Sleep in people with and without Intellectual Disabilities

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Systematic Review

Sleep in people with and without intellectual disabilities: a systematic review and meta-analysis

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Abstract

Background   Sleep problems are regularly reported in people with intellectual disabilities. Recent years have seen a substantial increase in studies comparing sleep in people with intellectual disabilities to control participants, with an increase in the use of validated, objective measures. Emerging patterns of differences in sleep time and sleep quality warrant pooled investigation.

Methods   A systematic search was conducted across three databases (Ovid Embase, PsycInfo and Medline) and returned all papers comparing sleep in people with intellectual disabilities to a control group, published since the last meta-analysis on the topic. A quality framework was employed to rate the risk of bias across studies. Separate meta-analyses of sleep duration and sleep quality were conducted. Subgrouping compared findings for those studies with participants with genetic syndromes or neurodevelopmental conditions and those with heterogeneous intellectual disability.

Results   Thirteen new papers were identified and combined with those from the previous meta-analysis to provide 34 papers in total. Quality of studies was generally rated highly, though sampling provided risk of bias and adaptive functioning was rarely measured. People with intellectual disability associated with genetic syndromes or neurodevelopmental conditions sleep for shorter time periods (standardised mean difference = .26) and experience worse sleep quality (standardised mean difference = .68) than their peers. People with intellectual disability of heterogeneous origin show no difference in sleep time but have poorer sleep quality. There was some evidence that age moderated these effects.

Conclusions   People with intellectual disability have poorer sleep than those without. Subtle patterns suggest that aetiology of intellectual disability moderates the topography of these difficulties, with further work needed to differentiate common and distinct mechanisms across groups.

Keywords   Genetic syndrome, Intellectual disabilities, Intellectual disability, Learning disability, Meta-analysis, Sleep, Systematic review

Introduction

Intellectual disability (ID) is a neurodevelopmental condition that encompasses deficits to an individual's adaptative and intellectual functioning (World Health Organization 1992; American Psychiatric Association 2013). ID affects approximately 1–3% of the population (Purugganan 2018). Causes of ID are

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heterogeneous, for example, genetic syndromes, perinatal insult and brain injury (Vasudevan & Suri 2017; Ilyas et al. 2020; Lee et al. 2022). Children and adults with ID have long been considered to experience worse sleep than their neurotypical peers (e.g. van de Wouw et al. 2012; Bassell et al. 2015; Harper et al. 2023).

Difficulties with sleep can be defined in a number of ways, including by the presence or absence of a diagnosable sleep disorder (American Academy of Sleep Medicine 2014). Here, we focus on the continuous measurement of night-time sleep duration and sleep quality (Surtees et al. 2018). Such continuous data provide for sensitive comparison across groups: Quantitative measurement allows for participants who may have meaningful sleep difficulties that do not meet criteria for a sleep disorder *and* for differences to be identified between those who do meet that threshold. Consideration of measurable parameters also avoids the inherent difficulties with disorders being identified and defined primarily with respect to neurotypical populations.

People with ID are not a homogeneous group (Patel et al. 2020), so to generalise their sleep as such can be problematic (Didden & Sigafoos 2001). Poor sleep experienced by people of different ID subgroups is likely explained by different aetiologies (Wiggs 2001; Surtees et al. 2018). Disaggregating data according to the origins of a person's ID provides important insights on sleep time and quality. Many genetic syndromes and neurodevelopmental conditions associated with ID predict poor sleep, including, but not limited to Smith–Magenis syndrome (SMS; Smith et al. 2019), Williams syndrome (WS; Axelsson et al. 2013) and autism (Goodlin-Jones et al. 2008). There are a broad range of reasons why different genetic syndromes and developmental conditions associated with ID all confer higher rates of sleep difficulty, with contributions from genetic, biological, neurological, psychological, physiological and medical factors (Bissell et al. 2021). For a fuller consideration of prevalence and why this may be notable across different genetic syndromes, see Agar et al. (2021).

Further, ID of heterogeneous origin is often associated with co-morbid physical health conditions, which vary across people with ID, such as epilepsy (Stefanski et al. 2021) and/or functional deficits (Lee et al. 2022). Each of these factors likely contributes to sleep difficulties observed in people with ID.

Based on a search conducted in 2015, Surtees et al. (2018) report a meta-analysis investigating sleep differences between people with and without ID. An overall difference was found in both sleep duration and a broad parameter of sleep quality – encompassing sleep efficiency measurements, as well as scores from standardised questionnaires. People with ID were shown to sleep for shorter periods and have poorer quality sleep. Data were markedly heterogeneous. Part of this heterogeneity was explained by differences between those with and without an identified genetic syndrome or neurodevelopmental condition. Those with a known associated genetic syndrome or neurodevelopmental condition slept for shorter periods *and* had poorer quality than their peers. Those with ID of heterogeneous origin did not show a difference in their sleep time and showed a very marginal reduction in sleep quality. Notably, these important subgroup differences were concluded on the basis of a small number of studies in people with heterogeneous ID.

The 8 years since the previous meta-analysis search was conducted (Surtees et al. 2018) has seen a massive growth in research comparing sleep time and quality in people with ID to that of typically developing (TD) controls. This provides a substantial improvement in power to test how robust reported differences are. An expansion in recent research on individuals with ID of heterogeneous origin suggests a possible new direction of findings, particularly regarding their sleep duration. Newer studies across multiple age groups report significantly longer sleep for people with ID than age-matched controls (Chan et al. 2019; Bohmer et al. 2020). Conversely, new data on the sleep quality of this subgroup seem to strengthen the original finding that individuals with idiopathic ID experience poorer sleep quality than TD controls (Surtees et al. 2018; Chan et al. 2019). Further studies have also been published including people with ID and an associated genetic syndrome or neurodevelopmental condition, a number of which have suggested that original subgroup conclusions may not be congruous (Gunes et al. 2019; Smith et al. 2019; Trickett et al. 2019).

Rationale

Though sleep differences have been repeatedly identified between people with ID and their peers, the consistency of these findings across different parameters and subgroups have often been poorly defined. A previous meta-analysis confirmed a substantial overall effect of poor sleep in people with ID, but the small number of studies meant that conclusions about subgroup and parameter-level differences were necessarily tentative (Surtees *et al*. 2018). This update review examines recent research alongside the articles included in the original meta-analysis (Surtees *et al*. 2018), which provides substantially increased power to test those differences. We aim to identify and describe studies published since 2015 and combine with data from before this time to create pooled standardised mean difference estimates in sleep duration and sleep quality between people with and without ID. We further aim to conclusively identify whether such differences are moderated by ID subgroup (i.e. ID from genetic syndrome or neurodevelopmental condition, or from heterogeneous origin).

Methods

Study identification procedures identified papers quantifying sleep differences between people with and without ID published since the previous systematic search was conducted, as part of the last meta-analysis on the topic (Surtees *et al*. 2018). Search terms and screening processes were taken directly from this original review, with a restricted set of dates included to avoid duplication. Detailed information on the new studies is identified throughout, including assessment of their quality, alongside summary data across the whole sample.

Search strategy

Following PRISMA guidance for meta-analyses (see Table S3), a systematic literature search was performed using OVID Medline, Embase and PsycInfo (1806–1966 and 1967–2021; for a full search strategy and Boolean operators used, see Supporting Information, S1). Search terms were replicated from Surtees *et al*. (2018). For ID, search terms included intellectual disability, intellectual disturbance, learning disability, mental retardation, mental handicap, mental deficiency, mental disorder, mental incapacity, idiocy, Down syndrome, oligophrenia and variants thereof. For sleep, terms included sleep, insomnia, dyssomnia, parasomnia, somnolence, hyposomnia and variants thereof. Search terms were mentioned in the abstract, keywords or title of the article. Restrictions required that papers were written in English and empirically peer-reviewed. Date restrictions were placed, such that papers were only included if they were published in 2015 or later.

Paper selection

Paper selection for the final review was independently completed by the two first authors before final papers were agreed upon. Titles, abstracts and full-text articles were screened in a systematic process. Articles were excluded at any stage if they met any of the exclusion criteria or did not explicitly report the inclusion of participants with ID/a related disorder or a measure of sleep time/quality. If this was not the case, the article was carried through to the final selection stage where the full text was retrieved, and a final inclusion/exclusion decision was made. Papers were excluded at any stage if they were found to violate inclusion criteria. If articles met all inclusion criteria but otherwise contained an insufficient dataset, the appropriate data were requested from the corresponding author. Two communication attempts via email were made in each instance. Final inclusion decisions were collectively agreed between the authors of this update review.

Exclusion criteria

Exclusion criteria were retained from Surtees *et al*. (2018). Studies were excluded if they (1) did not report primary data, (2) included fewer than 10 participants, (3) included fewer than five participants with an ID, (4) if all participants in the study had epilepsy, (5) if all participants in the study had a brain injury, (6) if the study did not measure sleep or (7) if the study did not report sleep by variability in function (i.e. did not report sleep separately for ID and comparison groups). Criteria for measuring sleep time and/or quality required data from an ID group and a TD comparison group. Papers were also excluded after two correspondence attempts if they failed to report appropriate data for the meta-analysis, such as reporting median and interquartile range

values instead of means and standard deviations. For a more detailed overview of the reasons for using the exclusion criteria in turn, refer to the original meta-analysis (Surtees et al. 2018; see also Surtees 2016).

Data extraction and management

For the final review, data were extracted from the new papers by the two first authors and combined with previously extracted data from the original 21 articles (Surtees et al. 2018). Extracted data included participant demographics such as age, gender and average intelligence quotient (IQ) of the group, where reported. Methodological inclusion/exclusion information was also extracted, such as genetic syndrome classification, exclusion of autistic people without concomitant ID, medication usage and reported sleep interventions. Further methodological information, such as recruitment processes, was noted. The two primary dependent constructs were sleep duration and sleep quality; variables that matched these constructs were extracted. For sleep duration, the number of hours slept (measured objectively or subjectively reported) qualified for extraction. Where studies objectively measured sleep quality, sleep efficiency (the percentage of time in bed that was spent asleep) was extracted as the primary variable. Where this was not the case, the broadest measure of sleep was selected. Objective measures were favoured and extracted over subjective reports in studies that reported multiple measures for either construct. Data from newly identified papers were extracted separately by the two first authors – with any differences discussed with the third author. Data from the original meta-analysis (Surtees et al. 2018) were re-extracted, and no errors noted.

Quality review

A quality framework (see Table 1) was used to weight the contribution of studies of varying quality in the analysis. The framework used was the same as that used in Surtees et al. (2018). In line with best practice (Higgins et al., 2011), this framework reflects the key threats to internal validity for the specific question addressed in this review. This approach reflects prioritising identifying genuine risk of bias in the reviewed studies (Higgins et al., 2011), over-rating the quality of reporting of the study. The benefit of this is

that it allows for effective weighting of conclusions in the light of threats to validity. The potential cost is that it can make it more difficult to compare the quality of methodology and reporting to that in other research domains.

Each participant group, when recruited differently, received independent quality ratings and for each dependent variable where appropriate. This included a consideration of the appropriateness of matching of ID and control groups, including differences in demographic characteristics such as age (see Table 1). The framework was based on three factors thought to reflect the key threats to internal and external validity. Each of the framework measures were equally weighted, though calculated across differing numbers of sub-questions. For a more detailed overview, including justifications for using this measure of study quality rather than a more formal measure, refer to the original meta-analysis (see Surtees et al. 2018).

The quality review was run twice by the two first authors, independent of one another. Results were compared after the second instance, and an excellent level of inter-rater reliability was obtained for the whole scale ($\alpha = .81$), with individual item ratings varying between good (for matching of samples, $\alpha = .55$) and excellent (for reliability/validity of measurement of level of intellectual functioning, $\alpha = 1.0$; measurement of adaptive functioning, $\alpha = 1.0$). A consensus between authors was then reached to ensure all items in the quality review had a score that reached the excellence threshold.

Data analysis

Following Surtees et al. (2018), analysis was completed separately for sleep time and sleep quality. Sleep time measures included any report of total sleep time. Sleep quality included any reported measure of sleep quality. In the case of questionnaires, this included measures of sleep quality or sleep problems/disturbance. In the case sleep quality was reported negatively, a linear transformation was applied. In the case of actigraphy, sleep efficiency was taken as a broad measure of sleep quality.

For each analysis, the generic inverse variance model was employed to estimate the effect. Quantile–quantile plots were created and reviewed to consider normality of fixed and random effects models (REM) distributions. In each case, the REM

Table 1 Quality framework used to assess studies

Item	Poor (0)	Adequate (1)	Good (2)	Excellent (3)
Sample Identification of ID sample	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	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
Matching of ID and control group	• 1 sample recruited with a pre-specified sleep problem • Significant differences in demographic characteristics of groups	• ID group recruited through restricted means, while control group recruited randomly • Different exclusion criteria for two groups • Identification unknown in one or both samples, but no evidence of differences between groups		• Two samples recruited by equivalent means (e.g. random samples from a GP practice, fliers given to the general population, through local schools drawing from the same population, or siblings of ID children being asked to participate)
Measurement of intellectual disability Reliability/validity of measurement of level of intellectual functioning	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	Unspecified	• Clinician judgement • 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 Behaviour Scales

Table 1. *(Continued)*

Item	Poor (0)	Adequate (1)	Good (2)	Excellent (3)
Reliability/validity of sleep measure	Response to a single question	Validated sleep questionnaire; note any form of validation is applicable (e.g. 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
Measurement of sleep				

A total score based on the average across these three 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*.

was ultimately chosen, and the restricted estimator of maximum likelihood employed, to be conservative in the case of deviations from normality. Standardised mean difference was calculated for all studies and reported as Hedges' *g*. Heterogeneity was calculated using Higgins I^2, with returned values over 75% considered as problematic variance (Higgins *et al.* 2003).

Due to the core conjecture of this paper, that groups of participants with identified genetic syndromes or neurodevelopmental conditions may experience different sleep from those with heterogeneous ID, a subgroup analysis was conducted initially to examine this hypothesis. In the event that this analysis produced a significant difference in effect, all subsequent analysis was conducted on the two groups separately.

Contingent upon substantial heterogeneity, a leave-one-out analysis was conducted (separately for subgroups, if indicated by subgroup analysis), with Baujat plots drawn to identify papers that were substantially discrepant *and* influential. In the event that such studies were identified, papers were reviewed against two further criteria. Firstly, whether the paper was identified to present with substantial risk of bias, as determined by the overall score on the quality framework (discrepant and influential papers with a high risk of bias were removed). Secondly, whether the paper differed notably from other papers in broad methodological choices, such as participant or measurement characteristics. When an influential and discrepant paper with relatively low risk of bias was identified, it was only removed if it was a notable outlier in its methodological choices. Subsequent to conclusions of the leave-one-out analysis, quality effects models were calculated to estimate the effect weighting studies by their score on the quality framework (as per Surtees *et al.* 2018).

Funnel plots were used to identify possible influence of publication bias or small sample sizes. In the event that publication bias was identified, this was corrected by imputing additional studies using a trim-and-fill procedure (Duval & Tweedie 2000). To analyse the likelihood that unpublished studies would result in a different conclusion, Rosenthal's 'Failsafe' number was calculated (Rosenthal 1979).

Given that one of the functions of the paper was to serve as an 'update' review, meta-regression of year of publication was included to identify changes in effect

size over time. To examine the impact of participant age, meta-regression of average age of sample by paper was conducted.

Results

Figure 1 reports the results of the search. Five thousand, two hundred and three papers were returned for screening from two separate searches in June 2021 and January 2023. Thirteen new papers were identified across both searches, which were combined with the 21 papers from Surtees *et al.* (2018) to leave 34 papers in total (see Fig. 1). This represented an additional 62% of papers in

comparison to the original meta-analysis (Surtees *et al.* 2018).

Participant characteristics

The thirteen new papers added to the final analysis included 1485 participants across nineteen groups of people with ID (see Table 2 for a full description of the papers, including participant demographics, study methodology and quality ratings). These comprised four groups of people with ID of heterogeneous origin, three with SMS, two with autism (and ID), two with Angelman syndrome, one with Down syndrome, one with Williams syndrome, one with
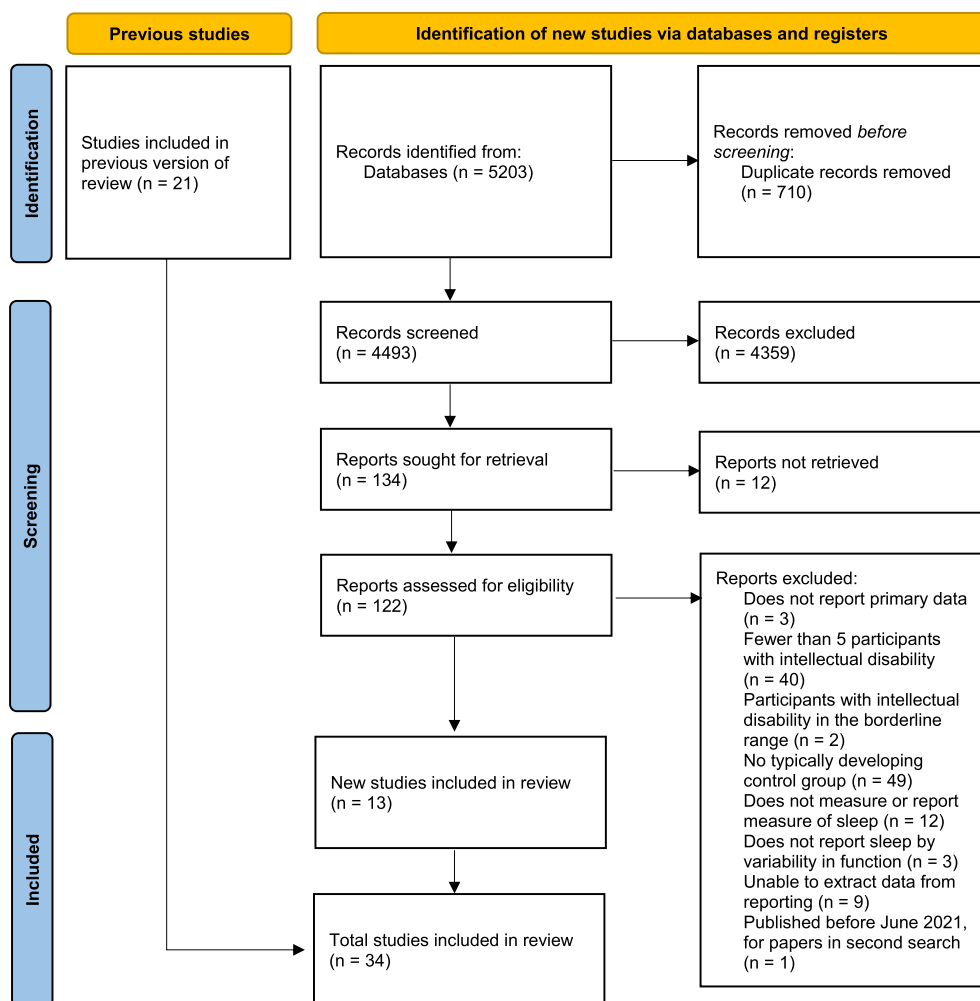
Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analysis flow chart, demonstrating the search and screen process.

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Table 2 Demographics, methodology and quality ratings for all studies

Study	Quality			N	% male	Mean age (range)	Group description
	Sample	ID measures	Sleep measures				
Al-Farsi et al. 2018	3	2	1	81	48	8.9 (3–14)	ID
Anders et al. 2012†	2	3	3	57	74	3.8 (2–5.8)	ID
Annaz et al. 2011†	2	1	1	64	44	8.2 (6.2–12.5)	Williams syndrome
Ashworth et al. 2013†	2	1	3	22	50	9.4 (6.1–12.2)	Down syndrome
	2	1	3	24	50	9.6 (6.1–12.6)	Williams syndrome
Ashworth et al. 2015	2	2	3	22	50	9.42 (6.09–12.23)	Down syndrome
	1	2	3	22	45	9.24 (6.08–12.58)	Williams syndrome
Axelsson et al. 2013†	1	1	1	18	—	2.5 (1.3–4.0)	Williams syndrome
Ballester et al. 2019	2	2	3	41	78	33 (27–39)	Autism and ID
Bohmer et al. 2020	2	3	3	501	52	62.02	ID
Buckley et al. 2010†	2	1	0	37	54	— (2.3–14.8)	Angelman syndrome
	0	0	3	13	54	4.3 (2.7–7.1)	Developmental delay
Chan et al. 2019	2	2	1	375	63	12.59 (6–18)	ID (weekday sleep)
	2	2	1	368	63	12.59 (6–18)	ID (weekend sleep)
	2	2	1	377	63	12.59 (6–18)	ID
	2	1	0	15	53.3	9.0 (3–16)	Down syndrome
	2	1	0	17	76.5	11.6 (3–18)	Prader–Willi syndrome
Cotton & Richdale 2010†	0	1	2	34	74	7.2 (3.5–14)	Autism and ID
	0	1	2	12	58	8.7 (3–13)	Down syndrome
	0	1	2	24	71	7.1 (4–14)	ID
	0	1	2	12	83	9.4 (3–15)	Prader–Willi syndrome
Cotton & Richdale 2010†	1	2	3	10	70	18.2 (12–24)	Autism and ID
	0	2	3	8	75	22.5 (17–31)	Down syndrome
Dimitriou et al. 2014†	2	2	3	14	36	8.8	Williams syndrome
Diomedi et al. 1999†	1	2	3	10	70	18.2 (12–24)	Autism and ID
	0	2	3	8	75	22.5 (17–31)	Down syndrome
Elia et al. 2000†	0	1	3	7	100	9.9 (5.6–16.7)	Fragile X syndrome
Evans et al. 2016	1	3	1	34	62	12.86 (2.8–51.1)	Mowat–Wilson syndrome
Fraser et al. 2005†	2	1	2	141	-	13.5 (0–40)	Sanfilippo syndrome
Fukuma et al. 1974†	1	2	3	10	50	12.3 (7–17)	Down syndrome
	1	2	3	10	50	11.3 (7–17)	ID
Ghanizadeh & Faghih 2011†	2	1	1	58	42	11.1	ID and medical condition
	2	1	1	75	44	11.8	ID with no medical condition
Gombos et al. 2011†	2	1	3	9	33	20.5 (14–28)	Williams syndrome

Table 2. *(Continued)*

Study	Quality			N	% male	Mean age (range)	Group description
	Sample	ID measures	Sleep measures				
Goodlin-Jones et al. 2008†	2	3	3	68	81	3.9 (2.3–5.7)	Autism and ID
	2	3	3	57	74	3.8 (2–6.8)	Developmental delay
Gunes et al. 2019	1	3	–	66	72	8.06 (2–18)	Autism and ID
Kara et al. 2018	2	2	–	43	74	- (4–6)	ID
Levanon et al. 1999†	1	1	3	23	-	4.8 (1.7–8.0)	Down syndrome
Maaskant et al. 2013†	2	2	3	501	50	62 (50–92)	ID
	1	2	0	22	55	16.3 (11–25)	ID and generalised anxiety disorder
Miano et al. 2004†	1	1	3	6	33	12 (9–17)	Angelman syndrome (older)
	1	1	3	9	56	4 (3–5)	Angelman syndrome (younger)
Miano et al. 2008†	1	2	3	9	89	13.8 (8–20)	Down syndrome
Richdale & Prior 1995†	1	2	3	14	100	13.1 (7–25)	Fragile X syndrome
	1	2	2	12	58	9.1 (2.7–19)	Autism
	1	2	2	52	67	7.7 (1.8–19)	ID
	1	2	–	91	—	— (6–16)	Cerebral palsy and ID
Smith et al. 2019	2	1	3	24	54	5.1 (2–9)	Smith–Magenis syndrome (<10 years)
	2	1	3	7	29	13.2 (11–14)	Smith–Magenis syndrome (10–15 years)
Smith-Hicks et al. 2021	3	1	–	47	49	12.7	Phelan–McDermid syndrome
	3	1	–	64	48	8.4	SYNGAP1-related ID
Sniecinska-Cooper et al. 2015†	2	1	3	21	48	7.3 (4.5–11.0)	Williams syndrome
Tawfik et al. 2009†	1	2	3	16	100	10.8 (6–18)	Fragile X syndrome
Trickett et al. 2018	1	1	–	26	62	8.54 (2–15)	Smith–Magenis syndrome
	1	1	–	70	46	8.64 (2–15)	Angelman syndrome
	1	1	–	20	55	7.2 (2–15)	Tuberous sclerosis
Trickett et al. 2019	1	2	3	20	40	9.43 (4–15)	Angelman syndrome
Trickett et al. 2020	1	2	3	20	45	8.7 (4–15)	Smith–Magenis syndrome

Table 2. (Continued)

Study	Measure of ID	Measure of sleep	Sleep time variable	Sleep quality variable	Average IQ (range)	Quality weighting
Al-Farsi et al. 2018	Not stated	Childhood Sleep Habits Questionnaire (Arabic)	—	Total score	—	.61
Anders et al. 2012†	Mullen Scales of Early Learning	Actigraphy	Total sleep time	—	55.0 (49–74)	.89
Annaz et al. 2011†	None reported (syndrome)	Childhood Sleep Habits Questionnaire	—	Total score	—	.44
Ashworth et al. 2013†	None reported (syndrome)	Actigraphy	Total sleep time	Sleep efficiency	—	.67
					—	.67
Ashworth et al. 2015	Raven's Coloured Progressive Matrices	Actigraphy	Actual sleep time	Sleep efficiency	—	.74
					—	.70
Axelsson et al. 2013†	None reported (syndrome)	Questionnaire	Night sleep	—	—	.33
Ballester et al. 2019	Not stated	Ambulatory circadian monitoring	Total sleep time	—	<70	.65
Bohmer et al. 2020	Clinician report	Actigraphy	Total sleep time	—	—	.85
	None reported (syndrome)	Questionnaire	—	—	(80 to <25)	.33
Buckley et al. 2010†	Not stated	PSG	Total sleep time	Sleep efficiency	58.1 (9.6–76.6)	.33
Chan et al. 2019	Not stated	Childhood Sleep Habits Questionnaire & The Hong Kong Children Sleep	Actual sleep duration	Sleep-related QoL	—	.50
	Parent or clinician report	Questionnaire	—	—	—	.50
	Parent or clinician report	Questionnaire	—	—	—	.50
Cotton & Richdale 2010†	Questionnaire	Diary	Total sleep time	Sleep quality	—	.33
Cotton & Richdale 2010†	Questionnaire	Diary	Total sleep time	Sleep quality	—	.33
	Psychological Educational Profile	PSG	—	Sleep efficiency	23.3	.56
					19.2	.56
Dimitriou et al. 2014†	Not reported (syndrome)	Actigraphy	Total sleep time	Sleep efficiency	—	.78
Diomedi et al. 1999†	Psychological Educational Profile	PSG	—	Sleep efficiency	<30	.56
Elia et al. 2000†	Not reported (syndrome)	PSG	Total sleep time	Sleep efficiency	—	.56
	Not reported (Psychological Educational Profile)					.44
Evans et al. 2016	None reported (syndrome)	Sleep Disturbance Scale for Children	—	Total score	—	.59
Fraser et al. 2005†	Not reported (syndrome)	Questionnaire	—	Sleep disturbance	—	.56
Fukuma et al. 1974†	Suzuki–Binet	PSG	Total sleep time	—	31.1	.61
					45.8	.56
Ghanizadeh & Faghih 2011†	Schooling	Childhood Sleep Habits Questionnaire	—	Bed-time resistance and	—	.44
				Sleep-Disturb-ance	—	.44

Table 2. (*Continued*)

Study	Measure of ID	Measure of sleep	Sleep time variable	Sleep quality variable	Average IQ (range)	Quality weighting
Gombos et al. 2011†	None reported (syndrome)	PSG	Total sleep time	Sleep efficiency	—	.61
Goodlin-Jones et al. 2008†	Mullen Scales of Early Learning	Actigraphy	Total sleep time	Sleep efficiency	60.3	.89
					55.2	.89
Gunes et al. 2019	WISC-R	Children's Sleep Habits Questionnaire	—	Total score	—	.52
Kara et al. 2018	None stated	Children's Sleep Habits Questionnaire	—	Total score	—	.46
Levanon et al. 1999†	None reported (syndrome)	PSG	Total sleep time	Sleep efficiency	—	.56
Maaskant et al. 2013†	Care home and patient notes WAIS or WISC-R	Actigraphy Interview		Intra-daily variability —	— 55.7 (50–67)	.67 .28
Miano et al. 2004†	None reported (syndrome)	PSG	Total sleep time	Sleep efficiency	—	.56
					—	.56
Miano et al. 2008†	WAIS-R or WISC-R	PSG	Total sleep time	Sleep efficiency	—	.61
					—	.61
Richdale & Prior 1995†	Leiter/Bailey scales Parent report WISC-III	Diary Questionnaire Questionnaire (SDSC)	Total sleep time	— — —	<55 <70	.50 .28 .44
Smith et al. 2019	None reported (syndrome)	Actigraphy	Total sleep time	Sleep efficiency	—	.70
					—	.70
Smith-Hicks et al. 2021	None reported (syndrome)	Children's Sleep Habits Questionnaire		Total score	—	.52
					—	.52
Sniecinska-Cooper et al. 2015†	None reported (syndrome)	Actigraphy		Sleep efficiency	—	.67
Tawfik et al. 2009†	WISC-III	PSG	Total sleep time	—	61	.67
Trickett et al. 2018	None reported (syndrome)	Modified Version of Simonds and Parraga's Sleep Questionnaire	—	Total score	—	.37
					—	.37
					—	.37
					—	.37
Trickett et al. 2019	None reported (syndrome)	Actigraphy	Total sleep time	Sleep efficiency	—	.70
Trickett et al. 2020	None reported (syndrome)	Actigraphy	Total sleep time	Sleep efficiency	—	.70

ID, intellectual disability; PSG, polysomnography.

Total quality weighting was considered poor if QW < .17, adequate if .17 < QW < .5, good if .5 < QW < .83 and excellent if .83 < QW.

†Studies that were taken from the original meta-analysis on this topic (Surtees et al. (2018).

Mowat–Wilson syndrome, one with Phelan–McDermid syndrome, one with SYNGAP1-related ID and one with tuberous sclerosis complex. Bohmer et al. (2020) reported on a sample of older adults (M = 62.2 years), and Ballester et al. (2019) reported on an adult sample (M = 33 years). All other samples reported an average age of less than 18 years (average ages: 7.2–12.86 years). There was a higher proportion of male than female participants across all samples, 60.23% (average of averages, not weighting for study sizes).

These data were combined with that from the 21 studies previously extracted in the original meta-analysis (see Surtees et al. 2018 for an overview of papers). In total, this meant 34 papers were included in the final analysis, with 2962 participants across 51 groups of people with ID (for full details, see Table 2).

Study quality

As noted in the method, effects from individual participant groups were given unique quality ratings. Using the criteria specified, no study effects were classified as 'excellent' overall, nine as 'good', 13 as 'adequate' and none as poor. Quality awarded for 'measurement of ID' was generally the poorest, with the mean rating being adequate. This reflected appropriately validated IQ measures not often being used and measures of adaptive functioning being even rarer, as most studies relied on presence within a syndrome group or provided no evidence at all for level of functioning. Impaired functioning is typically included in criteria for ID (DSM-V; American Psychiatric Association 2013; ICD-10; World Health Organization 1992) but appears to be often overlooked in research papers, even where it is clinically relevant (Quine 1992). 'Identification of samples' received higher ratings in most studies. In many cases, this was because participants were recruited from multiple or national non-random samples and with unaffected siblings often recruited as controls. Measurement of sleep time or quality was considered to be 'excellent' in 44% of cases, suggesting many studies favoured an objective measure of sleep (polysomnography or actigraphy) compared to years previously where studies predominantly relied on parent report (Didden & Sigafoos 2001).

These data were combined with that from the original meta-analysis (Surtees et al. 2018). Across the 51 populations of people with ID tested, three were rated as excellent overall, 14 as good, 34 as adequate and none as poor.[1]

Meta-analysis

Sleep time

Twenty-two studies reported a measure of sleep time (see Supporting Information for a table of these studies and means for each group; Table S1; see also openly available dataset on Open Science Framework; https://osf.io/znwae/). The papers reviewed contained a total of 32 groups of people with ID, meaning TD comparison groups were replicated on 10 occasions. The REM revealed a marginally non-significant difference (SMD = −.22, 95% CI = −.45–.01), with a trend for people with ID sleeping for shorter periods each night than people without ID. Substantial heterogeneity was identified, I^2 = 90%. A significant subgroup difference was noted between participants with and without an identified genetic syndrome or neurodevelopmental condition (X^2 = 13.91, P < .01). For groups of participants with an identified genetic syndrome or neurodevelopmental condition, a large difference was noted, such that they slept for shorter periods than participants without ID (SMD = −.43, 95% CI = −.67 to −.18). An acceptable level of heterogeneity was observed, I^2 = 69%. For groups of participants with ID of heterogeneous origin, the opposite effect was observed, such that they slept for longer periods than participants without ID (SMD = .32, 95% CI = .02–.62). For these studies, a high level of heterogeneity was observed, I^2 = 91%. Given the significant subgroup difference, data analysis proceeded separately for the two groups.

Genetic syndrome or neurodevelopmental condition. For participants with an identified genetic syndrome or neurodevelopmental condition, acceptable heterogeneity meant leave-one-out analysis was not conducted. The quality effects model evidenced no

[1]Numbers correct prior to the exclusion of Bohmer et al. (2020) and Fraser et al. (2005) for presumed age-related and syndrome-specific differences, respectively.

change in the estimate effect, and only a marginal change in confidence intervals ($SMD = -.43$, 95% $CI = -.69$ to $-.18$). Neither meta-regression on year of publication, $QM = 2.52$, $P = .11$, nor meta-regression on age of participants, $QM = 1.20$, $P = .27$, evidenced an effect. There was no evidence of significant funnel plot asymmetry ($t_{22} = 1.5$, $P = .15$; see Supporting Information, Fig. S1). Rosenthal's Failsafe test suggested a further 294 studies would be needed to reduce the effect to non-significant ($P = .05$).

Intellectual disability of heterogeneous origin. Given unacceptable heterogeneity for effects in participants with ID of heterogeneous origin, a leave-one-out analysis was conducted to understand the relative influence of, and discrepancy from, to the effect of each of the studies and is summarised in a Baujat plot (Supporting Information, Fig. S2). A number of studies were identified that contributed significantly to heterogeneity. One of these studies showed evidence of also contributing substantially to the estimate of the effect. Bohmer *et al.* (2020) was reviewed additionally to consider methodological quality and similarity to approaches employed in other studies. In terms of quality rating, Bohmer *et al.* (2020) was found to be one of the higher rated papers. Notably though, it differed from other studies in testing older adults with ID, while most other studies investigated the sleep of children with ID. Consequently, the paper was removed from subsequent analysis, as unlikely to be representative of the broader group. A revised estimate, omitting Bohmer *et al.* (2020), returned a notably lower standardised mean difference that was marginally non-significant, $SMD = .23$, 95% $CI = -.08$–.53. Heterogeneity was reduced, $I^2 = 87\%$. For participants with ID of heterogeneous origin, meta-regression on year of publication did not evidence an impact of this as a significant moderator, $QM = .54$, $P = .46$. Meta-regression on age of participants did evidence an effect, $QM = 5.60$, $P = .018$, with older participants showing larger effects. There was not evidence of funnel plot asymmetry (see Supporting Information, Fig. S3).

Omnibus analysis following outlier removal. Re-running the omnibus and subgroup analyses after removing Bohmer *et al.* (2020) also revealed a significant

difference ($SMD = -.26$, 95% $CI = -.48$ to $-.03$, $I^2 = 85\%$), with people with ID having shorter sleep time than people without ID (see forest plot; Fig. 2). A significant subgroup difference was noted between participants with and without an identified genetic syndrome or neurodevelopmental condition ($X^2 = 10.66$, $P < .01$), noting that the effect showed shorter sleep times for studies of those with identified genetic syndromes or neurodevelopmental conditions and a trend for longer sleep times in those without genetic syndromes or neurodevelopmental conditions.

Sleep quality

Twenty-nine studies reported a measure of sleep quality (see Supporting Information for a table of these studies and means for each group; Table S2). The papers reviewed contained a total of 43 groups of people with ID, meaning TD comparison groups were replicated on 14 occasions. The REM revealed a significant difference ($SMD = 1.37$, 95% $CI = .29$–2.46), showing people with ID experienced poorer quality sleep than people without ID. Substantial heterogeneity was identified, $I^2 = 94\%$. There was a marginally non-significant trend for a subgroup difference between participants with and without an identified genetic syndrome or neurodevelopmental condition ($X^2 = 3.44$, $P = .06$). Both participants with identified genetic syndromes or neurodevelopmental conditions, ($SMD = 1.76$, 95% $CI = .27$–3.24) and participants with ID of heterogeneous origin ($SMD = .34$, 95% $CI = .17$–.51) experienced poorer quality sleep than their peers without ID. This subgrouping explained a substantial proportion of heterogeneity, with those studies including participants with ID of heterogeneous origin returning acceptable levels, $I^2 = 74\%$, and those studies with participants with identified genetic syndromes returning unacceptable levels of heterogeneity, $I^2 = 95\%$. With this in mind, further analysis focussed on understanding heterogeneity in those studies.

A leave-one-out analysis was conducted to understand the relative influence of, and discrepancy from, to the effect of each of the studies and is summarised in a Baujat plot (Supporting Information, Fig. S4). One study was identified as clearly influential and discrepant. Given this, Fraser
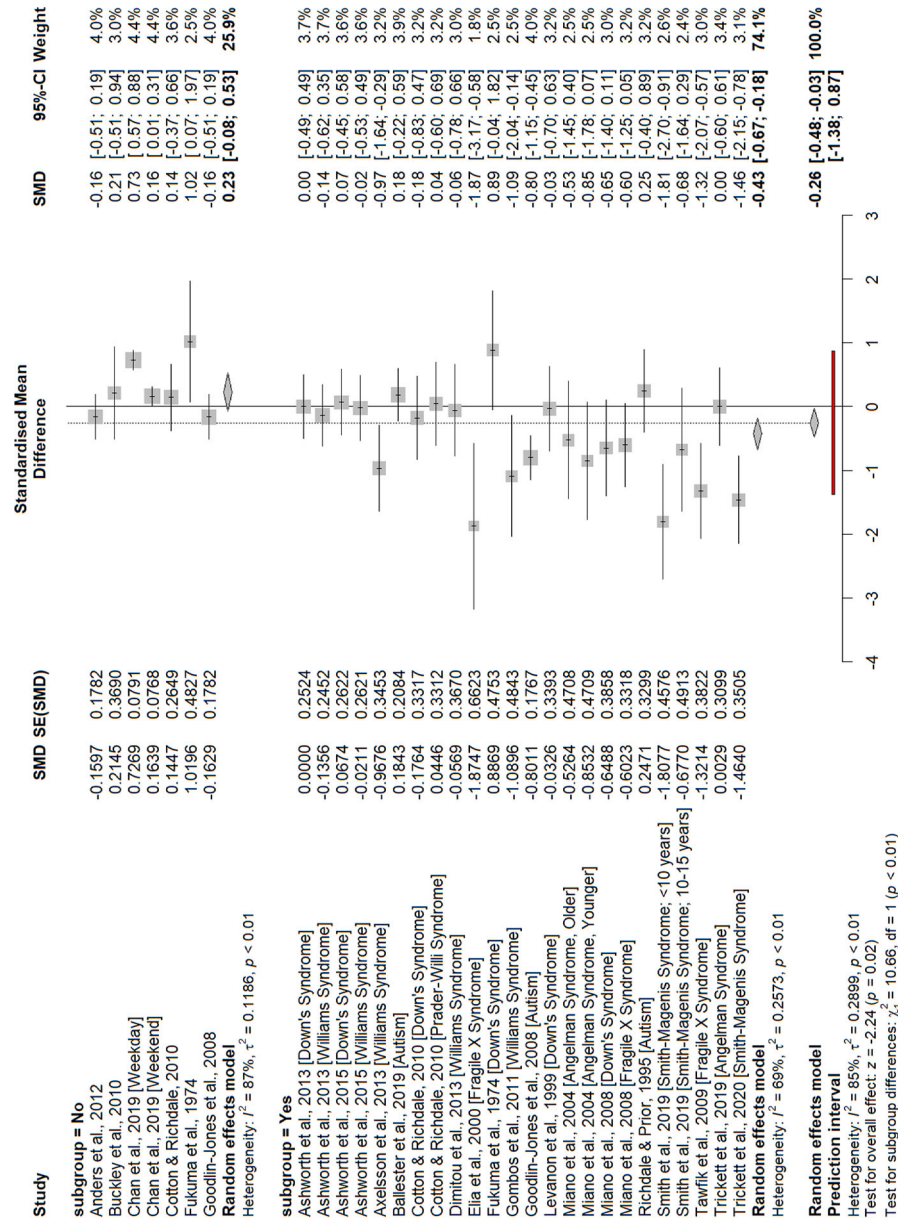
Study	SMD	SE(SMD)		SMD	95%-CI	Weight
subgroup = No						
Anders et al., 2012	-0.1597	0.1782		-0.16	[-0.51; 0.19]	4.0%
Buckley et al., 2010	0.2145	0.3690		0.21	[-0.51; 0.94]	3.0%
Chan et al., 2019 [Weekday]	0.7269	0.0791		0.73	[0.57; 0.88]	4.4%
Chan et al., 2019 [Weekend]	0.1639	0.0768		0.16	[0.01; 0.31]	4.4%
Cotton & Richdale, 2010	0.1447	0.2649		0.14	[-0.37; 0.66]	3.6%
Fukuma et al., 1974	1.0196	0.4827		1.02	[0.07; 1.97]	2.5%
Goodlin-Jones et al., 2008	-0.1629	0.1782		-0.16	[-0.51; 0.19]	4.0%
Random effects model				**0.23**	**[-0.08; 0.53]**	**25.9%**
Heterogeneity: $I^2 = 87\%$, $\tau^2 = 0.1186$, $p < 0.01$						
subgroup = Yes						
Ashworth et al., 2013 [Down's Syndrome]	0.0000	0.2524		0.00	[-0.49; 0.49]	3.7%
Ashworth et al., 2013 [Williams Syndrome]	-0.1356	0.2452		-0.14	[-0.62; 0.35]	3.7%
Ashworth et al., 2015 [Down's Syndrome]	0.0674	0.2622		0.07	[-0.45; 0.58]	3.6%
Ashworth et al., 2015 [Williams Syndrome]	-0.0211	0.2621		-0.02	[-0.53; 0.49]	3.6%
Axelsson et al., 2013 [Williams Syndrome]	-0.9676	0.3453		-0.97	[-1.64; -0.29]	3.2%
Ballester et al., 2019 [Autism]	0.1843	0.2084		0.18	[-0.22; 0.59]	3.9%
Cotton & Richdale, 2010 [Down's Syndrome]	-0.1764	0.3317		-0.18	[-0.83; 0.47]	3.2%
Cotton & Richdale, 2010 [Prader-Willi Syndrome]	0.0446	0.3312		0.04	[-0.60; 0.69]	3.2%
Dimitou et al., 2013 [Williams Syndrome]	-0.0569	0.3670		-0.06	[-0.78; 0.66]	3.0%
Elia et al., 2000 [Fragile X Syndrome]	-1.8747	0.6623		-1.87	[-3.17; -0.58]	1.8%
Fukuma et al., 1974 [Down's Syndrome]	0.8869	0.4753		0.89	[-0.04; 1.82]	2.5%
Gombos et al., 2011 [Williams Syndrome]	-1.0896	0.4843		-1.09	[-2.04; -0.14]	2.5%
Goodlin-Jones et al., 2008 [Autism]	-0.8011	0.1767		-0.80	[-1.15; -0.45]	4.0%
Levanon et al., 1999 [Down's Syndrome]	-0.0326	0.3393		-0.03	[-0.70; 0.63]	3.2%
Miano et al., 2004 [Angelman Syndrome, Older]	-0.5264	0.4708		-0.53	[-1.45; 0.40]	2.5%
Miano et al., 2004 [Angelman Syndrome, Younger]	-0.8532	0.4709		-0.85	[-1.78; 0.07]	2.5%
Miano et al., 2008 [Down's Syndrome]	-0.6488	0.3858		-0.65	[-1.40; 0.11]	3.0%
Miano et al., 2008 [Fragile X Syndrome]	-0.6023	0.3318		-0.60	[-1.25; 0.05]	3.2%
Richdale & Prior, 1995 [Autism]	0.2471	0.3299		0.25	[-0.40; 0.89]	3.2%
Smith et al., 2019 [Smith-Magenis Syndrome; <10 years]	-1.8077	0.4576		-1.81	[-2.70; -0.91]	2.6%
Smith et al., 2019 [Smith-Magenis Syndrome; 10-15 years]	-0.6770	0.4913		-0.68	[-1.64; 0.29]	2.4%
Tawfik et al., 2009 [Fragile X Syndrome]	-1.3214	0.3822		-1.32	[-2.07; -0.57]	3.0%
Trickett et al., 2019 [Angelman Syndrome]	0.0029	0.3099		0.00	[-0.60; 0.61]	3.4%
Trickett et al., 2020 [Smith-Magenis Syndrome]	-1.4640	0.3505		-1.46	[-2.15; -0.78]	3.1%
Random effects model				**-0.43**	**[-0.67; -0.18]**	**74.1%**
Heterogeneity: $I^2 = 69\%$, $\tau^2 = 0.2573$, $p < 0.01$						
Random effects model				**-0.26**	**[-0.48; -0.03]**	**100.0%**
Prediction interval					**[-1.38; 0.87]**	

Standardised Mean Difference

Heterogeneity: $I^2 = 85\%$, $\tau^2 = 0.2899$, $p < 0.01$
Test for overall effect: $z = -2.24$ ($p = 0.02$)
Test for subgroup differences: $\chi_1^2 = 10.66$, df = 1 ($p < 0.01$)

Figure 2. Forest plot showing the meta-analysis of sleep time, subgrouped by participant group. Yes, participants with intellectual disability associated with a genetic syndrome or neurodevelopmental condition; No, participants with intellectual disability of heterogeneous origin. Negative values reflect shorter sleep times for people with intellectual disability.
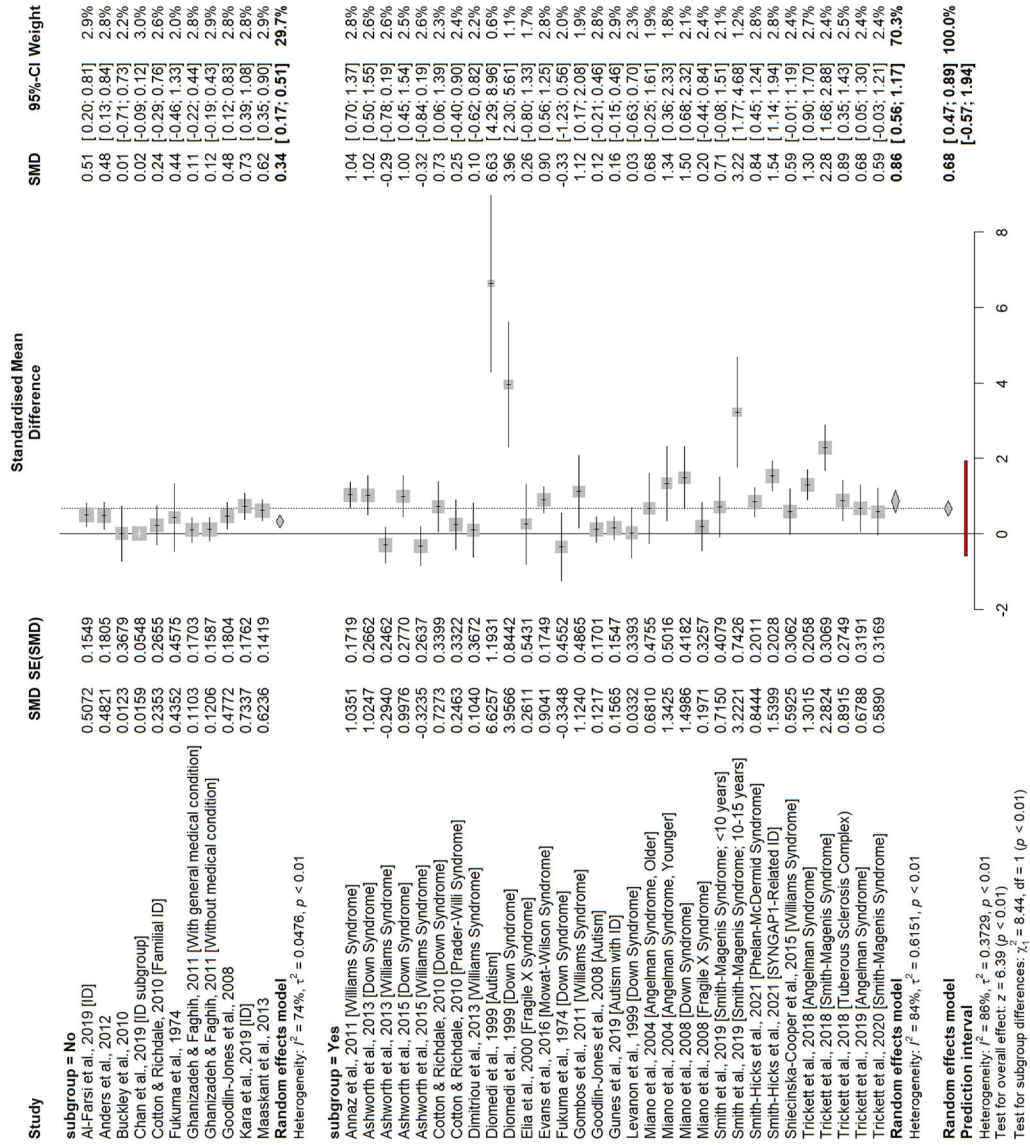
Figure 3. Forest plot showing the meta-analysis of sleep quality, subgrouped by participant group. Yes, participants with intellectual disability associated with a genetic syndrome or neurodevelopmental condition; No, participants with intellectual disability of heterogeneous origin. Negative values reflect poorer sleep quality for people with intellectual disability.

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et al. (2005) was reviewed additionally to consider methodological quality and similarity to approaches employed in other studies. In terms of quality rating, Fraser *et al.* (2005) was found to be within the moderate range of studies included in the review, with no items rated as poor. Notably though, it was the only study to consider participants with Sanfilippo syndrome and used a novel questionnaire. With this in mind, the paper was removed from subsequent analysis, as unlikely to be representative of the broader group. A revised estimate, omitting Fraser *et al.* (2005), revealed a significant difference (SMD = .68, 95% CI = .47–.89), showing people with ID experienced poorer quality sleep than people without ID. Substantial, but notably lower, heterogeneity was identified, I^2 = 86%. There was a significant subgroup difference between participants with and without an identified genetic syndrome or neurodevelopmental condition (X^2 = 8.44, $P <$ .01; Fig. 3). Both participants with identified genetic syndromes or neurodevelopmental conditions, (SMD = .86, 95% CI = .56–1.17) and participants with ID of heterogeneous origin (SMD = .34, 95% CI = .17–.51) experienced poorer quality sleep than their peers without ID. This subgrouping explained a substantial proportion of heterogeneity, with those studies with participants with ID of heterogeneous origin returning acceptable levels, I^2 = 74%, and those studies with participants with identified genetic syndromes or neurodevelopmental condition remaining high, I^2 = 84%. Given significant subgroup differences, investigation of a quality effects model, meta-regression and publication bias was conducted independently for groups of participants with ID of heterogeneous origin.

Genetic syndrome or neurodevelopmental condition. For participants with an identified genetic syndrome or neurodevelopmental condition, the quality effects model suggested a small reduction (5.8%) in the estimated effect size, SMD = .81, 95% CI = .50–1.13. Meta-regression on year of publication did not evidence an impact of this as a significant moderator, QM = .24, P = .63. Meta-regression on age of participants did evidence an effect, QM = 8.97, $P <$ .01, with a larger effect for samples with older participants. Funnel plot asymmetry was consistent with evidence of publication and/or small sample biases – studies with larger standard errors were more likely to return higher SMDs (see Fig. 4). A trim-and-fill process was conducted to correct for potential publication bias but did not impute any missing studies. Rosenthal's Failsafe test suggested 2494 further null results would be needed to produce a non-significant effect (P = .05).

Intellectual disability of heterogeneous origin. For participants with ID of heterogeneous origin, the quality effects model returned an increase (11.7%) in the estimate of the effect (SMD = .38, $95\%CI$ = .20–.56). Neither meta-regression on year of publication, $QM <$ .01, P = .99, nor meta-regression on age of participants, QM = 1.39, p = .24, evidenced an impact of significant moderation of the effect. There was not evidence of funnel plot asymmetry (Supporting Information, Fig. S5). Rosenthal's Failsafe test suggested 148 null studies would be required for the effect to revert to being non-significant (P = .05).

Discussion

This systematic review and meta-analysis updates a previous pooled estimate of the difference in sleep time and sleep quality between people with and without ID. The last 8 years has seen a substantial increase in the rate of research in this area, with 47% more studies providing eligible estimates of sleep duration and 61% more studies now providing eligible estimates of sleep quality. As well as providing an increasingly accurate pooled mean difference estimate, this has allowed for an appropriately powered comparison between the two broad groups encompassed in research in this area: people with ID associated with identified genetic syndromes or neurodevelopmental conditions and those with ID of heterogeneous origin.

Sleep time

For sleep time, our final omnibus model returned a marginally significant difference between people with and without ID, such that those with ID experienced shorter sleep times each night than their peers. A crucial subgroup difference was noted. For participants with an identified genetic syndrome or neurodevelopmental condition, this effect was a robust small–medium sized effect. For participants
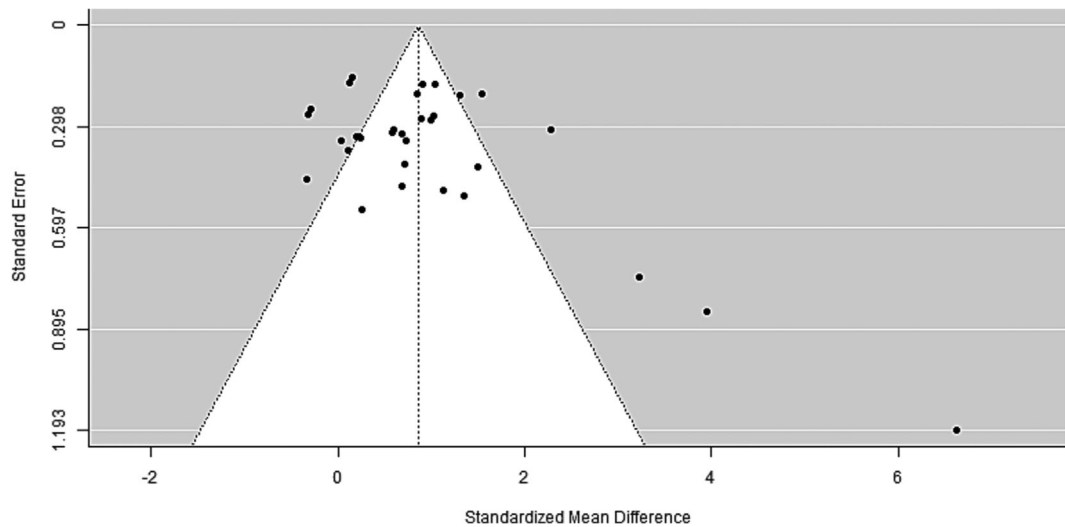
Figure 4. Funnel plot for meta-analysis of sleep quality in studies with people with genetic syndromes or neurodevelopmental conditions.

with ID of heterogeneous origin, there was no evidence for difference from TD control groups.

For participants with ID of heterogeneous origin, effects were larger in older participants. Related to this, Bohmer et al. (2020) was excluded from analysis after being reviewed for unusual discrepancies and contributing substantially to the estimate. This study examined sleep in older adults with ID and found a large effect for longer sleep times. This study was rated as excellent in quality overall and had a large sample size. More data are needed, but a reasonable hypothesis from the existing data is that young children with ID of heterogeneous origin may experience shorter sleep times, like their peers with identified syndromes or developmental conditions. Through to adulthood, however, this effect may reverse. One hypothesis for why this happens may relate to the nature of progressive physical health conditions (Prasher & Janicki 2019). A second may relate to the increasing move towards extra-familial supported accommodation (Cocks et al. 2016). A final possibility could relate to sampling biases; studies in children likely rely on parental interest for participation, whereas studies in adults or older adults more regularly sample across institutions. It is not clear, however, why these factors are not equally impacting those with identified genetic syndromes or neurodevelopmental conditions. More research is needed on age-related changes to sleep in people with ID more broadly, with

a startling number of studies choosing to focus on children, when problems clearly persist in adulthood.

Sleep quality

For sleep quality, evidence was clearer for poorer sleep in those with ID. Again, there was evidence of a significant subgroup effect, such that those with identified genetic syndromes or neurodevelopmental conditions slept more poorly than those with ID of heterogeneous origin. For those with genetic syndromes or neurodevelopmental conditions, a large effect was observed. This effect was moderated by age, such that older participants showed more difference from their TD peers. Fraser et al. (2005) was removed as an outlier, for contributing substantially to heterogeneity and the overall effect. Fraser et al. (2005) reported particularly low sleep quality in a group of children with Sanfilippo syndrome in comparison to controls. Though supporting data exist on sleep problems in this group (Ruijter et al. 2008; Mahon et al. 2014), these are not necessarily more pronounced than in some other genetic syndromes (Agar et al. 2021). More data are needed to understand if this study represents a statistical anomaly, whether Sanfilippo syndrome confers an unusually high risk of sleep disturbance in comparison to other syndromes or whether the questionnaire used is unusually sensitive to sleep

difference. For those with ID of heterogeneous origin, a small, but still significant, effect was calculated.

Understanding subgroup differences

In sum, the growth in research over the past 8 years has allowed stable estimates to be calculated, with continued support for ID being associated with poor sleep. Those children and adults with identified genetic syndromes or neurodevelopmental conditions, in addition to ID, sleep for shorter intervals per night *and* experience poorer quality sleep than their TD peers. This worse quality sleep seems to deteriorate further with age. Those children and adults with ID of heterogeneous origin also experience poorer quality sleep, but not shorter night sleep intervals, and they may even sleep for longer periods than their peers into adulthood and particularly as older adults. A plethora of reasons have been suggested as to why people with ID may sleep more poorly than their peers (Bissell *et al.* 2021). It is likely the case that no single factor is responsible, but rather a combination of biopsychosocial risk factors and perpetuators (Bissell *et al.* 2021).

We treat the evidence for difference between groups of people with associated syndromes or developmental disorders and those with ID of heterogeneous origin cautiously. By definition, both groups remain heterogeneous – data remain too scant to allow meta-analytic comparison between syndromes or to model the impact of developmental risk factors. Specific genetic syndromes and neurodevelopmental conditions have been associated with specific predictors for sleep problems. For example, the well-identified differences in circadian rhythms in people with SMS (De Leersnyder *et al.* 2003; Williams *et al.* 2012) and also separable biopsychosocial models, such as suggested in autism (Richdale & Schreck 2009). Given such differing mechanisms, it is perhaps not surprising that within the context of a large group-level difference, heterogeneity in these studies remained high. Further, it is likely that research in the area may follow those groups who present with the most substantial sleep problems. For people with ID of heterogeneous origin, the substantial difference in sleep quality across studies in particular is notable. Given the likely vast individual differences in cause and behavioural

phenotype in this group (Büttner & Hasselhorn 2011), that a clear effect persists remains notable.

Implications for research and practice of the identified subgroup differences should be treated cautiously. On the one hand, it is clear that identified syndromes and neurodevelopmental conditions are consistently found to have the worst sleep – therefore merit a particularly strong focus from research and clinical services. On the other, those with ID of heterogeneous origin likely represent the largest population but have been studied less frequently. It is clear that more research and support is needed on the sleep of people with ID more broadly.

Study quality and limitations

Using a systematic quality framework, the studies included generally performed well, with most being rated as good or adequate overall. This reflects an increased use of objective measures of sleep in people with ID – in particular a growth in actigraphy to measure habitual sleep patterns. Similarly, intellectual functioning tended to be measured through standardised IQ tests or reflected association with a syndrome with a well-described phenotype. Measures of adaptive functioning tend to be rarer, in spite of their equal role in ID diagnostic criteria.

The meta-analyses showed people with ID of heterogeneous origin to continue to be under-represented in research. Similarly, a high proportion of research has focussed on children. This is particularly notable, as, for sleep quality at least, there is some evidence that differences from TD peers increase with age. Additionally, most of the research in this area continues to be conducted in Western Europe and North America. Though biological determinants of poor sleep may be translatable across cultures, broader social differences mean gaining estimates from the developing world is important.

For the analysis itself, single studies provided multiple subgroups, which may have reduced confidence intervals marginally, though like Surtees *et al.* (2018), there was little evidence that this would have altered conclusions. Unexplained heterogeneity remained high for the analysis of sleep duration in those with ID of heterogeneous origin and the analysis of sleep quality in those with genetic syndromes or neurodevelopmental conditions. That much of this heterogeneity remained unexplained

likely reflects the multifactorial nature of study differences; participant groups were recruited through differing means, at different ages, using different methodologies.

Conclusion

Recent years have seen a substantial increase in research on the sleep of people with ID. Over one-third of papers that have compared sleep time or sleep quality between people with and without ID have been published since the last review eight years ago. Meta-analytic models clearly show that people with ID sleep for shorter periods and less well than their TD peers. This is most significant for those with associated genetic syndromes or neurodevelopmental conditions. Understanding the causes and consequences of this should remain a priority for researchers and clinical services alike.

Conflict of interest

We have no conflict of interest to disclose.

Data availability statement

The data that support the findings of this study are openly available in Open Science Framework at https://osf.io/znwae/.

References

Agar G., Brown C., Sutherland D., Coulborn S., Oliver C. & Richards C. (2021) Sleep disorders in rare genetic syndromes: a meta-analysis of prevalence and profile. *Molecular Autism* **12**, 18.

American Academy of Sleep Medicine (2014) International classification of sleep disorders—third edition (ICSD-3). *AASM Resource Library* **281**, 2313.

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**, 1572–80.

Ashworth A., Hill C. M., Karmiloff-Smith A. & Dimitriou D. (2015) The importance of sleep: attentional problems in school-aged children with Down syndrome and Williams syndrome. *Behavioural Sleep Medicine* **13**, 455–71.

Al-Farsi O., Al-Farsi Y., Al-Sharbati M. & Al-Adawi S. (2018) Sleep habits and sleep disorders among children with autism spectrum disorders, intellectual disabilities and typically developing children in Oman: a case-control study. *Early Child Development and Care* **189**, 1–11.

American Psychiatric Association (2013) *Diagnostic and Statistical manual of mental disorders*, 5th edn. American Psychiatric Association, Washington, DC.

Anders Y., Rossbach H., Weinert S., Ebert S., Kuger S., Lehrol S. et al. (2012) Home and preschool learning environments and their relations to the development of early numeracy skills. *Early Childhood Research Quarterly* **27**, 231–44.

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**, 164–9.

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**, 3988–96.

Ballester P., Martínez M. J., Javaloyes A., Inda M. D., Fernández N., Gázquez P. et al. (2019) Sleep problems in adults with autism spectrum disorder and intellectual disability. *Autism Research: Official Journal of the International Society for Autism Research* **12**, 66–79.

Bassell J. L., Phan H., Leu R., Kronk R. & Visootsak J. (2015) Sleep profiles in children with Down syndrome. *American Journal of Medical Genetics. Part A* **167A**, 1830–5.

Bissell S., Liew A., Richards C. & Surtees A. (2021) Sleep problems and developmental delay. In: *Pediatric Sleep Medicine: Mechanisms and Comprehensive Guide to Clinical Evaluation and Management* (eds D. Gozal & L. Kheirandish-Gozal), pp. 667–80. Springer International Publishing, Willowick, OH.

Bohmer M. N., Oppewal A., Bindels P. J. E., Tiemeier H., van Someren E. J. W. & Festen D. A. M. (2020) Comparison of sleep-wake rhythms in elderly persons with intellectual disabilities and the general population. *Sleep Medicine* **76**, 148–54.

Buckley A. W., Rodriguez A. J., Jennison K., Buckley J., Thurm A., Sato S. et al. (2010) Rapid eye movement sleep percentage in children with autism compared with children with developmental delay and typical development. *Archives of Paediatrics and Adolescent Medicine* **164**, 1032–7.

Büttner G. & Hasselhorn M. (2011) Learning disabilities: debates on definitions, causes, subtypes, and responses. *International Journal of Disability, Development and Education* **58**, 75–87.

Chan K. L., Lo C. K. M., Ho F. K. & Ip P. (2019) Disability-specific associations with child health and functioning. *International Journal of Environmental Research and Public Health* **16**, 1024–33.

Cocks E., Thoresen S. H., O'Brien P., McVilly K., Thomson A., Gadow F. et al. (2016) Examples of individual supported living for adults with intellectual disability. *Journal of Intellectual Disabilities* **20**, 100–8.

Cotton S. & Richdale A. (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**, 490–500.

De Leersnyder H., de Blois M. C., Bresson J. L., Sidi D., Claustrat B. & Munnich A. (2003) Inversion du rythme circadien de la mélatonine dans le syndrome de Smith-Magenis [Inversion of the circadian melatonin rhythm in Smith-Magenis syndrome]. *Revue Neurologique* **159**, 6S21–6.

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**, 255–72.

Dimitriou D., Karmiloff-Smith A., Ashworth A. & Hill C. (2014) Impaired sleep-related learning in children with Williams syndrome. *Pediatrics Research International Journal* 662275, 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**, 548–53.

Duval S. & Tweedie R. (2000) Trim and fill: a simple funnel-plot-based method of testing and adjusting for publication bias in meta-analysis. *Biometrics* **56**, 455–63.

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

Evans E., Mowat D., Wilson M. & Einfeld S. (2016) Sleep disturbance in Mowat Wilson syndrome. *American Journal of Medical Genetics. Part A* **170**, 654–60.

Fraser J., Gason A. A., Wraith J. E. & Delatycki M. B. (2005) Sleep disturbance in Sanfilippo syndrome: a parental questionnaire study. *Archives of Disease in Childhood* **90**, 1239–42.

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. *Folia Psychiatrica et Neurologica Japonica* **28**, 333–45.

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

Gombos F., Bódizs R. & Kovács I. (2011) Atypical sleep architecture and altered EEG spectra in Williams syndrome. *Journal of Intellectual Disability Research* **55**, 255–62.

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 and Adolescent Psychiatry* **47**, 930–8.

Gunes S., Ekinci O., Feyzioglu A., Ekinci N. & Kalinli M. (2019) Sleep problems in children with autism spectrum disorder: clinical correlates and the impact of attention deficit hyperactivity disorder. *Neuropsychiatric Disease and Treatment* **15**, 763–71.

Harper L., McAnelly S., Walshe I., Ooms A. & Tuffrey-Wijne I. M. (2023) Behavioural sleep problems in children and adults with intellectual disabilities: an integrative literature review. *Journal of Applied Research in Intellectual Disabilities* **36**, 916–28.

Higgins J. P. T., Altman D. G., Gotzsche P. C., Juni P., Moher D., Oxman A. D. et al. (2011) The cochrane collaboration's tool for assessing risk of bias in randomised trials. *BMJ* **343**, d5928.

Higgins J. P. T., Thompson S. G., Deeks J. J. & Altman D. G. (2003) Measuring inconsistency in meta-analyses. *British Medical Journal (Clinical Research Ed.)* **327**, 557–60.

Ilyas M., Mir A., Efthymiou S. & Houlden H. (2020) The genetics of intellectual disability: advancing technology and gene editing [version 1; peer review, 2 approved]. *F1000Research* **9**, 22.

Kara T., Alpgan Ö., Yılmaz S. & Akaltun I. (2018) Sleep habits as an indicator of social competence and behaviour in pre-schoolers in the context of neurodevelopmental disorders. *Psychiatry and Clinical Psychopharmacology* **29**, 68–75.

Lee K., Cascella M. & Marwaha R. (2022) Intellectual disability. In: *StatPearls*. StatPearls Publishing, Treasure Island, FL.

Levanon A., Tarasiuk A. & Tal A. (1999) Sleep characteristics in children with Down syndrome. *The Journal of Paediatrics* **134**, 755–60.

Maaskant M., van de Wouw E., van Wijck R., Evenhuis H. M. & Echteld M. A. (2013) Circadian sleep-wake rhythm of older adults with intellectual disabilities. *Research in Developmental Disabilities* **34**, 1144–51.

Mahon L. V., Lomax M., Grant S., Cross E., Hare D. J., Wraith J. E. et al. (2014) Assessment of sleep in children with mucopolysaccharidosis type III. *PLoS ONE* **9**, e84128.

Miano S., Bruni O., Leuzzi V., Elia M., Verrillo E. & Ferri R. (2004) Sleep polygraphy in Angelman syndrome. *Clinical Neurophysiology* **115**, 938–45.

Miano S., Bruni O., Elia M., Scifo L., Smerieri A., Trovato A. et al. (2008) Sleep phenotypes of intellectual disability: a polysomnographic evaluation in subjects with Down syndrome and fragile-X syndrome. *Clinical Neurophysiology* **119**, 1242–7.

Patel D. R., Cabral M. D., Ho A. & Merrick J. (2020) A clinical primer on intellectual disability. *Translational Paediatrics* **9**, S23–35.

Prasher V. P. & Janicki M. P. (2019) *Physical Health of Adults with Intellectual Developmental Disability*. Springer International Publishing, Cham.

Purugganan O. (2018) Intellectual disabilities. *Pediatrics in Review* **39**, 299–309.

Quine L. (1992) Severity of sleep problems in children with severe learning difficulties: description and correlates. *Journal of Community and Applied Social Psychology* **2**, 247–68.

Richdale A. L. & Prior M. R. (1995) The sleep/wake rhythm in children with autism. *European Child and Adolescent Psychiatry* **4**, 175–86.

Richdale A. L. & Schreck K. A. (2009) Sleep problems in autism spectrum disorders: prevalence, nature, & possible biopsychosocial aetiologies. *Sleep Medicine Reviews* **13**, 403–11.

Rosenthal R. (1979) The file drawer problem and tolerance for null results. *Psychological Bulletin* **86**, 638–41.

Ruijter G. J., Valstar M. J., van de Kamp J. M., van der Helm R. M., Durand S., van Diggelen O. P. et al. (2008) Clinical and genetic spectrum of Sanfilippo type C (MPS IIIC) disease in The Netherlands. *Molecular Genetics and Metabolism* **93**, 104–11.

Smith A. C. M., Morse R. S., Introne W. & Duncan W. C., Jr. (2019) Twenty-four-hour motor activity and body temperature patterns suggest altered central circadian timekeeping in Smith-Magenis syndrome, a neurodevelopmental disorder. *American Journal of Medical Genetics. Part A* **179**, 224–36.

Smith-Hicks C., Wright D., Kenny A., Stowe R. C., McCormack M., Stanfield A. C. et al. (2021) Sleep abnormalities in the synaptopathies-SYNGAP1-related intellectual disability and Phelan-McDermid syndrome. *Brain Sciences* **11**, 1229.

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

Stefanski A., Calle-López Y., Leu C., Pérez-Palma E., Pestana-Knight E. & Lal D. (2021) Clinical sequencing yield in epilepsy, autism spectrum disorder, and intellectual disability: a systematic review and meta-analysis. *Epilepsia* **62**, 143–51.

Surtees A. D. R. (2016) Sleep problems in children with developmental disorders. University of Birmingham. Clin. Psy.D. Available at: https://etheses.bham.ac.uk/id/eprint/6958/

Surtees A., Oliver C., Jones C. A., Evans D. L. & Richards C. (2018) Sleep duration and sleep quality in people with and without intellectual disability: a meta-analysis. *Sleep Medicine Reviews* **40**, 135–50.

Tawfik T. Z., Hashem S., Zaki M. A., El-Shazly N., Hegazy M. M., El-Meguid N. A. et al. (2009) Sleep disorders in fragile X syndrome. *The Egyptian Journal of Neurology, Psychiatry, and Neurosurgery* **46**, 445–53.

Trickett J., Heald M., Oliver C. & Richards C. (2018) A cross-syndrome cohort comparison of sleep disturbance in children with Smith-Magenis syndrome, Angelman syndrome, autism spectrum disorder and tuberous sclerosis complex. *Journal of Neurodevelopmental Disorders* **10**, 9.

Trickett J., Oliver C., Heald M., Denyer H., Surtees A., Clarkson E. et al. (2019) Multi-method assessment of sleep in children with Angelman syndrome: a case-controlled study. *Frontiers in Psychiatry* **10**, 874.

Trickett J., Oliver C., Heald M., Denyer H., Surtees A., Clarkson E. et al. (2020) Sleep in children with Smith-Magenis syndrome: a case-control actigraphy study. *Sleep* **43**, zsz260.

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**, 1310–32.

Vasudevan P. & Suri M. (2017) A clinical approach to developmental delay and intellectual disability. *Clinical Medicine* **17**, 558–61.

Wiggs L. D. (2001) Sleep problems in children with developmental disorders. *Journal of the Royal Society of Medicine* **94**, 177–9.

Williams S. R., Zies D., Mullegama S. V., Grotewiel M. S. & Elsea S. H. (2012) Smith-Magenis syndrome results in disruption of CLOCK gene transcription and reveals an integral role for RAI1 in the maintenance of circadian rhythmicity. *The American Journal of Human Genetics* **90**, 941–9.

World Health Organization (1992) *The ICD-10 classification of mental and behavioural disorders: Diagnostic criteria for research*. World Health Organisation.

Supporting Information

Additional Supporting Information may be found online in the supporting information tab for this article.

Table S1. Means of sleep **time** for experimental and control groups. Note differences in decimal places/significant figures reflect different reporting across original studies.

Table S2. Means of sleep **quality** for experimental and control groups. Note differences in decimal places/significant figures reflect different reporting across original studies.

Table S3. PRISMA 2020 Checklist.

Figure S1. Forest plot for the meta-analysis of sleep time in groups of people with genetic syndromes or neurodevelopmental conditions.

Figure S2. Baujat plot illustrating leave one out analysis for the meta-analysis of sleep time in groups of people with intellectual disability of heterogeneous origin.

Figure S3. Forest plot for the meta-analysis of sleep time in groups of people with intellectual disability of heterogeneous origin.

Figure S4. Baujat plot illustrating leave one out analysis for the meta-analysis of sleep quality in groups of people with genetic syndromes or neurodevelopmental conditions.

Figure S5. Forest plot for the meta-analysis of sleep quality in groups of people with intellectual disability of heterogeneous origin.