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REVIEW

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Behavioural sleep problems in children and adults with intellectual disabilities: An integrative literature review

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Abstract

Background: People with intellectual disabilities are more likely to experience sleep problems, which can affect quality of life, physical health, mental health and well-being.

Methods: An integrative literature review was conducted to investigate what is known about behavioural sleep disturbances in people with an intellectual disability. The search used the following databases: Scopus, PsycInfo and Cinahl, to find papers published since 2015.

Results: Within intellectual disability research, sleep appears as a common issue due to its high prevalence, negative relationships with an individual's physical and mental health, their quality of life, and impact of sleep problems on family or carers. The growing evidence base appears to support the use of behavioural, lifestyle and pharmacological interventions to improve sleep in people with an intellectual disability.

Conclusion: A wide array of literature provides evidence that people with intellectual disabilities are affected by and need support with their sleep.

KEYWORDS
intellectual disability, learning disabilities, sleep

1 | INTRODUCTION

People with intellectual disabilities are more likely to have sleep problems than people who do not have an intellectual disability (Bassell et al., 2015; Esbensen et al., 2018; Surtees et al., 2018). The main types of sleep problems in people with intellectual disabilities are behavioural sleep problems or problems as a result of sleep disordered breathing (Horne et al., 2019). Behavioural sleep problems can include sleeping for less time, finding it harder to initiate and stay asleep, waking early, feeling sleepy during the day, and feeling anxious about bedtimes. These sleep problems impact sleep efficacy, sleep quality and sleep quantity.

Sleep has been linked to cognitive performance on tasks requiring memory consolidation and learning (Gui et al., 2017), processing speed (Lim et al., 2017), procedural memory used when unconsciously applying cognitive and motor skills (Rångtell et al., 2017), attention (Lehto & Uusitalo-Malmivaara, 2014), decision making and creativity (Seeley et al., 2016). In addition, correlations between physical health conditions and impaired sleep have been reported in the literature. Health conditions include metabolic and cardiovascular diseases (Hege et al., 2018), inflammatory diseases (Ali & Orr, 2014), infectious diseases (Ibarra-Coronado et al., 2015), cancer (Hu et al., 2013; Ji et al., 2017) and depression (Paunio et al., 2015). In addition, sleep-reduced beta-amyloid has been identified in the development and progression of Alzheimer's disease (Vanderheyden et al., 2018). However, there is a difference between correlations and causations and future research is needed to examine if changes in sleep patterns could act as a marker to screen for and manage additional health concerns (Esbensen et al., 2016).

Sleep problems can affect health and well-being as well as quality of life of the individual and their families (Mori et al., 2019). It is therefore important to understand what is known about sleep in people with intellectual disabilities. Furthermore, as part of a holistic assessment and care plan relating to health and well-being, it is important to consider promoting good sleep. Therefore, the aim of this paper is to provide a synthesis of the most up-to-date evidence relating to sleep in people with intellectual disabilities. This paper takes a broad view across the available evidence, to consider the impact of poor sleep on health, well-being and quality of life, and the opportunity for lifestyle interventions to improve sleep, because research studies often tend to focus on one aspect.

2 | METHODS

An integrative review was used to develop a breadth of understanding of the area. A limitation of integrating a wide range of research designs adds complexity to the appraisal framework used. Therefore, as Webb and Roe (2007) explain, many reviewers who have published integrative reviews do not undertake any appraisal of the research. However, thoughts regarding the rigour of the research studies are included in the summary of findings table and within the summary below.

The literature presented here was located using the following databases: Scopus, PsycInfo and CINAHL. Database searches used a combination of the word 'sleep' with any one of the followings terms: 'Learning Dis*', 'Intellectual* Dis*', 'Mental* retar*', 'Down* Syndrome', 'Global dev* delay', OR 'Intellectual* retar*'. All papers reviewed had both 'sleep' AND one of these terms in the title and/or in the abstract of the article.

All peer-reviewed papers written in English and published between January 2015 and May 2022 were included. Due to the plethora of research available and a desiderate need to include the most contemporary research to understand the state of the art, only papers published after 2015 were included. Therefore, the most up-to-date research papers were reviewed to gain insight into the most recent evidence relating to behavioural sleep disorders in people with intellectual disabilities.

The exclusion criteria were as follows: participants reported as having a specific learning difficulty such as dyslexia, dyspraxia or dyscalculia, having a neuro-developmental disability such as autism or attention deficit hyperactivity disorder but who did not have an intellectual disability. Furthermore, studies were excluded that solely investigated sleep disordered breathing, epilepsy syndromes linked to nocturnal seizures, narcolepsy or restless leg syndrome as these did not meet the inclusion criteria of a behavioural sleep disorder. Finally, pharmacological interventions and studies focusing on children under the age of 5 were omitted. A two-stage approach was used to review the relevance of the literature following the PRISMA guidelines. Initially, the titles and abstracts were screened for relevance ($n = 2117$), followed by reviewing full text articles and papers ($n = 204$). The full text was reviewed for papers without an abstract to ascertain relevance. A total of 99 articles met the inclusion criteria (see Appendix 1).

A thematic analysis was used to develop prominent themes and a summary of findings (Webb & Roe, 2007). Thematic analysis is deemed appropriate to capture meaning and identify prominent themes within the literature from across a broad spectrum of research methodologies which includes papers that have explicit and demonstratable outcome variables to research with more semantic findings or implicit meaning.

The analysis involved re-reading the papers to enable familiarisation with the data, which allowed for a more inductive approach to inform the codes and themes. Thematic analysis can be either inductive or deductive, however, an inductive analysis enabled the researcher to code the data without trying to fit into a pre-existing frame allowing the analysis to be more firmly linked to the data itself (Braun & Clarke, 2006). The data was coded at a semantic level to identify initial themes and connections between papers and then themes were reviewed and papers re-coded, finally the three themes that were defined will be discussed (Braun & Clarke, 2022, pp. 70–71).

3 | FINDINGS

Three main themes emerged from the literature: (1) people with intellectual disabilities are more likely to have sleep problems than people who do not have an intellectual disability; (2) there is a relationship between poor sleep and physical health, mental health, and people's quality of life and poor sleep also negatively impacts their family or carers; and (3) behavioural, lifestyle and pharmacological interventions have been used to improve sleep in people with an intellectual disability.

3.1 | Relationship between intellectual disability and sleep quantity or sleep quality/efficacy

Forty-one papers addressed the relationship between having intellectual disabilities and sleep. Three sub-themes emerged within this theme, including evidence on the type of sleep problems, prevalence rates and assessment of sleep problems and the aetiology of sleep problems.

3.1.1 | Type of sleep problems

A meta-analysis conducted by Surtees et al. (2018) found a decreased sleep time of 18 minutes per night, as well as poorer sleep quality in people with intellectual disabilities. However, the difference found in sleep time was only significant when those with specific genetic syndromes were included in the analysis. Commonly reported behavioural sleep disturbances in people with intellectual disabilities include

problems initiating sleep, bedtime resistance, problems maintaining sleep, sleep anxiety, daytime sleepiness and waking up too early (Köse et al., 2017).

3.1.2 | Prevalence

Papers reviewed showed that persistent sleep problems appear to occur more frequently in children and adults with intellectual disabilities than in neuro-typical individuals (Bassell et al., 2015; Surtees et al., 2018).

Prevalence rates of behavioural sleep problems in people with intellectual disabilities range from 22.7% to 87.5% (Bishop-Fitzpatrick & Rubenstein, 2019; Esbensen et al., 2018). This large variance is largely explained by the assessment tools used and characteristics of individuals within the sample selected (e.g., specific syndromes and additional diagnoses). The validity and reliability of assessment tools need to be considered when interpreting data on prevalence rates of sleep problems in people with an intellectual disability. Pictorial rating scales for subjective markers of sleep have been shown to improve the utility of sleep questionnaires (Hill et al., 2020), although the majority of studies reported in this paper use questionnaires completed by family or carers. Furthermore, the wide variance in prevalence rates can be explained by the characteristics of individuals within the sample selected. Although outside of the scope of this review, previously published reviews on prevalence rates of insomnia have also questioned how communication difficulties can affect explanation of personal sleep experiences needed for diagnostic criteria, leading to a reliance on proxy reported measures (van de Wouw et al., 2012). More recently, the wide variation has been explained by van den Broek et al. (2022) who argues that difficulties in gaging prevalence rates is further complicated by a lack of a definitive definition of what is insomnia and what is a behavioural sleep problem that does not meet the diagnostic cut-off for insomnia. The variety of definitions used by researchers highlights how prevalence rates vary given that some include consideration of the clinical significance and impact on daytime functioning while others do not (van den Broek et al., 2022).

In addition, specific genetic conditions have been linked to a predisposition for sleep disturbance. Genetic conditions linked to sleep disturbances include people with Down's Syndrome with and without obstructive sleep apnoea (Al-Khudhairy et al., 2019; Bassell et al., 2015; Chawla, Howard, et al., 2021; Esbensen et al., 2018; Heubi et al., 2021; Stores, 2019; Worley et al., 2015), Tuberous sclerosis complex (Trickett et al., 2018), Mowat–Wilson Syndrome (Di Pisa et al., 2019; Evans et al., 2016), Cornelia de Lange Syndrome (Zambrelli et al., 2016), Angelman syndrome (den Bakker et al., 2018; den Bestern et al., 2021; Levin et al., 2022; Pearson et al., 2018; Trickett et al., 2018), Smith-Magenis syndrome (Mullegama et al., 2015; Rive Le Gouard et al., 2021; Smith et al., 2019; Trickett et al., 2020), Fragile X syndrome (Budimirovic et al., 2022; Mullegama et al., 2015). In addition to rare conditions which are linked to sleep disturbances such as Christianson Syndrome (Gruber et al., 2022), Dup15q Syndrome (Barstein et al., 2021), Pallister-Killian Syndrome (Fetta et al., 2022), Phelan-McDermid Syndrome and SYNGAP1-ID (Smith-Hicks et al., 2021), MBD5-associated neurodevelopmental disorder (Gandhi et al., 2021), Arboleda-Tham syndrome (Smith & Harris, 2021), Bosch-Boonstra-Schaaf optic atrophy syndrome (Rech et al., 2020) and emerging genetic variants including microdeletion syndrome at 2q22.1q22.3 and de novo DDX3X variants (Chen et al., 2020; Verhoeven et al., 2020). This shows that a wide array of specific intellectual disability syndromes are associated with sleep problems, although variation from night to night and within syndrome groupings is found (Trickett et al., 2019).

3.1.3 | Aetiological factors

Varying genetic mechanisms have been posed to explain the association between sleep problems and specific syndromes, this includes anatomical, physiological and neurological differences such as brain atrophy (Agar et al., 2021; Lee et al., 2020). For example, anatomical alterations are indicated by changes in alpha oscillations on electroencephalogram recordings in Down's syndrome and gamma oscillation in Angelman syndrome (Den Bakker et al., 2018; Ruiz-Mejias, 2019). Phase advanced sleep patterns and reduced sleep efficacy in people with Smith Magenis syndrome have been linked to melatonin and RA/1-related dysregulation of CLOCK genes (Smith et al., 2019). However, conditions such as constipation, epilepsy, obstructive sleep apnoea, gastro-oesophageal reflux disease, or conditions that cause pain often have a negative impact on sleep. Therefore, it is currently unclear to what extent sleep disturbances are related to intellectual disability syndromes or as a result of comorbid conditions, given the high prevalence rates of comorbid conditions in people with specific syndromes (Bassell et al., 2015; Chow et al., 2020; Esbensen et al., 2016; Evans et al., 2016; Fernandez et al., 2017; Gilbertson et al., 2021; Zambrelli et al., 2016).

As already discussed, several aetiological factors can cause sleep disturbances, including pathophysiological factors and physical and psychological health comorbidities (Stores, 2019). In addition, developmental, social, environmental and behavioural factors can also impact sleep. Developmental factors that can affect sleep in people with intellectual disabilities include physiological changes associated with maturation, with more sleep problems reported in younger children and in adults over 50 years (Böhmer et al., 2020). Environmental factors include sharing a bedroom or co-sleeping, which have frequently been reported to disrupt sleep (Köse et al., 2017). However, extrapolating conclusions from this is problematic due to studies lacking evidence of the reasons why parents feel the need for close proximity during the night-time (e.g., increased concern over the health state of their child). Furthermore, in children, parental knowledge of sleep and TV, phone or laptop screen time being limited to 3 h and in adults daily light exposure appear to moderate the relationship with nocturnal sleep duration and sleep problems (Böhmer et al., 2021; Halstead et al., 2021; Saletovic et al., 2021). This highlights the

influence of environmental and lifestyle factors which can be used as part of sleep hygiene interventions to improve sleep in people with intellectual disabilities. In application of this knowledge researchers and clinicians would need to consider participants' age, genetic syndromes and how these impact behavioural phenotypes, in addition to individual differences, given that each person has unique environmental, social and psychological variables (Pearson et al., 2018).

3.2 | Relationship between the sleep of people with intellectual disabilities on their cognition, emotional and psychological well-being, neurodevelopment, behaviour, physical health and quality of life

Forty-six papers addressed the relationship between sleep on measures of quality of life, cognition, emotional and psychological well-being, behaviour, neurodevelopment and physical well-being. The papers show that sleep problems are negatively correlated with people with intellectual disabilities cognitive, functional and social skills. In people with intellectual disabilities sleep has been linked to impulsivity, learning, recall, the ability to make plans and behaviours that challenge. However, it should be acknowledged that all the studies reported here present correlations and not causations; thus, bidirectional causality and confounding variables cannot be ruled out. For example, a bidirectional effect is likely to be present with sleep impacting and being impacted by aspects of temperament, such as effortful control and inhibitory control (Lukowski & Milojevich, 2017). Moreover, in people with intellectual disabilities sleep can improve mood and a preference for a more energetic day, and daily activity has been perceived as a determinant of sleep quality (Powers et al., 2021). Confounding variables include pain, prescribed medications and other physical or mental health conditions potentially impacting both sleep and behaviour (Poppes et al., 2016).

3.2.1 | Cognitive and functional ability

Poor sleep is linked to further deterioration in learning and applying skills and functional abilities that are needed for life and independence. For example, the functional daily life scores, fine dexterity skills and physical functioning of children with Down's syndrome negatively correlated with a sleep habit questionnaire, questionnaire on sleep disorders and objective sleep markers (Bertapelli et al., 2022; Chen & Ringenbach, 2019, 2022; Churchill et al., 2015). Support for this argument comes from evidence that disorders linked to initiating and maintaining sleep in people with Down's syndrome is associated with their visual-motor integration abilities (Fucà et al., 2021). Other significant associations were also reported in the accomplishment of responsibilities, relationships, community life, school, mealtimes, fitness and personal care (Churchill et al., 2015). However, Chawla, Bernard, et al. (2021) found no relationship between sleep and cognition, functional ability or behaviour. Suggesting that these problems are not solely driven by sleep issues, with confounding variables such as physical problems and pain potentially affecting both sleep and behaviour (Chow et al., 2020; Esbensen et al., 2021).

Sleep also effects cognitive ability. Evidence supports that sleep has a role in consolidating learning, restoring cells and synaptic plasticity (Trickett et al., 2018). People with Down's syndrome that have poor sleep have specific difficulties with expressive language, including word retention, recall and syntax (Ashworth et al., 2017; Edgin et al., 2015). However, contrary to these findings performance on word recall was not found to be related to long-term retention (Sakhon et al., 2018) and consistency in day-to-day sleep patterns has been negatively correlated with verbal and scene recall (Lovos et al., 2021). Therefore, the relationship between sleep and cognitive ability is likely to be more complex with multiple factors impacting the outcome.

The sleep phase involving rapid eye movement (REM) has been linked to memory consolidation and stabilisation. EEG recordings show that people with Down's syndrome spend less time in REM sleep, with evidence that brief intervals in REM sleep impairs learning (Spanò et al., 2018). Furthermore, people with Williams syndrome, who are also known to have reduced REM sleep, show improved procedural skill learning when task acquisition is followed by sleep (Joyce et al., 2019). Although Spanò et al. (2018) acknowledge that sleep apnoea is related to REM disruptions, information about sleep disordered breathing of the participants is not presented in the published paper. In comparison, Joyce et al. (2019) stipulated that they excluded people undergoing treatment for obstructive sleep apnoea, however no specific analysis of sleep apnoea was completed. Therefore, further research is required to corroborate if the results are limited to those with undiagnosed OSA or to people with Williams syndrome.

Executive functioning includes planning, attention, transitioning between tasks, working memory, inhibitory control and self-regulation. People with Down's syndrome with longer length of night awakenings performed worse on tasks involving episodic memory, executive functioning, motor planning and coordination (Codyy et al., 2020). Furthermore, parental reports of executive functioning abilities of their children with Down's syndrome have been linked to restless sleep. However, this was not confirmed by neuropsychological assessments completed on the same children (Esbensen & Hoffman, 2018a, 2018b). In addition, conflicting results have been reported regarding the impact of sleep quality on focussed attention in people with Down's syndrome and Williams syndrome (Ashworth et al., 2015).

3.2.2 | Neurodevelopmental, neurodegenerative, mental health and physical health

Conflicting evidence has emerged for an association between anxiety, depression and sleep. Bond et al. (2020) found that anxiety and depression were associated with sleep problems in adults with an intellectual disability who are aged over 40. However, Esbensen et al. (2018) found that correlations from parent-reported sleep disturbances with increased conduct problems and anxious behaviours were

no longer significant when sleep was objectively assessed using actigraphy (a non-invasive sensor typically worn around the wrist to measure rest cycles). Furthermore, not obtaining the recommended amount of sleep per day did not correlate with anxiety or depression in a recent study conducted by Whitney et al. (2019). These finding could be explained by outcome measures assessing different constructs. With dichotomous variables comparing the child's typical sleep pattern and sleep duration to the norm for their age being less effective to predict relationships between mental well-being and sleep, compared to parental reports that compare the night's sleep against the individual's usual sleep pattern. Therefore, it is important to assess individualised markers of sleep quality as opposed to a sole measure of sleep quantity in future research investigating the negative effect of sleep problems on mental well-being and quality of life.

Sleep disturbance has also been reported as a precursor to a diagnosis of psychiatric problems and dementia in people with Kleefstra syndrome and Down's syndrome (Dekker et al., 2018; Fleming et al., 2021; Vermeulen et al., 2017). The relationship between sleep and dementia in people with Down's syndrome is complex and thought to be linked to a destructive feedback loop where a lack of sleep adequacy increases beta amyloid burden in the striatal (the subcortical basal ganglia in the forebrain), the beta amyloid aggregation then disrupts the sleep cycle (Codyy et al., 2020).

People with additional diagnoses have increased likelihood of sleep issues, particularly those with autism (Ballester et al., 2019; Bishop-Fitzpatrick & Rubenstein, 2019; Elrod & Hood, 2015; Gunes et al., 2019; Köse et al., 2017), attention deficit hyperactivity disorder (Bakke et al., 2021; Chan et al., 2019; Esbensen, Vincent, et al., 2022), dementia or comorbid mental health conditions (Esbensen et al., 2018; Köse et al., 2017), comorbid physical health conditions (Esbensen et al., 2018), physical health conditions known to cause pain (Esbensen et al., 2021; Trickett et al., 2018) and sensory problems including hearing and visual disabilities (Stores, 2019).

3.2.3 | Behaviour

Sleep problems have been described as one of the most influential factors on behaviour problems (Leader et al., 2021). Sleep disturbances have been positively correlated with generalised behaviour problems and asocial behaviour problems. Recent literature shows correlations between sleep disturbance and self-injury, stereotypical, aggressive or destructive behaviour, socially offensiveness, withdrawal, inattention, attention deficit and hyperactivity scores and uncooperative behaviours (Cohen & Tsiouris, 2020; Esbensen et al., 2016; Evans et al., 2016; Kelmanson, 2017; Kurzius-Spencer et al., 2018; Poppes et al., 2016).

3.2.4 | Quality of life

Sleep and excessive daytime somnolence have been shown to be a prominent factor in quality-of-life scores, physical health and positive emotions (Downs et al., 2022; Jacoby et al., 2022; Reddihough et al., 2021). Furthermore, parental physical and emotional wellbeing are also negatively impacted when the child has frequent disturbed sleep. Cross-sectional and longitudinal effects have been found on reduced parental wellbeing, with greater deterioration following sleep disturbances that extended over a prolonged period of time (Choi et al., 2019; Esbensen et al., 2021; Mori et al., 2019).

3.3 | Impact of behavioural or lifestyle interventions on sleep

Twelve papers addressed the impact of behavioural or lifestyle interventions on sleep. A number of non-pharmacological treatments for sleep are found in the literature.

3.3.1 | Types of interventions: Behavioural training programs

Behavioural training programs aim to increase behaviours which promote sleep and reduce behaviours which interfere with sleep. According to the National Institute of Health and Care Excellence (2015) and (2022), behavioural therapy and full assessment and management of the causes of sleep problems (e.g., pain) should supersede medication for people with intellectual disabilities (with behaviours that challenge). Clinicians typically support non-pharmacological interventions as the first line of treatment (Bruni et al., 2018). In addition, many parents prefer behavioural sleep interventions due to potential side effects or interactions from polypharmacy (Esbensen et al., 2016; Sutton et al., 2019).

Several different behavioural approaches have been implemented with people with intellectual disabilities. Behavioural sleep interventions can include advising family/carers regarding sleep hygiene, reinforcement of behaviours that support sleep and extinction (a process of no longer reinforcing behaviours that have a negative impact on sleep) and changes to the environment depicted in sleep ecology models (Kirkpatrick et al., 2019).

In addition to behavioural interventions already discussed, research has looked at alternative approaches such as biofeedback using EEG recordings (Ogoshi et al., 2018), light therapy (Shanahan et al., 2019), and parents have experimented with approaches such as using weighted blankets (Sutton et al., 2019). These alternative approaches currently lack a robust evidence base and raise concerns regarding safety of using equipment when it is not closely monitored (Sutton et al., 2019). Precaution with using weighted blankets to improve sleep is particularly important with infants and young children, people with chronic circulatory or respiratory concerns including obstructed sleep apnoea and when the person lacks the dexterity to remove the blanket (Noyed, 2022). Therefore, new approaches to improve sleep in people with intellectual disabilities should be assessed for safety as well as efficacy, to ensure that professionals are guiding parents and carers using rigorous evidence-based practices.

3.3.2 | Efficacy of behavioural sleep programs

Positive effects of behavioural sleep interventions on sleep quality, quantity or sleep management are reported in 76% ($n = 68$) of studies and a further 17% ($n = 15$) report mixed results (Spruyt & Curfs, 2015). However, it is difficult to extrapolate these results for three reasons. First, generalisability of these results is limited given that most studies included within the review were case studies that had adapted interventions to the individuals. Lack of adherence to a systematic approach thus makes it problematic to reliably extrapolate results to the wider population. Second, most behavioural interventions used a combination of approaches including sleep ecology, sleep hygiene or operant conditioning. Limited research is available that investigates the efficacy of such interventions in isolation (Sutton et al., 2019), and an analysis of specific interventions is needed to make evidence-based recommendations. Finally, interventions may not be generalisable due to differences in the cause of the sleep problem. As mentioned previously sleep is more likely to be disrupted in those with specific syndromes or with comorbidities. Therefore, an individual with an intellectual disability who is in pain or who has different anatomical and physiological differences to their peers may not have the same response to the behavioural intervention used within a research study.

Parent training programs, either group-based or individual sessions, have consistently proven to be effective in managing behavioural sleep problems. A manualised sleep training program run over 5 weeks improved sleep outcomes, similarly the control involving a 5 week program that addressed topics of relevance including a discussion of sleep in week two also resulted in improvements in sleep, resulting in a lack of statistical difference between the intervention and the control on actigraph or parental report measures (Esbensen, Hoffman, et al., 2022). The improvements in sleep scores in both groups suggest that it can be sufficient to provide basic information on sleep management to individuals. However, standardised pamphlets have not been found successful (Kirkpatrick et al., 2019; Spruyt & Curfs, 2015; Stuttard et al., 2015). The positive effect of training programs is repeatedly reported in systematic reviews, despite the differences in methodologies used by the studies included. Significant gains in sleep quality and/or quantity have been found in single case experimental designs and pretest-posttest designs, using outcome data from sleep diaries or actigraphy (Esbensen, Hoffman, et al., 2022; Priday et al., 2017). Studies suggest that the improvements are sustained long-term, however this conclusion is based on a limited number of longitudinal studies with follow-up data available (Priday et al., 2017). In a systematic review of behavioural interventions, social validity from parents' feedback was promising, indicating that they found the intervention to be acceptable, relevant, and effective (Kirkpatrick et al., 2019). However, there was a lack of information regarding parents' adherence to the training, which makes it impossible to judge treatment fidelity. In contrast, Esbensen, Hoffman, et al. (2022) included treatment fidelity as an outcome measure and found high parental engagement and attendance at sessions and low drop-out rates. Thus, supporting the claim that parents are motivated and able to engage in behavioural sleep interventions.

4 | DISCUSSION

The aim of the review is to provide a synthesis of the most up-to-date evidence relating to sleep in people with intellectual disabilities. This review presents 99 research papers related to behavioural sleep problems in people with intellectual disabilities. The array of papers published highlights that this is an important, unmet and current need for people with intellectual disabilities and their families or carers. The plethora of research that is being conducted aims to advance knowledge of sleep problems and support people with intellectual disabilities to sleep well.

The key themes found include that the sleep–wake cycle is often disrupted in people with intellectual disabilities. Second, that there is a relationship between sleep and people with intellectual disabilities ability, mood and behaviour as well as quality of life. Behavioural interventions have been somewhat effective in treating behavioural sleep problems and further research could offer the opportunity to maximise the efficacy of behavioural interventions. These themes will now be discussed in more depth.

A vast majority of the papers have focussed on identifying sleep problems in people with intellectual disability. A wide array of specific intellectual disability syndromes are associated with sleep problems. Sleep problems in people with intellectual disabilities are associated with their quality of life, functional and cognitive abilities, social abilities, as well as their mood and mental well-being. Research has enabled greater understanding of how specific intellectual disability syndromes are associated with sleep problems and the importance of optimising quality and sufficient quantity of sleep. However, is not able to offer a defining feature that can be used for diagnostic purposes due to the individual differences across syndromes in relation to the sleep problems experienced.

Fewer research studies have focussed on interventions to improve sleep. This was partly due to exclusion of papers looking at pharmacological interventions to promote sleep. Many of the papers on pharmacological interventions specifically focussed on melatonin which has been used with some success in people with intellectual disabilities (Shanahan et al., 2019). However, pharmacological interventions should only be used if sleep hygiene and behavioural interventions are insufficient to improve sleep to an acceptable level for the individual (British National Formulary 2023) and thus a review of behavioural interventions was warranted. Behavioural interventions using a combination of sleep hygiene, sleep ecology and reinforcement appear to be effective. This finding supports previous systematic reviews on behavioural interventions for sleep problems (Priday et al., 2017) and improvements in sleep quality and quantity (Surtees et al., 2018). Alternative interventions that can be used in parallel with education programs on sleep hygiene are also emerging and further research would enable the credibility and reliability of these interventions to be evaluated.

The quality of the research was not systematically reviewed as part of the inclusion criteria. Despite evidence that several issues were present in the research conducted. Methodological weaknesses within studies include use of subjective and proxy reported measures of sleep without objective measures to corroborate results and provide a fuller understanding of sleep parameters. Second, the majority of studies that looked at relationships between specific syndromes and sleep or sleep and aspects of mental wellbeing presented correlations and not causations, and the lack of a control group was often highlighted as a weakness within the studies. Correlations between sleep problems and comorbid conditions are unable to provide evidence on the causation or direction of relationships. Thus, directional causality and confounding variables cannot be ruled out. For example, a bidirectional effect is likely to be present with sleep impacting and being impacted by aspects of temperament, such as effortful control and inhibitory control (Lukowski & Milojevich, 2017). A bidirectional relationship between sleep and comorbid conditions, quality of life, activity and social engagement are thought to exist, but further research is needed on the extent to which these influencing factors impact disturbed sleep. Confounding variables include pain, prescribed medications and other physical or mental health conditions, which potentially impact both sleep and behaviour (Poppes et al., 2016). Furthermore, for cross-sectional or cohort studies data analysis predominantly assessed changes in group means over time or across conditions for sleep duration and quality of sleep. However, as Fullagar and Bartlett (2016) point out research and evaluations on the effectiveness of sleep interventions, should include data at the individual level due to the variability of sleep across participants and variability in what they perceive as a 'good night' sleep.

Further research is therefore needed to understand the perspectives of people with an intellectual disability relating to their personal subjective experience of sleep and the interventions used to promote good sleep. Furthermore, in future studies, the use of objective data measurement is required to further examine how physiological differences across syndromes effect the usefulness of specific interventions to improve sleep quality and quantity, in order to identify which interventions are more likely to be effective for each person with an intellectual disability.

Examples from the literature that can be used to highlight how subjective and objective markers can be used in parallel include Esbensen, Hoffman, et al. (2022) and Trickett et al. (2020), who used combinations of sleep questionnaires, sleep diaries and actigraphy. Furthermore, both of these studies included control groups for the purpose of comparison, and to increase rigour Esbensen, Hoffman, et al. (2022) used random allocation of participants to the intervention or control group.

The evidence available has implications for practice as well as for families and carers supporting people with intellectual disabilities, including the need for monitoring of sleep and seeking professional support and guidance for sleep problems. The annual health checks offer an opportunity to review problems with sleep and the impact that these have on the person and their family. However, consideration of sleep problems within the annual health check currently focusses on sleep disordered breathing and as a sign of reflux oesophagitis (NHS England, 2017), with more thorough assessment of sleep problems needed. Following from a holistic assessment of the individual including review of their sleep pattern, care planning relating to health and well-being should consider behavioural interventions to promote good sleep.

This review has limitations and strengths. An integrative review enabled a wide range of research using different research designs to investigate what is already known about sleep in people with an intellectual disability. However, a limitation of integrating a wide range of research designs adds complexity to the appraisal framework used. Many authors who have published integrative reviews describe but do not appraise the research (Webb & Roe, 2007). In contrast, a systematic review where only quantitative papers are reviewed would lend itself to a more robust critical analysis of the research. Therefore, a limitation is that the quality of the papers included in the review were not systematically appraised. However, throughout this paper, limitations of the studies were presented and discussed.

A further limitation of this review is that searches only spanned the last 7 years, which could exclude seminal research conducted prior to this date. However, themes from contemporaneous research were identified following reviewing of the most recent articles from multiple database searches. A strength of the review is that it provides an overarching picture of the most recent knowledge about sleep in people with intellectual disabilities, to enable areas for further research to be elicited.

The inclusion of research involving both children and adults can be seen as a strength and as a limitation. This approach enables learning across the age spectrum as many of the findings would hold true from childhood to adulthood. However, differences across the wide range of ages should be acknowledged. Changes are known to occur at adolescence due to cultural norms, social and bioregulatory factors (Tarokh & Carskadon, 2013). A dissipation of sleep pressure and increase in use of electrical items that provide external light signals which are important for synchronising time cues result in a later phase preference (Misiunaite et al., 2020). In conjunction, work or educational demands dictating timing of the day may lead to weekday sleep deprivation and variability in weekday and weekend sleeping patterns (Tarokh & Carskadon, 2013). However, the rationale for including all participants over the age of five in this review is that adult sleep architecture typically occurs around the age of 5 years. At this age a biphasic sleep wake pattern occurs that is indicative of consolidation of an adult sleep pattern (Spruyt & Curfs, 2015). Therefore, research across the age spectrum and in different populations may offer potential insight or interventions but further research is needed to corroborate for other ages and the expected benefit in people with different diagnoses.

In conclusion, evidence reviewed suggests that improving sleep is likely to benefit not only the individual's quality of life, their physical and mental well-being but also that of their family and carers. The sleep patterns of people with intellectual disabilities should be monitored using a combination of objective and subjective assessment tools such as combinations of the children's sleep habits questionnaire

and the pictorial Epworth sleepiness scale, sleep diaries and actigraphy. A multi-disciplinary team should be involved to review sleep problems and causes of these problems (including a review of pain, medication, comorbidities, impact on quality of life) and provide appropriate support to enhance the quality and quantity of their sleep, as needed. Behavioural interventions such as education and advice on sleep hygiene should be included within the package of care provided and provided at the earliest opportunity to people with intellectual disabilities with sleep problems, their families and carers.

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DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available in Pure at https://researchportal.northumbria.ac.uk/. These data were derived from the following resources available in the public domain: - CINAHL, https://web.s.ebscohost.com/ehost/ - SCOPUS, https://www.scopus.com/ - PsychInfo, https://www.apa.org/pubs/databases/psycinfo.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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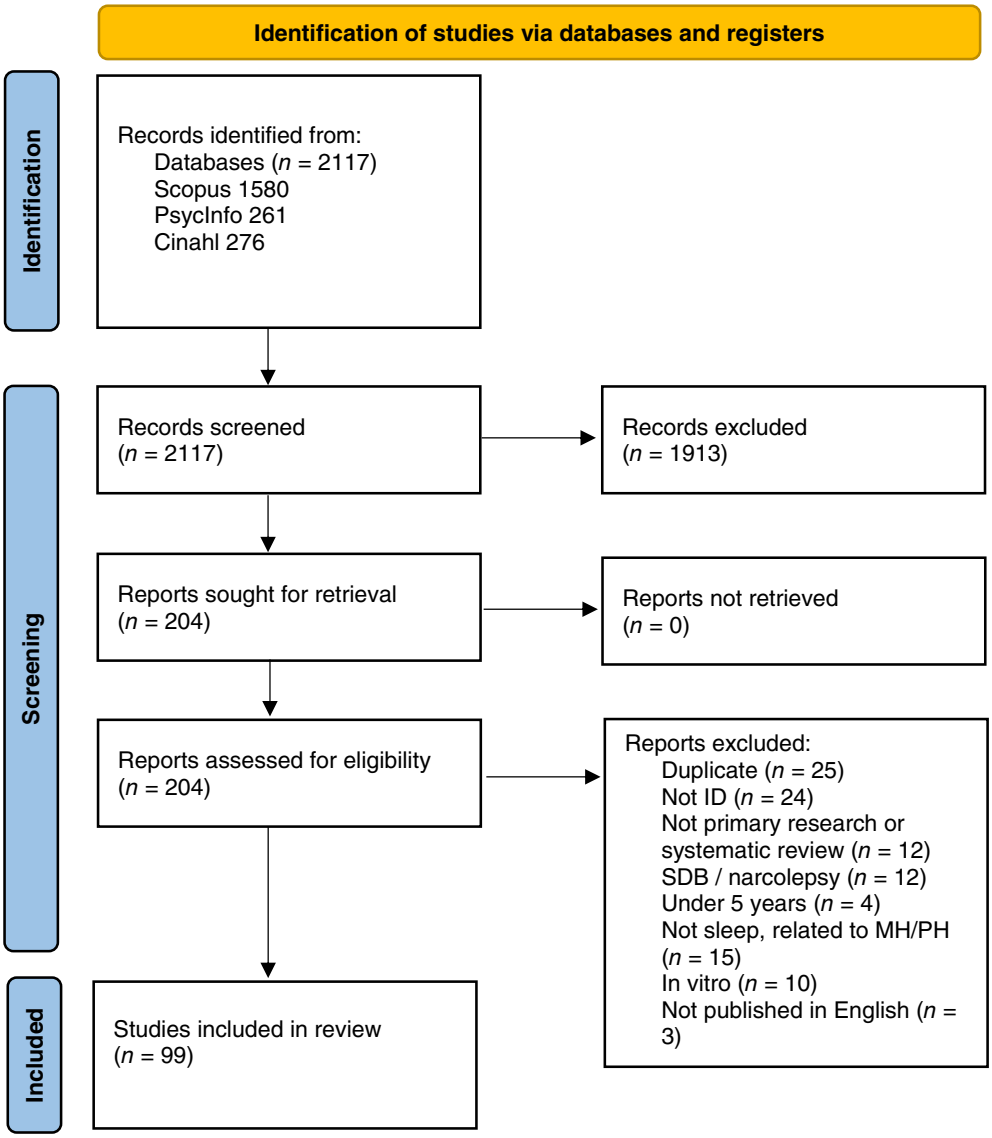
APPENDIX 1: PRISMA flow chart to show studies included at each stage of the search process



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