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Sleep duration and sleep quality in people with and without intellectual disability: A meta-analysis.

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Summary

This study provides the first meta-analysis of the purported differences in sleep time and sleep quality between people with and without intellectual disabilities. Twenty-one papers were identified that compared sleep time and/or sleep quality in people with and without intellectual disabilities. The meta-analysis of sleep time revealed that people with an intellectual disability slept for 18 minutes less, on average, than people without an intellectual disability. This significant difference was limited to those studies that tested groups of people with an identified genetic syndrome or developmental disorder. 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. There were no differences found between studies that measured sleep directly and those that used diary or questionnaire measures. Notably, most samples were drawn from populations of people with specified genetic syndromes or developmental disorders, rather than intellectual disability of heterogeneous origin. Similarly, most studies investigated sleep in children, although there was no evidence that the differences between the groups reduced 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

Intellectual disabilities are defined by co-occurring deficits in intellectual ability (often defined as an IQ < 70) and deficits in day-to-day functioning. [1,2] An estimated 2-3 % of people in the United States are thought to have an intellectual disability. [3] These people are by no means a homogenous group, with causes of intellectual disability including genetic syndromes, pre-natal complications, perinatal insult and later brain injury. [4] As well as deficits in intellectual ability and functioning, people with intellectual disability are at risk of a range of comorbid psychological and social difficulties [4], and of physical health and other developmental problems. [5]

Reports of experienced clinicians and a growing body of empirical evidence support the idea that children with intellectual disabilities more regularly experience poorer sleep than typically developing children.[6-12] The smaller body of evidence on sleep in adults with intellectual disabilities presents similar findings.[13-15] In spite of the growing number of studies conducted with these populations, reviews have suggested that there are significant limitations to knowledge of how sleep differs in people with and without intellectual disabilities.[10] Methodological differences between studies have made the prevalence of sleep problems hard to quantify, with estimates ranging from 13% to 86%.[8] 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. Among the most common reported problems with sleep in this population are shorter durations of sleep time and lower scores on various indices of quality of sleep [16,17]. One way of understanding sleep problems in this population, therefore, is to quantify the differences in sleep time and sleep quality between this group and people without intellectual disabilities.

The current systematic review includes the first meta-analysis of studies comparing sleep in people with and without intellectual disabilities. The search terms used identified measures of sleep time, estimates of sleep quality and frequency of sleep problems.

Challenges in Researching Sleep in People with Intellectual Disabilities

Understanding and Defining Sleep. One reason for the variability in findings across studies of sleep in people with intellectual disabilities has been the limitations to our understanding of sleep more broadly. The science of sleep has been developing rapidly over the past 30 years,[18] and large variability in sleep exists in people with seemingly healthy sleep patterns.[19] Unhealthy sleep has been categorised into two types. A person experiencing significantly reduced sleep time, may be categorised as having a *dyssomnia*. These reflect difficulties in initiating or maintaining sleep (including Insomnia, ICD-10;[2] American Sleep Disorders Association[20]). Unhealthy sleep does not, however, necessarily affect sleep time. *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,[8] but also evidence that the variability in quality of sleep can extend beyond the presence of a specified disorder[8]. Whether meeting diagnostic criteria for a sleep disorder or not, poor sleep can have an impact on people and their families and poor sleep is often inferred based on either shortened sleep time or reduced sleep quality.[8] In this review, *sleep time* and *sleep quality* are treated as distinct dependent variables. Advantages of this include allowing for broad conclusions and reducing the chance of type-1 error (through making fewer comparisons). One disadvantage is that sleep quality has been considered in numerous ways and therefore is not necessarily a homogenous construct.

Sleep time as a lay construct is defined in different ways- varying from the total amount of sleep across a day to the longest period of sleep within a day. In studies that measure sleep directly, the construct of Total Sleep Time (TST) is typically used. This includes the total time spent asleep over a defined night sleep period. Shorter sleep time has

been associated with poor functional outcomes for people with intellectual disability, including more sleepiness the following day and increased challenging behaviour.[21-22] For this reason, we have considered group differences evidencing shorter sleep time to reflect poorer sleep within that group, though we acknowledge that ideal sleep time may differ from one individual to the next. For the meta-analysis, measures of *sleep time* will include TST from direct measures of sleep, reported hours of sleep in 24 hours (where TST not available) and reported hours of from diaries (as appropriate).

Sleep quality is perhaps even harder to define and it is broadly accepted that sleep quality is not equivalent to sleep time, although the two are related. [8,23] For instance, fractured sleep time and regularly waking may compromise sleep quality beyond reducing the overall duration of sleep. [8] Our strategy for the current paper has been to include a measure of sleep quality where possible, acknowledging that this may mean combining studies that did not measure the same aspect of sleep quality (see discussion in Method).

Individual differences and potential confounds. Most research on sleep in people with intellectual disability has been undertaken with children[24]. Most parents of this group report that they believe poor sleep in their children is an inevitable result of intellectual disability or brain damage.[9] However, people with intellectual disabilities are by no means a homogenous group, so consequently problems with sleep are likely of varied aetiology[25]. Factors such as severity of intellectual disability[14,24] and poor social and communication skills[9] have been linked to poor sleep. This suggests that people with intellectual disabilities may be more or less likely to experience poor 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.[26,27] 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,[28] which greatly increases the likelihood of problems with sleep.[29]

People with intellectual disabilities are also more likely to have comorbid genetic and developmental disorders, many of which have been associated with poorer sleep. People with Smith Magenis syndrome often evidence inverted melatonin cycles,[30] 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 problems with sleep in people with Down syndrome,[31] perhaps associated with the physical differences linked to the condition predisposing sleep disordered breathing and sleep apnoea.[32] Additionally, poorer sleep is reported in children with Angelman syndrome,[33] Williams syndrome,[34] Fragile-X syndrome,[35] Prader- Willi Ssyndrome,[36] Rett syndrome,[37] Sanfilippo syndrome[38] and Jacobsen syndrome.[39] Sleep in people with autism spectrum disorders (ASD) has received more attention than other developmental disorders. [10] Interestingly, increased autistic symptomology predicts an increased likelihood of problems with sleep.[40,41] There is also some evidence that intellectual disability is further predictive of poorer sleep in people with ASD.[11] Given these reports, in the current study, sleep 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[8], or reports of staff in care homes, [14] rather than direct measurement. This clearly reduces methodological load on experimenters, individuals, their families and staff, often allowing for larger sample sizes. However, there is conflicting, yet significant, evidence showing parents

of typically[42] and atypically[43] developing children overestimate their child's sleep difficulties. Similarly, there is evidence of poor concordance between parent report and actigraphy in other developmental disorders, such as ADHD[44]. Evidence is less clear for adults with intellectual disabilities. Additionally, questionnaire measures, the most common tool for such studies, are rarely validated for populations of people with intellectual disabilities. [10] Similarly, most samples have been, at least to some degree, self-selecting (although systematic cohort samples have been collected[9,13]). In the current review, supplementary analyses investigate whether any group differences are reflected when only studies using direct measures are analysed. Furthermore, a set of independent quality criteria against which to weight studies based on the strength of methodology is implemented.

Rationale

Over the past 25 years, there have been a significant number of studies reporting comparisons of sleep time and sleep quality in populations of people with and without intellectual disabilities. However, these studies typically have comparatively few participants and often 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. This meta-analysis, examining the evidence for whether people with intellectual disabilities do have a shorter duration and poorer quality of sleep than people without intellectual disabilities, is timely. We provide further, exploratory analysis on 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. 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 and colleagues' recent systematic review of sleep in adults with intellectual disability. [14] This recent review was aimed only at adults and specifically at sleep problems, but also included more generic terms for sleep. For full search terms see supplementary materials (S1). Terms for intellectual disability included: intellectual disability, intellectual disturbance, learning disability, mental retardation, mental handicap, mental deficiency, mental disorder[i], mental incapacity, idiocy, down syndrome, oligophrenia and variants thereof. Terms for sleep 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. The final search returned 1590 results. The reference lists of three recent systematic reviews on similar topics[10, 12, 14] were also screened for papers that were not returned by the original search.

Paper Selection

Paper selection was completed by the first author. Figure 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) or a measure of

[i] The term "mental disorder" may have unnecessarily elevated the number of studies returned, as it can describe mental health conditions, as well as being used as an archaic term for intellectual disability. Our exclusion criteria, however, meant that this did not affect the final selection of papers.

sleep time/ quality. If this was not the case, the full paper was retrieved and included/ excluded based on the same criteria.

++++++++++++++++Insert figure 1 about here++++++++++++++++++++++

++++++++++++++++Insert table 1 about here++++++++++++++++++++++

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[ii]. 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 disability, see table 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 intellectual disability,[46] 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 disability was reported, as only approximately 55% of people with ASD are thought to have an intellectual disability.[47] 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

[ii]Studies with fewer than five participants in both groups were felt more likely to reflect small-N case studies and single case experimental designs, rather than group-level comparisons. Readers interested in these studies may be interested in a recent meta-analysis of Single Case Experimental Designs analysing behavioural interventions for sleep in people with intellectual disabilities[45].

functioning, as well as cognitive performance.[1,2] A measure of functioning was not required for the study 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)[27], but excluding participants with 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, sleep questionnaires and direct measurement through polysomnography or actigraphy. 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 first 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 the 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[iii]. 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. 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 "sleep quality" and "sleep problems". Sleep problems were understood to be inversely related to sleep quality. Using a wide variety of measures of sleep quality is problematic in potentially combining different factors of the same construct. However, given the relatively small literature in this area, it was felt that this was more beneficial than excluding data (or treating it within separate analyses). 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.[50]

[iii] Sleep over a 24-hour period was used from Goodlin-Jones et al.[48] and Anders et al.[49]. Though this might provide more variable data through including day-time naps, it was felt that the cost of this extra variability was out-weighed by the benefit of including a broader range of studies. The two studies that did include naps as well as night sleep tested pre-school children.

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" indicates 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 2) was adapted from Richards and colleagues,[51] 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.[2] Quality of measurement of sleep reflected the use of indirect or 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 and Black[52] 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 author (DE) also completed ratings using 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 adaptive functioning, α = 1.0).

++++++++++++++++Insert table 2 about here++++++++++++++++++

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 S2). The 21 papers in the analysis included 33 groups of people with intellectual disabilities (see table S2 for a full description of the papers, including participant demographics, study methodology and quality ratings) 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 and colleagues[53] reported on an adult sample, Maaskant and colleagues[54] on a sample of older adults, and all other samples had an average age of less than 18 (average ages: 2.54-13.5 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 intellectual disability by ICD-10 criteria.[2]

Study Quality.

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. Studies in which children with a specified genetic disorder were recruited via an internally held database of self-selecting families and compared to a group of typically developing children recruited from a local school scored poorly under the criteria, but reflect 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 parents 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,[1,2] 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. [9]

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[8] reviewed papers and concluded most were based on parent report. Alternatively, it could be the case that studies that employ a control group are also more likely to use a direct measure of sleep.

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 (REM) was tested. The REM 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 (QEM) was employed. The QEM 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 control data are replicated for comparison with multiple syndrome groups then this increases the probability of a type one error by increasing the end size of the comparison 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 intelligenece quotient (IQ; or lowest IQ recorded within the literature, see table 1) was chosen. For one study the "younger children" group was selected over the older as this was closer to the average age across the whole analysis.[33]

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.[55] 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. Analysing syndrome groups alongside groups of people with intellectual disability of heterogeneous aetiology allows for the best representation of the broad population of people with intellectual disabilities (in which both groups often access the same services indiscriminantly). Analysing them separately allows for understanding of whether any differences are likely the result of intellectual disability itself or likely the result of other factors which are regularly associated with intellectual disability. If one of these analyses revealed a group difference, but the other did not, a t-test was undertaken to investigate whether this apparent difference was supported statstically. Finally, correlations between weighted mean difference (WMD) and, age, gender and IQ were undertaken to investigate whether the evidence differed across these demographic factors.

Sleep time

Primary Analysis. Fifteen studies reported a measure of sleep time (see supplementary materials for a table of these studies and means for each group; S3). 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 REM (table 3) revealed a significant difference, such that groups of people with intellectual disabilities slept for shorter periods each night than did people without intellectual disabilities (see forest plot, figure 2). The QEM revealed that weighting studies by their quality did not have an impact on the significance of the model (figure 3). The mean difference equated to 18 minutes less per night for people with intellectual disabilities, ranging from 52

minutes more to 106 minutes less across studies. Lower sleep time was obtained for people with intellectual disabilities in all instances, except for Richdale and Prior,[56] Fukuma and colleagues,[57] Buckley and colleagues[58] and two of the three groups from Cotton and Richdale.[59] 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 REM, but remained significant in the QEM.

Secondary analysis. The effect remained significant when only studies measuring sleep time directly were included, for the REM and for the QEM. Splitting the analysis, revealed that the effect was significant for studies that reported on specified genetic or developmental disorders, for the REM and for the QEM. Data from those (relatively few) studies that reported on a group of people with undifferentiated intellectual disability or developmental delay did not evidence a significant pooled effects in either the REM or the QEM. 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) = -.328, $p = .10$), the proportion of male participants (r_s (18) = -.018, $p = .93$), nor the average IQ of the group (r_s (6) = .60, $p = .12$).

++++++++++++++Insert table 3 about here++++++++++++++++++++++

++++++++++++++Insert figures 2 and 3 about here++++++++++++++++++

Sleep quality

Primary analysis. Eighteen studies reported a measure of sleep quality (see supplementary materials for a table of these studies and means for each group; S4). These contained 27 experimental groups of people with intellectual disabilities, so typically developing comparison groups were repeated on nine occasions. Annaz and colleagues[60] reported a total score from the childhood sleep habits questionnaire, Cotton and Richdale[59] reported a questionnaire measure of sleep quality, Fraser and colleagues[61] a questionnaire subscale on Sleep Disturbance, Ghanizadeh and Faghih[62] on bedtime resistance and sleep duration, Maaskant and colleagues[54] reported intradaily variability and all others reported sleep efficiency (the proportion of time spent in bed actually asleep). The REM (table 4) revealed a significant difference, such that people with intellectual disabilities experienced significantly poorer sleep than people without intellectual disability (see figure 4). Ashworth and colleagues[34] group of children with Williams Syndrome and Fukuma and colleagues[57] 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 QEM, however, did not show a significant effect (figure 5). The change in significance was particularly driven by one, large and high quality study[54], which received over 50% of the weighting in this model, and if removed reinstated the significant effect. When only one group of people with intellectual disability was included, the effect remained significant for the REM and non significant for the QEM.

***Secondary* analysis.** Excluding studies in which sleep quality was measured indirectly did not affect the significance of the effect, for the REM and for the QEM. Splitting the analysis, revealed that the effect was significant for studies that reported on specified genetic or developmental disorders, for the REM and for the QEM. Data from those (relatively few) studies that reported on an undifferentiated intellectual disability group produced a significant pooled effect for the REM and a marginally significant effect for the QEM.

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) nor the average IQ of participants (r_s (6) = .60, p = .12).

++++++++++++++Insert table 4 about here++++++++++++++++++++++

++++++++++++++Insert figures 4 and 5 about here++++++++++++++++++

Sleep Problems

Five studies only reported the frequency of sleep problem in groups of people with and without intellectual disabilities (figure 6). Only one of these studies found people without intellectual disabilities to experience more problems than people with intellectual disabilities and in this study the both groups were recruited on the basis of a diagnosis of Generalized Anxiety Disorder.[63]

++++++++++++Insert figure 6 about here++++++++++

Discussion

Summary

Research on sleep in people with intellectual disabilities and/ or developmental disorders has blossomed over the last 15 years. [12] 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. These findings were clearest in those people with

specified genetic syndromes or developmental disorders: Evidence for problematic sleep in people with intellectual disability of heterogeneous origin was limited to impairments in *sleep quality*, whilst those with specific genetic syndromes or developmental disorders evidenced both shorter duration and poorer quality sleep. That the data were for the most part robust in both the REM and QEM suggested that findings were not influenced by a small number of studies with poorer methodology. Only five papers were identified that allowed for comparison of the proportion of people with sleep problems in each group. These also supported the conclusion that sleep quality was poorer in people with intellectual disabilities.

Sleep time and quality in people 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 than do people who are typically developing. Across the fifteen studies included in the analysis, 446 people with intellectual disabilities averaged 18 minutes less sleep each night than did 391 people drawn from typically developing populations. The significant finding was shown to be evident when only those studies reporting on genetic syndromes were included (a mean difference of 33 minutes less sleep in the experimental group), but not when only participants with intellectual disability of heterogeneous aetiology were included (a mean difference of 3 minutes less sleep in the experimental group), with a statistical difference identified between these groups. Given only 5 studies were identified that tested people with intellectual disability of heterogeneous origin, this may, in part be due to lack of power. 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.

Shorter sleep time is not evidence of a clinical problem with sleep. However, shorter sleep durations in people with intellectual disabilities have been associated with increased day-time

challenging behaviour,[20] poorer attention[64] and increased parent stress.[65] This suggests that understanding this difference further remains an important task in improving the lives of people with intellectual disabilities and their families.

One caveat to conclusions drawn 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 REM). This reflected a widening of confidence intervals (rather than a substantial change to the WMD), suggestive that further research may be required to add to the power of the evidence base.

Sleep quality. Using a REM, 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 asleep. Further studies, however, reported summary variables from questionnaires. This may account for the large degree of heterogeneity identified in the analysis. The QEM for sleep quality showed no significant difference. Here one study[64] had a significant impact on the outcome. Interestingly, this study itself reported a significant difference, suggesting that the result from the QEM 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 REM). Again, there was no evidence that age, IQ or gender made a difference to the data.

The descriptive data from the five studies that measured the proportion of participants with sleep problems were consistent with the other findings: 80% of the studies found a greater proportion of people with intellectual disabilities experienced sleep problems. The one that did not,

included a control group with a comorbid mental health problem for which poor sleep is a diagnostic criterion (people with Generalized Anxiety Disorder) and so may be thought atypical.

Clinical relevance

The scope of this meta-analysis has not allowed for the statistical comparison of the number of people with and without intellectual disabilities who meet criteria for a diagnosable sleep disorder[20]. Criteria for sleep disorders are not typically validated for people with intellectual disabilities and may miss other differences that have an impact on functional outcomes. The two variables tested here "sleep time" and "sleep quality" were chosen to reflect differing aspects of what might be considered good sleep. Robinson and Richdale[16], for instance, found that 58% of reported sleep problems in children with intellectual disabilities related to settling, night-waking or both, which might be expected to impact on sleep time. Importantly, Robinson and Richdale also showed that children with a sleep problem slept for significantly shorter periods each night (by more than one hour), suggesting that even if shortened sleep time might not be the identified sleep problem, it may still be affected. Shortened sleep time has also been found to be linked to negative functional outcomes, such as day-time sleepiness and challenging behaviour [21,22]. Throughout this review, we have defined sleep quality broadly, to incorporate a range of findings. Whilst it is not possible to conclude based on our data that people with intellectual disabilities experience a greater prevalence of clinical problems with sleep, we can conclude that sleep quality is typically lower. Again, it is worth noting that poor sleep quality is related to other poor outcomes in this group, such as parent stress[11,65]

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 and Hood's[66] recent meta-analysis comparing sleep in children with ASD to children who develop typically. There have, however, been several reviews on the topic. Didden and Sigafoos[8] 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[8] cited only a single study comparing sleep in people with intellectual disabilities to people without intellectual disabilities directly. [11] 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. We also, draw more fine-grained distinctions, for instance between people with intellectual disabilities of heterogeneous origin (for whom we found evidence of poorer sleep quality) and people with specified genetic syndromes or developmental difficulties (for whom we found evidence of poorer quality *and* shorter duration sleep). More recently, Richdale and Baker[10] 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 likely to have poorer sleep. Much of this literature comprised reviews, case-studies, treatment studies or studies with no typically developing group for comparison. Tietze and colleagues[12] noted the evidence for sleep disturbances in children with genetic syndromes and/ or intellectual disabilities and made the case for further investigation into children with multiple disabilities. Van de Wouw and colleagues[14] completed the first review of sleep in adults with intellectual disability. The authors concluded that evidence in this cohort was weak and largely based on "subjectively derived data" (Van de Wouw et al.[14], p1). The current review is consistent with this, in finding only two studies on adult populations. There is no evidence from those studies examined to suggest that age impacts on group differences between people with and without intellectual disability.

Limitations of this review

All the papers included in the review were rated as adequate or better using the quality framework. Methodological procedures have advanced greatly since Didden and Sigafoos[8] concluded that most studies relied on parent report, which is evidently open to bias. 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. Consequently, 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. One concern about the move to direct measures, however, is that people with intellectual disabilities may find such methods hard to tolerate[67, 68], thus either biasing the sample or actively affecting the quality of their sleep. Quality of sampling, however, was less good and 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.

As well as being limited by the scope of the extant literature, this review is limited by the methodological and analytic processes undertaken. 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[9, 13, 15]. 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 der Wouw and colleagues,[14] due to its relatively high prevalence.[69] 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. Furthermore, where IQ tests were reported, we included studies in which the range of IQs was less than 85 (or in some cases not reported), if the mean group IQ was reported as less than 70. This again may have meant a small number of participants may not have met diagnostic criteria for an 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 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 need to be treated with caution. Though again, this limitation would favour the null hypothesis.

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, each of which may require in-depth future research. 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. While choosing from specific syndrome groups can make samples more homogenous and make understanding mechanisms for impairment easier, most local services are aimed at populations of people with intellectual disability of heterogeneous aetiology. It is worth noting that broader prevalence studies have tended to focus on these groups, [9, 13] but this has been combined with the use of indirect measures. Furthermore, syndromes that have previously been associated with poor sleep did not contribute a paper to this analysis, due to lack of studies including typically developing comparison groups.[36,37, 39]

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[14-15] and the analysis in this review suggests there is no evidence to believe difficulties with sleep in people with intellectual disability recede as they get older. Given the changes to sleep over developmental time, it may have been preferable to analyse data from adults and children separately. However, given the lack of studies for adults, this was not possible. Similarly, understanding the relationship between severity of intellectual disability and poor sleep was not possible with the current state of the literature. Though researchers have suggested that sleep quality decreases with severity of disability, [9, 15, 70] only seven studies reported IQ for their participants. More stark was the paucity of reporting of adaptive functioning. That impairments to functioning retain a key place in diagnosing intellectual disabilities,[1,2] but rarely feature in research papers remains a problem that is likely to bias conclusions.

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 conclusions are supportable, to some degree. Significant limitations exist, most notably the proportion of research based on child participants and the lack of studies based on people with intellectual disability of heterogeneous origin. Similarly, it is the authors' 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.

Practice points

1) Clinician's should be aware that problems with sleep quality are more likely in people with Intellectual Disabilities.
2) A range of genetic syndromes have been understood to confer an increased risk shorter sleep duration and poorer sleep quality.
3) Uncertainty over mechanisms for sleep problems in people with intellectual disabilities, may mean individual formulation is often indicated.

Research agenda

1) Further comparison studies of sleep between adults with and without intellectual disabilities in different contexts.
2) Further studies investigating sleep in people with intellectual disability of heterogeneous origin.
3) Measuring adaptive behaviour alongside intellectual functioning when working with populations of people with intellectual disability.

Abbreviations

ASD:	Autism spectrum disorders
CI:	Confidence interval
IQ:	Intelligence quotient
QEM:	Quality effects model
REM:	Random effects model
WMD:	Weighted mean difference

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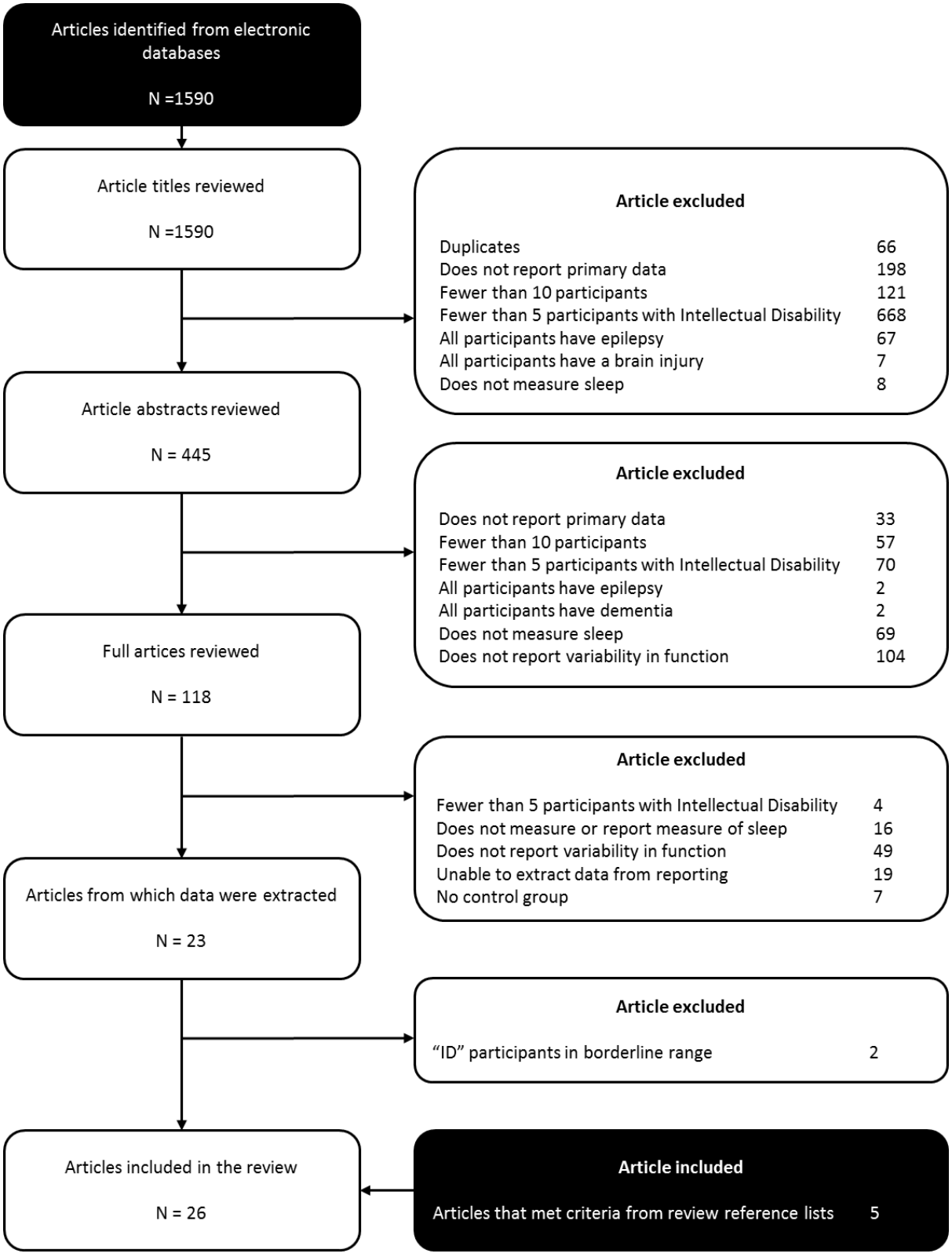

Figure 1. Flow chart for the selection of articles.

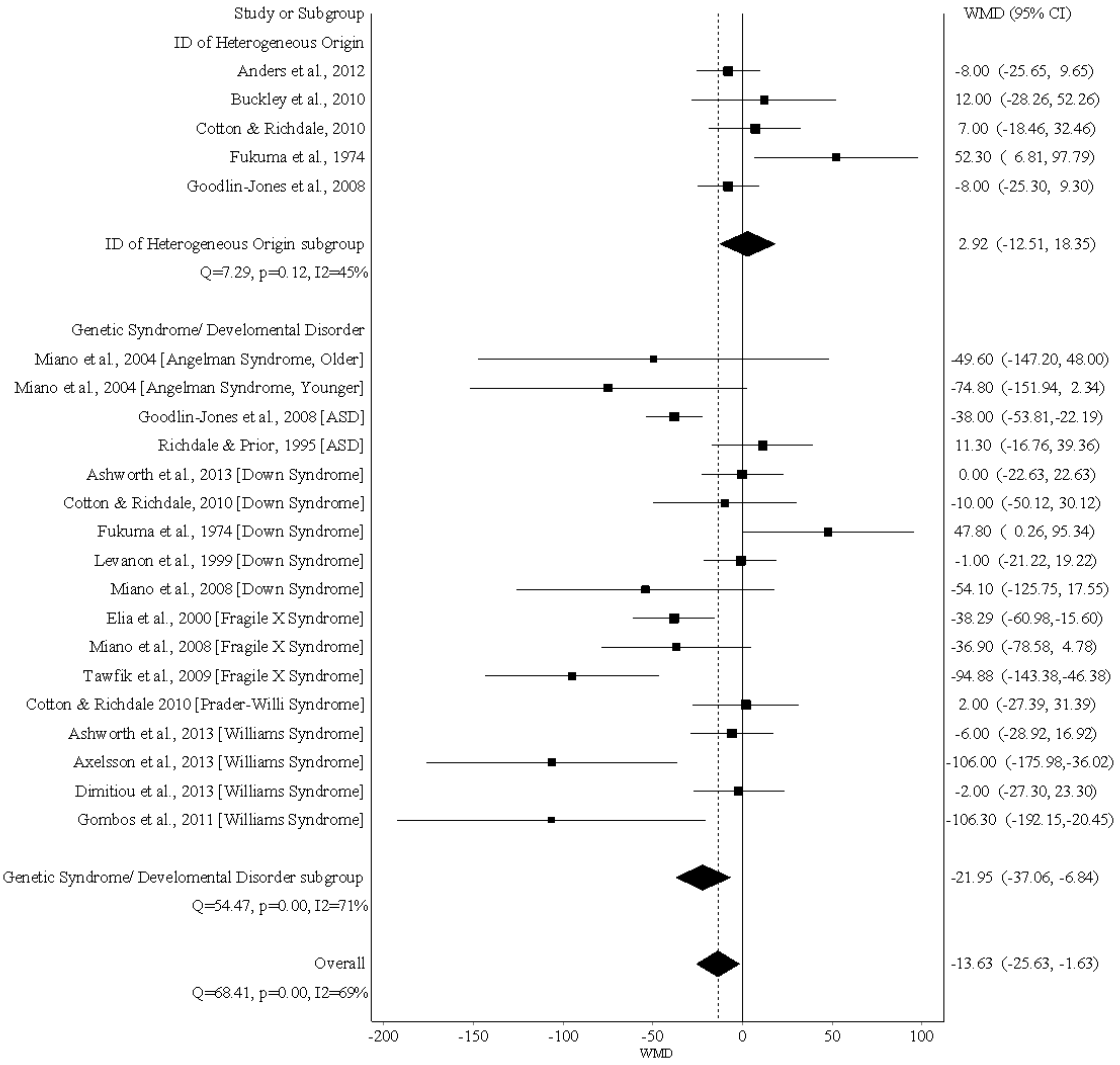

Figure 2. Forest plot of the Random Effects Model for the meta-analysis of sleep time in people with intellectual disabilities compared to controls.

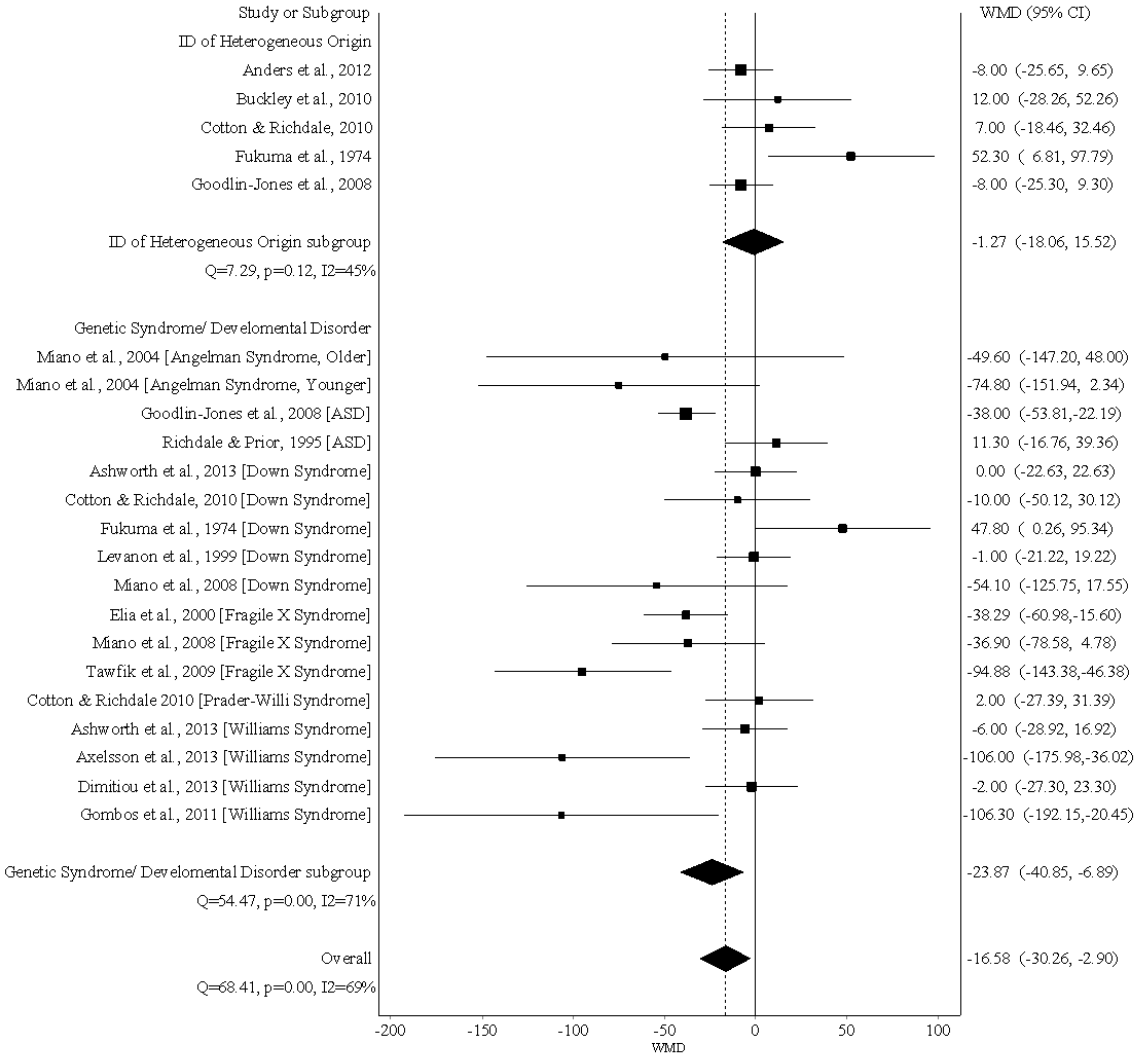

Figure 3. Forest plot of the Quality Effects Model for the meta-analysis of sleep time in people with intellectual disabilities compared to controls.

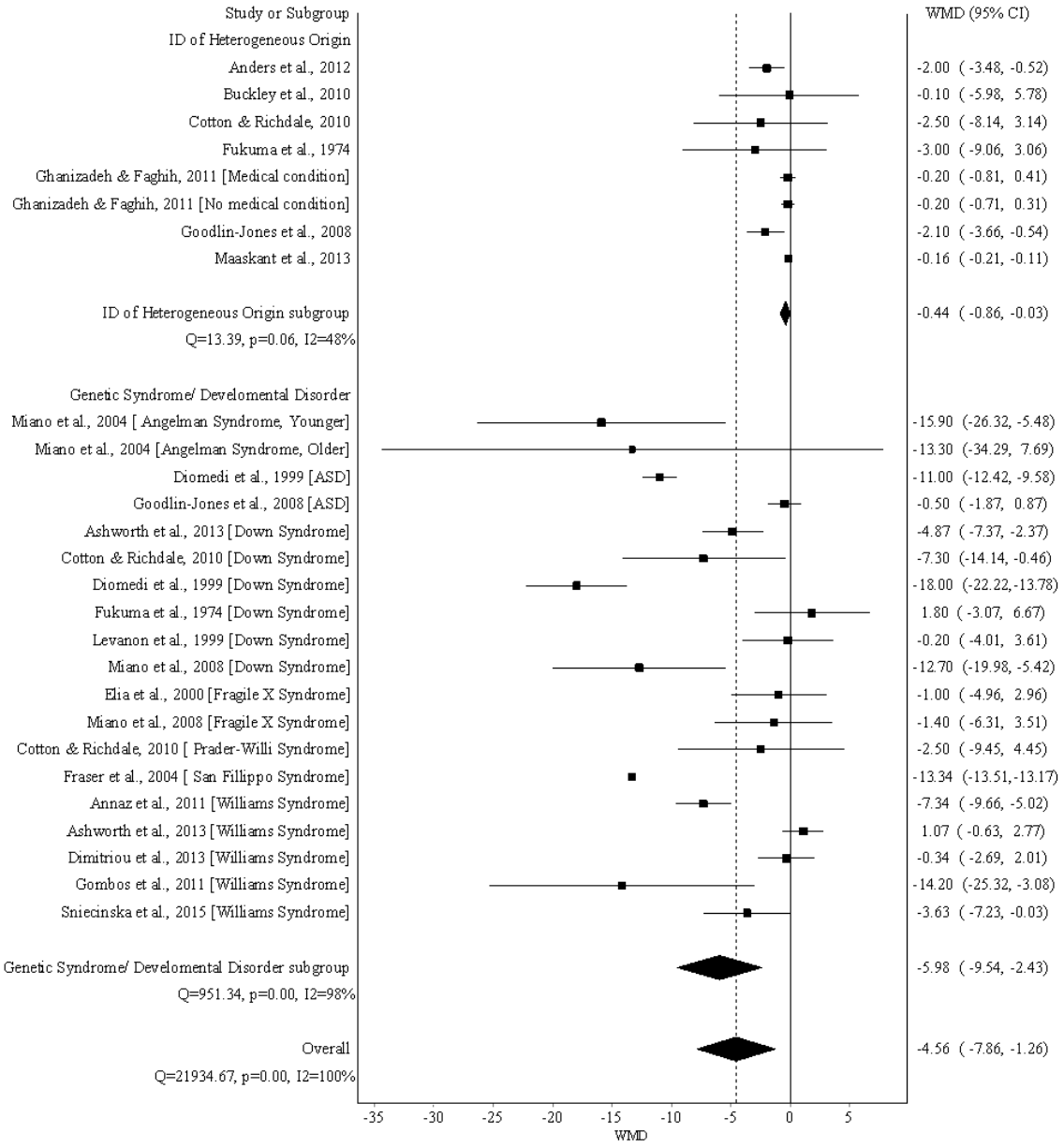

Figure 4. Forest plot of the Random Effects Model for the meta-analysis of sleep quality in people with intellectual disabilities compared to controls.

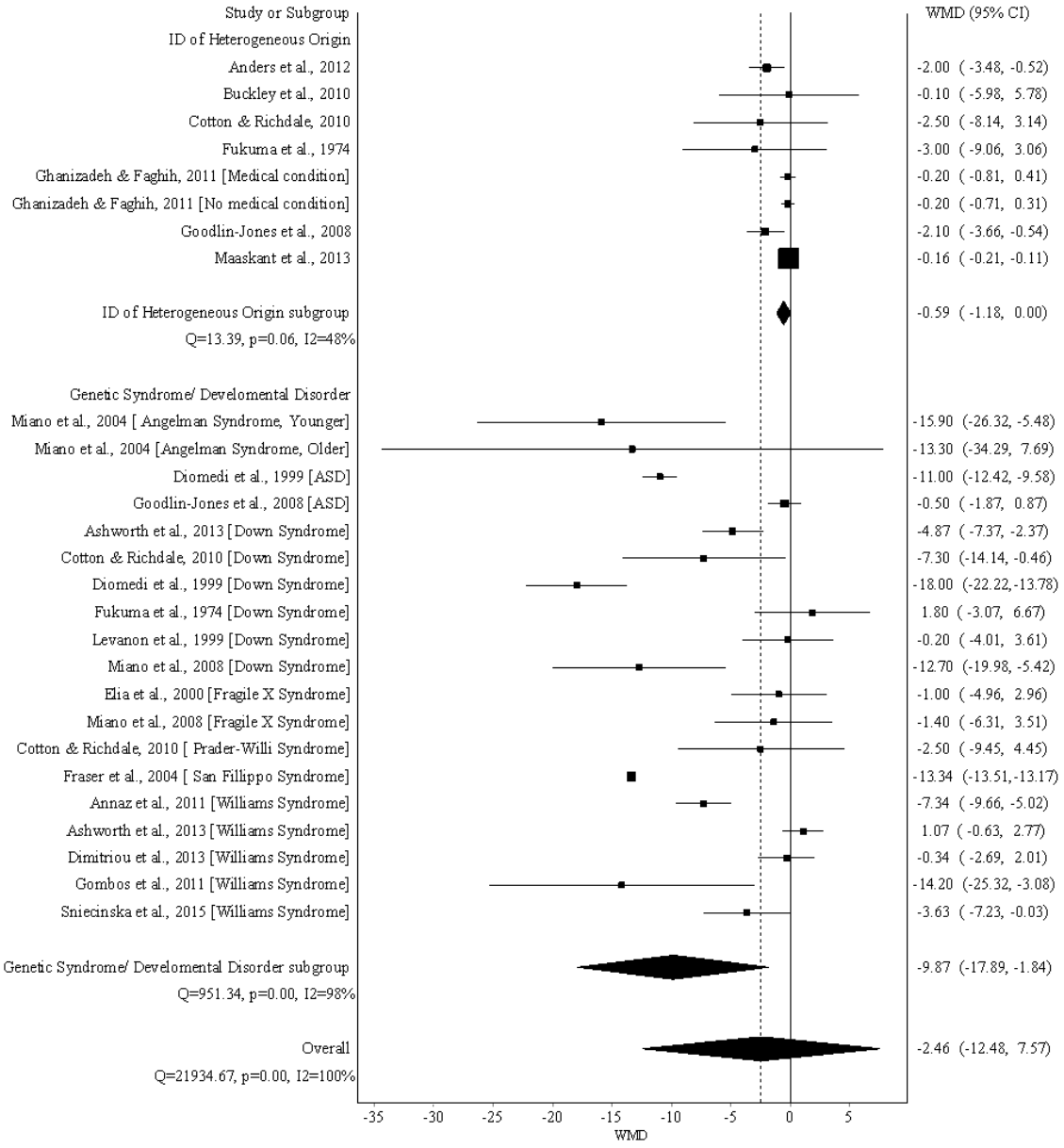

Figure 5. Forest plot of the Quality Effects Model for the meta-analysis of sleep quality in people with intellectual disabilities compared to controls.

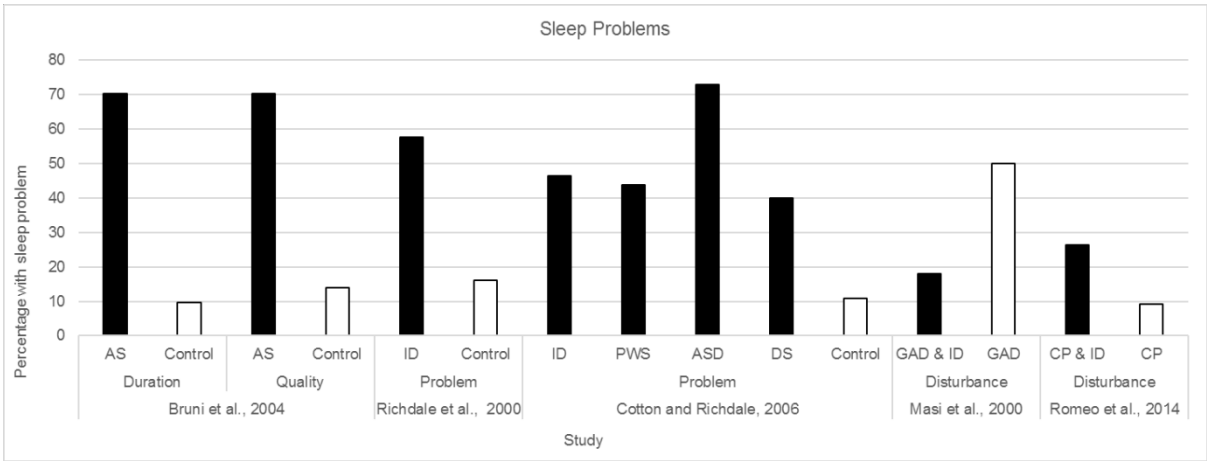

Figure 6. Studies investigating the percentage of participants reported as having a problem with specified aspect of sleep. AS = Angelman Syndrome, ID = Intellectual Disbaility, PWS = Prader-Willi Sydrome, DS = Down Syndrome, ASD = Autism Spectrum Disorder, GAD = Generalized Anxiety Disorder, CP = Cerebral Palsy.

Table. 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
Angelman Syndrome	Mental age 0-2[t1]
Down Syndrome	Approximately 50, with wide variability [t2]
Fragile-X Syndrome	96% have Intellectual Disabilities or Developmental Delay [t3]
Prader-Willi Syndrome	Mean IQ approximately 60 [t4]
Sanfilippo Syndrome	Majority have mental age 0-2, modal group < 3 months [t5]
Williams Syndrome	IQ of approximately 56 (range: 50–70) [t6,t7]

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Table 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) *Coded Red*	Adequate (1) *Coded Orange*	Good (2) *Coded Yellow*	Excellent (3) *Coded Green*
Sample α = .82	**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

Table 3. Results of the meta-analysis of sleep time: REM = Random Effects Model, QEM = Quality Effects Model. * Indicates a significant difference between intellectual disability and control 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%

Table 4. Results of the meta-analysis of sleep quality: REM = Random Effects Model, QEM = Quality Effects Model * Indicates a significant difference between intellectual disability and control 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%