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Concordance of objective and subjective measures of sleep in children with neurodevelopmental conditions: A systematic review and meta-analysis

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ABSTRACT

The purpose of this systematic review and meta-analysis is to delineate the concordance of objective and subjective measures of sleep in children with neurodevelopmental conditions (NDCs). A systematic literature search identified 31 studies that compare objective and subjective estimates of sleep parameters in autism, ADHD or rare genetic syndromes associated with intellectual disability. The meta-analyses revealed smaller mean differences and larger correlations indicative of greater concordance for parameters associated with sleep scheduling compared to parameters associated with sleep duration and night awakenings. Relative to objective measures, subjective measures produced: 1) greater estimates of total sleep time, sleep efficiency and time in bed; and 2) lower estimates of wake after sleep onset and number of night awakenings. Subgroup analyses also revealed differences in concordance between measurement comparison types (e.g., stronger correlations between actigraphy and sleep diaries, compared to actigraphy and questionnaires) and NDC diagnostic groups. The results predominantly replicate concordance trends observed in typically-developing samples, although some NDC-specific patterns of concordance were identified. This indicates that objective and subjective sleep measures retain broadly similar properties across populations, although researchers and clinicians should be cautious of the impact of NDC-related characteristics on sleep parameter estimates. These findings should inform sleep assessment design and the interpretation of sleep parameter estimates in NDCs, increasing the rigour of sleep parameter description across research and clinical settings.

1. Introduction

Poor sleep is common amongst children with neurodevelopmental conditions (NDCs). Sleep difficulty prevalence rates range from 50 to 80% in autistic children [1], 25-70% in children with ADHD [2], and approximately 19-95% of individuals with rare genetic syndromes associated with intellectual disability experience 'general' sleep difficulties [3]. The prevalence of poor sleep exceeds that observed in typically-developing (TD) children, likely due to biological, behavioural, psychological and environmental risk factors engendered by the presence of NDCs [4–6]. Poor sleep is a significant burden for children with NDCs as it is associated with several deleterious daytime outcomes including increased challenging behaviour [7,8], depressed mood and anxiety [9–11] and poorer adaptive functioning [12,13]. Poor sleep in NDC populations may also negatively impact caregiver wellbeing and family functioning [14–16]. The prevalence and impact of poor sleep in children with NDCs demonstrates the need to better understand children's sleep in research and clinical settings.

Poor sleep can be conceptualised in terms of sleep disorders and their behavioural, physiological and experiential symptoms, which are defined by diagnostic manuals such as the International Classification of Sleep Disorders – Third Edition (ICSD-3 [17]). However poor sleep and sleep disorders can also be understood in terms of

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Abbreviations

A	Autism
AASM	American academy of sleep medicine
ACT	Actigraphy
ADHD	Attention-deficit hyperactivity disorder
AMI	Ambulatory monitoring, INC
AS	Asperger syndrome
BISQ	Brief infant screening questionnaire
CI	Confidence interval
CSHQ	Children's sleep habits questionnaire
CSQ	Child sleep questionnaire
DS	Down syndrome
DSM	Diagnostic & statistical manual
ECG	Electrooculography
EEG	Electroencephalography
EMG	Electromyography
ESS	Epworth sleepiness scale
FISH	Family inventory of sleep habits
ICD	International classification of disorders
MESS	Modified Epworth sleepiness scale
MSLT	multiple sleep latency test
MSPSQ	Modified Simmonds & Parraga sleep questionnaire
NDC	Neurodevelopmental condition
PI	Prediction interval
PKS	Pallister-Killian syndrome
PSG	Polysomnography
PSQ	Pediatric sleep questionnaire
PSQI	Pittsburgh sleep quality index
Q	Questionnaire
QEM	Quality-effects model
R&K	Rechtschaffen and Kales
REM	Random-effects model
RS	Rett syndrome
SADS	Schedule for affective disorders and schizophrenia
SD	Sleep diary
SDB	Sleep-disordered breathing
SDSC	Sleep disturbance scale for children
SE	Sleep efficiency
SOL	Sleep onset latency
TD	Typically-developing
TIB	Time in bed
TST	Total sleep time
WASO	Wake after sleep onset
WS	Williams syndrome

sleep parameters; these describe structural aspects of sleep, including the quantity (e.g. sleep onset latency (SOL) and wake after sleep onset (WASO)) and timing (e.g. time of sleep onset and sleep offset) of sleep and wake, both of which are associated with poor sleep, individually and collectively.

Sleep parameters are estimated using objective and subjective measurement techniques. The most implemented objective techniques in NDC pediatric research include actigraphy and polysomnography (PSG) [18,19]. Actigraph devices are typically worn on the wrist or ankle, and measure acceleration generated by body movements via accelerometers; acceleration data are processed to estimate sleep and wake patterns. Polysomnography is considered the 'gold-standard' measure of sleep, integrating measures of brain activity via electroencephalography with other physiological signals, including electromyography, electrooculography and pulse oximetry, to code sleep stages and sleep architecture, and detect sleep disorders [20]. With regard to subjective measures, caregiver-completed sleep diaries and questionnaires are conventional throughout NDC pediatric research [18], thus capturing caregivers' perceptions of children's sleep. Sleep diaries prompt daily recordings of specific aspects of children's sleep, such as sleep schedules, night awakenings and daytime naps [19]. Diaries may also measure sleep-related behaviours, for example bedtime resistance [21]. Questionnaires ask caregivers to reflect on children's typical sleep/wake patterns and sleep-related behaviours over a recent time period, and rate these on predefined categorical or continuous scales (e.g. Likert-type scales, yes/no questions), or open-ended questions regarding time and length of sleep behaviours. Questionnaires are completed at one time point, and thus provide global estimates of children's sleep parameters.

Despite the heterogeneity of objective and subjective sleep measures, these are frequently used to measure the same sleep parameters. Established research with TD children has identified differences between objective and subjective sleep parameter estimates, and revealed patterns in the magnitude and direction of these differences across parameters. Most notably, caregiver-completed sleep diaries and questionnaires often underestimate the frequency and duration of children's night awakenings, and subsequently overestimate total sleep time (TST) and sleep efficiency (SE), compared to actigraphy [22–25] and PSG [26,27]. In contrast, closer agreement is observed for sleep scheduling parameters, including time of sleep-onset and sleep offset [22,28]. The concordance of objective and subjective sleep estimates also depends on the specific measures being compared. Compared to actigraphy-questionnaire comparisons, stronger correlations and smaller differences in absolute estimates are indicated across all sleep parameters for actigraphy-sleep diary comparisons [23,25,29]. This may be due to the lower precision of questionnaires conferred by global estimates of sleep, categorical scales and greater vulnerability to recall biases [30].

Delineating concordance between objective and subjective sleep measures is beneficial as this reveals whether the different designs of sleep measures influence the quantification of sleep parameters. These insights can inform the interpretation of objective and subjective sleep data, providing more precise insights into children's sleep/wake patterns. Understanding measurement concordance may also inform the design of sleep assessments necessary to obtain more representative accounts of children's sleep parameters. These insights are crucial as researchers and clinicians may preferentially employ subjective measures to describe children's sleep, as these are more affordable and do not require technical expertise to administer [31,32].

Although the concordance of objective and subjective measures is well-established in TD children, this knowledge lags behind in children with NDCs. This disparity needs to be resolved as NDC-related characteristics may affect the properties, and subsequent concordance, of objective and subjective sleep measures. Firstly, poorer concordance of objective and subjective sleep measures has been observed in individuals with sleep difficulties and disorders, relative to those with good sleep quality [33,34]. Although previous findings predominantly relate to adult samples, this effect may extend to NDC populations and thus skew concordance trends relative to TD peers. Secondly, higher rates of co-sleeping and "signalling" behaviours have been observed in autistic children [35–37] and children with intellectual disabilities [35,38–40] than in TD children. Evidence suggests these behaviours may improve

the concordance of objective and subjective sleep parameter estimates, possibly by increasing child-caregiver interactions throughout the night [41–43]. Thirdly, caregivers of children with NDCs are likely to experience poor sleep and heightened stress [14,44], which are associated with deficits in retrospective memory (i.e. recall of information) and prospective memory (i.e. recall of intentions to complete a task) [45–47]. Memory impairments may impede the accuracy of subjective sleep estimates, and potentially inhibit timely completion of sleep diaries, further increasing recall biases in sleep estimates. Fourthly, compared to TD children, there is greater night-to-night instability of sleep in autistic children [48,49], and children with ADHD [50–54] and rare genetic syndromes [55,56], compared to TD peers. This instability reduces the representativeness of global sleep parameter estimates from questionnaires [56], and may increase discordance between sleep measures administered over few nights (e.g. PSG) and those administered over many (e.g. sleep diaries). Finally, previous NDC research suggests that challenging behaviour around bedtime and during night awakenings may impact caregivers' estimates of children's sleep parameters [57–61]. However, objective sleep measures are not sensitive to these behaviours, potentially driving further discordance between objective and subjective sleep parameter estimates. Together this evidence highlights the need to investigate the concordance of objective and subjective sleep measures specifically within NDC populations.

Although previous reviews [19,62,63] and meta-analyses [64,65] have explored differences between objective and subjective sleep parameter data in children with NDCs, none have rigorously examined the concordance of these measures. Firstly, previous meta-analyses have not pooled studies that collect objective and subjective sleep data, instead these have evaluated the consistency of significant findings between studies that utilize objective measures, and studies that utilize subjective measures [64,65]. Secondly, only questionnaires were included as subjective measures of sleep despite widespread use of sleep diaries in NDC research and evidence that sleep diaries and questionnaires differ in concordance with objective measures [23,25,29]. Finally, whilst objective and subjective sleep measures are used throughout rare genetic syndrome research [18], no meta-analysis has addressed the concordance of objective and subjective sleep measures in this population specifically. To comprehensively address the concordance of objective and subjective sleep measures in children with NDCs, existing studies that objectively and subjectively measure sleep in NDC samples should be synthesized, including those that utilize sleep diaries and recruit children with rare genetic syndromes associated with intellectual disability.

In summary, the concordance of objective and subjective sleep parameter estimates is poorly understood in children with NDCs. In response, a systematic review and meta-analysis of studies that employ both objective and subjective sleep measures in NDC samples will be conducted, including those that utilize sleep diaries and recruit children with rare genetic syndromes. This meta-analysis was conducted with the following aims:

1) Synthesise available data on concordance between objective and subjective estimates of sleep parameters, in children with NDCs.
2) Explore sources of heterogeneity in the concordance of objective and subjective measures of sleep in children with NDCs, including those identified in previous TD research (method comparisons) and specific to NDC populations (NDC diagnosis).
3) Compare concordance in children with NDCs to existing concordance trends from TD research.

2. Methods

The meta-analysis was pre-registered prior to the search (https://www.crd.york.ac.uk/prospero/display_record.php?ID=CRD42022307499), and was conducted in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines [66].

2.1. Search strategy

Six databases were searched on February 3, 2022: Ovid PsycINFO (1806 to January 2022 Week 5), Ovid MEDLINE(R) and In-Process, In-Data-Review & Other Non-Indexed Citations (1946 to 2nd February 2022), Ovid Embase (1974 to 2nd February 2022), ProQuest Dissertations & Theses Global (all years), PubMed (all years) and World Cat (all years). No other search limits were imposed. These databases encompassed peer-reviewed studies, pre-prints and grey literature subject to review processes (e.g. conference abstracts, theses and dissertations), mitigating against publication bias.

Search strategies used subject headings and keywords (see Table 1 for full list of search terms). Full descriptions of the search strategies for each database are outlined in S1. No search terms were implemented for NDC status to ensure that the breadth of the child literature was identified. This also allowed the search to capture TD concordance studies: those that employed reasonably large samples ($n \geq 75$) were extracted during screening for later comparisons with meta-analysed NDC concordance estimates.

To identify studies not included in databases, reference lists of previous relevant systematic reviews (e.g. Refs. [3,64,67]) and of eligible studies from database searches were also hand-searched.

2.2. Study selection

Fig. 1 outlines the search process and reasons for exclusion. Initial database searches produced 10,752 results. After duplicates were removed, 7007 papers were screened at title and abstract level, using the inclusion and exclusion criteria outlined in Table S2.

Following title and abstract screening, 6563 papers were manually removed (see Table S3 for comprehensive list of papers that met exclusion criteria). Alongside the criteria in Table S2, the full-texts of the remaining 444 papers were screened following criteria outlined in Table S4. Two authors (ROS and AH) independently screened 25% of the papers at title and abstract (1752 papers) and full-text (111 papers) levels. Inter-rater reliability analyses revealed 'almost perfect' agreement for title and abstract screening ($\kappa = 0.837$, $p<.001$) and 'substantial agreement' for full-text screening ($\kappa = 0.737$, $p<.001$). The authors discussed discrepancies and reached consensus for inclusion/exclusion before ROS continued with the remaining 75% of papers.

2.3. Data extraction and quality review

Study characteristics and meta-analysed data were extracted from the 31 eligible studies. Sleep parameters reported in number of hours were converted into number of minutes. Data extraction was completed independently by ROS and a research assistant, intraclass correlation efficient analyses revealed good-to-excellent inter-rater reliability (ICC = 0.93, 95% CI [0.90; 0.95]). Discrepancies were discussed and resolved between the authors.

A quality criteria checklist was adapted from previous meta-analyses [3,68] and the Mixed Methods Appraisal Tool Version 2018 (MMAT [69]) to review the internal and external validity of studies (see Table S5). For each study, sampling technique, complete outcome data, confirmation of NDC and sleep measures were

Table 1
Search terms for central concepts of the research aims.

Sleep	"Sleep*" OR "TST" OR "WASO" OR "night waking*" OR "night awakening*" OR "SE" OR "SOL" OR "time to fall asleep" OR "time in bed" OR "bed time" OR "wake* time" OR "get* time" OR "nap*" OR "fatigue*" OR "tired*" OR "hypersom*" OR "excessive somnol*" OR "narcolepsy" OR "insomnia" OR "circadian rhythm disorder*" OR "CRSD" OR "CRSWD" OR "N24HSWD" OR "non-24-h circadian rhythm disorder" OR "parasomnia*" OR "confusional arousal" OR "somn*" OR "night* terror*" OR "nightmare*" OR "OSA" OR "CSA" OR "hypoventilation*" OR "hypopnea*" OR "night bruxism*" OR "teeth grind*" OR "restless legs syndrome" OR "periodic limb movement syndrome"
Subjective	"Sleep log" OR "sleep diar*" OR "sleep journal" OR "parent* report*" OR "parent* log" OR "parent* diar*" OR "parent* journal" OR "parent-report" OR "caregiver report" OR "caregiver-report" OR "caregiver log" OR "caregiver diar*" OR "caregiver journal" OR "self report" OR "self-report" OR "survey" OR "diar*" OR "instrument*" OR "questionnaire"
Objective	"Actigraph*" OR "actograph*" OR "actimet*" OR "actiwatch*" OR "acceleromet*" OR "actomet*" OR "sleepwatch*" OR "activity sleep monitor*" OR "activity monitor*" OR "polysomnogra*" OR "PSG" OR "electroencephalogra*" OR "EEG" OR "sleep recording*" OR "sleep monitor*" OR "sleep stage*" OR "sleep architecture" OR "objective sleep" OR "videosomnography" OR "VSG" OR "auto-videosomnography" OR "home-videosomnography" OR "videopolysomnography" OR "video-polysomnography" OR "video polysomnography" OR "video recordings of sleep"
Children	"child*" OR "adolescen*" OR "pediatric" OR "pediatric" OR "young people" OR "teen*" OR "youth"

'Sleep*' term captures sleep parameters (e.g. sleep onset latency, total sleep time, sleep efficiency) and disorders (e.g. central sleep apnea, sleep-disordered breathing), therefore these were not specified in the search terms.

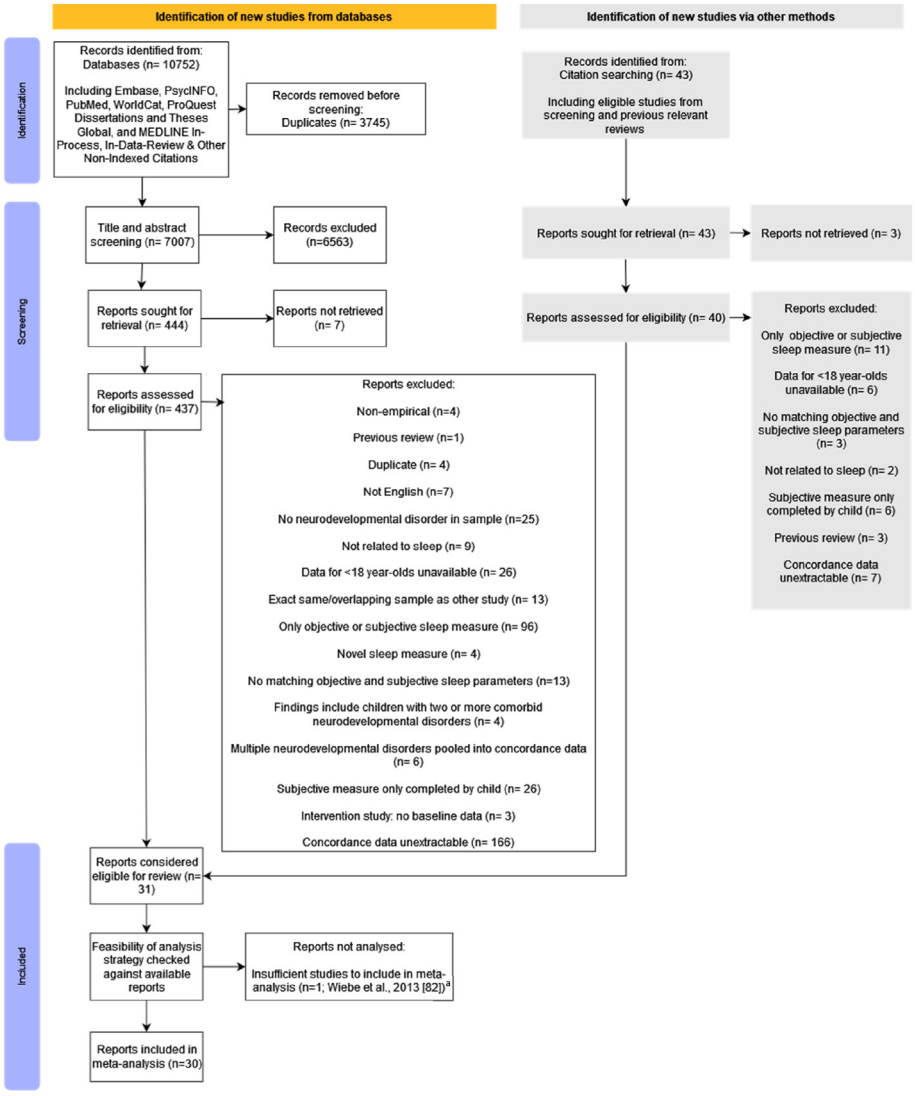

Fig. 1. The search process in accordance with PRISMA 2020 guidelines. *Note.* [a] Wiebe et al. (2013) [82] reported correlations for daytime sleepiness scores from the multiple sleep latency test and a questionnaire.

scored on a scale from 0 (poor) to 3 (excellent). Each concordance estimate involved two sleep measures, and each measure received a score, therefore the mean sleep measure score was taken with total scores ranging from 0 to 12. This was divided by 12 to generate a quality rating, ranging from 0 to 1.

Quality ratings were independently assigned by ROS and AH for all included studies. Intraclass correlations revealed good-to-

excellent inter-rater reliability for the final quality ratings (ICC = 0.88, 95% CI [0.75; 0.94]), and ratings for sampling technique (ICC = 0.90, 95% CI [0.79; 0.95]), confirmation of NDC diagnosis (ICC = 0.92, 95% CI [0.84; 0.96]) and subjective measures (ICC = 0.88, 95% [0.75; 0.94]). Moderate-to-excellent reliability was observed for complete outcome data (ICC = 0.81, ICC [0.61; 0.91]) and objective measure ratings (ICC = 0.80, 95% CI [0.59; 0.91]). Discrepancies were discussed and resolved between the authors.

2.4. Data analysis

Mean values for objective and subjective sleep parameter estimates, and Pearson's r correlation coefficients, were extracted from the 31 eligible studies. Where studies reported Spearman's rank-order correlation coefficients, these were converted to Pearson's r coefficients via formulae described by Pearson [70]; these formulae estimate Pearson's r correlation coefficients with minimal bias [71,72].

Statistical analyses were conducted in R version 3.6.0, using the *meta* package [73], and Microsoft Excel, using the *MetaXL* 5.3 add-in Ref. [74]. Meta-analytic models were conducted for raw mean differences and Pearson's r correlation coefficients, with corresponding 95% confidence intervals and 95% prediction intervals, for models with 3 or more studies [75]. The correlation coefficients were pooled following Fisher's r-to-z transformation, these were subsequently back-transformed and interpreted using guidelines by Hinkle et al. [76]. Random-effects (REM) and quality-effects models (QEM) were calculated for individual sleep parameters. Random-effects models were chosen as these assume between-study differences in sampling error, sample characteristics and study design influence true effect sizes [77]. Given the between-study diversity of sample characteristics and methodology, REMs were considered more appropriate than fixed-effect models. However, REMs do not account for credibility-related heterogeneity between studies, therefore QEM were calculated to additionally weight studies by credibility. Analyses were conducted with the restricted maximum-likelihood estimator (REML) instead of the DerSimonian-Laird estimator, which can produce false positives when meta-analyses contain few studies and heterogeneity is high [78], as was the case in this analysis.

To explore factors that impact concordance of sleep parameter estimates, subgroup analyses were conducted for NDC diagnosis and method comparison type, for example actigraphy versus sleep diary and actigraphy versus questionnaire. Other factors shown to impact concordance (e.g. weekend/weekday data collection [79]) were not assessed due to limited data. A minimum of two studies per subgroup was required and differences in subgroup effect sizes were explored in REMs via *Q*-tests. To compare concordance in NDC and TD populations, QEMs were calculated for individual sleep parameters, with separate models for each method comparison type, and these results were plotted against findings from previous robust TD studies (n≥75).

Heterogeneity was tested with Baujat plots, Cochrane's *Q* and Higgins I^2 (with 95% confidence intervals). Where models contained ≥10 studies, risk of publication bias was tested with visual inspection of contour-enhanced funnel plots and Egger's test for funnel plot asymmetry [80]. Leave-one-out sensitivity analyses were conducted to determine the influence of individual studies on overall model effect size estimates, and the rigour of these estimates. Outliers were assessed for models/subgroups with ≥3 studies via externally studentized residuals, following the recommendations of Viechtbauer and Cheung [81]. Studies with residuals >1.96 were classified as outliers. Outliers were removed where these substantially affected overall model effect size estimates, determined by leave-one-out analyses, and could be attributed to poor methodology.

3. Results

3.1. Study characteristics

Study characteristics and quality ratings are presented in Table 2. Of the 31 eligible studies, one was not meta-analysed due to insufficient data [82]. Of those studies meta-analysed, fourteen conducted actigraphy-sleep diary comparisons, eight conducted actigraphy-questionnaire comparisons, two conducted PSG-sleep diary comparisons, and seven conducted PSG-questionnaire comparisons. Across all studies, actigraphy and sleep diaries were completed simultaneously, and sleep diaries were completed immediately prior to PSG. The administration of questionnaires and objective sleep measures was less well described, two studies indicated PSG and questionnaires were completed on the same night [98,99], and one study indicated questionnaires were completed approximately seven days prior to actigraphy [57].

Quality ratings revealed most studies had 'fair'-to-'good' sample recruitment (25/31) and confirmation of NDC diagnosis (22/31). With the exception of questionnaires, sleep measures received lower ratings. Sleep diaries and PSG were rated as 'poor' in the majority of studies (12/16 and 5/9 respectively), primarily as the number of valid nights of data and PSG derivations were unspecified. Of the 21 actigraphy studies, 13 received a 'fair' rating and four were rated as 'poor', due to poor descriptions of concomitant sleep diaries, data cleaning procedures, missing data procedures, and when activity markers were pressed. Ratings for complete outcome data were also generally low, with 7/31 and 14/31 studies rated as 'poor' and 'fair', respectively.

3.1.1. Concordance between objective and subjective sleep parameter estimates in children with NDCs

To address the first aim of the meta-analysis, raw mean differences and Pearson's r correlation coefficients were pooled for individual sleep parameters. These analyses combined all objective-subjective method comparison types (e.g. actigraphy-questionnaire, polysomnography-questionnaire). Outliers were identified in the raw mean difference models for WASO [102], TST [103], sleep offset time [60], sleep onset time [88], number of night awakenings [53,94] and SE [100]. Outliers were also identified in the correlation models for sleep offset time [83] and TST [58]. All outliers remained in the models, either due to little influence on the pooled model effect, determined by leave-one-out analyses, or sound methodology consistent with the methods in the remaining estimates. The final QEM forest plots are presented in Figs. S6–S21.

The REMs revealed significantly greater mean values for TST, SE and time in bed (TIB), and lower mean values for sleep onset time, WASO and number of night awakenings, from subjective measures compared to objective measures (Table 3 and Figs. 2 and 3). Large effects were observed across the sleep parameters, with objective and subjective TST, TIB and WASO estimates differing by over 50 min. Sleep offset time and SOL mean values did not significantly differ between objective and subjective measures. The QEMs produced very similar pooled estimates to the REMs, aside for sleep onset time which was no longer statistically significant ($p = 0.054$). Evidence of heterogeneity was found for all models except sleep offset time and TIB (see Table 3).

The REMs also revealed positive correlations for all sleep parameters aside from number of night awakenings, which was marginally negative (see Table 4 and Fig. 4). From strongest to weakest, significant correlations were observed for sleep offset time, SOL, sleep-disordered breathing (SDB) and TST ($r = 0.803$,

Table 2
Sample characteristics and quality criteria for included papers.

Author	NDC	Sample characteristics			Sleep measurement		Quality criteria				
		N	Male:Female	M Age ±SD (Range)	Objective measure *Scoring algorithm/criteria*	Subjective measure *Method of evaluation*	Recruitment	Complete data	Confirmation	Objective[a]	Subjective[a]
Allik, 2006 [83]	AS/ autism	32	28:4	10.8 (8.5–12.8)	Actigraphy (Actiwatch, CamNtech) *Actiwatch Sleepwatch algorithm*	Sleep diary (study specific)	2	3	1	1	3
Ashworth et al., 2013 [84]	DS	20	10:10	9.42 ± 1.98 (6.09–12.23)	Actigraphy (Actiwatch Mini, CamNtech) *Unspecified algorithm*	Questionnaire (CSHQ) *Rating scale - subscale scores*	2	2	3	1	3
Ashworth et al., 2013 [84]	WS	24	12:12	9.55 ± 2.09 (6.08–12.58)	Actigraphy (Actiwatch Mini, CamNtech) *Unspecified algorithm*	Questionnaire (CSHQ) *Rating scale - subscale scores*	2	3	3	1	3
Chin et al., 2018 [85]	ADHD	71	54:17	8.83 ± 1.86	PSG (Laboratory) *AASM International criteria*	Questionnaire (PSQ) *Rating scale - subscale scores*	1	3	3	0	2
Choi et al., 2010 [86]	ADHD	27	24:3	8.97 ± 2.1	PSG (Laboratory) *R&K criteria*	Questionnaire (CSHQ) *Single open-ended items*	1	0	2	1	3
Cipolla et al., 2010 [87]	Autism	15	–	–	Actigraphy (Mini-MotionLogger, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Questionnaire (CSHQ) *Rating scale - subscale scores*	2	1	1	2	3
Corkum, 1999 [57]	ADHD	25	20:5	9.12 ± 1.42	Actigraphy (Mini-MotionLogger, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Questionnaire (CSQ) *Single open-ended items*	1	1	2	2	1
Corkum, 1999 [57]	ADHD	25	20:5	9.12 ± 1.42	Actigraphy (Mini-MotionLogger, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	1	1	2	2	2
Corkum et al., 2008 [88]	ADHD	21	15:6	8.5 ± 1.63 (6.08–12.08)	Actigraphy (Mini-MotionLogger, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (included in previous studies by author)	1	1	3	2	3
Cortesi et al., 2012 [89]	Autism	134	–	–	Actigraphy (unspecified, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	1	0	2	1	0
Diaz-Roman et al., 2019 [90]	ADHD	20	–	–	PSG (Home) *R&K criteria*	Sleep diary (study specific)	3	0	1	1	0
Esbensen et al., 2018 [58]	DS	44	–	–	Actigraphy (MMML, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Questionnaire (CSHQ) *Rating scale - subscale scores*	0	2	0	0	3
Fetta et al., 2021 [91]	PKS	14	5:9	8.39 ± 6.14 (1–17.33)	PSG (Laboratory) *AASM International criteria*	Questionnaire (SDSC) *Rating scale - subscale scores*	2	3	3	0	1
Gringras et al., 2014 [92]	Autism	67	54:13	(5–16.83)	Actigraphy (MMML, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	2	0	1	1	0
Gruber et al., 2012 [59]	ADHD	26	17:9	8.46 ± 1.5 (7–11)	PSG (Home) *AASM International criteria*	Questionnaire (CSHQ) *Rating scale - subscale scores*	3	3	2	1	3
Gruber et al., 2011 [93]	ADHD	11	7:4	8.7 ± 1.3	Actigraphy (ActiWatch 64, MMI) *Actiware Sleep algorithm*	Sleep diary (study specific)	3	0	2	0	0
Gwilliam et al., 2020 [94]	WS	11	–	2.55 ± 0.05	Actigraphy (MotionWatch8, CamNtech) *Not specified*	Questionnaire (BISQ) *Single open-ended items*	2	1	1	1	1
Hagebeuk et al., 2012 [95]	RS	7	0:7	6.86 ± 3.53	PSG (Laboratory) *AASM International criteria*	Questionnaire (SDSC) *Rating scale - subscale scores*	0	1	1	0	1
Hering et al., 1999 [60]	Autism	8	7:1	8.0 ± 3.0	Actigraphy (unspecified, AMInc) *Sadeh* et al. *(1994) algorithm* [106]	Questionnaire (study specific) *Single open-ended items*	1	1	1	1	0
Hvolby et al., 2008 [53]	ADHD	45	37:8	Median 8.33 (5.75–10.92)	Actigraphy (Mini-MotionLogger, AMI) *Not specified*	Sleep diary (study specific)	1	3	2	0	0
Lambert et al., 2016 [26]	Autism	11	–	10.27 ± 2.24	PSG (Laboratory) *R&K criteria*	Sleep diary (study specific)	1	3	2	0	0
Merbler et al., 2018 [96]	RS	12	0:12	9.34 ± 4.35 (1.66–17.08)	Actigraphy (Actiwatch 2, Philips) *Not specified*	Sleep diary (study specific)	1	2	3	1	2
Mughal et al., 2020 [97]	Autism	17	–	–	Actigraphy (MotionWatch8, CamNtech) *Not specified*	Sleep diary (study specific)	2	1	1	0	0
Ng, 2018 [98]	DS	20	12:8	Median 11.5 IQR 5.4	PSG (Laboratory) *AASM International criteria*	Questionnaire (PSQ) *Rating scale - subscale scores*	1	1	0	1	2
Pabary et al., 2019 [99]	DS	35	–	Median 6 (2–16)	PSG (Laboratory) *AASM International criteria*	Questionnaire (PSQ) *Rating scale - subscale scores*	1	0	0	0	2
Sanabra et al., 2021 [100]	ADHD	60	34:26	9.32 ± 2.82	Actigraphy (ActiSleep, ActiGraph) *Not specified*	Sleep diary (study specific)	1	3	3	1	0
Sniecinska, 2014 [101]	WS	21	–	–	Actigraphy (Actiwatch Mini, CamNtech) *Not specified*	Questionnaire (CHSQ) *Rating scale - subscale scores*	2	1	3	1	3

Surtees, 2016 [102]	Autism	16	10:6	9.81 ± 2.40 (5–13)	Actigraphy (Actiwatch, Philips) *Not specified*	Sleep diary (study specific)	3	1	2	2	0	
Tse et al., 2020 [103]	Autism	78	62:16	10.05 ± 1.08	Actigraphy (GT3X, ActiGraph) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	2	0	1	1	0	
Tse et al., 2019[c] [104]	Autism	19	14:5	10.11 ± 1.2	Actigraphy (GT3X, ActiGraph) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	2	1	1	1	0	
Tse et al., 2019[d] [104]	Autism	21	18:3	9.81 ± 1.17	Actigraphy (GT3X, ActiGraph) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	2	1	1	1	0	
Veatch, 2016 [105]	Autism	80	64:16	–	Actigraphy (Actiwatch Spectrum, Philips) *Philips Respironics algorithm*	Questionnaire (CSHQ) *Single open-ended items*	2	1	2	1	3	
Wiebe et al. 2013[b] [82]	ADHD	20	13:7	9.2 ± 1.6	MSLT	Questionnaire (MESS) *Rating scale - total score*	2	2	-[e]	2	1	
Wiggs et al., 2005 [61]	ADHD	71	63:8	8.8 ± 2.6	Actigraphy (Mini Motionlogger, AMI) *Sadeh* et al. *(1994) algorithm* [106]	Sleep diary (study specific)	2	3	2	1	0	

Abbreviations: AASM: American Academy of Sleep Medicine. AMI: Ambulatory Monitoring, Inc. AMInc: American Military, Inc. AS: Asperger syndrome. BISQ: brief infant screening questionnaire. CSHQ: children's sleep habits questionnaire. CSQ: child sleep questionnaire. DS: Down syndrome. MESS: modified Epworth sleepiness scale. MMI: MiniMitter, Inc. MMML: Micro-Mini Motionlogger. MSLT: multiple sleep latency test. PKS: Pallister-Killian syndrome. PSG: Polysomnography. PSQ: pediatric sleep questionnaire. R&K: Rechtschaffen and Kales. RS: Rett syndrome. SDSC: sleep disturbance scale for children. WS: Williams syndrome.

[a] Mean average of ratings for objective and subjective sleep measures included in overall quality rating scores.

[b] Not included in meta-analysis due to insufficient studies.

[c] One of two subgroups of autistic children: "physical activity intervention".

[d] One of two subgroups of autistic children: "control".

[e] Quality rating not assigned as quality criteria not developed for multiple sleep latency test.

high, to 0.494, moderate). Non-significant correlations were observed for sleep onset time, WASO, SE and number of night awakenings. The QEMs produced closely replicated the REM estimates, with sleep offset time and SDB as notable exceptions. Substantial heterogeneity was observed for all models aside from SE, number of night awakenings and SDB (see Table 4).

Sensitivity analyses revealed influential studies in the raw mean difference model for sleep onset time, and correlation models for number of night awakenings, SE, SDB, sleep offset time, sleep onset time and WASO. No influential studies were identified in the remaining models.

3.1.2. Subgroup analyses exploring sources of heterogeneity in the concordance of objective and subjective sleep parameter estimates

To address the second aim of the meta-analysis, subgroup analyses were conducted to explore the effect of method comparison type and NDC diagnosis on concordance of objective and subjective sleep parameter estimates. Given limited data, subgroup analyses were only conducted for a subset of method comparison types, NDC diagnoses, and sleep parameters.

3.2. Method comparison type

Differences between TST means did not vary between actigraphy and sleep diary, actigraphy and questionnaire, and PSG and sleep diary comparisons ($Q = 0.46$, $p = 0.79$). Similarly, sleep offset time mean differences did not diverge across actigraphy and sleep diary, and actigraphy and questionnaire comparisons ($Q = 0.83$, $p = 0.36$). There was no significant difference in SE mean differences between actigraphy and sleep diary, and PSG and sleep diary comparisons ($Q = 1.27$, $p = 0.26$).

In contrast to mean difference analyses, significant differences in TST correlations were observed between method comparison types ($Q = 10.24$, $p = 0.001$), with stronger positive correlations for sleep diaries and actigraphy ($r = 0.68$, 95% CI [0.58; 0.76]) compared to questionnaires and actigraphy ($r = 0.31$, 95% CI [0.05; 0.53]). Similarly, correlations of SOL estimates were significantly stronger for sleep diaries and actigraphy ($r = 0.78$, 95% CI [0.66; 0.87]) compared to questionnaires and actigraphy ($r = 0.51$, 95% CI [0.20; 0.73], $Q = 4.69$, $p = 0.03$). Non-significant differences in WASO correlations were noted between sleep diaries and actigraphy, and questionnaires and actigraphy ($Q = 0.07$, $p = 0.79$).

3.2.1. NDC diagnosis

Due to limited studies, rare genetic syndromes and several sleep parameters (SE, TIB, sleep onset time, sleep offset time, WASO, and number of night awakenings) were excluded from the between-NDC comparisons. Differences between objective and subjective SOL means did not vary between autistic children and children with ADHD ($Q = 1.64$, $p = 0.20$). However, differences between objective and subjective TST means were significantly greater for children with ADHD (MD = 98.24, 95% CI [68.36; 128.11]) compared to autistic children (MD = 46.43, 95% CI [12.28; 80.58], $Q = 5.01$, $p = 0.03$).

3.2.2. Comparisons of concordance between children with NDCs and TD children

To accomplish the third aim of the meta-analysis, the results of previous TD studies ($n \geq 75$) were plotted against QEM pooled estimates, controlling for specific method comparisons (Figs. 5-7). The figures indicate broadly similar trends of concordance between objective and subjective sleep measures in NDC and TD pediatric populations. Notable discrepancies between the groups are only evident for actigraphy and questionnaire TST mean differences, and correlations between PSG and questionnaire SDB estimates.

3.2.3. Publication bias

Only the TST raw mean difference and correlations models had sufficient studies (k≥ 10) to test publication bias. Visual inspection of the contour-enhanced funnel plots did not indicate asymmetry, although a typical funnel shape was not produced in either plot (see complete funnel plots in Figs. S22 and S23). Egger's test identified non-significant effects, confirming the lack of asymmetry for the TST mean difference model ($p = 0.78$) and TST correlation model ($p = 0.36$).

4. Discussion

4.1. Summary of findings

This review was the first to delineate the concordance of objective and subjective sleep measures in children with NDCs, including autism, ADHD and rare genetic syndromes associated with intellectual disability. The findings extend those of previous meta-analyses [64,65] by pooling studies that employed both objective and subjective sleep measures, including sleep diaries, as well as those that recruit rare genetic syndrome samples. Additionally, this review was the first to explore the factors that impact concordance in NDC populations, and whether concordance trends were consistent between NDC and TD populations.

To address the first aim of the meta-analysis, objective and subjective sleep parameter estimates were compared via mean differences and correlation coefficients. The results varied substantially between sleep parameters, with smaller mean differences and stronger correlations for parameters associated with children's sleep schedules, such as SOL and sleep offset time, compared to sleep duration and night awakening parameters, including TST, SE, WASO, TIB and number of night awakenings. Sleep onset time was the only exception to this trend; however this should be interpreted cautiously given these models included three or fewer studies, had broad prediction intervals, and the pooled effects were susceptible to poor methodological quality. The results also revealed specific patterns of concordance for nocturnal sleep parameters, with subjective measures producing greater estimates of TST (+73.24 min), TIB (+53.36 min) and SE (+5.34%), and lower estimates of WASO (-50.81 min) and number of night awakenings (-23.07 awakenings), compared to objective measures.

Against expectations, the findings broadly replicated patterns of concordance observed in previous TD research (e.g. Refs. [22–27]), with similarities in the strength and direction of effects. Notable exceptions include the large mean differences for WASO and number of night awakenings, which are typically smaller in TD samples [22,24,26]. This discrepancy may be explained by retrospective memory difficulties incurred by stress and sleep difficulties in caregivers of children with NDCs [14,44–47]. Alternatively, challenging behaviours during night awakenings may influence caregivers' estimates of night awakening parameters [57,58,60,71]. The broad similarity of concordance between TD and NDC populations indicates consistencies in the properties of sleep measures and mechanisms of concordance between the populations. As suggested in TD research, strong concordance may be observed for sleep scheduling parameters due to high caregiver involvement around children's sleep/wake routines and easier interpretability and calculation of these parameters [62,79]. With regard to nocturnal sleep parameters, caregivers may underestimate the duration and frequency of children's night awakenings,

Table 3
Summary of random-effects and quality-effects mean difference meta-analyses for objective and subjective sleep parameter estimates. Pooled mean differences and confidence intervals are presented, as well as prediction intervals and heterogeneity statistics (I^2 and Q-test) for random-effects models.

Sleep parameter (total pooled concordance estimates)	Concordance estimates per method comparison (NDC diagnosis for each estimate)				REM mean difference *95% CI*	QEM mean difference *95% CI*	*95% PI*[a]	I^2[a] *95% CI*	*Q*-test[a]	*p value*
	ACT + SD	ACT + Q	PSG + SD	PSG + Q						
Sleep onset latency (k = 9)	6 (4 ADHD, 2 A)	1 (1 WS)	1 (A)	1 (1 ADHD)	4.60 *-1.74; 10.94*	4.37 *-2.01; 10.75*	*-16.08; 25.28*	82.5% *68.1%; 90.4%*	45.66	< 0.001
Sleep onset time (k = 3)[b]	1 (1 ADHD)	2 (1 ADHD, 1 A)	0	0	**-50.29** ***-94.58; -5.99***	-44.96 *-90.68; 0.77*	*-562.87; 462.29*	76.9% *24.7%; 92.9%*	8.66	0.01
Sleep offset time (k = 5)[b]	3 (1 ADHD, 2 A)	2 (1 ADHD, 1 A)	0	0	3.26 *-6.65; 13.17*	5.04 *-4.97; 15.06*	*-12.84; 19.36*	50.2% *0.00%; 81.7%*	8.02	0.09
Time in bed (k = 3)	2 (1 ADHD, 1 A)	0	1 (1 ADHD)	0	**53.63** ***39.59; 67.68***	**53.36** ***39.20; 67.54***	*-37.42; 144.68*	0.0% *0.00%; 89.6%*	1.18	0.55
Total sleep time (k = 17)[b]	12 (4 ADHD, 7 A, 1 RS)	2 (1 ADHD, 1 A)	2 (1 ADHD, 1 A)	1 (1 ADHD)	**71.95** ***47.21; 96.69***	**73.24** ***47.53; 98.96***	*-35.74; 179.63*	94.4% *92.4%; 95.9%*	285.98	< 0.001
Wake after sleep onset (k = 4)	3 (2 A, 1 RS)	1 (1 WS)	0	0	**-52.62** ***-70.14; -35.09***	**-50.81** ***-68.96; -32.66***	*-133.96; 28.73*	91.5% *81.5%; 96.1%*	35.43	< 0.001
Sleep efficiency (k = 7)	5 (1 ADHD, 4 A)	0	2 (1 ADHD, 1 A)	0	**5.38** ***2.10; 8.66***	**5.34** ***1.95; 8.73***	*-6.24; 17.00*	96.6% *94.9%; 97.8%*	179.07	< 0.001
Number of night awakenings (k = 4)	2 (2 ADHD)	1 (1 WS)	1 (1 A)	0	**-22.53** ***-34.13; -10.93***	**-23.07** ***-34.73; -11.41***	*-78.83; 33.77*	99.1% *98.7%; 99.4%*	350.30	< 0.001

Abbreviations: A: autism. ACT: actigraphy. ADHD: attention-deficit hyperactivity disorder. CI: confidence intervals. DS: Down syndrome. NDC: neurodevelopmental condition. PI: prediction intervals. PSG: polysomnography. Q: questionnaire. QEM: quality-effects model. REM: random-effects model. RS: Rett syndrome. SD: sleep diary. WS: Williams syndrome. Statistically significant mean differences (p< 0.05) are in bold.
[a] Values for random-effects models. [b] Corkum (1999) [56] reported actigraphy, sleep diary and questionnaire means for TST, and sleep onset and offset time. Only actigraphy and questionnaire data were included to avoid double-counting actigraphy data, and enable method comparison subgroup analyses.

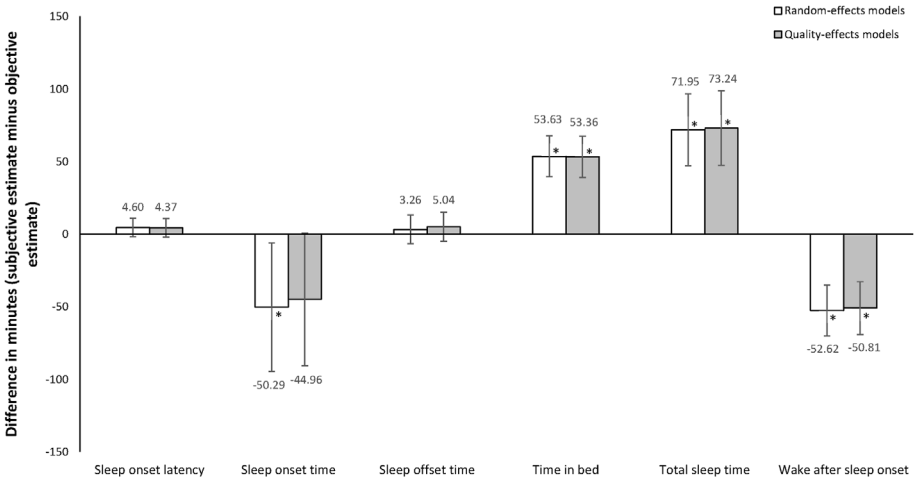

Fig. 2. Random-effects and quality-effects pooled differences between mean sleep parameter estimates from objective and subjective sleep measures, with 95% confidence intervals. Differences should be interpreted in reference to typical values for each sleep parameter (e.g. sleep onset latency is typically fewer minutes than time in bed, leading to smaller mean differences). Note. Asterisks indicate statistically significant effects.

and subsequently overestimate TST and TIB, due to limited awareness of children's night awakenings [120] and difficulties accurately recalling the children's night awakenings [79]. To confirm and extend the current findings, future research should test directly the impact of NDC-related characteristics, such as co-sleeping and signalling behaviours, on the concordance of objective and subjective sleep measures.

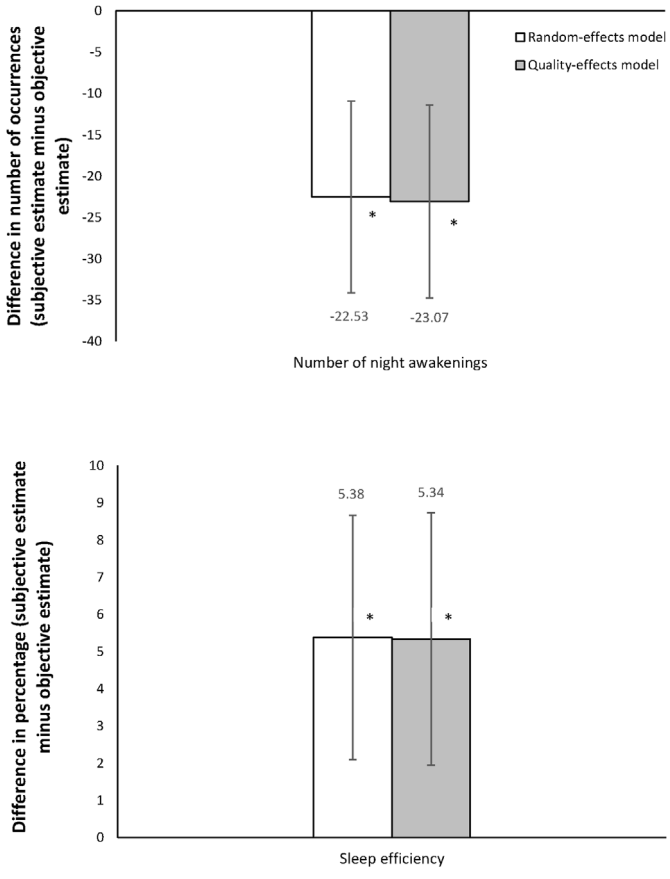

Fig. 3. Random-effects and quality-effects pooled differences between mean sleep parameters estimates from objective and subjective sleep measures, with 95% confidence intervals. Note. Asterisks indicate statistically significant effects.

To address the second aim of the meta-analysis, factors that may affect the concordance of objective and subjective sleep measures were explored, including the specific measures being compared. The findings revealed comparable mean differences between actigraphy-sleep diary and actigraphy-questionnaire subgroups; in contrast, with the exception of WASO, significantly stronger correlations were observed between actigraphy and sleep diaries compared to actigraphy and questionnaires. These results replicate those of previous TD research [23,25,29], further supporting the similar properties of objective and subjective sleep measures between these populations. Greater comparability of actigraphy and sleep diaries may be because these measures, unlike questionnaires, provide daily estimates of sleep parameters [19]. Whilst correlation analyses are sensitive to daily estimates, tests of mean differences are not, possibly underpinning the invariable mean differences between actigraphy-sleep diary and actigraphy-questionnaire comparisons. Future research should further explore the role of daily sleep parameter estimates on the concordance of objective and subjective sleep measures.

To the authors' knowledge, the concordance of PSG and sleep diaries has not yet been compared with that of actigraphy and sleep diaries in a sample of children. The non-significant differences between these method comparisons is surprising, given actigraphy and PSG produce disparate sleep parameter estimates in NDC samples [58,121–124]. However, the results align with previous adult research indicating similar mean differences and correlations between PSG-sleep diary and actigraphy-sleep diary comparisons across several sleep parameters [125,126].

Subgroup analyses for NDC diagnosis revealed significantly greater differences between objective and subjective TST means for children with ADHD compared to autistic children. In contrast, SOL mean differences did not differ between the groups. These findings may be attributable to high rates of co-sleeping in autistic children [35,36], which may improve the concordance of objective and subjective estimates of TST, but not SOL [42]. Co-sleeping has not been similarly documented in children with ADHD [127]. These findings provide preliminary support for the impact of specific NDC diagnoses, and associated characteristics, on the concordance of subjective and objective sleep measures. Given the novelty of this finding, replication studies are required.

Table 4
Summary of random-effects and quality-effects correlation meta-analyses for objective and subjective sleep parameter estimates. Pooled correlation coefficients and confidence intervals are presented, as well as prediction intervals and heterogeneity statistics (I^2 and Q) for random-effects models.

Sleep parameter (total pooled concordance estimates)	Concordance estimates per method comparison (NDC diagnosis for each estimate)				REM correlation *95% CI*	QEM correlation *95% CI*	*95% PI*[a]	I^{2a} *95% CI*	*Q*-test[a]	p value
	ACT + SD	ACT + Q	PSG + SD	PSG + Q						
Sleep onset latency (k = 9)	3 (3 A)	4 (1 DS, 3 WS)	1 (A)	1 (1 ADHD)	**0.66** ***0.49; 0.78***	**0.66** ***0.47; 0.78***	*0.02; 0.91*	72.9% *47.1%; 86.2%*	29.57	< 0.001
Sleep onset time (k = 2)	1 (1 ADHD)	1 (1 WS)	0	0	0.80 *-0.83; 1.00*	0.20 *-0.99; 1.00*	-	97.4% *93.6%; 99.0%*	39.01	< 0.001
Sleep offset time (k = 4)	3 (1 ADHD, 2 A)	1 (1 A)	0	0	**0.87** ***0.44; 0.98***	**0.87** ***0.17; 0.99***	*-0.99; 1.00*	95.8% *92.0%; 97.8%*	70.95	< 0.001
Total sleep time (k = 11)	5 (4 A, 1 RS)	6 (1 A, 2 DS, 3 WS)	0	0	**0.49** ***0.31; 0.64***	**0.51** ***0.30; 0.67***	*-0.22; 0.86*	82.0% *68.9%; 89.5%*	55.47	< 0.001
Sleep efficiency (k = 2)	2 (2 A)	0	0	0	0.13 *-0.23; 0.46*	0.08 *-0.29; 0.43*	-	45.4%	1.83	0.18
Wake after sleep onset (k = 5)	3 (2 A, 1 RS)	2 (1 A, 1 DS)	0	0	0.31 *-0.13; 0.65*	0.38 *-0.13; 0.72*	*-0.87; 0.96*	88.3% *75.4%; 94.5%*	34.31	<0.001
Number of night awakenings (k = 2)	0	2 (1 WS, 1 DS)	0	0	-0.06 *-0.31; 0.19*	-0.13 *-0.41; 0.16*	-	0.00%	0.88	0.35
Sleep-disordered breathing (k = 4)	0	0	0	4 (2 DS, 1 RS, 1 PKS)	**0.57** ***0.22; 0.79***	**0.68** ***0.34; 0.87***	*-0.76; 0.98*	55.1% *0.0%; 85.1%*	6.68	0.08

Abbreviations: A: autism. ACT: actigraphy. ADHD: attention-deficit hyperactivity disorder. CI: confidence intervals. DS: Down syndrome. NDC: neurodevelopmental condition. PI: prediction intervals. PKS: Pallister-Killian syndrome. PSG: polysomnography. Q: questionnaire. QEM: quality-effects model. REM: random-effects model. RS: Rett syndrome. SD: sleep diary. WS: Williams syndrome. Statistically significant mean differences ($p< 0.05$) are in bold. Statistically significant correlations ($p< 0.05$) are in bold.
[a] Values for random-effects models.

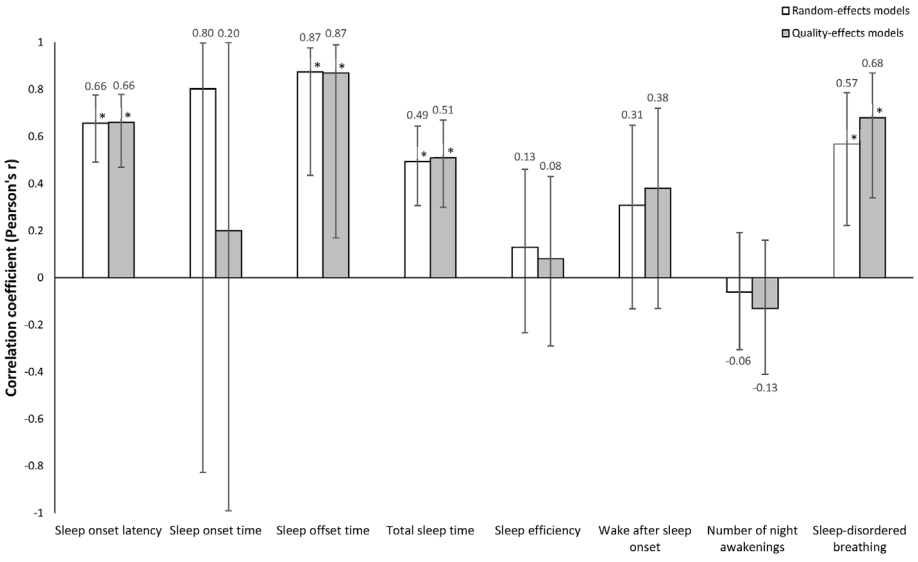

Fig. 4. Random-effects and quality-effects pooled correlations between sleep parameter estimates from objective and subjective sleep measures, with 95% confidence intervals. Note. Asterisks indicate statistically significant effects.

To address the third aim of the meta-analysis, the meta-analytic pooled effects were plotted against results from previous TD studies. These comparisons demonstrated broad similarity of concordance trends between TD and NDC populations, further supporting the similarity of subjective and objective sleep measurement properties across NDC and TD populations. Only two subtle discrepancies emerged, including larger actigraphy and questionnaire TST mean differences, and larger SDB correlations, for the NDC group. The discrepant TST mean differences may be explained by the instability of sleep in NDCs [49,54,56,128], decreasing the precision of global questionnaire sleep parameter estimates. Discrepancies in SDB correlations may be explained as the NDC studies explored rare genetic syndromes commonly associated with SDB [91,129,130]. The greater prevalence of SDB

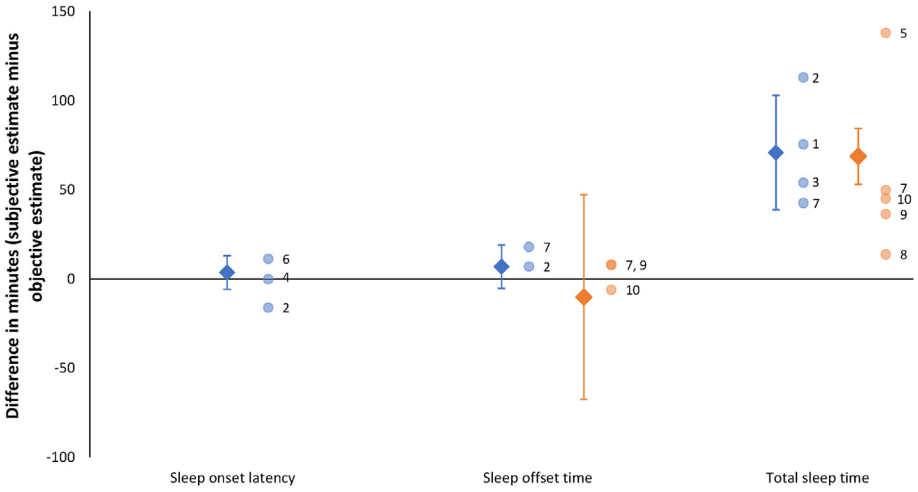

Fig. 5. Quality-effects pooled differences between mean sleep parameter estimates from objective and subjective sleep measures, with 95% confidence intervals, plotted for specific measurement comparisons and against results from previous robust TD studies (n≥ 75). Blue diamonds represent the pooled actigraphy-sleep diary mean differences, blue circles the previous TD actigraphy-sleep diary mean differences. Orange diamonds represent pooled actigraphy-questionnaire mean differences, orange circles the previous TD actigraphy-questionnaire mean differences. [1] Belanger et al. (2014) [107]. [2] Dayyat et al. (2011) [108]. [3] Dewald et al. (2012) [109]. [4] Gaina et al. (2005) [110]. [5] Holzhausen & Hagen (2021) [111]. [6] Hvolby et al. (2008) [53]. [7] Li et al. (2021) [28]. [8] Martinez et al. (2014) [112]. [9] Mazza et al. (2020) [79]. [10] Short et al. (2013) [113]. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

Fig. 6. Quality-effects pooled differences between mean sleep efficiency estimates from actigraphy and sleep diaries, with 95% confidence intervals, plotted against results from previous robust TD studies (n≥ 75). The blue diamond represents the pooled actigraphy-sleep diary mean difference, blue circles the previous TD actigraphy-sleep diary mean differences.
[1] Belanger et al. (2014) [107]. [2] Dewald et al. (2012) [109]. [3] Tse et al. (2020) [103]. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

symptoms in these populations has been suggested to impact questionnaire SDB estimates and strengthen associations PSG estimates [99]. These discrepancies indicate that NDC-related characteristics may impact the concordance of objective and subjective sleep measures, albeit for specific sleep parameters and method comparisons.

Researchers and clinicians should consider the meta-analytic findings during the interpretation of sleep parameter estimates and design of sleep assessments. For sleep parameters with divergent estimates, such as night awakening parameters, researchers and clinicians should consider the impact of measurement characteristics, for example the validity of measures for NDC populations or possible informant recall biases, when interpreting children's sleep data. Given the broad similarity of concordance trends between TD and NDC populations, guidance for sleep assessment design based on established TD research may be extended to NDC populations (e.g. [31, 120]). In particular, objective techniques are critical for robust estimates of sleep duration and night awakening parameters as these continuously monitor children's sleep and are resistant to error incurred by caregivers' limited awareness of night awakenings and recall biases [62,120]. In contrast, more convergent estimates of parameters associated with children's sleep schedules (e.g. sleep offset time) suggest single sleep measures may obtain sufficiently reliable estimates of these parameters. Where possible, researchers and clinicians should avoid the sole use of questionnaires as these are significantly less concordant with objective measures relative to sleep diaries, indicating greater effects of subjective biases on questionnaire data. In the context of NDC populations, the simultaneous use of both objective and subjective sleep measures is recommended. Objective measures are not free from error [120,131] and only subjective measures assess sleep-related behaviours and contexts that are necessary to fully understand sleep parameter data [31]. Additionally, objective measures are resource intensive and may be difficult for children with NDCs to tolerate [132]. In selecting measures for sleep assessments, researchers and clinicians should also consider the effect of NDC-related characteristics and threats to validity. For example, many questionnaires are not validated for children with specific NDCs and actigraphy data can be compromised by sleep-related movement disorders which are commonly observed in NDCs [133,134].

5. Limitations of this review

The conclusions of the meta-analysis should be interpreted in light of several limitations of the meta-analysed data. Firstly, mean differences and correlation coefficients have been criticised as poor estimators of absolute concordance [135,136]. However, these metrics have been pooled in previous concordance-related meta-analyses (e.g. Refs. [137–139]), and gold-standard tests of concordance such as Bland-Altman plots and intraclass correlations are rare throughout NDC research (e.g. Ref. [140]). Secondly, the meta-analysis only included subjective measures completed by caregivers, limiting the generalizability of the findings to self-report assessments. This was necessary to include younger children and

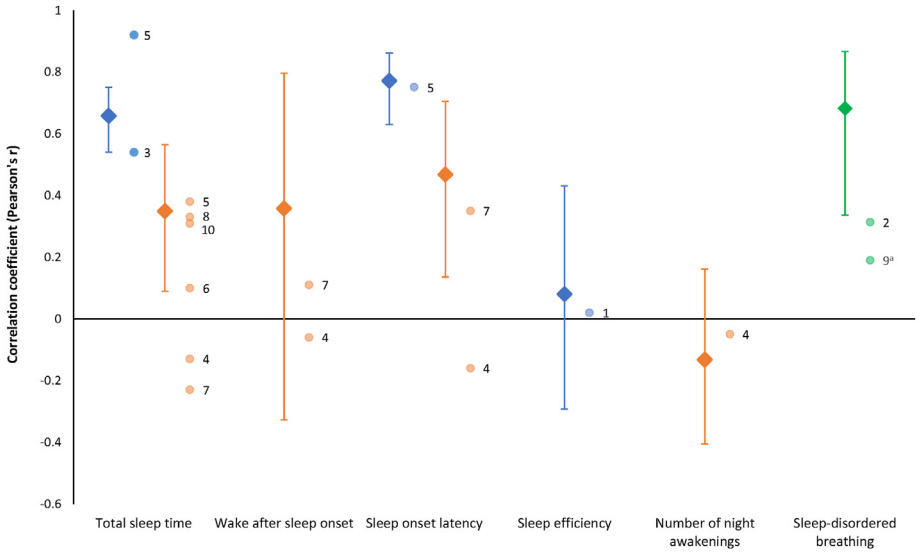

Fig. 7. Quality-effects pooled correlations between sleep parameter estimates from objective and subjective sleep measures, with 95% confidence intervals, plotted for specific measurement comparisons and against results from previous robust TD studies (n≥ 75). Blue diamonds represent the pooled actigraphy-sleep diary correlations, blue circles the previous TD actigraphy-sleep diary correlations. Orange diamonds represent pooled actigraphy-questionnaire correlations, orange circles the previous TD actigraphy-questionnaire correlations. Green diamonds represent pooled PSG-questionnaire correlations, green circles the TD PSG-questionnaire correlations. [1] Belanger et al. (2014) [107]. [2] Bertran et al. (2015) [114]. [3] Chang et al. (2011) [115]. [4] Duraccio et al. (2015) [116]. [5] Gaina et al. (2005) [110]. [6] Gunn et al. (2019) [117]. [7] Holley et al. (2010) [118]. [8] Martinez et al. (2014) [112]. [9] Masoud et al. (2022) [119]. [10] Mazza et al. (2020) [79]. [a] Spearman's rank correlation converted to Pearson's r correlation following the same formula as the meta-analyses. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

children with intellectual disabilities and social communication differences for whom self-report may be less feasible [21,141]. Thirdly, by grouping objective measures into single meta-analytic models, actigraphy- and PSG-specific concordance trends may have been obscured. However, this grouping approach was necessitated by the limited available studies, and the subgroup analyses refuted differing concordance between actigraphy-sleep diary, and PSG-sleep diary comparisons. Finally, the scarcity of data limited many meta-analytic models to two or three studies, reducing the power of these analyses. Limited data was largely attributable to poor reporting of sleep parameter means and correlations, especially for sleep diaries, accounting for 166/415 exclusions during full-text screening. This highlights poor reporting practices as a barrier to understanding measurement properties of sleep measures in NDC populations, which future research should rectify.

5.1. Methodological considerations

Assessment of study quality revealed further limitations in reporting practices throughout NDC sleep research. In particular, studies utilizing actigraphy and sleep diaries often did not specify how many nights of data were included in the analyses, did not define sleep parameters or outline how these were calculated, and omitted how missing data and artefacts were handled. This information is crucial to gauge the rigour of sleep parameter estimates. Standardized reporting practices have been previously outlined for actigraphy [142,143], addressing the issues above, and basic reporting guidelines are provided for PSG and sleep diaries below. Additionally, aside a few notable exceptions [96,98,99], studies did not specify the timescale within which questionnaires and objective sleep measures were completed. Therefore, it is unclear whether questionnaire and objective sleep parameter estimates pertained to the same nights of sleep. This information is necessary to ensure subjective and objective data appropriately correspond to each other, especially given the high night-to-night variability of sleep amongst children with NDCs [49,54,56,128].

6. Conclusion

This meta-analysis is the first to directly address the concordance of objective and subjective sleep parameter estimates in children with NDCs, and explore factors that impact this concordance. The findings reveal variation in concordance between sleep parameters and specific method comparisons, broadly emulating previous TD research, and provide preliminary insights into the impact of NDC diagnoses on concordance. This review may inform the design of sleep assessments and interpretation of sleep data for NDC populations, throughout research and clinical settings, however the findings require replication in future research for which this review has highlighted the necessity of improved reporting practices.

7. Practice points

- For sleep parameters with divergent objective and subjective estimates, researchers and clinicians should proactively consider the properties of objective and subjective measures when interpreting these parameter estimates. For sleep parameters with more convergent objective and subjective estimates, these estimates may be reliably interpreted at face value.
- Researchers and clinicians should consider objective and subjective sleep measures as complimentary, and utilize both where possible, to obtain robust and comprehensive descriptions of children's sleep parameters and behaviours.
- Researchers and clinicians should consider threats to validity when selecting measures for sleep assessments (e.g. sleep-related movement disorders or poor psychometric properties).
- If only one sleep measure can be feasibly implemented, researchers and clinicians should avoid questionnaire techniques and prioritize objective measures as these are posited to obtain more rigorous estimates of night awakening and sleep duration parameters.

8. Research agenda

- Objective and subjective sleep parameter data should be systematically reported throughout future publications, whether through descriptive statistics or open access to raw datasets.
- Future research should extend the current findings with gold-standard tests of concordance (e.g. Bland-Altman plots or intraclass correlations), or provide the data necessary to conduct these tests.
- Studies should directly examine the influence of NDC-related characteristics, such as co-sleeping and signalling behaviours, on the concordance of objective and subjective measures of sleep.
- Future research should improve the transparency of data collection and analytic procedure for all sleep measures, following reporting criteria below:
    - o Polysomnography:
    - Specify the number of nights of data analysed for each participant, and whether an adaptation night was employed.
    - Specify polysomnography electrode derivations (e.g. EEG, EMG, EOG derivations).
    - Specify the guidelines used to score sleep parameters (e.g. American Academy of Sleep Medicine criteria, or Rechtschaffen and Kales criteria), and guidelines used to acquire PSG data.
    - Specify whether a single or multiple individuals scored the sleep parameters, and whether reliability was checked between multiple individuals' scores.
    - o Actigraphy
    - Refer to checklists by Meltzer et al. (2012) [142] and Scoch et al. (2021) [143].
    - o Sleep diaries
    - Specify the number of nights of data analysed for each participant
    - Provide a definition for each sleep parameter assessed by the diary, and outline any calculation procedures used to estimate sleep parameters (e.g. total sleep time = minutes between sleep onset time and sleep offset time, minus minutes awake after sleep onset).
    - Specify who completed the diary, and whether multiple individuals completed the diary.
    - o Questionnaires
    - Specify timescale within which questionnaires and objective sleep measures are completed (e.g. on same night, questionnaire completed 2 days prior to actigraphy assessment).

Author contributions

Study design and concept: ROS, CR, SB, AB. Acquisition of data: ROS, AH. Analysis of data: ROS. Interpretation of data: ROS, CR, SB, AB. Drafting the manuscript: ROS, CR, SB, AB.

Declaration of competing interest

The authors declare no conflict of interest in relation to this work.

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Appendix A. Supplementary data

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* The 10 most important references are denoted by an asterisk