inclusion criteria.

Criteria for the *dependent variable* included measuring sleep time and/or quality in groups of people with intellectual disabilities *and* the typically developing comparison group. Measures of sleep were considered to include parent reports, diaries, direct measurement through polysomnography or actigraphy and sleep questionnaires. Studies investigating solely sleep apnoea, sleep disordered breathing or other parasomnias were not included. Although it was assumed that these factors could impact on sleep time and quality, they do not represent a measure of either sleep time or quality as such. Studies in which the only sleep data came from participants who had been given sleep medication were not included, but studies that measured sleep before the onset of medication as a control condition were. Papers were excluded if they failed to report data in a form that was appropriate for the analysis, such as reporting means but not standard deviations or not reporting the sleep of people with intellectual disabilities separately from those without intellectual disabilities.

Data extraction and management

From the 26 papers included, data were extracted by the author on the number and nature of the participants. This included important demographics about the groups of participants, such as age, gender and average IQ if reported. Furthermore, methodological inclusions/ exclusions were extracted, such as genetic syndrome classification, exclusion of people with ASD, exclusion of people with epilepsy and current or previous reported interventions to improve sleep. Further methodological information, such as how participants were recruited and means of identifying the groups, such as through cognitive or genetic testing, was also recorded. As the two primary dependent constructs were sleep time and sleep quality, those variables that matched these constructs were selected from any reported. For Sleep Time, any direct or indirect measurement of the number of hours slept was extracted. In most cases, this figure reported night sleep, but if this was not reported, a measure of total sleep in a 24-hour period was extracted. For sleep quality, where studies reported measuring sleep directly, sleep efficiency, equal to the percentage of time in bed that was spent asleep, was extracted as the primary variable (Didden & Sigafoos, 2001). Whilst factors such as settling difficulties and waking after sleep onset would clearly impact on sleep quality, sleep efficiency was deemed to be the broadest measure of overall quality, at least in respect to expected and/ or desired quality of sleep. Where studies did not measure sleep directly, the broadest measure of sleep was selected, this included two "sleep quality" and "sleep problems". Sleep problems were understood to be inversely related to sleep quality. Data were extracted from studies that only reported a categorical percentage of participants with sleep problems, rather than a continuous score, but not included in the meta-analysis. Where studies reported multiple measures for one or more constructs, direct measures were chosen over indirect measures, as these are considered a "gold standard" in

the field (Michaelson, Allan, Chaney & Mair, 2006); see appendix 1.1.2 for list of dependent variables not chosen for the analysis.

Quality Review

A Quality Framework was developed to weight the contribution of studies of varying quality in the analysis and remove studies of poor quality. For the purpose of this review, "quality" relates to the methodological constraints of the study in relation to answering the specific question asked within this meta-analysis, rather than an objective measure of the overall quality of the study per se. Studies received independent quality ratings for each participant group when they were recruited through different means and for each dependent variable where appropriate. The quality framework (table 1.1.2) was adapted from Richards, Jones, Groves, Moss, & Oliver (2015), with studies that were considered poor overall removed before the analysis. The framework was based on three factors, thought to reflect the key threats to internal and external validity. Key threats to internal validity emanated from unreliable or incomplete measurement of intellectual disability or sleep. Determining intellectual disability was understood to include measurement of two factors, cognitive functioning and adaptive functioning (World Health Organisation, 1992). Quality of measurement of sleep reflected the use of indirect and direct measures, as well as how they were applied. Furthermore, construct validity was threatened by the fact that sleep quality is often defined broadly and measured in very different ways, meaning different studies may genuinely be assessing different aspects of sleep. In addition to these, the key threat to external validity came from how well the sample reflected the population from which it was drawn. Each of these measures was allocated equal weighting, though calculated across

differing numbers of sub-questions. More formal and comprehensive measures of study quality, such as those proposed by Downs & Black (1998), were considered, however, such scales give weight to less influential factors, such as the inclusion of a structured abstract at the expense of factors likely to have a fundamental impact on the reliability and validity of the findings, such as how samples were recruited. Similarly, such frameworks would not be sensitive to factors specific to sleep research in intellectual disabilities, for instance the relative merits of parent report and standardised measures of cognitive functioning or of polysomnography and a questionnaire that had not been validated.

For 28.5% of papers a second researcher also completed ratings using the framework. An additional item, on the degree to which samples matched, was removed at this stage, after showing an unacceptable level of reliability (α = -.89). Discussion between the two raters indicated that the lack of reliability was likely because many papers reported information very differently. For the items that remained in the framework an excellent level of reliability was obtained, (α = .94) for the whole scale, with individual item ratings varying between good (for identification of sample, α = .82) and excellent (for measurement of intellectual functioning, α = 1.0).

Table 1.1.2. Quality framework used to assess studies. A total score based on the average across these 3 domains was also calculated and awarded an overall quality, such that 0-0.5 = Poor, 0.5-1.5 = adequate, 1.5-2.5 = good, 2.5-3 = excellent.

	Item (Reliability)	Poor (0)	Adequate (1)	Good (2)	Excellent (3)
Sample	**Identification of ID sample (α = .82)**	Unspecified	-Single restricted or non-random sample e.g., a specialist clinic or previous research study -Single regional sample e.g., a regional parent support groups	-Multiple restricted or non-random samples e.g., multi-region specialist clinics, multiple schools -National non-random sampling e.g., national parent support groups	Random sample
	Identification of TD sample (α = .95)	Unspecified	-Single restricted or non-random sample e.g., a specialist clinic or previous research study -Single regional sample e.g., a regional parent support groups -Recruited through friends and family of researchers	-Multiple restricted or non-random samples e.g., multi-region specialist clinics, multiple schools	Random sample
Measurement of Intellectual Disability	**Reliability/ Validity of measurement of level of Intellectual Functioning (α = 1.0)**	Unspecified	-Syndrome group known to be associated with ID Self/parent report -Recruited from specialist ID school/ support group	-Self/parent report with well validated measure	-Formal IQ test (Wechsler Intelligence Scale for Children etc.)
	Adaptive functioning (α = .95)	Unspecified	-Clinician judgment -Self/Parent report -Syndrome group known to be associated with ID	-Self/Parent report, with well validated measure	-Formal measure, such as the Vineland Adaptive Behavior Scales
Measurement of sleep	**Reliability/ Validity of Sleep Measure (α = .94)**	Response to a single question	-Validated sleep questionnaire, note any form of validation is applicable (for instance clinician judgement to make adaptations for population)	-Self/parent monitoring through diaries -Atypical use of polysomnography/ actigraphy	-Polysomnography (following at least 1 day for adaptation) -Actigraphy of 7 days or more

Results

Overview of paper content and quality

Participant Characteristics.

In spite of the search returning over 1500 papers, only 21 were included in the final analyses. In addition, five studies only reported comparisons of the frequency of sleep problems between groups of people with and without intellectual disabilities; these are considered at the end of the results section (and included in table 1.1.3). The 21 papers in the analysis included 33 groups of people with intellectual disabilities (see table 1.1.3) and a total of 1377 participants. These comprised eight groups of people with intellectual disability of heterogeneous aetiology, six with Down Syndrome, five with Williams Syndrome, four with ASD (and intellectual disabilities), three with fragile-X Syndrome, one with Angelman Syndrome, one with Prader-Willi Syndrome and one with Sanfilippo Syndrome. Diomedi et al., 1999 reported on an adult sample, Maaskant et al. (2013) on a sample of older adults, and all other samples had an average age of less than 18 (average ages: 2.5-13.5 years). Consistent with the profiles of a number of syndromes recruited across the studies, a higher proportion of male than female participants were reported, 60.37% (average of averages, not weighting for study size). Ranges of intellectual disability, where reported, varied with average IQ ranging from Profound to Mild by ICD-10 criteria (World Health Organisation, 1992).

Study Quality.

Table 1.1.3 indicates the quality ratings assigned to each study included in the analysis, with appendix 1.1.3 reporting the score on each item of the quality framework. Using the criteria specified, two studies were classified as “excellent” overall, 13 as “good”, one as

"adequate"/ "good", five as "adequate" and none as poor. Quality awarded for "sample" was generally the poorest, with the mean rating being adequate. This commonly reflected practical difficulties in recruitment alongside study aims. A study in which children with a specified genetic disorder from a database were recruited via their families and a typically developing comparison group from a local school would score poorly under the criteria, but was a very common methodology adopted in the literature. This may be because researchers wish to compare the extent of difficulties in these families to what parents of typically developing children experience or because of the practical and ethical difficulties of recruiting children with a rare genetic syndrome randomly. Definition of intellectual disability received higher ratings in most studies. In many cases, this was because appropriately validated IQ measures were employed. On the other hand, measures of adaptive functioning were much rarer, with most studies either relying on presence within a syndrome group or providing no evidence at all for level of functioning. Impaired functioning is common in criteria for intellectual disability (American Psychiatric Association, 2013; World Health Organisation, 1992), but appears to be regularly ignored in research papers. In a consideration of sleep disorders this is particularly important as functioning has been proposed as a mediator between intellectual ability and poor sleep (Quine, 1991). Measurement of sleep time or quality was considered to be "excellent" in the majority of cases. This was because of the preponderance of studies employing a direct measure of sleep (polysomnography or actigraphy). Interestingly, this may reflect a change in the nature of the research since Didden and Sigafoos (2001) reviewed papers and concluded most were based on parent report. Alternatively, it could be the case that studies that employ a typically developing comparison group are also more likely to use a direct measure of sleep.

Table 1.1.3. Demographics, methodology and quality ratings for all studies. *studies that reported only frequency of sleep problems

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2012				57	74	3.8 (2-5.8)	Intellectual Disability	Mullen Scales of Early Learning	Actigraphy	Total Sleep Time	-	55.0 (49-74)	.89
Annaz, Hill, Ashworth, Holley, & Karmiloff-Smith, 2011				64	44	8.2 (6.2-12.5)	Williams Syndrome	None reported (associated syndrome)	CSHQ	-	Total Score	-	.44
Ashworth et al., 2013				22	50	9.4 (6.1-12.2)	Down Syndrome	None reported (associated syndrome)	Actigraphy	Total Sleep Time	Sleep Efficiency	-	.67
				24	50	9.6 (6.1-12.6)	Williams Syndrome					-	.67
Axelsson, Hill, Sadeh, & Dimitriou, 2013				18	-	2.5 (1.3-4.0)	Williams Syndrome	None reported (associated syndrome)	Questionnaire	Night Sleep	-	-	.33
Bruni et al., 2012*				37	54	- (2.3-14.8)	Angelman Syndrome	None reported (associated syndrome)	Questionnaire	-	-	-	.33

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Buckley et al., 2010				13	54	4.3 (2.7-7.1)	Developmental delay	Not stated	Polysomnography	Total Sleep Time	Sleep Efficiency	58.1 (9.6-76.6)	.33
Cotton et al., 2006*				37	75.7	7.1 (3.5-14)	Autism Spectrum Disorder and Intellectual Disability	Parent or clinician report	Questionnaire	-	-	-	.33
				15	53.3	9.0 (3-16)	Down syndrome	Parent or clinician report					.33
				17	76.5	11.6 (3-18)	Prader-Willi Syndrome	Parent or clinician report					.33
				29	72.4	7.0 (4-14)	Intellectual Disability	Parent or clinician report					.33
Cotton & Richdale, 2010				34	74	7.2 (3.5-14)	Autism Spectrum Disorder and Intellectual Disability	Questionnaire	Diary	Total Sleep Time	Sleep Quality	-	.33
				12	58	8.7 (3-13)	Down Syndrome						.33

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Cotton & Richdale, 2010				24	71	7.1 (4-14)	Intellectual Disability	Questionnaire	Diary	Total Sleep Time	Sleep Quality	-	.33
				12	83	9.4 (3-15)	Prader-Willi Syndrome						.33
Dimitriou, Karmiloff-Smith, Ashworth, & Hill, 2013				10	70	18.2 (12-24)	Autism Spectrum Disorder and Intellectual Disability	Psychological Educational Profile	Polysomnography	-	Sleep Efficiency	23.3	.56
				8	75	22.5 (17-31)	Down Syndrome					19.2	.56
				14	36	8.8	Williams Syndrome	Not reported (related syndrome)	Actigraphy	Total Sleep Time	Sleep Efficiency	-	.78
Elia et al., 2000				7	100	9.9 (5.6-16.7)	Fragile-X Syndrome	Not reported (Psychological Educational Profile not used to make groups).	Polysomnography	Total Sleep Time	Sleep Efficiency	-	.44
Fraser, 2005				141	-	13.5 (0-40)	Sanfilippo Syndrome	Not reported (associated syndrome)	Questionnaire	-	Sleep Disturb-ance	-	.56

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Fukuma, Umezawa, Kobayashi, & Motoike, 1974				10	50	12.3 (7-17)	Down Syndrome	Suzuki-Binet	Polysomnography	Total Sleep Time	-	31.1	.61
				10	50	11.3 (7-17)	Intellectual Disability					45.8	.56
Ghanizadeh & Faghih, 2011				58	42	11.1	Intellectual Disability and medical condition	Schooling	Childhood Sleep Habits Questionnaire	-	Bed-time Resistance and Sleep-Disturb-ance	-	.44
				75	44	11.8	Intellectual Disability with no medical condition					-	.44
Gombos, Bódizs, & Kovács, 2011				9	33	20.5 (14-28)	Williams Syndrome	None reported (associated syndrome)	Polysomnography	Total Sleep Time	Sleep Efficiency	-	.61
Goodlin-Jones, Tang, Liu, & Anders, 2008				68	81	3.9 (2.3-5.7)	Autism Spectrum Disorder and Intellectual Disability	Mullen Scales of Early Learning	Actigraphy	Total Sleep Time	Sleep Efficiency	60.3	.89
				57	74	3.8 (2-6.8)	Developmental Delay					55.2	.89

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Levanon, Tarasiuk, & Tal, 1999				23	-	4.8 (1.7-8.0)	Down Syndrome	None reported (associated syndrome)	Polysomnography	Total Sleep Time	Sleep Efficiency	-	.56
Maaskant et al., 2013				501	50	62 (50-92)	Intellectual Disability	Care home and patient notes	Actigraphy		Intra-daily variability	-	.67
Masi et al., 2000*				22	55	16.3 (11-25)	Intellectual Disability and Generalized Anxiety Disorder	WAIS or WISC-R	Interview	-	-	55.7 (50-67)	.28
Miano et al., 2004				6	33	12 (9-17)	Angelman Syndrome (Older)	None reported (associated Syndrome)	Polysmnography	Total Sleep Time	Sleep Efficiency	-	.56
				9	56	4 (3-5)	Angelman Syndrome (Younger)					-	.56
Miano et al., 2008				9	89	13.8 (8-20)	Down Syndrome	Wechsler Intelligence Scales for Children (WISC)-Revised/ Wechsley Adult Intelligence Scales-Revised	Polysmnography	Total Sleep Time	Sleep Efficiency	-	.61
				14	100	13.1 (7-25)	Fragile-X Syndrome					-	.61

Study	Quality			N	% Male	Mean Age (Range)	Group Description	Measure of ID	Measure of sleep	Sleep Time Variable	Sleep Quality Variable	Average IQ (range)	Quality weighting
	Sample	ID Measures	Sleep Measures										
Richdale & Prior, 1995				12	58	9.1 (2.7-19)	Autism Spectrum Disorder	Leiter/Bailey scales	Diary	Total Sleep Time	-	< 55	.50
Richdale et al., 2000*				52	67	7.7 (1.8-19)	ID	Parent report	Questionnaire	-	-	-	.28
Romeo et al., 2014*				91	-	- (6-16)	Cerebral Palsy + ID	WISC-III	Questionnaire (SDSC)	-	-	< 70	.44
Sniecinska-Cooper et al., 2015				21	48	7.3 (4.5-11.0)	Williams Syndrome	None reported (associated syndrome)	Actigraphy	-	Sleep Efficiency	-	.67
Tawfik et al., 2009				16	100	10.8 (6-18)	Fragile-X Syndrome	WISC-III	Polysomnography	Total Sleep Time	-	61	.67

Meta-Analysis

Analysis Strategy

Primary analysis. Separate meta-analyses were conducted on group means and standard deviations of Sleep Time and Sleep Quality. Firstly, a Random Effects Model was tested. The Random Effects Model weights the effect of a study proportional to the number of participants it contributes to the meta-analysis. Such a weighting is problematic as the quality of studies varied dramatically. With this in mind, a Quality Effects Model was employed. The Quality Effects Model weights studies on methodological quality *as well as* number of participants. Analysis included studies in which data were reported for more than one group of people with intellectual disabilities, with the same group of typically developing people acting as a comparison on multiple occasions. This was done to avoid losing important data from an already narrow field. However, if the comparison data are replicated for comparison with multiple syndrome groups then this increases the probability of the type one error by increasing the end size of the comparison group and therefore reducing the estimate of variability in this group. To account for this, further analysis was conducted, in which only one group of people with intellectual disabilities was selected per study. If a group of people with intellectual disability of heterogeneous origin (with no syndrome or other criteria) was available, this was selected as the single group. Where this was not the case, the syndrome group with the lowest reported IQ (or lowest IQ recorded within the literature, see table 1.1.1) was chosen. For one study (Miano et al., 2004) the "younger children" group was selected over the older as this was closer to the average age across the whole analysis.

Secondary analysis. In addition to the global analyses, studies using direct measures were analysed independently as they are widely considered to have greater validity: Polysomnography has been considered a "gold standard" in the measurement of sleep, with actigraphy showing good levels of correlation to this (Van De Water, Holmes, & Hurley, 2011). Studies in which participants were identified as having specific genetic or developmental disorders were analysed separately, as were those in which participants had an intellectual disability of heterogeneous aetiology. Finally, correlations between Weighted Mean Difference and, age, proportion of male participants and IQ were undertaken to investigate whether the evidence differed across these demographic factors.

Total sleep time

Primary Analysis. Fifteen studies reported a measure of Sleep Time. The papers reviewed contained a total of 22 groups of people with intellectual disabilities, meaning typically developing comparison groups were replicated on seven occasions. The Random Effects Model (table 1.1.4) revealed a significant difference, such that groups of people with intellectual disabilities slept for shorter periods each night than did people without intellectual disabilities (Weighted Mean Difference (WMD) = -13.63, 95% Confidence Interval (CI) [-25.63, -1.63], see forest plot, figure 1.1.2). The Quality Effects Model revealed that weighting studies by their quality did not have an impact on the significance of the model (WMD = -16.58, 95% CI [-30.26, -2.90], figure 1.1.3). The mean difference equated to 23 minutes less per night for people with intellectual disabilities, ranging from 52 minutes more to 106 minutes less across stuides. Lower total sleep time was obtained for people with intellectual disabilities in all instances, except for Richdale and Prior (1995), Fukuma et al., (1974), Buckley et al., (2010) and two of the three groups from Cotton and Richdale

(2010). Each of these studies was relatively small, total experimental N = 81. When only one group per study was included, the effect became marginally non-significant, for the Random Effects Model (WMD = -13.32, 95% CI [-27.85, 1.20]), but remained significant in the Quality Effect Model (WMD = -16.58, 95% CI [-30.26, -2.90]).

Secondary analysis. The effect remained significant when only studies measuring Sleep Time directly were included, for the Random Effects Model (WMD = -14.50, 95% CI [-27.59, -1.40]) and for the Quality Effects Model (WMD = -17.47, 95% CI [-32.85, -2.08]). Splitting the analysis, revealed that the effect was significant for studies that reported on specified genetic or developmental disorders, for the Random Effects Model (WMD = -21.95, 95% CI [-37.06, -6.84]) and for the Quality Effects Model (WMD = -23.87, 95% CI [-40.85, -6.89]). Data from those (relatively few) studies that reported on a group of people with undifferentiated intellectual disability or developmental delay did not evidence significant pooled effects in either the Random Effects Model (WMD = 2.92, 95% CI [-12.51, 18.35]) or the Quality Effects Model (WMD = -1.27, 95% CI [-18.06, 15.52]). Statistical comparison showed that this represented a statistically significant difference between genetic syndrome and heterogenous intellectual disability groups ($t(20) = 2.10$, $p = .048$), though again note the relative paucity of studies that included a hetrogenous intellectual disability group.

There was no significant correlation between the size of the effect identified and the average age of participants in the study (r_s (24) = -.33, $p = .10$), the proportion of male participants (r_s (18) = -.018, $p = .93$), nor the average IQ of the group (r_s (6) = .60, $p = .12$).

Table 1.1.4. Results of the meta-analysis of sleep time: REM = Random Effects Model, QEM = Quality Effects Model. * Indicates a significant difference between intellectual disability and typical developing comparison groups

Analysis	Number of studies	Model	Number of experimental groups	Weighted Mean Difference, [95% CI]	Heterogeneity statistics	
					Cochran's Q (p)	Higgins I^2
All studies	15	REM	22	-13.63* [-25.63, -1.63]	68.41 (< .01)	69%
	15	QEM	22	-16.58* [-30.26, -2.90]	68.41 (< .01)	69%
Direct Measures Only	12	REM	17	-15.74* [-29.52, -1.95]	55.14 (< .01)	71%
	12	QEM	17	-17.47* [-32.85, -2.08]	55.14 (< .01)	71%
Heterogeneous ID	5	REM	5	2.92 [-12.51, 18.35]	7.29 (.12)	45%
	5	QEM	5	-1.27 [-18.06, 15.52]	7.29 (.12)	66%
Genetic syndromes/ developmental disorders	13	REM	17	-21.95* [-37.06, -6.84]	54.47 (< .01)	71%
	13	QEM	17	-23.87* [-40.85, -6.89,]	54.47 (< .01)	71%
Only 1 ID group per study	15	REM	15	-13.32 [-27.85, 1.20]	53.84 (< .01)	74%
	15	QEM	15	-16.58* [-30.26, -2.90]	53.84 (< .01)	74%

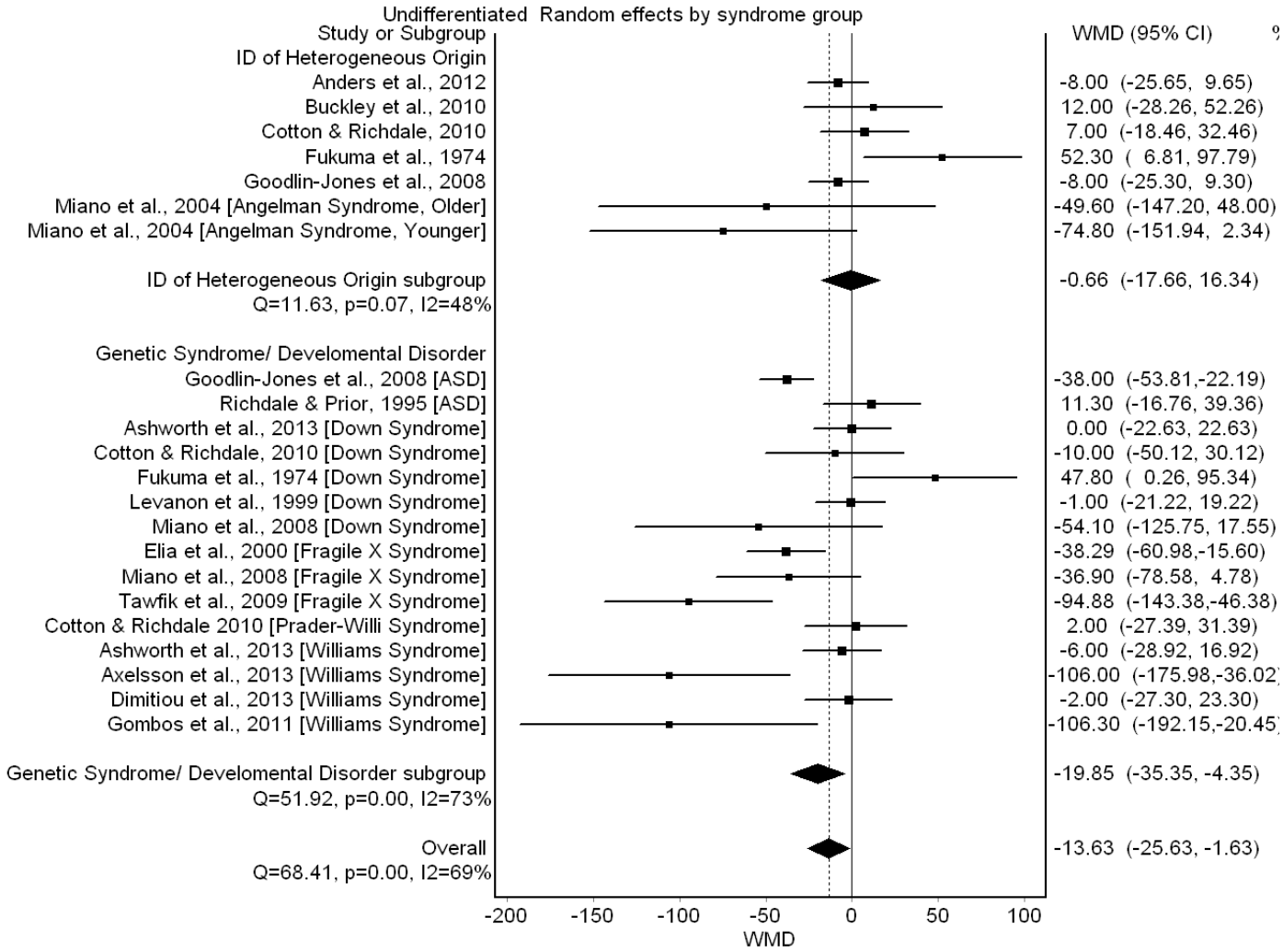


Figure 1.1.2. Forest plot of the Random Effects Model for the meta-analysis comparing sleep time in people with and without intellectual disabilities.

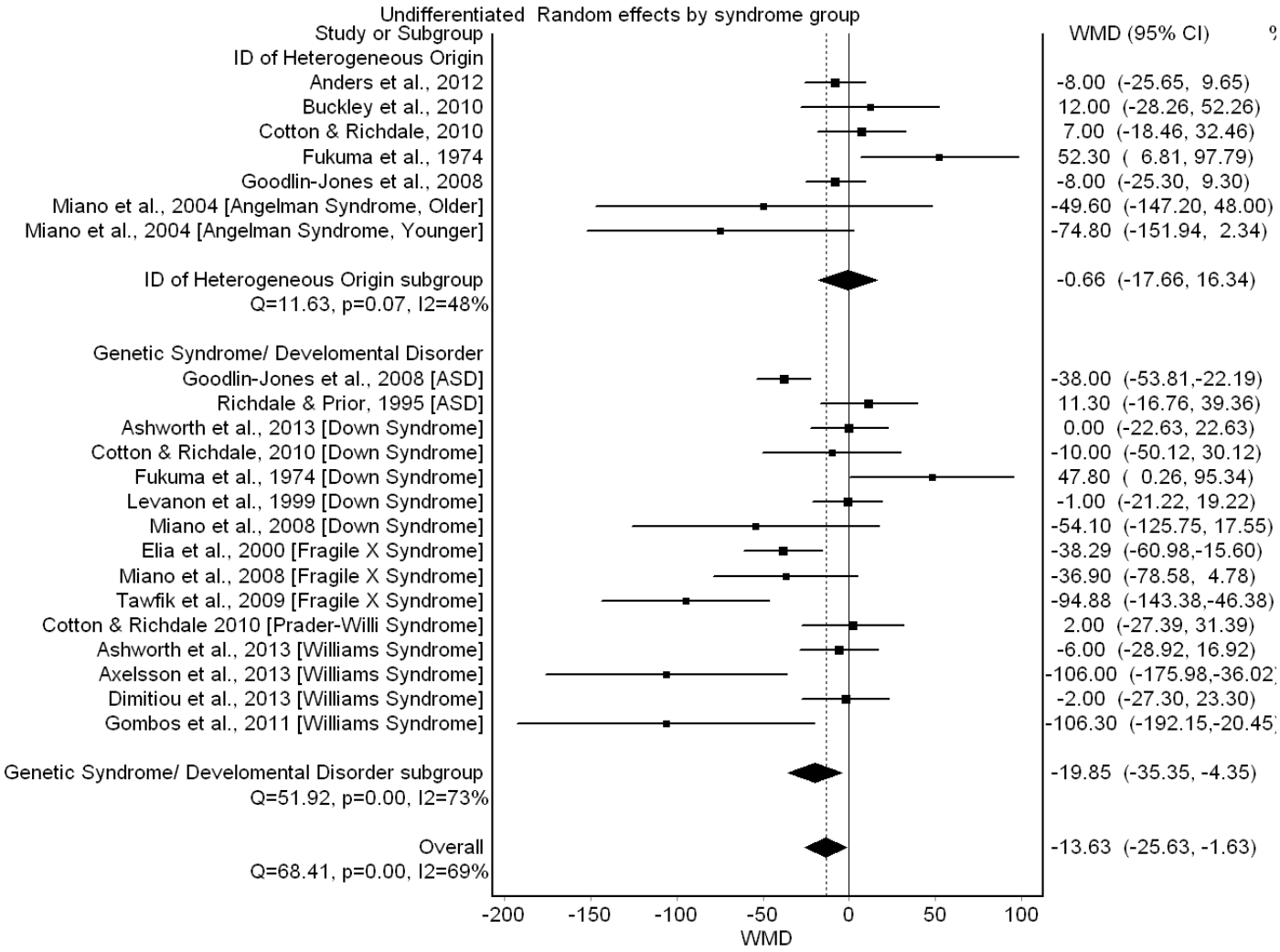


Figure 1.1.3. Forest plot of the Quality Effects Model for the meta-analysis comparing sleep time in people with and without intellectual disabilities

Sleep Quality

Primary analysis. Eighteen studies reported a measure of sleep quality. These contained 27 experimental groups of people with intellectual disabilities, so typically developing comparison groups were repeated on nine occasions. Annaz et al (2011) reported a Total Score from the Childhood Sleep Habits Questionnaire, Cotton et al., (2010) reported a questionnaire measure of Sleep Quality, Fraser et al., (2005) a questionnaire subscale on Sleep Disturbance, Ghanizadeh & Faghih (2011) on Bedtime Resistance and Sleep Duration, Maaskant et al., (2013) reported intradaily variability and all others reported Sleep Efficiency (the proportion of time spent in bed actually asleep). The Random Effects Model (table 1.1.5) revealed a significant difference, such that people with intellectual disabilities experienced significantly poorer sleep than people without intellectual disabilities (WMD = -4.56, 95% CI [-7.86, -1.27], see figure 1.1.4). Ashworth et al's (2013) group of children with Williams Syndrome and Fukuma et al's (1974) group of children with Down Syndrome were the only groups that were recorded as having better Sleep Quality than typically developing comparison groups. Notably, the data were particularly heterogeneous, Higgins I^2 = 100%, suggesting that Sleep Quality varied substantially across experimental groups. The Quality Effects Model, however, did not show a significant effect (WMD = -2.46, 95% CI [-12.48, 7.57], figure 1.1.5). The change in significance was particularly driven by one, large and high quality study (Maaskant et al., 2013), which received over 50% of the weighting in this model, and if removed reinstated the significant effect (WMD = -8.28, 95% CI [-16.00, -.56]). When only one group of people with intellectual disabilities was included, the effect remained significant for the Random Effects Model (WMD = -4.76, 95% CI [-8.91, -.60]) and non significant for the Quality Effect Model (WMD = -2.68, 95% CI [-12.34, 6.98]).

***Secondary* analysis.** Excluding studies in which Sleep Quality was measured indirectly did not affect the significance of the effect, for the Random Effects Model (WMD = -3.81, 95% CI [-5.75, -1.86]) and for the Quality Effects Model (WMD = -1.73, 95% CI [-6.84, 3.37]). Splitting the analysis, revealed that the effect was significant for studies that reported on specified genetic or developmental disorders, for the Random Effects Model (WMD = -5.98, 95% CI [-9.54, -2.43]) and for the Quality Effects Model (WMD = -8.98, 95% CI [-17.89, -1.84]). Data from those (relatively few) studies that reported on an undifferentiated intellectual disability group produced a significant pooled effect for the Random Effects Model (WMD = -.44, 95% CI [-.86, -.03]), and a marginally significant effect for the Quality Effects Model (WMD = -.59, 95% CI [-1.18, 0]). Statistical comparison showed this to represent a significant difference between the groups ($t(25) = 2.26$, $p = .033$).

There was no significant correlation between the size of the effect identified and the average age of participants in the study (r_s (25) = -.21, $p = .29$), the proportion of male participants in the sample (r_s (23) = -.039, $p = .85$) or the average IQ of participants (r_s (6) = .60, $p = .12$).

Table 1.1.5. Results of the meta-analysis of sleep quality: REM = Random Effects Model, QEM = Quality Effects Model * Indicates a significant difference between intellectual disability and typically developing comparison groups.

Analysis	Number of studies	Model	Number of experimental groups	Weighted Mean Difference, [95% CI]	Heterogeneity statistics	
					Cochran's Q (p)	Higgins I^2
All studies	18	REM	27	-4.56* [-7.86, -1.26]	21934.67 (< .01)	100%
	18	QEM	27	-2.46 [-12.48, 7.57]	21934.67 (< .01)	100%
Direct measures only	14	REM	20	-3.81* [-5.75, -1.86]	352.69 (< .01)	95%
	14	QEM	20	-1.73 [-6.84, 3.37]	352.69 (< .01)	95%
Heterogeneous ID	7	REM	8	-.44* [-.86, -.03]	13.39 (.06)	48%
	7	QEM	8	-.59* [-1.18, 0]	13.39 (.06)	48%
Genetic syndromes/ developmental disorders	15	REM	19	-5.98* [-9.54, -2.43]	951.34 (< .01)	98%
	15	QEM	19	-8.98* [-17.89, -1.84]	951.34 (< .01)	98%
Only 1 ID group per study	18	REM	18	-4.76* [-8.91, -.61]	21725.16 (< .01)	100%
	18	QEM	18	-2.47* [-13.18, -8.23]	21725.16 (< .01)	100%

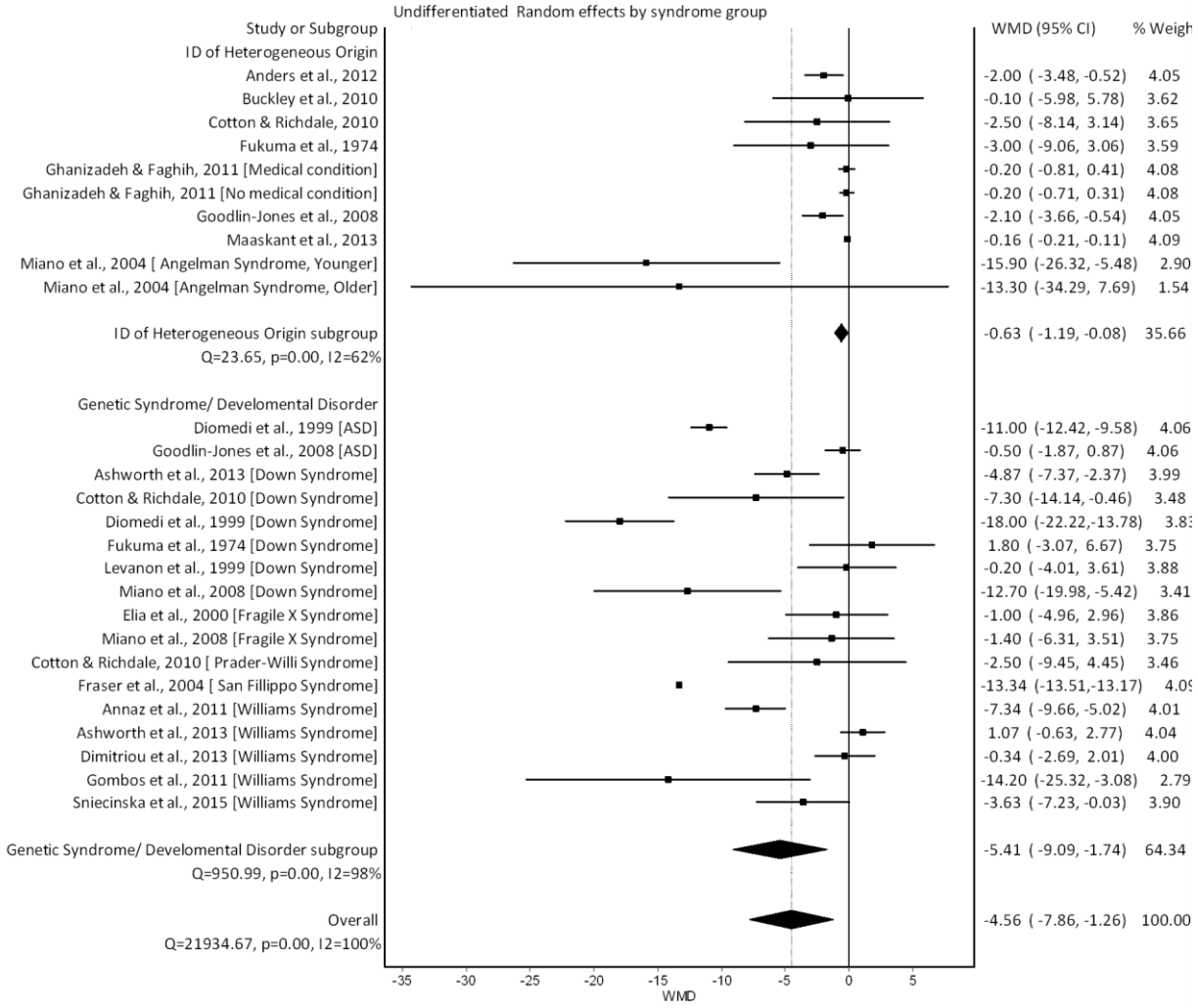


Figure 1.1.4. Forest plot of the Random Effects Model for the meta-analysis comparing sleep quality in people with and without intellectual disabilities

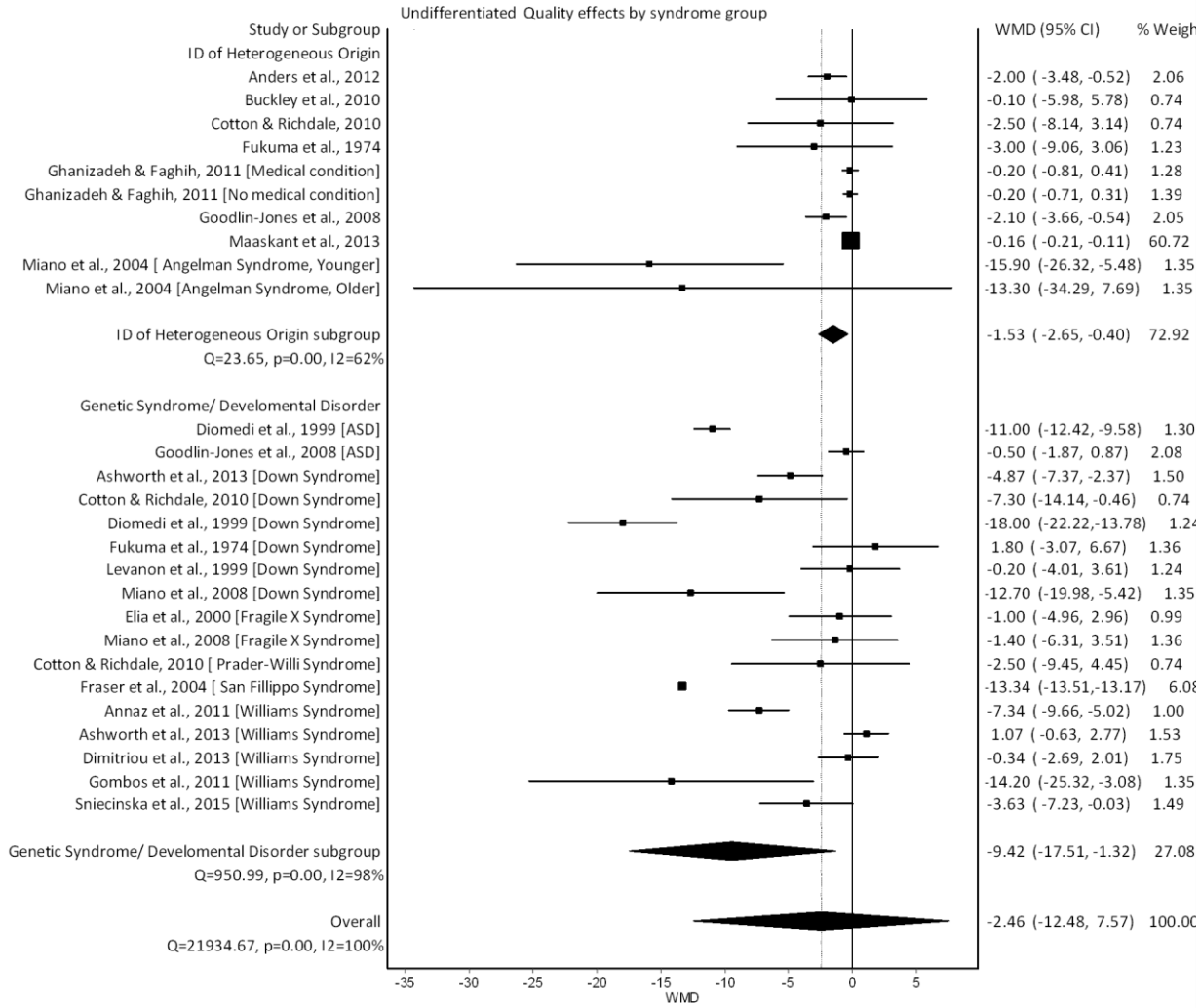


Figure 1.1.5. Forest plot of the Quality Effects Model for the meta-analysis comparing sleep quality in people with and without intellectual disabilities.

Sleep Problems

Five studies only reported comparing the frequency of sleep problems in groups of people with and without intellectual disabilities (figure 1.1.6). Only one of these studies found people without intellectual disabilities to experience more problems than people with intellectual disabilities and in this study both groups were recruited on the basis of a having diagnosis of Generalized Anxiety Disorder (Masi, Favillia, & Mucci, 2000).

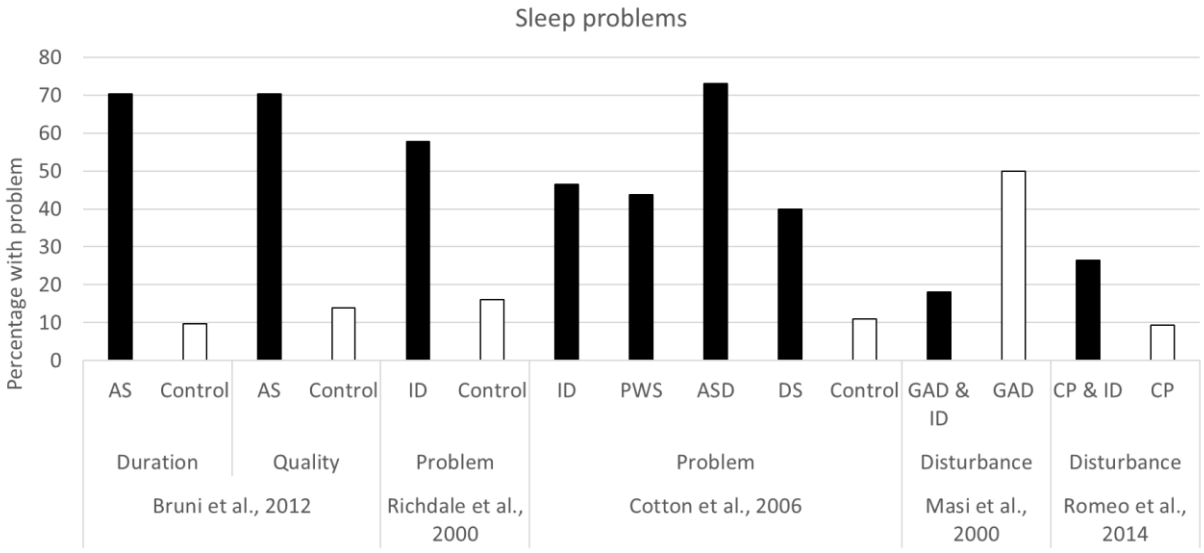


Figure 1.1.6. Percentage of participants reported as having a problem with specified aspect of sleep. AS = Angelman Syndrome, ID = Intellectual Disbaility, PWS = Prader-Willi Sydrome, DS = Down Syndrome, ASD = Autism Spectrum Disorder, GAD = Generalized Anxiety Disorder, CP = Cerebral Palsy.

Discussion

Research on sleep in people with intellectual disabilities and/ or developmental disorders has blossomed over the last 15 years (Tietze et al., 2012). The viability of this meta-analysis highlights that fact, including data from 26 different studies in which authors compared the duration and/ or quality of sleep in people with intellectual disabilities to those without intellectual disabilities. Findings of the meta-analysis suggested that there was evidence to support the hypothesis that people with intellectual disabilities experience significant deficits in both the duration and quality of their sleep. That this finding was for the most part robust in both the Random Effects and Quality Effects Models, suggested that it was not influenced by a small number of studies with poorer methodology. The nature of the papers reviewed, however, also served to highlight that a number of groups have been neglected in the literature. Similarly, reviewing the quality of the literature identified a number of consistent weaknesses in the ability of studies in this area to answer the question posed by this review.

Sleep Time and Quality in Children with Intellectual Disabilities: The state of the evidence

Sleep Time.

The meta-analysis provided evidence that the current literature supports the hypothesis that people with intellectual disabilities sleep for, on average, shorter periods each night than do people who develop typically. Across the fifteen studies included in the analysis, 446 people with intellectual disabilities averaged 23 minutes less sleep each night than did 391 people drawn from typically developing populations. Whilst Sleep Time is a rather crude measure of sleep, this simple statistic summarises and confirms the conclusions of authors and clinicians that sleep in populations of people with intellectual disabilities differs from that in

populations of people who are typically developing (Bartlett et al., 1985; Didden & Sigafoos, 2001; Quine, 1991). Shorter sleep durations in people with intellectual disabilities have been associated with increased day-time challenging behaviour (Adams, 2014), poorer attention (Lufi, 2004) and increased parent stress (Meltzer, 2007). The significant finding was shown to be evident when those studies reporting on genetic syndromes only were included, but not when only participants with intellectual disability of heterogeneous aetiology were included, with a statistical difference identified between these groups. Evidence suggested that the effect found was independent of the age of the participants tested, their IQ and of the proportions of each gender within the sample. Note, though, that an average IQ was only reported in a small number of studies.

One caveat to the conclusion that people with intellectual disabilities sleep for, on average, shorter periods than do their typically developing counterparts was that the analysis included the replication of typically developing comparison groups on multiple occasions. This was felt to be most appropriate in this case, given the small literature available. When only one group per study was analysed, the effect became marginally non-significant (for the Random Effects Model). This reflected a widening of confidence intervals (rather than a substantial change to the weighted mean difference), suggestive that further research may be required to add to the power of the evidence base.

Sleep Quality.

Using a Random Effects Model, the findings of the analysis of sleep quality showed it to be poorer in people with intellectual disabilities. Here the data were drawn from a broad range of dependent variables. Most studies reported measuring sleep efficiency directly: the proportion of time spent in bed actually asleep. Further studies, however, reported

summary variables from questionnaires. This may account for the large degree of heterogeneity identified in the analysis. That poor sleep in people with intellectual disabilities was identified from the quality variables as well as from total sleep time is further suggestive of the significance of the problems experienced. The Quality Effects Model in this case showed no significant difference. Here one study, Maaskant et al. (2013), had a significant impact on the outcome. Interestingly, this study itself reported a significant difference, suggesting that the result from the Quality Effects Model may be the result of the substantial heterogeneity of the studies, rather than higher quality studies not evidencing group differences.

For Sleep Quality the evidence of difference was apparent for both groups of people with genetic disorders/ developmental disabilities and for those of people with intellectual disability of heterogeneous origin (for the Random Effects Model). Again there was no evidence that Age, IQ or Gender made a difference to the data.

Convergence with other reviews and meta-analysis.

This is the first meta-analysis of sleep time and quality in people with intellectual disabilities, though note Elrod & Hood's (2015) recent meta-analysis comparing sleep in children with ASD to children who develop typically. There have, however, been a number of reviews on the topic. Didden and Sigafoos (2001) reviewed the literature to highlight the evidence for increased sleep problems in people with intellectual disabilities, but also noted the limitations of the literature at the time. Didden and Sigafoos (2001) cited only a single study comparing sleep in people with intellectual disabilities to people without intellectual disabilities directly (Richdale et al., 2000). Furthermore, they noted the reliance of studies on parental report and the possible bias this engenders. The current review favours the

broad conclusion that people with intellectual disabilities regularly have problems with sleep, but demonstrates the growth in literature in the intervening 15 years. More recently, Richdale and Baker (2014) reviewed all articles on sleep in developmental or intellectual disabilities from 2012 to 2013. Notably, here, nearly two thirds of papers focussed solely on ASD. Again the conclusions were consistent with those in the current review; that children with intellectual or developmental disabilities are more likely to experience problems with sleep. Much of this literature comprised reviews, case-studies, treatment studies or studies with no typically developing group for comparison. Tietze et al. (2012) noted the evidence for sleep disturbances in children with genetic syndromes and/ or intellectual disabilities and also made the case for further investigation into children with multiple disabilities. Van de Wouw et al., (2012) completed the first review of sleep in adults with intellectual disabilities. The authors concluded that evidence in this cohort was weak and largely based on "subjectively derived data" (Van de Wouw et al., 2012, p1). The current review is consistent with this, in finding only two studies on adult populations. On the other hand, showing no clear change in weighted mean differences across age groups does not support the hypothesis that sleep problems in people with intellectual disabilities significantly abate in adulthood.

Why is Sleep different in Intellectual Disabilities?

Various mechanisms have been proposed for why sleep may be impaired in people with intellectual disabilities. As with sleep in ASD, it is likely that biological, psychological and social factors all affect sleep (Richdale & Schreck, 2009). Biological factors that influence the likelihood of sleep problems include inverted melatonin cycles in people with Smith Magenis Syndrome (De Leersnyder, 2006), and proposed irregular expression of clock genes in ASD (Bourgeron, 2007). Similarly, anatomical correlates of disorders can have consequent

difficulties, such as the association between Down Syndrome and Sleep Disordered Breathing or Apnoea (Chen et al., 2013). Additionally, biological factors may include the greater likelihood of people with intellectual disabilities of having conditions such as epilepsy (Doran et al., 2002) and physical health conditions, known to have an impact on sleep quality and/ or duration. Psychological factors include evidence that sleep correlates with social and communication skills in people with intellectual disabilities (Quine,1991). Furthermore, people with intellectual disabilities are more likely to experience comorbid psychiatric conditions, such as anxiety and depression (Emerson, 2003), which have been linked to sleep problems independently (Cortesi, Giannotti, Ivanenko, & Johnson, 2010). Social routes to increased prevalence of sleep problems include parental stress (Richdale et al., 2000), which may be perpetuated by sleep problems, but may also impact on the application of sleep hygiene routines. Though not evidence for a causal role in the development of sleep problems, the efficacy of behavioural interventions for sleep disorders in children with intellectual disabilities (Wiggs & France, 2000) suggests including social factors within a formulation may be beneficial to clinicians. This meta-analysis cannot quantify the contribution of independent biopsychosocial factors, however the broad consistency of findings across different syndromes and disorders suggests that if sleep problems were a direct result of disorder-specific factors only, many of these remain unclear. With this in mind, individualized case conceptualisation may be indicated.

Scope of the findings and gaps in the literature: a manifesto for future research

The studies reviewed in this analysis investigated sleep in a broad range of syndromes and developmental disorders, suggestive of difficulties across several different conditions, which may each require in-depth future research. As referenced earlier, researchers in this field are required to make difficult choices around gaining representative samples. What was

notable was that relatively few studies reported a group collected from a broad population of people with intellectual disabilities. Whilst choosing from specific syndrome groups can make samples more homogenous and make understanding mechanisms for impairment easier, the majority of local services are aimed at populations of people with intellectual disability of heterogeneous aetiology. That no clear conclusion can be drawn for this group suggests further research is needed. It is worth noting that broader prevalence studies have tended to focus on these groups (Quine, 1991; Boyle et al., 2010), but this has been combined with the use of indirect measures. Furthermore, a number of syndromes that have previously been associated with poor sleep did not contribute a paper to this analysis as there were no studies in which sleep in these groups was compared to typically developing comparison groups (De Leersnyder, 2006; Maas et al., 2012; Young et al., 2007).

Only two of the studies analysed focused on a sample of *adults* with intellectual disabilities. The lack of research on adults from this group remains a clear deficit in the literature (van der Wouw., 2013) and the analysis in this review suggests there is currently no evidence to believe difficulties with sleep in people with intellectual disabilities recede over developmental time. Similarly, understanding the relationship between severity of intellectual disability and sleep problems was not possible with the current state of the literature. Though researchers have suggested that sleep problems increase with severity of disability (Didden et al., 2002; Quine, 1991; van de Wouw, et al., 2013), only seven studies reported IQ for their participants. This was likely because the majority of studies reported on specified syndromes, but even here an IQ score would be beneficial in understanding the nature of the group. More stark, was the paucity of reporting of adaptive functioning. That impairments to functioning retain a key place in diagnosing intellectual disabilities (American Psychiatric Association, 2013; World Health Organization, 1992), but rarely feature in research papers remains a problem that is likely to bias conclusions.

Methodological Concerns

All the papers included in the review were rated as adequate or better using the quality framework. It is clear that methodological procedures have advanced greatly since Didden and Sigafoos (2001) concluded that the majority of studies relied on parent report, which is evidently open to bias (Didden & Sigafoos, 2001). Direct measurement of sleep, through actigraphy or polysomnography has become the most common method to quantify similarities and differences in sleep quality and duration between people with and without intellectual disabilities. With this in mind, quality ratings for the measurement of sleep were generally high, suggesting that a high degree of confidence is warranted with respect to the difference found between the groups as identified in the papers. How representative these groups were, however, was thought to be less clear. No study was rated as "excellent" in identifying a sample. Identifying a fully random sample in these populations remains difficult, particularly where the sample in question relates to a rare genetic syndrome.

Limitations of this review

As well as being limited by the scope of the extant literature, this review is limited by the methodological and analytic processes undertaken. Firstly, by choosing only to examine studies in which a typically developing comparison group was included, many studies were not applicable to the research question. Whilst this has the obvious advantage of allowing for understanding of how sleep is different in people with intellectual disabilities, it ignores high quality research that has looked at, for example: individual differences in sleep in people with intellectual disabilities (Boyle et al., 2010; Quine, 1992; Van de Wouw-van Dijk, 2013) or compared prevalence of difficulties to published samples in the literature. The search was limited by focussing specifically on terms for intellectual disability. In doing so, it

may have missed papers relating to specific syndromes associated with intellectual disability (though note papers were added from recent reviews). Down syndrome was included as a search term, following van de Wouw and colleagues (2012), due to its relatively high prevalence (Sherman, Allen, Bean, & Freeman, 2007). Similarly, the requirement to cite sleep within the title, abstract or keyword may have meant missing papers which focussed on broader surveys of health. This could be more concerning as this could include papers in which sleep was measured, but not highlighted in the title, abstract or keywords if no significant difference was obtained. In including syndromes associated with intellectual disabilities (even in the absence of stated IQ testing), it is possible that some of the participants tested did not meet criteria for intellectual disability. Similarly, a choice could have been made to exclude papers that did not measure adaptive functioning. These choices meant the inclusion of more data and would both favour the null hypothesis, which was rejected in most cases. In analysis, a major methodological limitation was to include multiple groups from some studies, comparing against a single typically developing comparison group. In a broader literature, with more studies, this may have been undesirable; here it was felt important to reflect the literature. Finally, the heterogeneity of the Sleep Quality variables means any conclusions here need to be treated with caution. Though again, this limitation would favour the null hypothesis.

Conclusions

More than 30 years of research has suggested that people with intellectual disabilities experience poorer quality and shorter duration sleep than their typically developing peers. This is the first meta-analysis of the literature to examine this research question. This analysis suggests that both of these conclusions are supportable. Significant limitations exist, most notably the proportion of research based on child participants. Similarly, it is the

author's view that whilst several attractive proposals exist, the mechanism for understanding poor sleep in intellectual disabilities is not clearly identifiable from the literature as it stands.

Implications for Knowledge and Practice in Clinical Psychology

Clinical psychologists regularly work with people with intellectual disabilities, both in specialist and in universal services. Psychological models and research evidence indicate a role for poor sleep as both a potential consequence and a potential cause of psychological distress, challenging behaviour and poor day-time functioning. Sleep also interacts with broader psycho-social outcomes, for example by contributing to parent stress. Identifying and understanding reduced sleep time and poorer sleep quality in people with intellectual disabilities is therefore important for clinical practice. The review drew the following important conclusions:

- **People with intellectual disabilities sleep for shorter periods than people without intellectual disabilities.**
- **People with intellectual disabilities have poorer sleep quality than people without intellectual disabilities.**
- **Evidence of differences in sleep between people with and without intellectual disabilities is less clear in adults than children.**
- **Evidence of differences in sleep between people with and without intellectual disabilities is clearer in people with intellectual disabilities and an associated genetic syndrome or developmental disorder than those with intellectual disability of heterogeneous origin.**

References

Adams, H. L., Matson, J. L., & Jang, J. (2014). The relationship between sleep problems and challenging behavior among children and adolescents with autism spectrum disorder. *Research in Autism Spectrum Disorders*, *8*(9), 1024–1030.

American Psychiatric Association. (2013). *DSM-5 American Psychiatric Association*. Arlington: American Psychiatric Publishing.

American Sleep Disorders Association. (1997). *The international classification of sleep disorders, revised: diagnostic and coding manual*. Rochester, MN: American Sleep Disorders Association.

Anders, T., Iosif, A.-M., Schwichtenberg, A. J., Tang, K., & Goodlin-Jones, B. (2012). Sleep and Daytime Functioning: A Short-term Longitudinal Study of Three Preschool-age Comparison Groups. *American Journal on Intellectual and Developmental Disabilities*, *117*(4), 275–290.

Annaz, D., Hill, C. M., Ashworth, A., Holley, S., & Karmiloff-Smith, A. (2011). Characterisation of sleep problems in children with Williams syndrome. *Research in Developmental Disabilities*, *32*(1), 164–169.

Ashworth, A., Hill, C. M., Karmiloff-Smith, A., & Dimitriou, D. (2013). Cross syndrome comparison of sleep problems in children with Down syndrome and Williams syndrome. *Research in Developmental Disabilities*, *34*(5), 1572–1580.

Axelsson, E. L., Hill, C. M., Sadeh, A., & Dimitriou, D. (2013). Sleep problems and language development in toddlers with Williams syndrome. *Research in Developmental Disabilities*, *34*(11), 3988–3996.

Bailey, D. B., Raspa, M., Olmsted, M., & Holiday, D. B. (2008). Co-occurring conditions associated with *FMR1* gene variations: Findings from a national parent survey. *American Journal of Medical Genetics Part A*, *146A*(16), 2060–2069.

Bellugi, U., Wang, P. P., & Jernigan, T. L. (1994). Williams syndrome: An unusual neuropsychological profile. *Atypical Cognitive Deficits in Developmental Disorders: Implications for Brain Function*, *23*, 23–56.

Blair, P. S., Humphreys, J. S., Gringras, P., Taheri, S., Scott, N., Emond, A., … Fleming, P. J. (2012). Childhood Sleep Duration and Associated Demographic Characteristics in an English Cohort. *SLEEP*.

Bourgeron, T. (2007). The possible interplay of synaptic and clock genes in autism spectrum disorders. In *Cold Spring harbor symposia on quantitative biology* (Vol. 72, pp. 645–654). Cold Spring Harbor Laboratory Press. Retrieved from http://symposium.cshlp.org/content/72/645.abstract

Boyle, A., Melville, C. A., Morrison, J., Allan, L., Smiley, E., Espie, C. A., & Cooper, S.-A. (2010). A cohort study of the prevalence of sleep problems in adults with intellectual disabilities. *Journal of Sleep Research*, *19*(1-Part-I), 42–53.

Brylewski, J., & Wiggs, L. (1999). Sleep problems and daytime challenging behaviour in a community-based sample of adults with intellectual disability. *Journal of Intellectual Disability Research*, *43*(6), 504–512.

Buckley, A. W., Rodriguez, A. J., Jennison, K., Buckley, J., Thurm, A., Sato, S., & Swedo, S. (2010). Rapid eye movement sleep percentage in children with autism compared with children with developmental delay and typical development. *Archives of Pediatrics & Adolescent Medicine*, *164*(11), 1032–1037.

Cassidy, S. B., Mckillop, J. A., & Morgan, W. J. (1990). Sleep disorders in Prader-Willi syndrome. *Dysmorphology and Clinical Genetics*, *4*(1), 13–17.

Charman, T., Pickles, A., Simonoff, E., Chandler, S., Loucas, T., & Baird, G. (2011). IQ in children with autism spectrum disorders: data from the Special Needs and Autism Project (SNAP). *Psychological Medicine*, *41*(03), 619–627.

Chen, C.-C. (Jj), Spanò, G., & Edgin, J. O. (2013). The impact of sleep disruption on executive function in Down syndrome. *Research in Developmental Disabilities*, *34*(6), 2033–2039.

Colville, G. A., Watters, J. P., Yule, W., & Bax, M. (1996). Sleep problems in children with Sanfilippo Syndrome. *Developmental Medicine & Child Neurology*, *38*, 538–544.

Cortesi, F., Giannotti, F., Ivanenko, A., & Johnson, K. (2010). Sleep in children with autistic spectrum disorder. *Sleep Medicine*, *11*(7), 659–664.

Cotton, S. M., & Richdale, A. L. (2010). Sleep patterns and behaviour in typically developing children and children with autism, Down syndrome, Prader-Willi syndrome and intellectual disability. *Research in Autism Spectrum Disorders*, *4*(3), 490–500.

De Leersnyder, H. (2006). Inverted rhythm of melatonin secretion in Smith–Magenis syndrome: from symptoms to treatment. *Trends in Endocrinology & Metabolism*, *17*(7), 291–298.

Derry, C. P., Duncan, J. S., & Berkovic, S. F. (2006). Paroxysmal Motor Disorders of Sleep: The Clinical Spectrum and Differentiation from Epilepsy. *Epilepsia*, *47*(11), 1775–1791.

Didden, R., Korzilius, H., Aperlo, B. van, Overloop, C. van, & Vries, M. de. (2002). Sleep problems and daytime problem behaviours in children with intellectual disability. *Journal of Intellectual Disability Research*, *46*(7), 537–547.

Didden, R., & Sigafoos, J. (2001). A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Research in Developmental Disabilities*, *22*(4), 255–272.

Dimitriou, D., Karmiloff-Smith, A., Ashworth, A., & Hill, C. (2013). Impaired Sleep-Related Learning in Children with Williams Syndrome. *Pediatrics Research International Journal*, 1–10.

Diomedi, M., Curatolo, P., Scalise, A., Placidi, F., Caretto, F., & Gigli, G. L. (1999). Sleep abnormalities in mentally retarded autistic subjects: Down's syndrome with mental retardation and normal subjects. *Brain and Development*, *21*, 548–553.

Doi, S. A. R., Barendregt, J. J., Khan, S., Thalib, L., & Williams, G. M. (2015). Simulation Comparison of the Quality Effects and Random Effects Methods of Meta-analysis: *Epidemiology*, *26*(4), e42–e44.

Doran, S. M., Harvey, M. T., & Horner, R. H. (2006). Sleep and Developmental Disabilities: Assessment, Treatment and Outcome Measures. *Mental Retardation*, *44*(1), 13–27.

Downs, S. H., & Black, N. (1998). The feasibility of creating a checklist for the assessment of the methodological quality both of randomised and non-randomised studies of health care interventions. *Journal of Epidemiology and Community Health*, *52*(6), 377–384.

Duker, P. C., Driel, S. van, & Bercken, J. van de. (2002). Communication profiles of individuals with Down's syndrome, Angelman syndrome and pervasive developmental disorder. *Journal of Intellectual Disability Research*, *46*(1), 35–40.

Elia, M., Ferri, R., Musumeci, S. A., Del Gracco, S., Bottitta, M., Scuderi, C., … Grubar, J.-C. (2000). Sleep in subjects with autistic disorder: a neurophysiological and psychological study. *Brain and Development*, *22*(2), 88–92.

Elrod, M. G., & Hood, B. S. (2015). Sleep differences among children with autism spectrum disorders and typically developing peers: A meta-analysis. *Journal of Developmental & Behavioral Pediatrics*, *36*(3), 166–177.

Emerson, E. (2001). *Challenging behaviour: Analysis and intervention in people with severe learning disabilities*. Cambridge University Press, Cambridge.

Emerson, E. (2003). Prevalence of psychiatric disorders in children and adolescents with and without intellectual disability. *Journal of Intellectual Disability Research*, *47*(1), 51–58.

Fraser, J. (2005). Sleep disturbance in Sanfilippo syndrome: a parental questionnaire study. *Archives of Disease in Childhood*, *90*(12), 1239–1242.

Fukuma, E., Umezawa, Y., Kobayashi, K., & Motoike, M. (1974). Polygraphic study on the nocturnal sleep of children with Down's syndrome and endogenous mental retardation. *Psychiatry and Clinical Neurosciences*, *28*(4), 333–345.

Ghanizadeh, A., & Faghih, M. (2011). The impact of general medical condition on sleep in children with mental retardation. *Sleep and Breathing*, *15*(1), 57–62.

Gombos, F., Bódizs, R., & Kovács, I. (2011). Atypical sleep architecture and altered EEG spectra in Williams syndrome: Sleep architecture and EEG spectra in Williams syndrome. *Journal of Intellectual Disability Research*, *55*(3), 255–262.

Goodlin-Jones, B. L., Tang, K., Liu, J., & Anders, T. F. (2008). Sleep Patterns in Preschool-Age Children With Autism, Developmental Delay, and Typical Development: *Journal of the American Academy of Child & Adolescent Psychiatry*, *47*(8), 930–938.

Hering, E., Epstein, R., Elroy, S., Iancu, D. R., & Zelnik, N. (1999). Sleep patterns in autistic children. *Journal of Autism and Developmental Disorders*, *29*(2), 143–147.

Hoffman, C. D., Sweeney, D. P., Gilliam, J. E., Apodaca, D. D., Lopez-Wagner, M. C., & Castillo, M. M. (2005). Sleep problems and symptomology in children with autism. *Focus on Autism and Other Developmental Disabilities*, *20*(4), 194–200.

Levanon, A., Tarasiuk, A., & Tal, A. (1999). Sleep characteristics in children with Down syndrome. *The Journal of Pediatrics*, *134*(6), 755–760.

Lufi, D. (2014). The relationship between sleep patterns and attention levels. *Neurocase*, *20*(5), 591–598.

Maas, A. P. H. M., Didden, R., Korzilius, H., & Curfs, L. M. G. (2012). Exploration of differences in types of sleep disturbance and severity of sleep problems between individuals with Cri du Chat syndrome, Down's syndrome, and Jacobsen syndrome: A case control study. *Research in Developmental Disabilities*, *33*(6), 1773–1779.

Masi, G., Favillia, L., & Mucci, M. (2000). Generalized Anxiety Disorder in Adolescents and Young Adults with Mild Mental Retardation. *Psychiatry*, *63*(1), 54–64.

Maulik, P. K., Mascarenhas, M. N., Mathers, C. D., Dua, T., & Saxena, S. (2011). Prevalence of intellectual disability: A meta-analysis of population-based studies. *Research in Developmental Disabilities*, *32*(2), 419–436.

McGuire, B. E., Daly, P., & Smyth, F. (2010). Chronic pain in people with an intellectual disability: under-recognised and under-treated? *Journal of Intellectual Disability Research*, *54*(3), 240–245.

Mervis, C. B., & Klein-Tasman, B. P. (2000). Williams syndrome: Cognition, personality, and adaptive behavior. *Mental Retardation and Developmental Disabilities Research Reviews*, *6*(2), 148–158.

Miano, S., Bruni, O., Elia, M., Scifo, L., Smerieri, A., Trovato, A., … Ferri, R. (2008). Sleep phenotypes of intellectual disability: A polysomnographic evaluation in subjects with Down syndrome and Fragile-X syndrome. *Clinical Neurophysiology*, *119*(6), 1242–1247.

Miano, S., Bruni, O., Leuzzi, V., Elia, M., Verrillo, E., & Ferri, R. (2004). Sleep polygraphy in Angelman syndrome. *Clinical Neurophysiology*, *115*(4), 938–945.

Pilcher, J. J., Ginter, D. R., & Sadowsky, B. (1997). Sleep quality versus sleep quantity: Relationships between sleep and measures of health, well-being and sleepiness in college students. *Journal of Psychosomatic Research*, *42*(6), 583–596.

Quine, L. (1992). Severity of sleep problems in children with severe learning difficulties: description and correlates. *Journal of Community & Applied Social Psychology*, *2*(4), 247–268.

Randall, D. (2013). *Dreamland: Adventures in the Strange Science of Sleep*. London: W. W. Norton & Company.

Richards, C., Jones, C., Groves, L., Moss, J., & Oliver, C. (2015). Prevalence of autism spectrum disorder phenomenology in genetic disorders: a systematic review and meta-analysis. *The Lancet Psychiatry*, *2*(10), 909–916.

Richdale, A., Francis, A., Gavidia-Payne, S., & Cotton, S. (2000). Stress, behaviour, and sleep problems in children with an intellectual disability. *Journal of Intellectual and Developmental Disability*, *25*(2), 147–161.

Richdale, A. L. (1999). Sleep problems in autism: prevalence, cause, and intervention. *Developmental Medicine & Child Neurology*, *41*(01), 60–66.

Richdale, A. L., & Prior, M. R. (1995). The sleep/wake rhythm in children with autism. *European Child & Adolescent Psychiatry*, *4*(3), 175–186.

Richdale, A. L., & Schreck, K. A. (2009). Sleep problems in autism spectrum disorders: Prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews*, *13*(6), 403–411.

Roizen, N. J., & Patterson, D. (2003). Down's syndrome. *Lancet*, *361*, 1281–1289.

Roth, T. (2007). Insomnia: definition, prevalence, etiology, and consequences. *Journal of Clinical Sleep Medicine: JCSM: Official Publication of the American Academy of Sleep Medicine*, *3*(5 Suppl), S7.

Schreck, K. (2004). Sleep problems as possible predictors of intensified symptoms of autism*1. *Research in Developmental Disabilities*, *25*(1), 57–66.

Sherman, S. L., Allen, E. G., Bean, L. H., & Freeman, S. B. (2007). Epidemiology of Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews*, *13*(3), 221–227.

Smith, M. T., Perlis, M. L., Smith, M. S., Giles, D. E., & Carmody, T. P. (2000). Sleep quality and presleep arousal in chronic pain. *Journal of Behavioral Medicine*, *23*(1), 1–13.

Sniecinska-Cooper, A. M., Iles, R. K., Butler, S. A., Jones, H., Bayford, R., & Dimitriou, D. (2015). Abnormal secretion of melatonin and cortisol in relation to sleep disturbances in children with Williams syndrome. *Sleep Medicine*, *16*(1), 94–100.

Stores, G., & Stores, R. (2013). Sleep disorders and their clinical significance in children with Down syndrome: Review. *Developmental Medicine & Child Neurology*, *55*(2), 126–130.

Sutton, D. A., Moldofsky, H., & Badley, E., M. (2001). Insomnia and health problems in Canadians. *Sleep*, *24*(6), 665.

Tamakoshi, A., & Ohno, Y. (2004). Self-reported sleep duration as a predictor of all-cause mortality: results from the JACC study, Japan. *SLEEP-NEW YORK THEN WESTCHESTER-*, *27*(1), 51–54.

Tani, P., Lindberg, N., Nieminen-von Wendt, T., Von Wendt, L., Alanko, L., Appelberg, B., & Porkka-Heiskanen, T. (2003). Insomnia is a frequent finding in adults with Asperger syndrome. *BMC Psychiatry*, *3*(1), 1.

Tawfik, T. Z., Hashem, S., Zaki, M. A., El-Shazly, N., Hegazy, M. M., El-Meguid, N. A., & Hashem, H. S. (2009). Sleep Disorders in Fragile X Syndrome. *Egyptian Journal of Psychiatric Neurosurgery*, *46*(2), 445–453.

Tietze, A.-L., Blankenburg, M., Hechler, T., Michel, E., Koh, M., Schlüter, B., & Zernikow, B. (2012). Sleep disturbances in children with multiple disabilities. *Sleep Medicine Reviews*, *16*(2), 117–127.

Valstar, M. J., Marchal, J. P., Grootenhuis, M., Colland, V., & Wijburg, F. A. (2011). Cognitive development in patients with Mucopolysaccharidosis type III (Sanfilippo syndrome). *Orphanet J Rare Dis*, *6*, 43.

Van De Water, A. T. M., Holmes, A., & Hurley, D. A. (2011). Objective measurements of sleep for non-laboratory settings as alternatives to polysomnography - a systematic review: A systematic review of objective sleep measures. *Journal of Sleep Research*, *20*(1pt2), 183–200.

van de Wouw, E., Evenhuis, H. M., & Echteld, M. A. (2012). Prevalence, associated factors and treatment of sleep problems in adults with intellectual disability: A systematic review. *Research in Developmental Disabilities*, *33*(4), 1310–1332.

Van de Wouw-van Dijk, E. van de. (2013). *Sleep and Sleep-wake Rhythm in Older Adults with Intellectual Disabilities*. s.n.], S.l.

Whittington, J., Holland, A., Webb, T., Butler, J., Clarke, D., & Boer, H. (2004). Cognitive abilities and genotype in a population-based sample of people with Prader–Willi syndrome. *Journal of Intellectual Disability Research*, *48*(2), 172–187.

Wiggs, L., & France, K. (2000). Behavioural treatments for sleep problems in children and adolescents with physical illness, psychological problems or intellectual disabilities. *Sleep Medicine Reviews*, *4*(3), 299–314.

Wiggs, L., & Stores, G. (1996). Wiggs_et_al-1996-Journal_of_Intellectual_Disability_Research.pdf. *Journal of Intellectual Disability Research*, *40*(6), 518–528.

World Health Organisation. (1992). *The ICD-10 classification of mental and behavioural disorders: clinical descriptions and diagnostic guidelines.* Geneva: World Health Organisation.

Young, D., Nagarajan, L., de Klerk, N., Jacoby, P., Ellaway, C., & Leonard, H. (2007). Sleep problems in Rett syndrome. *Brain and Development*, *29*(10), 609–616.

CHAPTER II: SLEEP PROBLEMS IN AUTISM SPECTRUM DISORDERS: A COMPARISON TO SLEEP IN TYPICALLY DEVELOPING CHILDREN USING ACTIGRAPHY, DIARIES AND QUESTIONNAIRES

Abstract

Background. Prevalence rates of sleep problems in children with Autism Spectrum Disorders (ASD) are estimated at between 44 and 83%. Direct measurement of sleep affords the opportunity for quantifying differences between sleep in children with and without ASD and enables the examination of the behavioural consequences of poor sleep. In this study, we use actigraphy to compare sleep in children with and without ASD and examine the relationship between poor sleep, daytime sleepiness, impulsivity and overactivity.

Method. Sixteen children with ASD, performance IQ within the normal range and a parentally-reported sleep problem were compared to children without ASD, matched for age and gender. Five to seven days of actigraphy data were collected for both groups, alongside sleep diaries and questionnaires on sleep, day-time functioning and behaviour.

Results. Questionnaire data revealed that children with ASD had a higher prevalence of sleep problems than children without ASD. Although there were no differences between the groups on actigraphy data or diary measures of sleep, significant differences were noted in problems with parasomnias (a frequent problem for 79% of the children with ASD), sleep onset (43%) and day-time sleepiness (64%). Linear Mixed Effects modelling showed that while sleepiness predicted impulsivity/overactivity on the same day, sleepiness and impulsivity/overactivity were not predicted by sleep during the previous night.

Conclusions. Although there is good evidence that children with ASD sleep for shorter intervals and with poorer quality than children without ASD, evidence in children with ASD and without intellectual disability is equivocal. Here, even though children with ASD were recruited on the basis of a parent-reported sleep problem, actigraphy found no differences between their sleep and that of typically developing controls. Parent reports of sleep problems often reflected problems with parasomnias, sleep onset or sleepiness.

Introduction

Background

Sleep problems in children with Autism Spectrum Disorders (ASD) are commonly reported by their parents (ASD; Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Didden & Sigafoos, 2001; Höglund Carlsson et al., 2013; Richdale & Schreck, 2009; Wiggs & Stores, 2004) and are among the most prevalent comorbid conditions experienced by children with ASD (Xue Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2007). Comparison studies report that children with ASD experience worse sleep than their typically developing (TD) peers (Allik, Larsson, & Smedje, 2006; Elrod & Hood, 2015; Richdale & Schreck, 2009). Estimates for the prevalence of sleep problems in children with ASD vary from 44-83%, in comparison to only 9-50% in TD comparison groups (Elrod & Hood, 2015; Richdale & Schreck, 2009). Patzold, Richdale, & Tonge (1998) noted problems with sleep onset and maintenance as particularly widespread, but also significant levels of sleep-disordered breathing and parasomnias. Sleep problems are not only more prevalent in ASD, but also vary systematically with autistic symptomology in high functioning groups, such that severity of ASD symptoms predicts poor sleep (Hoffman et al., 2005; Schreck, 2004).

Methods for measuring sleep in children have developed from parent report via questionnaires or diaries, to the use of direct measures gained from polysomnography and actigraphy. Whilst initial evidence from parent report and diaries helped identify genuine parent concern, parent report has been understood to allow for significant overestimating of sleep problems (Goodlin-Jones, Tang, Liu, & Anders, 2008; Hering, Epstein, Elroy, Iancu, & Zelnik, 1999). One proposal has been that sleep is actually similar in children with and without ASD (Schreck & Mulick, 2000), with differences being a manifestation of the overall strain on parents of children with ASD (Hering et al., 1999; Wiggs et al., 2005). Measuring

sleep directly is likely to provide a more accurate representation of the prevalence and severity of sleep problems in children with ASD (Goldman et al., 2009).

In spite of often sharing a single diagnosis (particularly under new DSM-V criteria, American Psychiatric Association, 2013), children with ASD are far from being a homogeneous group and many experience comorbid conditions. Most notably, 44-70% of people with ASD also have a comorbid intellectual disability (Fombonne, Quirke, & Hagen, 2011; La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004). This is particularly relevant to sleep because of the documented relationship between sleep problems and intellectual disability (Bartlett, Rooney, & Spedding, 1985; Berkman, 2006; Quine, 1992; Richdale, Francis, Gavidia-Payne, & Cotton, 2000; Richdale & Baker, 2014; Surtees, Oliver, Jones, Evans, & Richards, submitted, Chapter 1; Tietze et al., 2012). With this in mind, one hypothesis is that increased sleep problems identified in the broader ASD population are an artefact of the increased number of children with intellectual disability in this group. Alternatively, there is some evidence from parent report data that suggests that sleep problems are more prevalent in children with ASD (of varying cognitive abilities) than samples of children with developmental delay (Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008). This conflict makes measuring sleep of children with ASD and no comorbid intellectual disability directly particularly important.

Current evidence from direct measurement of sleep in ASD

A recent systematic review (Elrod & Hood, 2015) identified studies that used direct measures to compare sleep in children with and without ASD. The studies identified included those using polysomnography (in six cases), actigraphy (in four cases) and one study in which both were employed. Polysomnography uses measure brain activity (using

electroencephalography), oxygen saturation in the blood, respiratory rate, heart-rate and movement and is considered the gold standard within sleep research (Michaelson, Allan, Chaney, & Mair, 2006), but is limited by measuring across a short time period. In all but one study in children with ASD (Goldman et al., 2009), only a single night was used in the final analysis. Studies employing actigraphy measure only movement, typically with a small watch-like device on the wrist. Actigraphy is considered less accurate than polysomnography, particularly because of its poor sensitivity in identifying restful waking periods (Sadeh & Acebo, 2002). Conversely, actigraphy is easier to employ for longer periods (Allik et al., 2006; Goodlin-Jones et al., 2008; Souders et al., 2009; tested for the recommended five nights or more) and for testing in the child's natural environment. Restricted preferences are a diagnostic feature of ASD, predictive of specific difficulties in adapting to new environments. With this in mind, employing actigraphy, as recommended for 5-7 nights in the child's home, may be a more sensitive and accurate measure of typical sleep patterns for children with ASD. However, polysomnography would, of course, remain more valid for identifying time spent in different stages of sleep, apnoea, sleep-disordered breathing and parasomnias.

Elrod and Hood's (2016) meta-analysis of comparisons between sleep in children with ASD and TD controls showed that across the 11 studies, there were significant differences between children with ASD and TD on Total Sleep Time (on average 32.8 minutes per day shorter in ASD), Sleep Latency (10.9 minutes longer per day in ASD) and Sleep Efficiency (1.9% per day). Elrod and Hood (2016) also tested whether the effect was moderated by the method used or the exclusion of children with intellectual disabilities, children on medications or children with seizure disorders. The only significant moderator identified was the effect of excluding children with intellectual disabilities on Total Sleep Time. When the analysis only included the three studies which did not include children with

intellectual disabilities (two using polysomnography and one actigraphy), there were no significant differences in Total Sleep Time. This finding is perhaps surprising, given the well-documented relationship between ASD and insomnia (Richdale & Shreck, 2009). There were differences identified in Sleep Latency and Sleep Efficiency, but these could be explained by parents of children with ASD setting earlier bedtimes for their children[1] (Allik et al., 2006). Alternatively, parents of children with ASD may *have* to set earlier bedtimes to achieve equivalence of Total Sleep Time *because* of long sleep latencies and poor sleep efficiency. With this in mind, an actigraphy study measuring the sleep of children with parent-identified sleep problems is timely.

Allik et al. (2006) completed the only published study employing actigraphy for five to seven nights for a group of children with ASD and no comorbid intellectual disability (Goldman et al., 2009, only employed actigraphy for two nights). Nineteen children with Asperger's Syndrome and 13 with High-Functioning Autism (age range eight to 13) were recruited pseudo-randomly from specialist services in Sweden and compared to 32 TD controls. Interestingly, in spite of not recruiting children with intellectual disabilities, the majority of the children with ASD still attended "classes or schools for children with various special needs" (Allik et al., 2006, p588). Within the sample of children with ASD, 59.2% reported sleep problems, in comparison to only 9% in the control group. Consistent with Elrod and Hood's (2016) meta-analysis, they identified no significant difference in Total Sleep Time: averaging 511 minutes (on school days) to 514 minutes (on weekends) for children with ASD, in comparison to 523 (weekend) to 522 (weekday) minutes for controls. However, children with and without ASD differed on both Sleep Efficiency: 87% for ASD, 90% for controls, and Sleep Latency: 22-32 (weekday-weekend) minutes for ASD, 11-16 (weekday-

[1] If bedtime is set earlier, children may take longer to get to sleep and spend longer in bed overall, thus decreasing sleep efficiency.

weekend) minutes for TD. Whilst Allik et al.'s study (2006) provides evidence for similarities and differences between children with ASD with no concurrent intellectual disability and TD controls, there remain limitations. Firstly, there was no evidence provided of the children's scores on cognitive testing (though these were assessed in patient notes), on a standardized assessment for ASD or on a measure of adaptive functioning. This limits understanding of the typicality of the group and does not allow for understanding how autistic symptomology, cognitive abilities or adaptive functioning may have related to sleep. Secondly, the authors do not report what proportion of children may have experienced a specified sleep problem. Finally, it is difficult to draw firm conclusions from a single study of this kind.

Explaining poor sleep in ASD

Whilst researchers broadly agree on the increased prevalence of sleep problems in children with ASD, the mechanism underlying these problems is less clear (Cortesi et al., 2010). Richdale and Schreck (2009) proposed a model that emphasises biological, psychological *and* social factors that predispose children with ASD to experiencing sleep problems and further perpetuate them. Biological mechanisms proposed include circadian rhythm dysfunction, irregular expression of clock genes and abnormal melatonin release or synthesis (Bourgeron, 2007; Richdale & Schreck, 2009). Importantly, it has been noted that night-time melatonin levels were inversely related to severity of autistic symptoms (Tordjman, Anderson, Pichard, Charbuy, & Touitou, 2005).

Proposed psychological mechanisms have mainly focussed on the psychological and behavioural features of ASD: deficits in communication, social interaction and restricted interested/ repetitive behaviours. This hypothesis is supported by the evidence that poor sleep correlates not only with overall ASD symptomatology (Hoffman et al., 2005; Malow et

al., 2006; Schreck et al., 2004), but also communication (Quine, 1992), social skills (Hoffman et al., 2005) and stereotypic behaviours (Schreck et al., 2004) independently. There is little evidence, however, for the direction of causality of this effect and poor sleep could precipitate an increase in symptom severity. One possibility is that sleep problems exacerbate difficulties in ASD symptoms (Schreck et al., 2004), perhaps mediated through established links between poor sleep and executive function (Jones & Harrison, 2001). Another is that poor sleep may be precipitated by increased challenging behaviour associated with more severe ASD symptoms (Wiggs & Stores, 1996). ASD symptomology is also confounded with intellectual disability, which predicts poor sleep, so may mean that the association between ASD symptomology and sleep is in no way causal. Further psychological mechanisms proposed have included potential comorbidities with mood disorders, such as anxiety and/ or depression (Cortesi et al., 2010) and Attention Deficit/ Hyperactivity Disorder (ADHD; Liu, Hubbard, Fabes, & Adam, 2006).

The contribution of social mechanisms to sleep problems has received less attention in the literature (Richdale & Schreck, 2009). Richdale (1999) noted that children with ASD and sleep problems often have unusual sleep routines, particularly around settling at bed-times. There is also good evidence that parents of children with ASD experience high levels of stress (higher even than parents of children with other disabilities; Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001) and parent stress levels are correlated with poor sleep (Richdale et al., 2000); though again direction of causality is unclear.

Richdale and Schreck's (2009) comprehensive and clinically instructive biopsychosocial model hypothesises an important interaction between genetic, biological, psychological and environmental factors and also presents an interconnected framework that views sleep problems both as a result and a perpetuator of individual and interpersonal

difficulties. For example, the abnormal expression of clock genes may lead to circadian rhythm irregularities that may precipitate poor sleep. In turn, poor sleep may exaggerate social deficits that then impact on circadian rhythms.

Associations between poor sleep and day-time functioning

Sleep problems present a significant clinical problem for children and families who experience them (Shang, Gau, & Soong, 2006). In children who develop typically, there is evidence of a link between poor sleep and detrimental effects on children's mood, cognition and behaviour (Malow et al., 2012). Children with ASD and sleep problems have been suggested to have an increased likelihood of mood disorders (Tani et al., 2003) and daytime challenging behaviour (Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2012; Didden, Korzilius, Aperlo, Overloop, & Vries, 2002; Hoshino, Watanabe, Yashima, Kaneko, & Kumashiro, 1984; Rzepecka, McKenzie, McClure, & Murphy, 2011; Wiggs & Stores, 2004). As indicated above, though, inferring causality between poor sleep and day-time behaviour is difficult. This is particularly the case in children with ASD, for whom complex networks are likely evident, linking physiological states and traits, core components of ASD, secondary cognitive, affective and behavioural features prevalent in ASD, comorbid conditions and also, familial and environmental interactions to sleep quality.

As well as examining sleep itself, the current study focusses on links to daytime sleepiness, impulsivity and overactivity. One reason to predict a relationship between sleep, and overactivity and impulsivity is that sleep problems have been identified in numerous studies in children with ADHD, for which overactivity and impulsivity are diagnostic criteria (Ball, Tiernan, Janusz, & Furr, 1997; Harvey, Bapker, Horner, & Blackford, 2001; LeBourgeois, Avis, Mixon, Olmi, & Harsh, 2004). It has also been suggested that daytime sleepiness is

more common in this group (Lecendreux, Konofal, Bouvard, Falissard, & Mouren-Simeoni, 2000; Palm, Persson, Bjerre, Elmqvist, & Blennow, 1992). Symptoms of ADHD are common in children with ASD (Leitner, 2014) and so the relationship between sleep, sleepiness, and impulsivity and overactivity is potentially an important one. Pavonen et al. (2008) reported higher levels of day-time sleepiness in children with ASD (Paavonen et al., 2008), suggestive of a simple link to potential for impairments in day-time functioning. Interestingly, there have also been links made between poor sleep and hyperactivity in children with ASD (Goldman et al., 2010), which could be linked to sleepiness and inattention. Goldman et al. (2010) found a correlation between night-time wakings and parentally-reported hyperactivity (on the Parental Concerns Questionnaire). An open question is how, or if, these difficulties are linked temporally. A correlation cannot distinguish between the hypothesis that hyperactivity precipitates poor sleep (and resultant daytime sleepiness) and the hypothesis that poor sleep precipitates day-time sleepiness, which predicts hyperactivity.

Rationale

A body of research has examined the prevalence, severity, nature and cause of sleep problems in children with ASD. However, very few of these studies have measured sleep directly, through actigraphy or polysomnography. The majority of studies that have done so have included children with a range of cognitive and functional abilities. On the one hand this is important in understanding sleep across the broader population of children with ASD. Conversely, it is difficult to identify the independent and complimentary roles for intellectual disability and ASD in predicting the prevalence and severity of poor sleep. The current study used actigraphy to compare sleep in children with ASD to a TD comparison group. Only

children with a parent-reported sleep problem were recruited to the ASD group to understand the nature of these problems (rather than estimate the prevalence of sleep problems in people with ASD more broadly). None of the participants tested had a comorbid intellectual disability, defined as performance IQ < 70. Parent diaries were also undertaken to compare parent reports of sleep in the two groups and evaluate their accuracy using actigraphy. Sleep questionnaires investigated parent reports of specific problems with sleep. All children with ASD were tested using the Wechsler Abbreviated Scale of Intelligence, the Vineland Adaptive Behavior Scale and a range of questionnaires, to identify the relationship between poor cognitive functioning, adaptive behaviour, daytime functioning and sleep. Finally, daytime sleepiness and impulsivity/overactivity were measured as two potential day-time difficulties that may relate to poor sleep.

Aims

This study compared children with ASD who had a parent reported sleep problem and typically developing children in three key areas:

I. Similarities and differences in sleep, measured directly by actigraphy.
II. Similarities and differences in parent reports of sleep.
III. Frequency of different parent-reported sleep problems.

Data from children with ASD were further examined to investigate:

IV. Similarities and differences between actigraphy measures and parent reports.
V. Individual differences in sleep quantity and quality using correlations.
VI. The temporal relationship between Sleep, Sleepiness and Overactivity/ Impulsivity.

Method

Participants.

Sixteen children aged five to 13 years (mean age = 9.8; 63% male), with Autism Spectrum Disorders (table 1.2.1) were matched on age and gender to 16 typically developing children (table 1.2.2). All families of children with ASD on a variety of databases[2] were contacted and invited to take part if their children fulfilled three criteria: an existing diagnosis of ASD, a current sleep problem and being aged five-15. Three further children were recruited to the original sample, but were excluded from this study. Two of these children were excluded because sleep diaries were incomplete. The final child was excluded after scoring below the normal range on cognitive testing. The final sample included four families in which more than one child with ASD took part (total N = 10). Parents of two children in the final sample did not return questionnaires within the identified time period and so are not included in this part of the analysis.

Typically developing children were recruited through friends, families and acquaintances of researchers and students at the university. A larger sample of 44 children were recruited, with the final sample selected to match for age and gender. All typically developing children selected scored below cut-off on the Social Communication Questionnaire (Rutter, Bailey & Lord, 2003; indicative of potential ASD).

[2] Databases included: A local area database of children with ASD in the West Midlands (UK), a research centre database (including children with a variety of genetic syndromes and developmental disorders), a database of children with ASD from a second research group and a small database of parents who attended a charity-led workshop on sleep problems in developmental disorders.

Table 1.2.1. Participant characteristics of children with ASD recruited for the study

Participant	Age (Years)	Gender	Performance IQ[1]	Adaptive Behaviour[2]
1	12	Female	112	68
2	7	Female	95	89
3	8	Male	131	75
4	13	Male	89	76
5	14	Male	94	79
6	11	Female	111	78
7	8	Male	110	80
8	12	Male	110	74
9	10	Male	112	62
10	9	Male	70	68
11	10	Male	76	66
12	7	Female	77	126
13	11	Female	112	68
14	10	Male	127	97
15	5	Male	>103	65
16	10	Female	84	67

[1]Score on the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999)
[2]Standard Score on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984)

Table 1.2.2 Group-level participant characteristics

	ASD Group, Mean (Range)	TD Group, Mean (Range)	*t*	*p*
Age (Years)	9.8 (5-13)	9.5 (5-14)	.83	.22
Gender (%Male)	62.5%	62.5%	-	-
Social Communication Questionnaire	27.62 (15-35)	3.29 (0-13)	11.17	< .001
Performance IQ	100.6 (70-131)	-	-	-
VABS Adaptive Level	77.4 (62-126)	-	-	-

Procedure

On recruitment, children with ASD and their parents attended a research centre for direct assessments of cognitive abilities and autistic symptomology. An assessment of adaptive behaviour was completed with parents by interview over the telephone. All assessments were completed in the 15 weeks prior to the week in which sleep was measured directly using actigraphy. Direct assessments of IQ, autistic symptomology and adaptive behaviour were not completed for typically developing children. All children in the typically developing comparison group attended mainstream primary or secondary schools.

Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000).

Diagnosis of Autism/ Autism Spectrum Disorder was corroborated by completion of the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2000).

Mullen Scales of Early Learning (Mullen, 1995).

To provide consistency of testing across a sample that varied in age and were initially of unknown cognitive ability, all children completed the Mullen Scales of Early Learning (Mullen, 1995). The Mullen scales are aimed at children younger than 68 months, but are often also used as a measure for children with intellectual disabilities. Children were considered to have reached ceiling on this measure if it was not possible to gain a ceiling estimate of their abilities on any of the subscales.

Wechsler Abbreviated Scale of Intelligence (WASI, Wechsler, 1999)

All children who achieved ceiling scores on the Mullen were tested using the performance subscales of the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999). All children who reached ceiling on the receptive language or expressive language subscales of the Mullen scales also completed the verbal subscales of the WASI. The WASI (Wechsler, 1999) is a widely used measure of cognitive ability in children (aged six-18) and adults, that included four individual tests. The two subscales are well-validated and can be combined into an overall IQ score reliably. One five-year old boy was below the minimum age suggested for the WASI, but scored above average for a six-year old on the test, suggesting he had no cognitive impairment (his performance IQ was not included in examination of individual differences in sleep).

Vineland Adaptive Behavior Scales (VABS, Sparrow et al., 1984).

The Vineland Adaptive Behavior Scales (Sparrow et al., 1984) measure adaptive behaviour and functioning in children of all ages. They provide individual subscales on visual reception, fine motor, gross motor, expressive language and receptive language skills, as well as an overall standard score.

Actigraphy.

Each child wore an Actiwatch (Phillips- Actiware) on the wrist for a continuous period of seven to eight days, in line with Acebo et al.'s (1999) guidance on obtaining reliable measures of sleep through actigraphy. Children and their parents were directed that, if possible, the watch should be worn at all times. Sleep intervals were calculated automatically using the Actiware software. Data cleaning was undertaken to remove

artefacts that can make actigraphy data unreliable (Acebo et al., 1999). Sleep intervals were altered if the watch was removed or the interval missed a significant period of sleep within the child's reported time in bed (for full details of data cleaning and coding see appendix 1.2.1). Variables extracted from the actigraphy measure included Bed-time (BT; the time at which children entered a restful state), Get-up time (GT; the end time of the final period of sleep in the morning), Onset Latency (OL; the time between BT and the first period encoded as sleep), Wake After Sleep Onset (WASO; The amount of time after first period of sleep spent awake), Time in Bed (TiB; the time between BT and GT), Total Sleep Time (TST; the actual recorded time spent asleep each night) and Sleep Efficiency (SE; the percentage of each sleep period spent actually asleep). An estimate of Sleep Latency was calculated as the time between the time the parent reported turning their child's light off and actigraphy recording that sleep had commenced.

Diary measures (appendix 1.2.2 comprises a blank diary).

Parents completed a diary for the period over which sleep was measured. This diary included questions about their child's sleep: time they went to bed, time lights were turned out, time parents felt their child awoke, time they got out of bed, time they took to get to sleep, day-time naps, night-time awakenings, difficult behaviours around bed-times and also details of their own interventions to promote sleep. The diary also included twice daily ratings of their child's sleepiness and impulsivity. Further information on their child's challenging behaviour and irritability were collected and are analysed elsewhere (Clarkson et al., in prep). Variables were calculated from diaries to match those from actigraphy. In most cases, this was transposed directly from parent report. In addition, three composite variables were calculated: Time in bed = Time out of Bed – Bed Time; Total Sleep Time =

Wake-up Time – Lights Out Time– Total Waking Time – Time to get to Sleep; Sleep Efficiency = 100 x (Total Sleep Time / Time in Bed).

Questionnaires.

Parents completed a pack of questionnaires within a week of direct assessment of sleep. For background, all parents completed questionnaires on demographic and health information. All parents completed the Social Communication Questionnaire (Rutter et al., 2003) to measure ASD symptomology in children with ASD and exclude children with potential ASD symptomology in the group of typically developing children. All parents completed a range of sleep questionnaires to examine group differences and correlate with direct measurement of sleep. Parents of children with ASD completed a questionnaire assessing overactivity/ impulsivity to examine correlations with sleep.

Modified Simonds and Parraga Sleep Questionnaire (Simonds & Parraga, 1982; Wiggs & Stores, 2004)

The Simmonds and Parraga Sleep Questionnaire is a broad ranging parent-report measure of sleep in children (Simmonds & Parraga, 1982). The modified version was adapted for children with developmental disabilities (Wiggs & Stores, 2004). The measure can be used to calculate an overall measure of sleep problems (Johnson, Turner, Foldes, Malow, & Wiggs, 2012), which correlates well with the Childhood Sleep Habits Questionnaire (Owens et al., 2000). It can be broken down into seven subscales: Bed-time Resistance, Sleep Onset Delay, Night Wakings, Sleep Anxiety, Parasomnias, Sleep Disordered Breathing and Daytime Sleepiness.

Obstructive Sleep Apnoea (Mindell & Owens, 2003)

The Obstructive Sleep Apnoea Questionnaire (Mindell & Owens, 2003) is used as a screening measure, to consider whether children require further assessment. With this in mind it contains both broad level questions about sleep and sleepiness, such as "Is it hard to wake your child up in the morning?" and more specific apnoea-related questions, such as "When sleeping does your child have trouble breathing, or struggle to breathe?". All items require yes/no responses and an average score (0-1) calculated, with a cut-off advised at >.33.

Family Inventory of Sleep Habits (Malow et al., 2008)

The Family Inventory of Sleep Habits (FISH, Malow et al., 2008) is a broad measure of sleep hygiene, focussing on an individual child's routine. The FISH shows good test-retest reliability in children with ASD (r = .82) over three months and negative correlations with measures of sleep problems from the CSHQ (Malow et al., 2008).

Modified Paediatric Epworth Sleepiness Scale (Williams, Scheimann, Sutton, Hayslett, & Glaze, 2008).

The Epworth Sleepiness Scale (Epworth, 1991) measures daytime sleepiness through asking people how likely they are to "doze" in a range of situations. The modified paediatric version (Williams et al., 2008) differs in asking for a parent response and in removing situations that are less likely to be experienced by children (such as having drank alcohol). The adult version of the scale demonstrates a good level of internal consistency (α =.88; Johns, 1992) and reliable test-retest figures over a period of months (r = .82: Johns, 1992).

Social Communication Questionnaire (Rutter et al., 2003).

The Social Communication Questionnaire asks parents to report whether or not the child demonstrates a range of different social behaviours, it is often used as a quick scale parent-report measure of ASD traits. The SCQ has been shown to be a good predictor of children's likelihood of having ASD (AUC = .90, Charmon et al., 2007) and has good agreement with the Autism Diagnostic Interview- Revised (Rutter et al., 2003).

The Activity Questionnaire (TAQ, Burbidge et al., 2010).

The Activity Questionnaire is a parent-report measure of over-activity and impulsivity for people with developmental disorders. It provides independent subscales for overactivity, impulsivity and impulsive speech. It has an excellent level of internal consistency (α =.93-.95 in verbal children; Burbidge et al., 2010) and good test-retest reliability (r = .75) over two weeks.

Data Analysis

Group Comparisons and correlations

Outcome variables from actigraphy, sleep diaries and questionnaires were compared between the children with ASD and the TD comparison group using independent samples t-tests. Relationships between actigraphy measures and parent report were analysed using Pearson's correlations. The relationship between individual differences in sleep time and sleep efficiency, and other variables were examined using Spearman's correlations due to some questionnaire data differing significantly from a normal distribution. For all statistical tests, $p < .01$ was used for significance to accommodate multiple comparisons with an

acceptable risk of type-1 and type-2 error. Results on which $.01 \leq p < .05$ were considered trends to lower the risk of not reporting potentially clinically significant results.

Temporal relationships

Three linear mixed effects regression models were considered to evaluate the relationships between sleep, sleepiness and impulsivity/ overactivity using R (R Core Team, 2012) and lme4 (Bates, Maechler & Bolker, 2012). In all cases, models were fitted using robust M estimators (maximum likelihood). Robust M estimators are generally considered to be preferable to sum of squares indicators as they are less reliant upon strict parametric assumptions. Random Effects terms included individual child differences (Participant herein), whether the day was a weekday or a weekend day and the time of the report (am vs. pm). Modelling Random Effects terms helps to control for the associated intraclass correlations (as described by Pinheiro & Bates; 2006), in this case individual differences between children and, time and day of measurement. For all three models, visual inspection of residual plots suggested no violation of Homoscedacity or Normality in the data.

Results

Data were analysed to address each of the aims of the study. Differences in sleep between the groups of children with and without ASD were investigated by analysing differences in actigraphy data. Differences in parent reports of sleep between the groups children with and without ASD were tested using data from sleep diaries. Frequency of different parent-reported sleep problems in the groups of children with and without ASD were identified using the sleep questionnaires and their subscales. Similarities and differences between actigraphy measures and parent reports were then analysed. Individual differences in sleep

quantity and quality were identified by correlating outcome variables from actigraphy with demographics and questionnaire totals. Finally, the temporal relationship between Sleep, Sleepiness and Overactivity/ Impulsivity was examined using Linear Mixed Effects Regression Modelling.

Group differences in sleep, measured by actigraphy.

In order to investigate differences in sleep between the groups of children with and without ASD, independent samples t-tests were undertaken comparing the two groups on each of the dependent variables taken from actigraphy. No significant group differences on any of the measures were identified, see table 1.2.3. Trends were identified for the children with ASD getting up earlier in the morning and having a shorter onset latency than children in the typically developing comparison group.

Table 1.2.3. Group mean scores and differences for measurements from actigraphy. *$p < .05$

	ASD Mean (SD)	TD Mean (SD)	*t*	*p*
Bed Time (hh:mm)	21:16 (1:08)	21:37 (0:51)	1	.32
Get-up Time (hh:mm)	06:40 (0:55)	07:15 (0:38)	2.15	.04*
Time in Bed (hh:mm)	09:19 (0:40)	09:38 (0:46)	.91	.39
Total Sleep Time (hh:mm)	08:09 (0:35)	08:13 (0:38)	.58	.79
Onset Latency (hh:mm)	00:09 (0:09)	00:17 (0:11)	2.12	.04*
Sleep Efficiency (%)	86.79 (2.60)	85.45 (4.26)	1.07	.30
Wake After Sleep Onset (hh:mm)	00:51 (0:17)	00:48 (0:16)	.45	.66
Sleep Latency (hh:mm)	00:52 (0:36)	00:32 (0:31)	1.55	.13

Group differences in parent reports of sleep

To compare parent reports of sleep time in their children, responses from the sleep diaries of the two groups were compared. Again, independent samples t-tests found no significant differences between the groups (see table 1.2.4). There were trends for the children with ASD being reported to wake up and get out of bed earlier than the TD children.

Table 1.2.4. Group mean scores and differences for measurements from sleep diaries. *p <. 05

	ASD Mean (SD)	TD Mean (SD)	t	p
Bed Time (hh:mm)	20:25 (0:58)	20:55 (0:58)	1.44	.16
Lights Out (hh:mm)	20:51 (1:05)	21:20 (1:09)	1.26	.22
Wake up time (hh:mm)	06:39 (0:47)	07:12 (0:46)	2.05	.05*
Time out of Bed (hh:mm)	06:52 (0:49)	07:31 (0:45)	2.30	.03*
Time to get to Sleep (hh:mm)	00:39 (0:29)	00:32 (0:19)	.81	.43
Wake After Sleep Onset (hh:mm)	00:11 (0:17)	00:03 (0:03)	1.77	.09
Time in Bed (hh:mm)	10:28 (0:51)	10:24 (0:49)	.16	.87
Total Sleep Time (hh:mm)	09:03 (1:07)	09:16 (1:22)	.45	.65
Sleep Efficiency (%)	86.5% (8%)	88.7% (10%)	.70	.49

Frequency of different parent-reported sleep problems in children with and without ASD.

To investigate the frequency and nature of parent-reported sleep problems, independent samples t-tests compared the two groups' scores on each of the questionnaires. Children in the ASD group scored significantly higher than children in the TD group on both measures related to sleep itself (the MSPSQ and the OSAQ; see table 1.2.5). There was no evidence of

group differences in sleep hygiene or day-time sleepiness (as measured by the FISH and the MPESS).

Table 1.2.5. Group mean scores and differences for questionnaires. **$p < .01$

Variable	ASD Mean (SD)	TD Mean (SD)	*t*	*p*
Modified Simonds and Parraga Sleep Questionnaire	73.05 (15.16)	53.29 (13.59)	3.77	.001**
Obstructive Sleep Apnoea Questionnaire	.41 (.14)	.13 (.09)	6.73	< .001**
Family Inventory of Sleep Habits	50.57 (11.06)	46.60 (5.02)	1.26	.22
Modified Paediatric Epworth Sleepiness Scale	2.21 (3.02)	2.50 (1.97)	.31	.76
The Activity Questionnaire	39.61 (18.37)	-	-	-

Specifying Sleep Problems

To investigate the likely sleep problems responsible for group differences on the MSPSQ total score, the subscales of the questionnaire (Johnson et al., 2012) were investigated. There were significant differences between the groups, such that the children in the ASD group scored higher than those in the TD group on Sleep Onset Delay ($t(1) = 2.89$, $p = .007$), Parasomnias ($t(1) = 3.22$, $p = .003$) and Day-Time Sleepiness ($t(1) = 3.44$, $p = .002$) and there was a trend for a difference on Night Waking ($t(1) = 2.68$, $p = .012$), see figure 1.2.1. There were no significant differences on other subscales ($ts(1) \leq 1.87$, $p \geq .07$). A list of individual item means for each group are included in appendix 1.2.4.

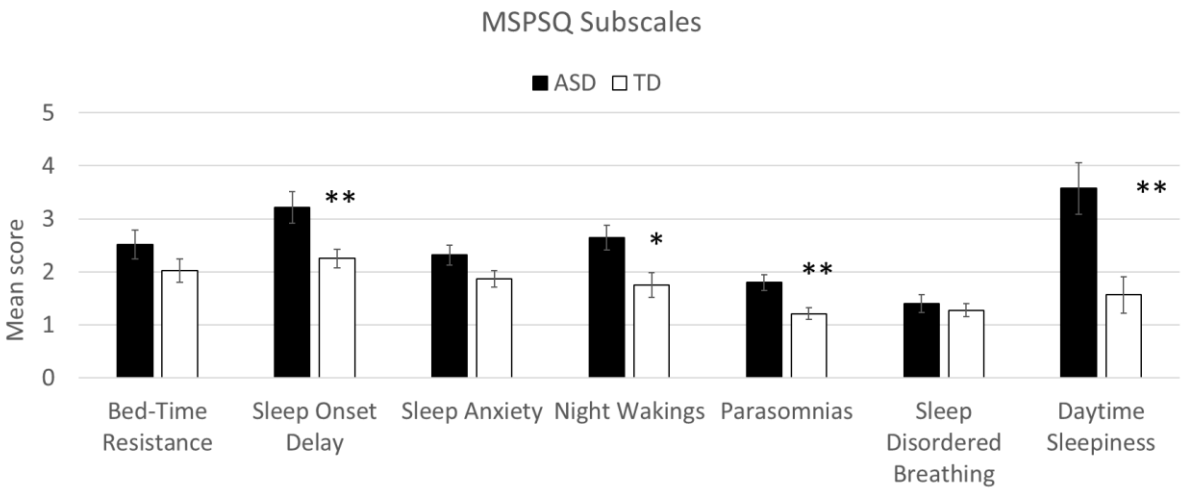


Figure 1.2.1 Comparisons between groups on subscales of the Modified Simmonds and Parraga Sleep Questionnaire. Error bars indicated standard error of the mean. ** $p < .01$, * $p < .05$.

Whilst diagnosing a sleep problem requires more in-depth clinical information than available in this study, the MSPSQ does afford the opportunity for identifying potential areas of concern. Johnson et al. (2012) suggest 56 as a cut-off on the MSPSQ total score as indicative of a sleep problem. Using this cut-off, 12 of 14 children (86%) with ASD were considered to have a sleep problem and only five of 16 typically developing children (31%). This represented a significant difference between groups ($X^2(1) = 9.02$ $p = .004$).

Wiggs & Stores (2004) cite any score above four on items of the MSPSQ as suggestive of a frequent problem that may be cause for concern. Table 1.2.6. reports the frequency of children who met this criterion on an individual subscale item (perhaps indicative of a specific problem) or as their median score for a subscale (perhaps indicative of a more pervasive problem).

Table 1.2.6. Possible sleep problems in the ASD and TD groups.

	Potential specific problem		Potential pervasive problem	
	ASD (N =14)	TD (N = 16)	ASD (N = 14)	TD (N = 16)
Bed-Time Resistance	12 (86%)	9 (56%)	2 (14%)	1 (6%)
Sleep Onset Delay	6 (43%)	0 (0%)	N/A	N/A
Sleep Anxiety	11 (79%)	11 (69%)	1 (7%)	0 (0%)
Night Waking	7 (50%)	2 (13%)	0 (0%)	1 (6%)
Parasomnias	11 (79%)	2 (13%)	0 (0%)	0 (0%)
Sleep Disordered Breathing	5 (36%)	3 (19%)	0 (0%)	0 (0%)
Day-Time Sleepiness	9 (64%)	3 (19%)	4 (29%)	0 (0%)

In sum, parents of the children with ASD reported higher frequencies of sleep problems on questionnaires (86% using the MSPSQ overall cut-off) than did parents of the children without ASD (39%). Significant differences related to subscales measuring sleep onset delay, parasomnias and day-time sleepiness.

Similarities and differences between actigraphy measures and parent reports.

Pearson's correlations were employed to investigate the relationship between equivalent variables measured by actigraphy and sleep diaries in children in the ASD group (table 1.2.7; for equivalent statistics from typically developing children, see appendix 1.2.3). There were significant positive correlations between diary reported bed-times, get-up times and sleep latencies and those found through actigraphy. Trends were identified for equivalent relationships in sleep time and time in bed. Paired samples t-tests were used to identify differences between actigraphy and parent report. In most cases, there were significant

differences between actigraphy recordings and parent report: Parents reported their children to go to bed earlier, spend longer in bed and get more sleep in total than recorded by actigraphy. Parents also recorded shorter waking times, with an overall more efficient sleep period than identified by actigraphy.

In sum, parent reports of sleep time (but not night-time wakings) reflected an accurate representation of individual differences. Differences between parent reports and direct measurement reflected parents overestimating their children's sleep time.

Table 1.2.7. Correlations and comparisons between actigraphy and Sleep diary measures for children with ASD (See appendix 1.2.3 for equivalent statistics in the TD group). Means are contained in tables 1.2.3 and 1.2.4. ***p* < .01, *p < .05.

Actigraphy Variable	Sleep Diary Variable	*r*	*p*	*t*	*p*
Bed Time	Bed Time	.81	< .001**	4.40	.001**
Get up time	Wake Time	.83	< .001**	.24	.82
Time in Bed	Time in Bed	.50	.05*	54.26	< .001**
Total Sleep Time	Total Sleep Time	.54	.03*	55.18	< .001**
Sleep Latency	Time to get to sleep	.54	.003**	1.12	.28
Sleep Efficiency	Sleep Efficiency	-.15	.57	131.90	< .001**
Wake After Sleep Onset	Wake After Sleep Onset	-.22	.42	2.27	.04*

Individual differences in Sleep Quantity and Quality

Demographic information and totals from the questionnaires were correlated against Total Sleep Time and Sleep Efficiency measures from actigraphy (table 1.2.8). Spearman's Rank correlations were used as some questionnaire data differed significantly from the normal distribution. The only significant correlation was a significant positive correlation between

Sleep Time and total score on the TAQ. There were trends for a negative correlation between Sleep Time and Age and a positive correlation between Sleep Efficiency and score on the SCQ.

Table 1.2.8. Correlation between dependent variable questionnaires and sleep outcome measures (from actigraphy) in the ASD group (See appendix 1.2.5 for the equivalent statistics from typically developing children). $^{**}p < .01$, $^{*}p < .05$.

Measure	Correlation with Total Sleep Time, *ρ (p)*	Correlation with Sleep Efficiency, *ρ (p)*
Age	-.60 (.01*)	.30 (.26)
Performance IQ	.05 (.85)	-.12 (.68)
Adaptive Behaviour (VABS)	.01 (.98)	-.32 (.22)
Sleep Problems (MSPSQ)	.28 (.34)	.14 (.63)
Sleepiness (PESS)	-.12 (.67)	-.17 (.57)
Social Communication (SCQ)	.30 (.32)	.60 (.03*)
Activity (TAQ)	.71 (< .01**)	-.11 (.71)
Family Sleep Habits (FISH)	.35 (.23)	-.10 (.74)

The temporal relationship between Sleep, day-time sleepiness and impulsivity/ overactivity

To model the temporal relationship between Sleep, Day-time Sleepiness and Impulsivity/ Overactivity, the data from each parent rating of Sleepiness and Impulsivity/ Overactivity (twice daily), and daily Actigraphy measurements of each of three variables were used.

Predicting Sleep Efficiency from Sleepiness and Impulsivity/ Overactivity on the previous afternoon

To predict Sleep Efficiency, Participant and Day (weekday vs. weekend) were entered as Random Effects Terms (modelled by intercept). A significant random effect was observed for Participant ($X^2(1) = 46.53$, $p < .01$), but not for Day ($X^2(1) = 0$, $p = 1.0$). Accordingly, the random effect for Day was removed from subsequent analyses. Fixed effects for Sleepiness and Impulsivity/ Overactivity (as rated in the afternoon) were then added to the model containing random effects for participant[3]. The addition of these fixed effects was not associated with a significant increase in the overall explained variance ($X^2(2)= 4.00$, $p = .13$; see table 1.2.9 for parameter estimates). The final model, therefore, had a single random effect to represent individual differences in the sleepiness of participants and a fixed effect to represent the mean of the dependent variable (i.e., the Sleep Efficiency score). Final Model = lmer(Sleep Efficiency ~ 1 + (1|Participant)).

Table 1.2.9. Fixed Effects model for predicting Sleep Efficiency

	Estimate	Std Error	t value
Intercept	89.43	1.39	64.28
Impulsivity/ overactivity	-.57	.39	-1.47
Sleepiness	-.46	.51	-.91

Predicting Sleepiness from previous night's sleep

To predict Sleepiness, Participant, Day (weekday vs. weekend) and Time of Day (am vs. pm) were entered as Random Effects Terms (modelled by intercept). A significant random effect was observed for Participant ($X^2(1) = 181.44$, $p < .01$), but neither for Day ($X^2(1) = 0$, $p = 1.0$), nor Time of Day ($X^2(1) = 0$, $p = 1.0$). Accordingly, the random effects for Day and Time of Day were removed from subsequent analyses. Fixed effects for Total Sleep Time, Wake after

[3] Model = lmer(Sleep Efficiency ~ 1 + Sleepiness + Impulsivity + (1|Participant)).

Sleep Onset and Sleep Efficiency were then added to the model containing random effects for participant[4]. The addition of these fixed effects was not associated with a significant increase in the overall explained variance ($X^2(3) = 1.80$, $p = .61$; see table 1.2.10 for parameter estimates). The final model, therefore, had a single random effect to represent individual differences in the sleepiness of participants and a fixed effect to represent the mean of the dependent variable (i.e., the Sleepiness score). Final Model = lmer(Sleepiness~ 1 + (1|Participant)).

Table 1.2.10. Fixed Effects model for predicting Sleepiness

	Estimate	Std Error	t value
Intercept	2.30	1.64	1.40
Total Sleep Time	-.00	.00	-1.21
Wake After Sleep Onset	.00	.00	.43
Sleep Efficiency	.00	.02	.15

Predicting Impulsivity/ Overactivity from sleep and Sleepiness

To predict Impulsivity/ overactivity, Participant, Day (weekday vs. weekend) and Time of Day (am vs. pm) were entered as Random Effects Terms (modelled by intercept). A significant random effect was observed for Participant ($X^2 = 107.46$, $p < .01$), but neither for Day ($X^2 = 0.19$, $p = .67$), nor Time of Day ($X^2 = .28$, $p = .59$). Accordingly, the random effects for Day and Time of Day were removed from subsequent analyses. Fixed effects for Total Sleep Time, Wake after Sleep Onset, Sleep Efficiency and Sleepiness were then added to the model containing random effects for participant[5]. Addition of the Fixed Effects was associated with a significant increase in the overall explained variance ($X^2(4) = 16.41$, $p < .01$; see table 1.2.11

[4]Model = lmer(Sleepiness ~ 1 + Total Sleep Time + Wake After Sleep Onset + Sleep Efficiency + (1|Participant)).
[5] Model = lmer(Impulsivity ~ 1 + Sleepiness + Total Sleep Time + Wake After Sleep Onset + Sleep Efficiency + (1|Participant)).

for parameter estimates). Removing each of the measures of sleep from the previous night individually did not decrease the overall explained variance: for Total Sleep Time ($X^2(1) = .06$, $p = .81$), for Wake After Sleep Onset ($X^2(1) = .71$, $p = .40$), for Sleep Efficiency ($X^2(1) = 1.80$, $p = .18$). The model with Sleepiness removed, however, was associated with a significant decrease in the overall explained variance, ($X^2(1) = 14.55$, $p < .01$). The final model, therefore, had a single random effect to represent individual differences in the impulsivity/ overactivity of the participants, a fixed effect to represent the mean of the dependent variable (i.e., the impulsivity/ overactivity score) and a fixed effect to represent the sleepiness at that point in time. Final Model = lmer(Impulsivity ~ 1 + Sleepiness + (1|Participant)), see figure 1.2.2.

Table 1.2.11. Fixed Effects model for predicting Impulsivity/ overactivity

	Estimate	Std Error	t value
Intercept	-.74	1.94	-.38
Total Sleep Time	-.00	.00	-.24
Wake After Sleep Onset	.00	.01	.84
Sleep Efficiency	.03	.02	1.35
Sleepiness	.31	.08	3.90

Figure 1.2.2. Predicted values for the final model predicting impulsivity/ overactivity. The only Random Effects Factor was participant and the only Fixed Effects Factor, sleepiness.

In sum, sleep efficiency, as measured by actigraphy was not predicted by parent ratings of either sleepiness or impulsivity/ overactivity on the previous day. Similarly, there was no evidence for a significant role for sleepiness in predicting child sleep that night. There was no evidence that sleep on the previous night predicted impulsivity/ overactivity either, but there was a significant positive relationship, such that parent rating of sleepiness at a given time predicted their rating of impulsivity/ overactivity at the same time.

Discussion

The sleep of children with ASD was compared to that of typically developing children using a comprehensive range of measures. All children with ASD were recruited on the basis of their parents considering them to have a sleep problem, whilst typically developing children were not recruited under this condition. The most objective measurements of sleep, through actigraphy, found no significant differences between the two groups. Subjective reports, through parent diaries, correlated well with these objective measurements, other than for wakings, and also showed no significant group differences. On questionnaire measures, however, there was evidence of group differences in sleep. Notably, children in the ASD group returned significantly higher scores on both the general sleep questionnaire (the MSPSQ; Wiggs & Stores, 2004) and the Screening Questionnaire for Sleep Apnoea (Mindell & Owens, 2003).

Measuring Sleep Directly through Actigraphy

Consistent and reliable reports have concluded that sleep problems are more common in children with ASD (44-83% of children with ASD experience sleep problems), than they are in the general population (9-50% of children more generally; Elrod & Hood, 2015; Richdale & Schreck, 2009). It is therefore surprising that measurements of children's sleep taken through actigraphy were not significantly different for children with ASD from children who were matched in age and gender, but did not have ASD. Such a conclusion is even more stark given that the children with ASD were recruited on the basis of a parent-reported sleep problem. Over a week of recordings, no significant differences between groups were observed in duration of sleep (as measured through Total Sleep Time) and quality of sleep (as measured through Wake After Sleep Onset and Sleep Efficiency). These findings are not

entirely inconsistent with previous findings in children with ASD *without* comorbid intellectual disabilities. A recent meta-analysis (Elrod & Hood, 2015) showed that the three studies that measured sleep directly, through polysomnography or actigraphy, found no overall difference in duration of sleep between children with and without ASD (and no comorbid intellectual disability). The findings here are consistent with that result. The meta-analysis (Elrod & Hood, 2015) did, however, find significant differences in sleep efficiency and sleep latency between groups. Here these were not observed, although the comparison using the sleep latency measure, calculated by combining the diary and the actigraphy, did approach significance for children with ASD having longer latencies. The onset latency measure, calculated automatically through the actigraphy software, showed mean differences in the opposite direction. Sleep Latency may be a difficult concept to measure in children with ASD using actigraphy as it will often be influenced by parental expectations, child restlessness and the ability to self-sooth.

The only other study using actigraphy in children with ASD without co-morbid intellectual disabilities (Allik et al., 2006) similarly found no group differences in overall sleep time, but did evidence differences in latency and efficiency. Interestingly, both groups in the study by Allik and colleagues (Allik et al., 2006), returned higher estimates of sleep efficiency (in spite of being of similar ages) than in this study. It is therefore possible that the control group here were unusually poor sleepers. Typically developing children were recruited through opportunity sampling by university staff and students, with social media being commonly used. It is possible that parents of children with irregular sleep patterns may have been more attracted by the opportunity to receive detailed feedback on their child's sleep (though note questionnaires suggested parents of children with ASD reported significantly more sleep problems than did those of TD children).

Parentally Reported Diaries

Parental reports of the sleep of their children with ASD correlated well with measures gained through actigraphy. Interestingly, comparisons between the two measures showed that parents actually overestimated their children's sleep. Evidence suggested that this was the result of underestimating the time their children were awake after going to sleep for the first time. These results suggest that parents of children with ASD were regularly unaware of the times at which their children were awake. Whilst direct measures of sleep are often preferred on the basis that parents may overestimate their children's sleep difficulties (Goodlin-Jones, Tang, Liu, & Anders, 2008; Hering, Epstein, Elroy, Iancu, & Zelnik, 1999), in this study, there is evidence for the opposite. This stands as further evidence for the preference of direct measures in both research and clinical practice. There were no significant differences between groups on diary-reported measures, although there were trends towards children with ASD going to bed earlier and rising earlier than their typically developing peers. As for actigraphy, it was surprising that data from children with ASD and a reported sleep problem did not significantly differ from that of their peers. This suggests that parents did not believe their child to have a sleep problem on the basis of underestimating their sleep duration or quality. One explanation is that they may have had had an unrealistic expectation of the sleep their child should be getting. Another is that parental experiences of sleep problems included broader difficulties with sleep than duration of sleeping and waking.

Questionnaires

Unlike measures from actigraphy and parent-report diaries, analysis of questionnaires did show differences between the two groups. This was consistent with children in the ASD

group being recruited on the basis of a reported sleep problem. Most notably, the children with ASD scored significantly higher on the MSPSQ (Wiggs & Stores, 2004) and the OSQ (Mindell & Owens, 2003). Both of these questionnaires assess sleep and night-time behaviour much more broadly than the measures collected in the diaries and measured through actigraphy. The MSPSQ (Wiggs & Stores, 2004), for instance, includes questions on bed-time resistance, day-time sleepiness and parasomnias, as well as questions on sleep latency and night-time waking. The OSQ (Mindell & Owens, 2003) includes items thought to be correlated with sleep apnoea, such as whether a child stops breathing at night, but also items associated with day-time sleepiness or inattention.

Subscales on the MSPSQ showed significant group differences on Sleep Onset, Parasomnias and Daytime Sleepiness. Diaries and actigraphy showed no difference in sleep latency, so differences in the sleep onset subscale were surprising, perhaps suggesting they reflected differences in parental perception. Similarly, differences in day-time sleepiness may be surprising as they were not identified on the broader scale of sleepiness (The Pediatric Epworth Sleepiness Scale). One reason may be that the Epworth scale focuses on a single criterion of the "likelihood of dozing", which may not reflect the broader experience of sleepiness. Differences on the parasomnias subscale were informative and report aspects of sleep that may not have been identifiable through actigraphy. Notably nearly 80% of the children with ASD experienced at least one form of parasomnia once a week or more (in comparison to less than 15% of the control group). Though dysomnias (difficulties in initiating or maintaining sleep) have received more attention in the literature, there is previous evidence suggestive of parasomnias being more prevalent in children with ASDs (Gail Williams, Sears, & Allard, 2004; Ming, Sun, Nachajon, Brimacombe, & Walters, 2009). In the only polysomnography study of parasomnias, Ming et al. (2009) reported a particularly high prevalence of disorders of partial arousal in children with ASD. Partial

arousals may indicate poorer sleep and pre-dispose children to sleep terrors and confused awakenings. Though cautioned by the possibility that children with ASD may have been more influenced by testing in a sleep laboratory, Ming et al. (2009) suggested that one reason for this could be greater fragmentation in sleep more generally. Further research in this area may be able to define more clearly the precise nature and prevalence of parasomnias in children with ASD, their likely bio-psychosocial precipitants and perpetuators and their impact on day-time functioning.

One caveat is that questionnaire measures of sleep in children can be criticised for measuring parent expectations as well as the child's actual sleep patterns. Day-time behaviour may impact parental stress and thus make sleep problems seem more severe. In this study, one reason to believe this may not have been the case was the reliability of parent-reports on the sleep diaries. Parents overestimated their children's sleep duration, but reported further sleep problems alongside this.

Impact on day-time functioning.

Poor sleep can impact on children's ability to function in the day-time (Dewald, Meijer, Oort, Kerkhof, & Bögels, 2010). In this study, links between poor sleep, sleepiness, impulsivity and overactivity were investigated. There was a significant correlation between total sleep and day-time activity. Children with higher total sleep were reported to be more active in the day. This was perhaps surprising given reports of sleep problems in children with ADHD (Ball et al., 1997; Harvey et al., 2001). Mixed-effects linear regression was used to model the relationships between night sleep, day-time sleepiness and impulsivity/ overactivity. Importantly, this method allowed for modelling the temporal relationship between sleep and day-time variables, as opposed to the individual differences methods typically employed

in correlational studies. Interestingly, there was no relationship between sleep, and sleepiness and overactivity/ impulsivity in the final model. This finding is consistent with results from a study in children with ADHD (LeBourgeois et al., 2004). Although parents of children with ADHD reported higher levels of sleep problems and higher day-time sleepiness in their children than did parents of typically developing children, Lebourgeois et al (2004) found no significant correlation between sleep problems and sleepiness.

All Random Effects Models were made significantly worse by removing the term for individual participant. This suggested that individual differences amongst children with ASD were stronger drivers of outcome variables than within-subjects variation through the week. The one fixed-effects term that was maintained was the role for sleepiness in predicting overactivity/ impulsivity. This should be interpreted with caution as both measures were taken from parents at a single time-point and parents may have used similar behaviours to determine both ratings. It does suggest, however, that in parental experience at least, day-time sleepiness may be a factor in the impulsivity or overactivity of children with ASD.

Limitations and future directions

Although a direct measure of sleep, actigraphy has significant limitations in comparison to polysomnography (Michaelson, et al., 2006). Actigraphy can often misrepresent restful waking as sleep (Sadeh & Acebo, 2002). It can also miss finer-grained distinctions in sleep cycles, such as time spent in REM sleep and evidence for sleep apnoea, both of which can be accurately recorded using polysomnography. The finding from this sample that estimates of parasomnias were high suggests that polysomnography may have more accurately represented the concerns parents had about their children's sleep. Only two studies have compared sleep in children with ASD and no comorbid intellectual disability using

polysomnography and each of these only recorded from a single night. Larger samples and longer testing periods of polysomnography may be needed to understand sleep problems in this group.

The study was also limited by sample size and nature. Only 16 children with ASD were recruited and these were drawn from a relatively broad age range and also a broad range of cognitive and adaptive abilities (though all were in the normal range on a standardized measure of performance IQ). That all children were recruited on the basis of their parents reporting them to have a sleep problem, limits the study in describing sleep problems in children with ASD, rather than a broad population of children with ASD.

Summary

Sleep problems in children with ASD have been well-documented and are cited as among the most common comorbid conditions for this group (Xue Ming et al., 2007). There have, however, been very few studies measuring sleep using direct measures that have compared sleep in children with ASD and no comorbid intellectual disability to children without ASD. Here, actigraphy was used to add to that literature. In support of a recent meta-analysis (Elrod & Hood, 2015), there was no evidence that the children with ASD, with no intellectual disability, slept for shorter periods. Interestingly, even though children were recruited on the basis of their parents considering them to have a sleep problem, there was no evidence of greater durations of waking or longer sleep latencies either. Diary measures suggested that parents were not underestimating the actual sleep their children got or overestimating their waking periods. Questionnaire data suggested that sleep problems in children with ASDs may reflect a broader range of sleep difficulties. There was no evidence that duration

or efficiency of sleep had a significant relationship with sleepiness or impulsivity on the following day.

Implications for Knowledge and Practice in Clinical Psychology

Clinical psychologists regularly work with children with ASD, both in specialist and in universal services. Sleep problems have been widely reported to be more prevalent in children with ASD than their typically developing peers. Sleep problems have been understood to be linked to broader case presentations through bio-psycho-social models. Difficulties in sleep in children with ASD have been associated with mental health problems, such as depression and anxiety, increased severity of autistic symptoms, increased challenging behaviour, increased parent stress and decreased day-time functioning. The findings of this empirical paper suggested that for children with ASD and no comorbid intellectual disability:

- **Duration of sleep, efficiency of sleep and night-time wakings in groups of children with ASD may not always differ significantly from typically developing children (even if parents identify a sleep problem).**
- **Parent reports through diaries may accurately reflect individual differences in their children's sleep, but over-estimate time slept.**
- **Parents of children with ASD may report a broad range of difficulties with sleep, which are not necessarily measurable through sleep diaries or direct recording.**

References

Acebo, C., Sadeh, A., Seifer, R., Tzischinsky, O., Wolfson, A. R., Hafer, A., & Carskadon, M. A. (1999). Estimating Sleep Patterns with Activity Monitoring in Children and Adolescents: How Many Nights Are Necessary for Reliable Measures? *SLEEP*, *22*(1), 95–103.

Allik, H., Larsson, J.-O., & Smedje, H. (2006). Insomnia in school-age children with Asperger syndrome or high-functioning autism. *BMC Psychiatry*, *6*(1), 18.

American Psychiatric Association. (2013). *DSM-5 American Psychiatric Association*. Arlington: American Psychiatric Publishing.

Anders, T., Iosif, A.-M., Schwichtenberg, A. J., Tang, K., & Goodlin-Jones, B. (2012). Sleep and Daytime Functioning: A Short-term Longitudinal Study of Three Preschool-age Comparison Groups. *American Journal on Intellectual and Developmental Disabilities*, *117*(4), 275–290.

Ball, J. D., Tiernan, M., Janusz, J., & Furr, A. (1997). Sleep patterns among children with attention-deficit hyperactivity disorder: a reexamination of parent perceptions. *Journal of Pediatric Psychology*, *22*(3), 389–398.

Bartlett, L. B., Rooney, V., & Spedding, S. (1985). Nocturnal difficulties in a population of mentally handicapped children. *The British Journal of Mental Subnormality*, *31*(60), 54–59.

Berkman, J. M. (2006). Sleep in children with developmental disabilities. *Medicine and Health Rhode Island*, *89*(3), 94.

Cortesi, F., Giannotti, F., Ivanenko, A., & Johnson, K. (2010). Sleep in children with autistic spectrum disorder. *Sleep Medicine*, *11*(7), 659–664.

Dewald, J. F., Meijer, A. M., Oort, F. J., Kerkhof, G. A., & Bögels, S. M. (2010). The influence of sleep quality, sleep duration and sleepiness on school performance in children and adolescents: A meta-analytic review. *Sleep Medicine Reviews*, *14*(3), 179–189.

Didden, R., Korzilius, H., Aperlo, B. van, Overloop, C. van, & Vries, M. de. (2002). Sleep problems and daytime problem behaviours in children with intellectual disability. *Journal of Intellectual Disability Research*, *46*(7), 537–547.

Didden, R., & Sigafoos, J. (2001). A review of the nature and treatment of sleep disorders in individuals with developmental disabilities. *Research in Developmental Disabilities*, *22*(4), 255–272.

Dunn, M. E., Burbine, T., Bowers, C. A., & Tantleff-Dunn, S. (2001). Moderators of stress in parents of children with autism. *Community Mental Health Journal*, *37*(1), 39–52.

Elrod, M. G., & Hood, B. S. (2015). Sleep differences among children with autism spectrum disorders and typically developing peers: A meta-analysis. *Journal of Developmental & Behavioral Pediatrics*, *36*(3), 166–177.

Fombonne, E., Quirke, S., & Hagen, A. (2011). Epidemiology of pervasive developmental disorders. In *Autism Spectrum Disorders* (pp. 90–111). New York: Oxford University Press.

Gail Williams, P., Sears, L. L., & Allard, A. (2004). Sleep problems in children with autism. *Journal of Sleep Research*, *13*(3), 265–268.

Goldman, S. E., Surdyka, K., Cuevas, R., Adkins, K., Wang, L., & Malow, B. A. (2009). Defining the Sleep Phenotype in Children With Autism. *Developmental Neuropsychology*, *34*(5), 560–573.

Goodlin-Jones, B. L., Tang, K., Liu, J., & Anders, T. F. (2008). Sleep Patterns in Preschool-Age Children With Autism, Developmental Delay, and Typical Development: *Journal of the American Academy of Child & Adolescent Psychiatry*, *47*(8), 930–938.

Harvey, M. T., Baker, D. J., Horner, R. H., & Blackford, J. U. (2001). Actigraphy and parental ratings of sleep in children with attention-deficit/hyperactivity disorder (ADHD). *Sleep*, *24*(3), 303.

Hering, E., Epstein, R., Elroy, S., Iancu, D. R., & Zelnik, N. (1999). Sleep patterns in autistic children. *Journal of Autism and Developmental Disorders*, *29*(2), 143–147.

Hoffman, C. D., Sweeney, D. P., Gilliam, J. E., Apodaca, D. D., Lopez-Wagner, M. C., & Castillo, M. M. (2005). Sleep problems and symptomology in children with autism. *Focus on Autism and Other Developmental Disabilities*, *20*(4), 194–200.

Höglund Carlsson, L., Norrelgen, F., Kjellmer, L., Westerlund, J., Gillberg, C., & Fernell, E. (2013). Coexisting Disorders and Problems in Preschool Children with Autism Spectrum Disorders. *The Scientific World Journal*, *2013*, 1–6.

Hoshino, Y., Watanabe, H., Yashima, Y., Kaneko, M., & Kumashiro, H. (1984). An investigation on sleep disturbance of autistic children. *Psychiatry and Clinical Neurosciences*, *38*(1), 45–51.

Johnson, C. R., Turner, K. S., Foldes, E. L., Malow, B. A., & Wiggs, L. (2012). Comparison of sleep questionnaires in the assessment of sleep disturbances in children with autism spectrum disorders. *Sleep Medicine*, *13*(7), 795–801.

Jones, K., & Harrison, Y. (2001). Frontal lobe function, sleep loss and fragmented sleep. *Sleep Medicine Reviews*, *5*(6), 463–475.

Krakowiak, P., Goodlin-Jones, B., Hertz-Picciotto, I., Croen, L. A., & Hansen, R. L. (2008). Sleep problems in children with autism spectrum disorders, developmental delays, and typical development: a population-based study. *Journal of Sleep Research*, *17*(2), 197–206.

La Malfa, G., Lassi, S., Bertelli, M., Salvini, R., & Placidi, G. F. (2004). Autism and intellectual disability: a study of prevalence on a sample of the Italian population. *Journal of Intellectual Disability Research*, *48*(3), 262–267.

LeBourgeois, M. K., Avis, K., Mixon, M., Olmi, J., & Harsh, J. (2004). Snoring, sleep quality, and sleepiness across attention-deficit/hyperactivity disorder subtypes. *SLEEP-NEW YORK THEN WESTCHESTER-*, *27*(3), 520–526.

Lecendreux, M., Konofal, E., Bouvard, M., Falissard, B., & Mouren-Simeoni, M.-C. (2000). Sleep and Altertness in Children with ADHD. *Journal of Child Psychology and Pschiatry*, *41*(6), 803–812.

Leitner, Y. (2014). The Co-Occurrence of Autism and Attention Deficit Hyperactivity Disorder in Children â€“ What Do We Know? *Frontiers in Human Neuroscience*, *8*.

Liu, X., Hubbard, J. A., Fabes, R. A., & Adam, J. B. (2006). Sleep Disturbances and Correlates of Children with Autism Spectrum Disorders. *Child Psychiatry and Human Development*, *37*(2), 179–191.

Lord, C., Risi, S., Lambrecht, L., Cook Jr, E. H., Leventhal, B. L., DiLavore, P. C., … Rutter, M. (2000). The Autism Diagnostic Observation Schedule—Generic: A standard measure of social and communication deficits associated with the spectrum of autism. *Journal of Autism and Developmental Disorders*, *30*(3), 205–223.

Malow, B. A., Marzec, M. L., McGrew, S. G., Wang, L., Henderson, L. M., & Stone, W. L. (2006). Characterizing sleep in children with autism spectrum disorders: a multidimensional approach. *SLEEP-NEW YORK THEN WESTCHESTER-*, *29*(12), 1563.

Michaelson, P. G., Allan, P., Chaney, J., & Mair, E. A. (2006). Ann Otol Rhinol Laryngol-2006-Michaelson-802-9.pdf. *Annals of Otology, Rhinology & Laryngology*, *115*(11), 802–809.

Ming, X., Sun, Y.-M., Nachajon, R. V., Brimacombe, M., & Walters, A. S. (2009). Prevalence of parasomnia in autistic children with sleep disorders. *Clinical Medicine Insights. Pediatrics*, *3*, 1.

Paavonen, E. J., Vehkalahti, K., Vanhala, R., von Wendt, L., Nieminen-von Wendt, T., & Aronen, E. T. (2008). Sleep in Children with Asperger Syndrome. *Journal of Autism and Developmental Disorders*, *38*(1), 41–51.

Palm, L., Persson, E., Bjerre, I., Elmqvist, D., & Blennow, G. (1992). Sleep and wakefulness in preadolescent children with deficits in attention, motor control and perception. *Acta Paediatrica*, *81*(8), 618–624.

Patzold, L. M., Richdale, A. L., & Tonge, B. J. (1998). patzold.pdf. *Journal of Pediatric Child Health*, *34*, 528–533.

Quine, L. (1992). Severity of sleep problems in children with severe learning difficulties: description and correlates. *Journal of Community & Applied Social Psychology*, *2*(4), 247–268.

Richdale, A., Francis, A., Gavidia-Payne, S., & Cotton, S. (2000). Stress, behaviour, and sleep problems in children with an intellectual disability. *Journal of Intellectual and Developmental Disability*, *25*(2), 147–161.

Richdale, A. L., & Baker, E. K. (2014). Sleep in Individuals with an Intellectual or Developmental Disability: Recent Research Reports. *Current Developmental Disorders Reports*, *1*(2), 74–85.

Richdale, A. L., & Schreck, K. A. (2009). Sleep problems in autism spectrum disorders: Prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews*, *13*(6), 403–411.

Rzepecka, H., McKenzie, K., McClure, I., & Murphy, S. (2011). Sleep, anxiety and challenging behaviour in children with intellectual disability and/or autism spectrum disorder. *Research in Developmental Disabilities*, *32*(6), 2758–2766.

Sadeh, A., & Acebo, C. (2002). The role of actigraphy in sleep medicine. *Sleep Medicine Reviews*, *6*(2), 113–124.

Schreck, K. (2004). Sleep problems as possible predictors of intensified symptoms of autism*1. *Research in Developmental Disabilities*, *25*(1), 57–66.

Schreck, K. A., & Mulick, J. A. (2000). Parental report of sleep problems in children with autism. *Journal of Autism and Developmental Disorders*, *30*(2), 127–135.

SHANG, C.-Y., GAU, S. S.-F., & SOONG, W.-T. (2006). Association between childhood sleep problems and perinatal factors, parental mental distress and behavioral problems. *Journal of Sleep Research*, *15*(1), 63–73.

Souders, M. C., Mason, T. B., Valladares, O., Bucan, M., Levy, S. E., Mandell, D. S., … Pinto-Martin, J. (2009). Sleep behaviors and sleep quality in children with autism spectrum disorders. *Sleep*, *32*(12), 1566–1578.

Surtees, A. D. R., Oliver, C., Jones, C., Evans, D., & Richards, C. R. (submitted). A meta-analysis comparing duration and quality of sleep in people with intellectual disabilities with controls.

Tani, P., Lindberg, N., Nieminen-von Wendt, T., Von Wendt, L., Alanko, L., Appelberg, B., & Porkka-Heiskanen, T. (2003). Insomnia is a frequent finding in adults with Asperger syndrome. *BMC Psychiatry*, *3*(1), 1.

Tietze, A.-L., Blankenburg, M., Hechler, T., Michel, E., Koh, M., Schlüter, B., & Zernikow, B. (2012). Sleep disturbances in children with multiple disabilities. *Sleep Medicine Reviews*, *16*(2), 117–127.

Tordjman, S., Anderson, G. M., Pichard, N., Charbuy, H., & Touitou, Y. (2005). Nocturnal excretion of 6-sulphatoxymelatonin in children and adolescents with autistic disorder. *Biological Psychiatry*, *57*(2), 134–138.

Wiggs, L., & Stores, G. (2004). Sleep patterns and sleep disorders in children with autistic spectrum disorders: insights using parent report and actigraphy. *Developmental Medicine & Child Neurology*, *46*(06), 372–380.

Xue Ming, Brimacombe, M., Chaaban, J., Zimmerman-Bier, B., & Wagner, G. C. (2007). Autism Spectrum Disorders: Concurrent Clinical Disorders. *Journal of Child Neurology*, *23*(1), 6–13.

CHAPTER III: PUBLIC DESEMINATION DOCUMENT

Literature Review

Background

Intellectual disabilities are characterised by impairments to intellectual functioning and adaptive behaviour and have a prevalence of approximately one in 100 people. Alongside diagnostic pre-requisites, people with intellectual disabilities also experience a greater likelihood of a range of physical, psychological and behavioural comorbidities. Among the most commonly reported difficulties are problems with sleep. Significant problems with sleep have been reported for over 30 years and are suggested to be greater than those in people without intellectual disabilities. In spite of the large number of research papers that have been published on sleep problems in people with intellectual disability, there are a number of areas of uncertainty:

- People with intellectual disabilities are a varied group and intellectual disabilities have a wide range of causes.
- Many studies have not compared sleep in people with intellectual disability to that in people without intellectual disabilities.
- Early studies relied on parent-report, that can be unreliable.
- Compromises in methodology can lead to biases in research in people with intellectual disabilities.

Approach and method

This review provided the first meta-analysis of studies comparing sleep time and sleep quality in people with and without intellectual disabilities. A systematic search returned 1590 papers, of which 26 papers met inclusion criteria. These papers were screened for

measures of sleep time and sleep quality and appropriate data were extracted. Estimates of sleep time included parent diaries and direct measurement through actigraphy or polysomnography. Estimates of sleep quality included sleep efficiency measures taken from actigraphy or polysomnography and outcomes of questionnaires. All papers were graded on quality using a bespoke quality framework. The meta-analysis used differences between the groups, number of participants tested and variability in outcomes to weight differences in sleep between the two groups. One analysis modelled differences between groups to be the result of random differences between procedures, another used the quality ratings of the studies to rate their importance. Analyses also examined the impact of the nature of the group and how sleep was measured.

Findings

Across 15 studies, people with intellectual disabilities were found to sleep on average 23 minutes less per night than people without intellectual disabilities (see figure 1.3.1). This represented a significant difference between groups in total. In groups of people with intellectual disability of unknown origin, this effect was not found to be significant. In those with specified genetic syndromes or developmental disorders, the effect was significant. The effect was even larger when only studies that measured sleep directly, rather than through questionnaires, were included. Evidence from measures of sleep quality was similar. When studies were only rated on the basis of the number of participants tested and the degree of variability between studies, there was evidence that people with intellectual disabilities experienced poorer quality sleep. When quality criteria were included, this effect became non-significant, though this seemed to be the result of one large high-quality study that was itself significant, but only marginally so. Studies that measured groups of people

with an intellectual disability of unknown origin and studies of people with genetic syndromes and developmental disorders both showed overall significant differences. Studies that reported the number of people with sleep problems or disorders also produced consistent results.

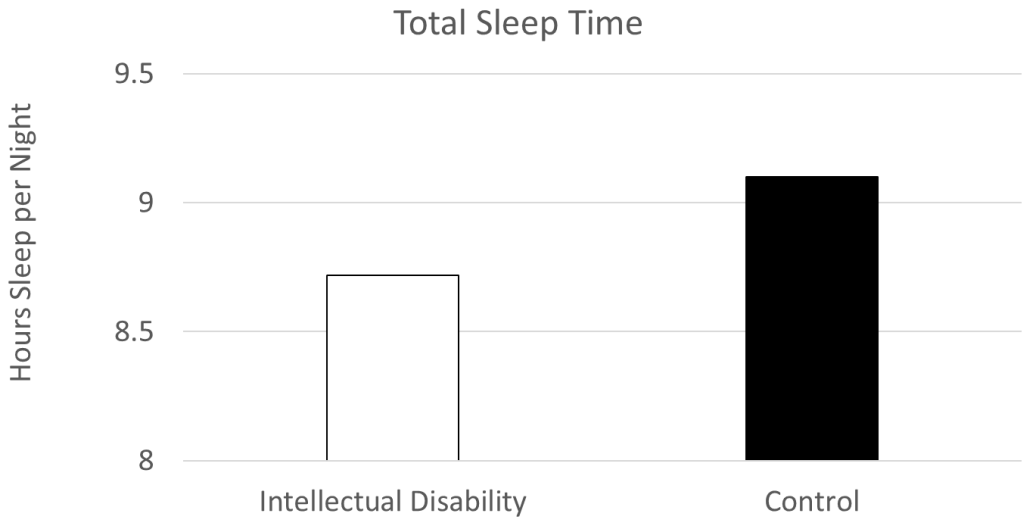


Figure 1.3.1. Mean sleep times in people with and without intellectual disability (mean of study means, not weighted for sample size)

Implications

Sleep problems predict poorer outcomes for health and well-being. This review concluded that people with intellectual disabilities slept for, on average, shorter periods per night than people without intellectual disabilities. Evidence also suggested that this sleep was of a lower quality. The review did not directly address the causes and consequences of this difference, but, there was evidence that:

- Differences in sleep time (but not quality) were only identified in studies of children with specified genetic syndromes or developmental disorders.
- Studies which used direct measures of sleep provided the clearest differences.

- There was no evidence that differences were affected by age, gender or severity of intellectual disability (though often these were poorly defined).

The current literature was significantly limited by the paucity of studies measuring sleep in adults with intellectual disabilities and people with intellectual disability of unknown origin.

Empirical Paper

Background

Autism spectrum disorders (ASD) are diagnosed on the basis of impairments to communication, social interaction and restricted or repetitive behaviours. Prevalence rates of ASD have been estimated at more than one in 100 people in the UK (National Autistic Society, 2016). As with intellectual disabilities, many people with ASD are affected by additional difficulties alongside core impairments. Among the most commonly reported problems are difficulties with sleep. Estimates for the prevalence of sleep problems in children with ASD vary from 44-83%, in comparison with only 9-50% in TD comparison groups (Elrod & Hood, 2015). Numerous studies have compared sleep in groups of people with ASD to those without. Most of these studies have, however, used parent reports or diaries, which can be subject to biases. Of those that have measured sleep directly, the majority have included broad samples of children with ASD, many of whom have intellectual disabilities (known to be linked to poor sleep). Only three previous studies have measured sleep directly in children with ASD and no intellectual disability and compared it to sleep in children without ASD. This is only the second study to have done this for a period of more than two nights.

Approach and method

Sixteen children with ASD, no intellectual disability and parent-reported sleep problems were recruited alongside 16 typically developing children. Sleep in all children was measured directly for at least seven nights using actigraphy. Actigraphy uses a small device a bit like a wristwatch to measure movement. An algorithm is then used to identify whether the person was asleep or awake from this movement. An actigram (figure 1.3.2) can be constructed from this information and shared with people and their families, when clinically useful. Estimates of that person's sleep duration and quality can also be calculated. Alongside these data, parents completed sleep diaries and a broad range of questionnaires.

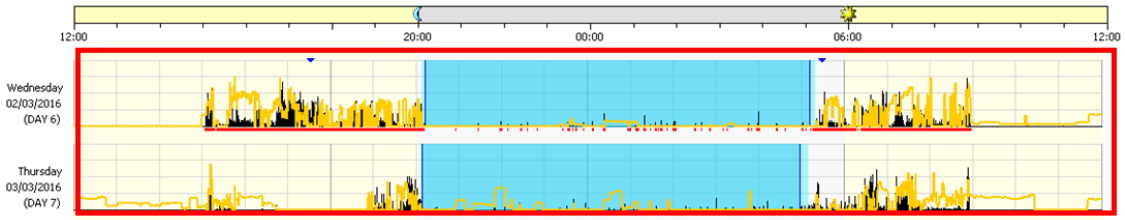


Figure 1.3.2 An example of an actigram.

Findings

Though children with ASD were recruited on the basis of having a sleep problem, neither direct measurement of sleep nor parent-recorded diaries identified differences between the groups in duration of sleep, efficiency of sleep or night wakings. Parent reports in diaries provided accurate information on individual differences in sleep between children, but overestimated sleep duration and efficiency significantly; most likely because they missed many incidents of night-waking. Questionnaires showed significant differences between groups. Children with ASD scored much higher on measures associated with sleep problems.

Subscale measures (see figure 1.3.3) suggested particular difficulties with a delayed onset to sleep, increased sleepiness in the day and increased parasomnias (such as night terrors, or bed-wetting).

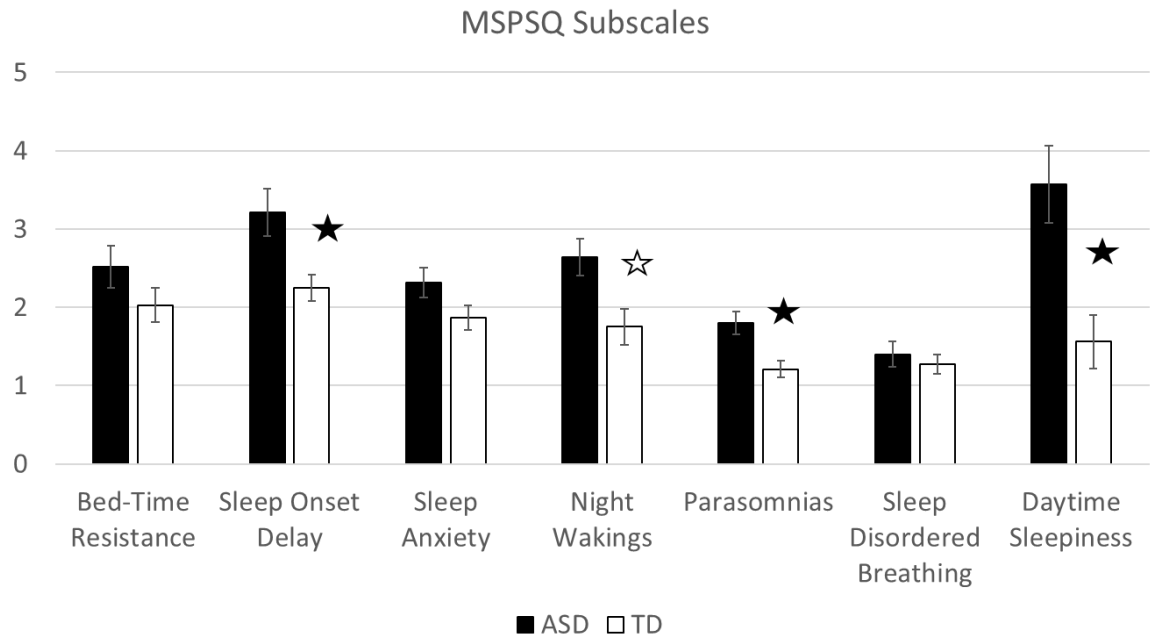


Figure 1.3.3 Subscale scores for different problems with sleep

Implications

That the two groups in the study did not differ on direct measures of their sleep (or parent diaries), was surprising. Many previous studies have suggested sleep problems are more prevalent in children with ASD and in this study the ASD group were recruited on the basis of parent-reported problems with sleep. One reason why no difference was found may be that the majority of previous studies have included children with ASD and a broad range of other conditions, including intellectual disabilities. The questionnaire data suggest alternative explanations, as parents did report significantly greater concerns on these measures:

- Problems may relate to broader difficulties than simply duration or efficiency of sleep (such as parasomnias or day-time sleepiness).
- Parents may successfully employ a range of strategies to manage sleep problems and so they may not be evident through direct measurement.
- Parents may over-estimate the problems their children experience because of increased stress.

The study provided further evidence that parents of children with ASD report a wide range of problems with sleep in their children. Direct measurement, though likely providing an accurate measure of sleep, did not describe parent concern.

References

Elrod, M. G., & Hood, B. S. (2015). Sleep differences among children with autism spectrum disorders and typically developing peers: A meta-analysis. *Journal of Developmental & Behavioral Pediatrics*, *36*(3), 166–177.

The National Autistic Society (2016). *Autism facts and history*. Retrieved from http://www.autism.org.uk/about/what-is/myths-facts-stats.aspx

APPENDICES FOR VOLUME I

List of Appendices

Appendices for Chapter I

Appendix 1.1.1 Full list of search terms.

Searches were made in OVID Medline, OVID Embase and OVID Psychinfo , with the following steps. In all cases .ab,kw,ti, refers to search being conducted within abstracts, keywords and titles. Adj. requires that the word appears near to the "adjacent" word.

Search Terms for Intellectual Disability

1. ((intellectual* or learning or development* or mental) adj (handicap* or retard* or disabilit* or deficien* or disturb* or disord* or incapac*)).ab,kw,ti.
2. idiocy.ab,kw,ti.
3. oligophrenia.ab,kw,ti.

Search Terms for Sleep or Sleep Problems

5. (sleep* or insomni* or dyssomni* or parasomni* or somnolen* or hypersomni*).ab,kw,ti.

Combine 2 terms

6. 1 or 2 or 3
7. 5 and 6

Limitations (note many of these only relevant in a subset of databases).
8. limit 20 to all journals
9. limit 21 to humans
10. limit 22 to disordered populations
11. limit 23 to journal article
12. limit 24 to english language
13. limit 25 to human
14. limit 26 to humans
15. limit 27 to english
16. limit 28 to 1800 quantitative study
17. limit 29 to peer reviewed journal
18. limit 30 to human
19. limit 31 to article
20. limit 32 to humans
21. remove duplicates from 33

Appendix 1.1.2 Full list of dependent variables recorded in papers

Author	Sleep Time Dependent Variable	Sleep Quality Dependent variable	Sleep problem % variable	Other Variables
Anders et al., 2012	Total sleep time			Bedtime, Sleep onset latency, WASO duration, WASO number, Nap duration, Days with naps per week, ESS, CSHQ- daytime sleepiness
Annaz et al., 2011	Total Score			CSHQ- Bedtime resistance, Sleep onset delay, Sleep duration, Sleep anxiety, Night waking, Parasomnias, Sleep disordered breathing, Daytime sleepiness
Ashworth et al., 2013	Total sleep in 24hours (hours)	Sleep efficiency		Bed time, Time in bed, Sleep latency, Assumed sleep time, Actual Sleep time, Night wakings, WASO duration, Moving time, Fragmentation, CSHQ-Bedtime resistance, Sleep onset delay, Sleep duration, Sleep anxiety, Night waking, Parasomnias, Sleep disordered breathing, Daytime sleepiness,
Axelsson et al., 2013	Night sleep	Subjective sleep quality		BISQ- Duration sleep night, Duration sleep day, Number night wakings, Duration night wakings, Settling duration, Time fall asleep. PSQJP- Sleep latency, Sleep duration, Habitual sleep efficiency, Sleep disturbance, Sleep medication, Daytime dysfunction, Global Score
Bruni et al., 2012			Sleep less than 8 hours, poor sleep quality	39 separate questions reported independently (all items from questionnaire on clinical-historical data and on sleep habits and disorders).

Author	Sleep Time Dependent Variable	Sleep Quality Dependent variable	Sleep problem % variable	Other Variables
Buckley et al., 2010	Total sleep time	Sleep Efficiency		Latency to sleep, WASO duration, Stae 1 sleep %, Stage 2 sleep %, Stage 3 or SWS %, REM sleep %, REM sleep latency
Cotton et al., 2006			Prevalence of sleep problem	Severity of sleep problem, type of sleep problem
Cotton & Richdale, 2010	Total sleep time	Sleep Quality		Visual analogue scales- Daytime(sleepiness, napping, excitement, energy, general behaviour), Bedtime (sleepiness, behaviour), night-time (night-wakening, restlessness), sleep (talking/muttering, co-sleeping, sleep quality), morning (forced to wake-up). Direct measures- Bedtime (Time lights out, time asleep, sleep latency), Night-time (time awake, total sleep during night), Morning (time wake up), Overall (total sleep time in 24hours)
Dimitiou et al., 2013	Total sleep time	Sleep efficiency		Actigraphy- Bed time, Assumed sleep time, Sleep efficiency, Actual sleep time, Sleep latency, Night wakings, Moving minutes, Fragmentation. CSHQ- Total score, Bedtime resistance, Sleep onset delay, Sleep duration, Sleep anxiety, Night waking, Parasomnias, Sleep disordered breathing, Daytime sleepiness
Diomedi et al., 1999		Sleep Efficiency		S1%, S2%, S3+S4%, REM%, Number of REM cycles, REM activity, REM density, US%, Wake %, Number of awakenings, Sleep efficiency index, R index, Tonic inhibition index, phasic inhibition index
Elia et al., 2000	Total sleep time	Sleep Efficiency		Time in bed, Sleep period time, total sleep time, sleep latency, Number of awakenings, Number of stage shifts, First REM latency, Sleep

Author	Sleep Time Dependent Variable	Sleep Quality Dependent variable	Sleep problem % variable	Other Variables
				efficiency index, Wakefulness after sleep onset %, S1%, S2%, S3-4%, SREM%, REM density, Density of twitches (S2, S3+4, SREM, Total)
Fraser et al., 2005		Inverse of sleep disturbance		Sleep problems (yes, no), Difficulty getting to sleep, Frequent night wakings, Early morning wakings, Disruptive behaviour, Dangerous behaviour, Sleeps during day, Total sleep disturbance
Fukuma et al., 1974	Total sleep time	Sleep efficiency		Total sleep time, S1%, S2%, S3%, S4%, REMP, IS, Wake, Sleep stage shifts, Number of awakenings
Ghanizadeh & Faghih, 2011		Bedtime resistance and sleep duration		Daytime sleepiness, Parasomnias, Sleep anxiety, other problems
Gombos et al., 2011	Total sleep time	Sleep Efficiency		WASO, Sleep latency, NREM (%), S1%, S2%, SWS%, REM%, REM latency, LM/hour, PLM index, Number of sleep cycles, Average REM period, Average sleep cycle
Goodlin-Jones et al., 2008	Total sleep time	Sleep Efficiency		Bedtime, Sleep start, Sleep end, Time in bed, Sleep onset latency, WASO duration, WASO number, Sleep %, 24-hour sleep, Nappers (number in sample), Nap duration, Number of naps, Sleep problem (%), CSHQ (total), RDC behavioural insomnia (sleep onset), RDC behavioural insomnia (night waking)
Levanon et al., 1999	Total sleep time	Sleep efficiency		Lights out, Time in bed, A/Aw Index, Jerks associated A/Aw, Respiritory associated A/Aw, Stage 2, SWS, REM sleep, Downward shifts

Author	Sleep Time Dependent Variable	Sleep Quality Dependent variable	Sleep problem % variable	Other Variables
Maaskant et al., 2013		Intradaily variability		Interdaily stability, relative amplitude
Masi et al., 2000			Sleep disturbances	
Miano et al., 2004	Total sleep time	Sleep efficiency		TIB, SPT, SOL, FRL, Stage shifts, Awakenings, MT/h, No of REMPs, WASO, S1, S2, SWS, REM, WASO%, S1%, S2%, SWS%, REM%
Miano et al., 2008	Total sleep time	Sleep efficiency		TIB, SPT, SOL, FRL, Stage shifts, Awakenings, No of REMPs, WASO%, S1%, S2%, SWS%, REM%
Richdale & Prior, 1995	Total sleep time			Day Sleepy, Napping, Sleep latency, Sleep onset, night waking, Total sleep per 24 hours, Wake time, Woken, excited/calm
Richdale et al., 2000			Current sleep problem	Past sleep problem, Daytime sleepiness, Daytime naps, Frequent night waking, Snoring, Bed wetting, Sudden night wakings, Breathing difficulties, Crying at night, Yelling at night, Wakes for toilet, Kitchen visits at night, Sleeps in another's bed, Teeth grinding, Sleep walking, Sleep talking
Romeo et al., 2014			% with scores ≥ 70 on SDSC	Subscales on SDSC: Difficulty in initiating or maintaining sleep, Sleep breathing disorders, Disorders of arousal, Sleep-wake transition disorders, Disorders of excessive somnolence, Sleep hyperhidrosis,
Sniecinska et al., 2015		Sleep Efficiency		Actigraphy- Time in bed, sleep latency, Wake time, night waking, Moving time, Sleep fragmentation index. CSHQ- Bedtime resistance,

Author	Sleep Time Dependent Variable	Sleep Quality Dependent variable	Sleep problem % variable	Other Variables
				Sleep onset delay, Sleep duration, Sleep anxiety, Night wakings, Parasomnias, Sleep disordered breathing, Daytime sleepiness, Total score.
Tawfik et al., 2009	Total sleep time			In Rem periods, Arousals, Spindles, Obstructive apnoeas, Central apnoeas, Hypopneas,

Appendix 1.1.3 Full list of Quality Ratings for each study

Paper	Measure	ID Sample	Sample		Measure of ID		Measure of Sleep	Total
			1a	1b	2a	2b	3	
Anders et al., 2012	Actigraphy	ID	2	2	3	3	3	.89
Annaz et al., 2011	CSHQ	WS	2	2	1	1	1	.44
Ashworth et al., 2013	Actigraphy	WS	2	2	1	1	3	.67
		DS	2	2	1	1	3	.67
Axelsson et al., 2013	BISQ	WS	2	0	1	1	1	.33
Bruni et al., 2012	Questionnaire	AS	2	2	1	1	0	.33
Buckley et al., 2010	Polysomnography	DD	0	0	0	0	3	.33
Cotton et al., 2006	Questionnaire	ID	2	2	1	1	0	.33
		PWS	2	2	1	1	0	.33
		ASD	2	2	1	1	0	.33
		DS	2	2	1	1	0	.33

Paper	Measure	ID Sample	Sample		Measure of ID		Measure of Sleep	Total
			1a	1b	2a	2b	3	
Cotton & Richdale, 2010	Questionnaire	ID	0	0	1	1	2	.33
		PWS	0	0	1	1	2	.33
		ASD	0	0	1	1	2	.33
		DS	0	0	1	1	2	.33
Dimitiou et al., 2013	Actigraphy	WS	2	2	3	1	3	.78
Diomedi et al., 1999	Polysomnography	ASD	1	0	3	0	3	.56
		DS	0	0	3	1	3	.56
Elia et al., 2000	Polysomnography	FX	0	0	1	1	3	.44
Fraser et al., 2005	Questionnaire	SS	2	2	1	1	2	.56
Fukuma et al., 1974	Polysomnography	DS	1	0	3	1	3	.61
		ID	1	0	3	0	3	.56
Ghanizadeh & Faghih, 2011	Questionnaire	ID + Medical condition	2	2	1	1	1	.44

Paper	Measure	ID Sample	Sample		Measure of ID		Measure of Sleep	Total
			1a	1b	2a	2b	3	
		ID + No medical condition	2	2	1	1	1	.44
Gombos et al., 2011	Polysomnography	WS	2	1	1	1	3	.61
Goodlin-Jones et al., 2008	Actigraphy	ASD	2	2	3	3	3	.89
		ID	2	2	3	3	3	.89
	Parent report on single question	ASD	2	2	3	3	0	.67
		ID	2	2	3	3	0	.67
Levanon et al., 1999	Polysomnography	DS	1	1	1	1	3	.56
Maaskant et al., 2013	Actigraphy	ID	2	2	1	1	3	.67
Masi et al., 2000	Parent response and interview	ID	1	1	3	0	0	.28
Miano et al., 2008	Polysomnography	FX	1	0	3	1	3	.61
	Polysomnography	DS	1	0	3	1	3	.61

			Sample		Measure of ID		Measure of Sleep	Total
Paper	Measure	ID Sample	1a	1b	2a	2b	3	
Miano et al., 2004	Polysomnography	AS	2	0	1	1	3	.56
Richdale et al., 2000	Question	ID	2	1	1	1	0	.28
Richdale and Prior,1999	Diaries	ASD-LF	2	0	3	0	2	.50
Romeo et al., 2014	Questionnaire	CP + ID	1	1	3	1	1	.44
Sniecinska et al., 2015	Actigraphy	WS	2	2	1	1	3	.67
	CSHQ	WS	2	2	1	1	1	.44
Tawfik et al., 2009	Polysomnography	FX	1	1	3	1	3	.67

Appendices for Chapter II

Appendix 1.2.1 Actigraphy Data Cleaning Procedure

Needed to complete cleaning

a) Open file on actiware
b) Child diary.

Step-1: Exclude any automatically-coded intervals which occur after the watch has been collected (E, Shift E, Ctrl E). This information can be gained from the sleep diary.

Step-2: Exclude any nights during which the parent identifies a time when the watch was taken off.

- Open child diary to the relevant night, and confirm if parent reports any times that watch was taken off between lights out and wake-up time. If there are any such times check against actigram for consistency (i.e. no movement at this time). If consistent, exclude this night from data in actigram (E, Shift E, Ctrl E). If parent reports that the watch has been removed, but this is not evident on the actigram (i.e. evidence of movement during this time), keep the existing interval as it is and continue to step 3.

Step-3: Exclude any nights during which the watch appears to have been taken off, but this was not noted in parent diary.

- Visually inspect each night on actigram. If on any night, there is no recorded activity (**0 in activity column** of data list) for a period of 2 hours or more, exclude this whole night from actigram (E, Shift E, Ctrl E).

Step-4: Clear any automatically-calculated sleep intervals from the day time and insert interval to night time

- Note any occasions on which the software has coded the sleep interval as in the day-time. Criteria for this is if the automatically-coded interval both starts and ends outside of period noted as sleep in parent diary. For any intervals on which this is the case, clear the sleep interval (Right click, clear interval).
- New interval should be inserted.
- To allocate start time of new interval: 1. Find first period of 20 minutes of sleep after lights out in diary (40 epochs coded as **0 in sleep/wake column** in data list). From there, go back to the last period of 10 minutes of activity (20 epochs coded as **1 in sleep/wake column**). Start time is first 0 after this.

- To allocate end time: 1. Find last period of 20 minutes of sleep before wake-up time in diary (40 epochs coded as **0 in sleep/wake column**). From there, go forward to the first period of 10 minutes of activity (20 epochs coded as **1 in sleep/wake column**). End time is first 0 before this.

Step-5: Extend any intervals that have not captured entire night sleep.

- Locate any 20 minute periods coded as sleep in the actigram (40 epochs coded as 0 in sleep/wake column), that are **not** found within the automatically calculated sleep interval, **but are** between lights out and wake-up in sleep diary.
- If period is after the automatically calculated interval, extend interval from sleep period to last point before 10 minutes coded as awake (20 consecutive scores coded as 1 in sleep/wake column on datalist). To do this, clear the original interval and add a new one with the original start time and the new end time.
- If period is before the automatically calculated interval, extend interval from sleep period to first point after 10 minutes coded as awake (20 consecutive epochs coded as 1 in sleep/wake column on datalist). To do this, clear the original interval and add a new one with the original end time and the new start time.

Step-6: Exclude any intervals that have twice the duration of the average Total Sleep Time

- Note any occasion where the software has created a sleep interval where the duration of Total Sleep Time and/or the sleep diary has stated that time between lights out and waking up time is twice that of the average TST. Exclude sleep interval.

Appendix 1.2.2 Sleep Diary

To be completed throughout the day				**Completed by (initials)**
Time Actiwatch Removed		Time Actiwatch Replaced		
Time Actiwatch Removed		Time Actiwatch Replaced		
Time Actiwatch Removed		Time Actiwatch Replaced		
To be completed in the evening				
Nap 1 Start time: End time:	**Nap 2** Start time: End time:	**Nap 3** Start time: End time:		
Please list any sedentary activities after 6pm **e.g. reading alone or with an adult, watching TV**				
Type of activity (select one) ☐ Watching TV ☐ Reading alone or with an adult ☐ Other- please state	**Start time of activity**	**End time of activity**		
Type of activity (select one) ☐ Watching TV ☐ Reading alone or with an adult ☐ Other- please state	**Start time of activity**	**End time of activity**		
Type of activity (select one) ☐ Watching TV ☐ Reading alone or with an adult ☐ Other- please state	**Start time of activity**	**End time of activity**		
Time got into bed:				
Time lights turned off: 				
Child's behaviour at bedtime (Select one): ☐ No behaviours of concern ☐ Will not stay in bed/wants to play ☐ Upset when caregiver leaves the room ☐ Become distressed – no obvious reason ☐ Destructive or self-injurious behaviour				
Response to child's behaviour at bedtime (Select one): ☐ Not applicable - No behaviours of concern ☐ Ignore ☐ Verbally reassure/cuddles etc. then leave the room ☐ Verbally remind child about bedroom expectations ☐ Stay in bedroom until child falls asleep ☐ Let child watch TV/play on tablet				

To your knowledge, was the event marker pressed at the correct time? (Please circle)		
Yes	**No**	

To be completed in the morning		
Time woke up: 		
Time got out of bed:		
Estimated time taken to fall asleep:		
To your knowledge, was the event marker pressed at the correct time? (Please circle)		
Yes	**No**	
Child's behaviour when getting out of bed: ☐ No behaviours of concern ☐ Refuses to get out of bed		
Response to child's behaviour (select one): ☐ Not applicable- no behaviours of concern ☐ Ignore behaviour ☐ Verbally remind child about morning routine expectations ☐ Suggest removal of preferred activity/item if will not get up		
Please rate how typical your child's sleep quality was **1 = Significantly better than usual, 3 = Typical and 5 = Significantly poorer than usual**		
1 2 3 4 5		
Do you think your child slept well? (Please circle)		
Yes	**No**	

Please record any times your child woke up during the night	
Waking 1	**Waking 2**
Time of waking: **End of waking:** **Perceived reason for waking (select one):** ☐ Wet/needing toilet ☐ Hungry/thirsty ☐ Pain/discomfort ☐ Anxiety ☐ Unknown ☐ Other – please state: **Child's behaviour during waking (select one):** ☐ No behaviours of concern ☐ Will not stay in bed/wants to play ☐ Become distressed ☐ Destructive or self-injurious behaviour **Response to child's behaviour (select one):** ☐ Not applicable - No behaviours of concern ☐ Ignore ☐ Verbally reassure/cuddles etc. then leave the room ☐ Verbally remind child about night-time expectations ☐ Stay in bedroom until child falls asleep ☐ Let child watch TV/play on tablet ☐ Give child a drink/take to the toilet etc.. but minimising attention	**Time of waking:** **End of waking:** **Perceived reason for waking (select one):** ☐ Wet/needing toilet ☐ Hungry/thirsty ☐ Pain/discomfort ☐ Anxiety ☐ Unknown ☐ Other – please state: **Child's behaviour during waking (select one):** ☐ No behaviours of concern ☐ Will not stay in bed/wants to play ☐ Become distressed ☐ Destructive or self-injurious behaviour **Response to child's behaviour (select one):** ☐ Not applicable - No behaviours of concern ☐ Ignore ☐ Verbally reassure/cuddles etc. then leave the room ☐ Verbally remind child about night-time expectations ☐ Stay in bedroom until child falls asleep ☐ Let child watch TV/play on tablet ☐ Give child a drink/take to the toilet etc.. but minimising attention

Waking 3	Waking 4
Time of waking: **End of waking:** **Perceived reason for waking (select one):** ☐ Wet/needing toilet ☐ Hungry/thirsty ☐ Pain/discomfort ☐ Anxiety ☐ Unknown ☐ Other – please state: **Child's behaviour during waking (select one):** ☐ No behaviours of concern ☐ Will not stay in bed/wants to play ☐ Become distressed ☐ Destructive or self-injurious behaviour **Response to child's behaviour (select one):** ☐ Not applicable - No behaviours of concern ☐ Ignore ☐ Verbally reassure/cuddles etc. then leave the room ☐ Verbally remind child about night-time expectations ☐ Stay in bedroom until child falls asleep ☐ Let child watch TV/play on tablet ☐ Give child a drink/take to the toilet etc.. but minimising attention	**Time of waking:** **End of waking:** **Perceived reason for waking (select one):** ☐ Wet/needing toilet ☐ Hungry/thirsty ☐ Pain/discomfort ☐ Anxiety ☐ Unknown ☐ Other – please state: **Child's behaviour during waking (select one):** ☐ No behaviours of concern ☐ Will not stay in bed/wants to play ☐ Become distressed ☐ Destructive or self-injurious behaviour **Response to child's behaviour (select one):** ☐ Not applicable - No behaviours of concern ☐ Ignore ☐ Verbally reassure/cuddles etc. then leave the room ☐ Verbally remind child about night-time expectations ☐ Stay in bedroom until child falls asleep ☐ Let child watch TV/play on tablet ☐ Give child a drink/take to the toilet etc.. but minimising attention

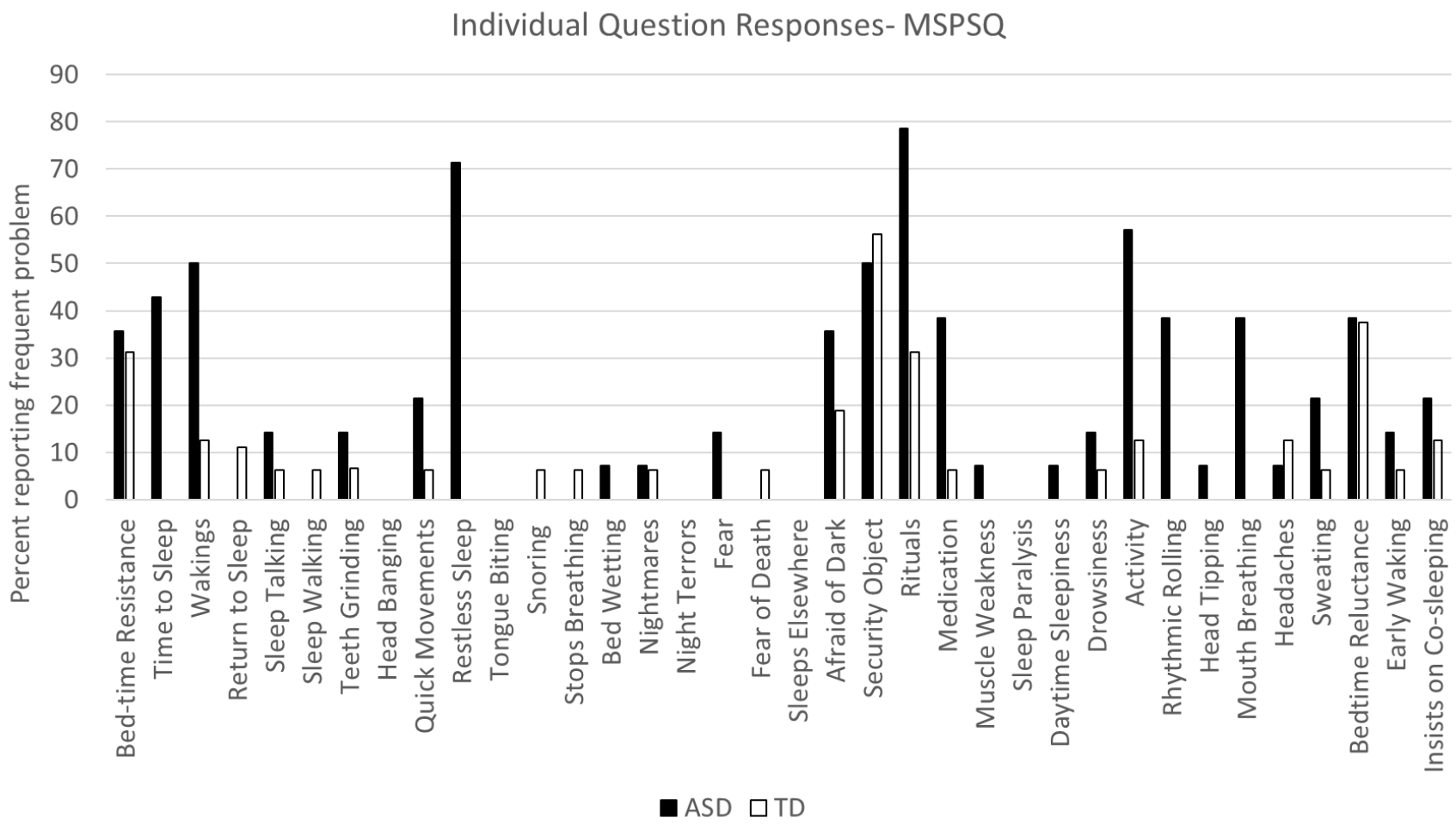
Any other notes:

……
……
……

Appendix 1.2.3 Correlations and comparisons between actigraphy and Sleep diary measures for children in the TD group

Actigraphy Variable	Sleep Diary Variable	*r*	*p*	*t*	*p*
Bed Time	Bed Time	.82	< .001	5.02	.001
Get up time	Wake Time	.70	.002	.33	.75
Time in Bed	Time in Bed	.78	< .001	5.92	< .001
Total Sleep Time	Total Sleep Time	.53	.04	3.60	.003
Sleep Latency	Time to get to sleep	.48	.08	.28	.78
Sleep Efficiency	Sleep Efficiency	-.07	.79	80.14	< .001
Wake After Sleep Onset	Wake After Sleep Onset	-.01	.96	2.27	.04

Appendix 1.2.4 Individual Question Responses on the MSPSQ



Appendix 1.2.5 Correlation between dependent variable questionnaires and sleep outcome measures (from actigraphy) in the TD group

Measure	Total Sleep Time, *ρ (p)*	Sleep Efficiency, *ρ (p)*
Age	-.32 (.22)	-.32 (.22)
Sleep	.29 (.27)	.13 (.62)
Sleepiness	.25 (.36)	-.10 (.71)
Social Communication	.16 (.59)	.09 (.75)
Family Sleep Habits	-.11 (.69)	-.26 (.35)