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Sleep in Neurodevelopmental Disorders

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Abstract

Individuals with intellectual and developmental disabilities (IDD) experience sleep problems at higher rates than the general population. Although individuals with IDD are a heterogeneous group, several sleep problems cluster within genetic syndromes or disorders. This review summarizes the prevalence of sleep problems experienced by individuals with Angelman syndrome, Cornelia de Lange syndrome, Cri du Chat syndrome, Down syndrome, fragile X syndrome, Prader-Willi syndrome, Smith-Magenis syndrome, Williams syndrome, autism spectrum disorder, and idiopathic IDD. Factors associated with sleep problems and the evidence for sleep treatments are reviewed for each neurodevelopmental disorder. Sleep research advancements in neurodevelopmental disorders are reviewed, including the need for consistency in defining and measuring sleep problems, considerations for research design and reporting of results, and considerations when evaluating sleep treatments.

Keywords

sleep; genetic syndrome; intellectual disability

Introduction

Individuals with intellectual and developmental disabilities (IDD) experience sleep problems at higher rates than the general population. Prevalence estimates vary, with estimates of reported sleep problems ranging from 34% to 86% (Bartlett, Rooney, & Spedding, 1985; Clements, Wing, & Dunn, 1986). Although the nature of sleep problems in individuals with IDD has been a focus of research for several decades, less research has been focused on treatment options and often assessed individuals with IDD as a homogenous group with little distinction for developmental changes with age. Several literature reviews in the last decade have drawn attention to sleep problems in specific genetic syndromes or disorders, specific age groups, or targeting issues with assessment and treatment (Churchill, Kieckhefer, Landis, & Ward, 2012; Didden & Sigafoos, 2001; Doran, Harvey, Horner, & Scotti, 2006; Richdale & Baker, 2014; Tietze et al., 2012; van de Wouw, Evenhuis, & Echteld, 2012). Given the prevalence of sleep problems in individuals with IDD, there is growing need to understand the types of sleep problems experienced by this heterogeneous group of individuals with IDD, how measurement of sleep problems impacts findings, and implications for appropriate treatment of sleep problems among individuals with IDD across the lifespan.

Several studies have compared sleep problems across syndrome groups in an effort to better characterize associated behavioral phenotypes and areas of concern specific to genetic syndromes (Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2013; Cotton & Richdale, 2006; Cotton & Richdale, 2010). These efforts are helpful in identifying potential neurobiological or craniofacial contributions to sleep problems. For example, facial and physical features may contribute to an increased risk for obstructive sleep apnea in individuals with Down syndrome (DS) and circadian timing may be altered in individuals with autism spectrum disorder (ASD) (Esbensen, 2016; Glickman, 2010). Additionally, previous studies highlight circadian rhythm problems in some children with Smith-Magenis syndrome (SMS) reporting higher endogenous melatonin levels during the day than at night (De Leersnyder et al., 2001; Potocki et al., 2000). However, the research rigor in studies of sleep problems in individuals with IDD or specific genetic syndromes varies, from (1) limited to case series, cohort studies, (2) well-conducted cohort studies or case control, and (3) few high-quality systematic reviews or cohort studies (van de Wouw et al., 2012). Sleep research in individuals with IDD is further hindered by diverse sleep assessment methods, small sample sizes, and heterogeneous groups with respect to disorders and age. This variability impacts the ability to draw conclusions about sleep problems in specific populations and the ability to effectively evaluate sleep treatment efficacy.

This review focuses on common sleep problems among individuals with different genetic syndromes or disorders, in an effort to identify research gaps and methods for improving research replicability and ultimately the sleep of individuals with IDD. Rather than compare the rates of problems across syndromes, we elected to focus on the specific sleep problems experienced within syndromes/disorders. The intention is to highlight sleep problems and commonalities within syndrome to better identify research gaps and inform sleep evaluation and treatment. The following sections will review sleep problems types, sleep assessment methods, review research targeted to specific syndromes and disorders, and identify areas for future research.

Common Sleep Problems

Prior to reviewing specific sleep problems experienced by individuals with IDD, a review of common sleep problems and their definitions is warranted. Prior classification systems for sleep disorders varied, contributing to the use of different terms to describe sleep problems in the research literature. Recent revisions to the International Classification of Sleep Disorders (ICSD-3) and Diagnostic and Statistical Manual of Mental Disorders (DSM-5) provide for more consistent definitions of sleep problems (Association, 2013; Medicine, 2014). Sleep problems are a heterogeneous set of problems, including difficulties with insomnia, sleep-related breathing disorders, excessive daytime sleepiness, circadian rhythm sleep disorders, parasomnias, and sleep-related movement disorders.

Insomnia includes difficulties with initiating or maintaining sleep, and can be behavioral or physiological. Behavioral insomnias often include bedtime refusals and difficulties settling associated with poor sleep hygiene. These behaviors often delay sleep onset, and/or can contribute to difficulties settling back to sleep after a night waking. Stressors may also exacerbate insomnia. Physiological causes of insomnia may include heightened arousal or

anxiety prior to bedtime, atypical melatonin profiles, pain, acid reflux, and heart dysfunction.

Sleep-related breathing disorders include a class of insomnia concerns that implicate the respiratory system. They include periodic difficulties with respiration during sleep, like obstructive and central sleep apnea, and sleep-related hypoventilation. Obstructive sleep apnea (OSA) is an upper airway obstruction that leads to cessation in breathing for between 20-200 seconds. Specific criteria for defining hypopnea have been developed and are a well-used tool in research (Medicine, 2007). Central sleep apnea is less common and refers to recurrent difficulties with respiratory effort during sleep. These apneas are often related to arousals and fractured sleep.

Excessive daytime sleepiness (EDS) includes hyper-somnolence and narcolepsy. EDS can involve an excess of total sleep time (at night or including daytime napping), reduced daytime arousal, and difficulty waking after a nap.

Circadian rhythm sleep disorders involve a repeated pattern of delayed or advanced sleep (generally off by more than 2 hours) in relation to the desired sleep-wake schedule. Circadian rhythm sleep disorder can result in symptoms of insomnia and EDS.

Parasomnias include events or arousals that accompany sleep, for example sleep-walking, sleep terrors, excessive nightmares. While seizures can be associated with parasomnias, they are not in themselves a parasomnia. Nocturnal enuresis (bed-wetting) falls under other parasomnias within ICSD-3 criteria, but fall under elimination disorders within the DSM-5 (Association, 2013; Medicine, 2014).

Sleep-related movement disorders include rhythmic or repeated movements of the legs, limbs, jaw, and/or neck during sleep. These disorders commonly include restless leg syndrome, bruxism, or sleep-related leg cramps.

Measurement

In addition to the heterogeneous set of sleep problems that are common concerns in individuals with IDD, there exists heterogeneity in the methods used to measure sleep behaviors and problems in individuals with IDD, which complicates the extraction of knowledge for these specialized populations. Different methods for assessing sleep were recently reviewed in relation to children with ASD (Hodge, Parnell, Hoffman, & Sweeney, 2012). Readers are encouraged to consult Hodge et al (2012) for a thorough review on how various measures of sleep have been used with this population. Briefly, we review the measurement of sleep and how they have been applied more generally with individuals with IDD.

Polysomnography (PSG) is the gold standard for assessing sleep problems, providing measures of brain, muscle, and heart activity via electro-encephalography, -oculography, -myography, -cardiography and pulse oximetry. PSG has been used to assess sleep in individuals with IDD for almost 50 years (Feinberg, Braun, & Shulman, 1969). However, access to PSG and the ability of all individuals with IDD to successfully complete a PSG is

hindered by several characteristics that may present when administered to individuals with IDD. Sensory sensitivity may reduce electrode compliance, and anxiety of being away at home or in a new environment contribute to some children not completing a PSG. Recent advancements in supporting PSG through sensitization techniques, social stories, and preparatory visits have increased the ability of individuals with IDD to successfully complete a PSG. However, accessibility and cost of PSGs likely make them prohibitive for some research studies or study populations (Primeau et al., 2015).

Actigraphy is commonly used as a measure of sleep. It has demonstrated reliability with PSG in the general population (Sadeh, Hauri, Kripke, & Lavie, 1995) and in small studies of children with IDD (Goldman, Bichell, Surdyka, & Malow, 2012). Benefits of actigraphy include the ability to measure sleep in the home setting over several nights, reduced cost, and increased compliance in comparison to PSG. Actigraphy is generally tolerated by children and adults with IDD (Ashworth et al., 2013; Goldman, Bichell, et al., 2012). However, a recent large study of older adults with IDD demonstrated that successful use was only obtained in 35% of adults, with unsuccessful use generally related to intolerance with wearing the device (van Dijk, Hilgenkamp, Evenhuis, & Echteld, 2012). To address sensory sensitivity to actigraphy watches worn on the wrist, their accuracy in measuring sleep has been evaluated when worn on the shoulder or ankle. Generally actigraphy worn on the shoulder demonstrate high correlation with wrist actigraphy for sleep onset, total sleep time, and sleep efficiency among children with ASD (Adkins et al., 2012). However, shoulder actigraphy demonstrates poor correlations with wrist actigraphy for wake-after-sleep-onset (WASO). Ankle worn actigraphy for individuals with IDD demonstrates higher rates of compliance and comparable estimates for sleep onset time and total sleep time but this placement does not appear to capture night awakenings well when compared to videosomnography (Sitnick, Goodlin-Jones, & Anders, 2008). Given the small samples in each of these studies, additional research is needed to strengthen the use of actigraphy in individuals with IDD, especially when considering shoulder or ankle placement.

Subjective measures of sleep include parent reported sleep diaries, and sleep questionnaires. *Sleep diaries* are often used in conjunction with actigraphy, where parents report on nightly sleep behaviors over a 1 or 2 week period. Parents generally are asked to report on time in bed, time asleep, time awake, and any night awakenings. Parents may also be asked about daytime behaviors, such as napping, and regarding behaviors that may influence sleep hygiene, such as exercise, screen time, and caffeine intake. Relatively few studies have addressed the accuracy or biases inherent in parent-report dairies for children or individuals with IDD but a recent study demonstrated that in families raising children with ASD parent-report diaries were just as accurate as those from families raising children with no known diagnosis (Schwichtenberg, Hensle, Honaker, Miller, & Ozonoff, in press). In general, parents in both groups were accurate reporters of their child's nighttime sleep; however, both groups underestimated daytime sleep (when compared to actigraphy estimates).

Questionnaire measures of sleep have been most commonly used to screen for sleep problems among individuals with IDD. However, sleep questionnaires have often been author designed or generally not standardized for use with individuals with IDD (Rosen, Lombardo, Skotko, & Davidson, 2011). Most commonly, studies have used the Children's

Sleep Habits Questionnaire (CSHQ) due to its reliability with and ability to make comparison to typically developing children, however, its psychometric properties have not been established with individuals with IDD (Ashworth et al., 2013). Others measures have been developed or adapted for use with children with IDD, such as the Behavioral Evaluation of Disorders of Sleep Scale (BEDS) and adapted Simonds-Parragan Sleep Questionnaire (Maas et al., 2011; Simonds & Parraga, 1982; Wiggs & Stores, 1996). However, questionnaire reports of sleep generally demonstrate poor reliability with actigraphy. Parent reports of sleep among school-age children with DS or Williams syndrome (WS) correlate well with total sleep time, but not with sleep onset latency, and number and duration of night wakings (Ashworth et al., 2013). While parent reports of sleep problems have been found to correlate with objective measures of restlessness and snoring in children with DS, these objective measures have not then been correlated with sleep quality (R. Stores & Stores, 2014). Further, among young children with DS, questionnaires based on parent report have demonstrated poor reliability with PSGs (Maris, Verhulst, Wojciechowski, Van de Heyning, & Boudewyns, 2016; Shott et al., 2006), such that the American Academy of Pediatrics recommends all children with DS receive a PSG prior to the age of 4 years of age (Bull & Genetics, 2011).

Meeting *standards* for a sleep condition based on the ICSD-3 or the DSM-5 are also used to document the presence of sleep problems (Association, 2013; Medicine, 2014). The challenge when comparing rates of sleep problems across different types of measurement tools are that they often result in different reported rates of sleep problems (Dohnt, Gradisar, & Short, 2012; Espie & Tweedie, 1991). This challenge is not a new concern or specific to just sleep problems, yet our research community has continued to frequently rely on questionnaire findings rather than diagnostic criteria. Screening tools, such as parent report questionnaires, are often an over-estimate of certain sleep problems such as difficulties with sleep onset or co-sleeping, and an under-estimate of other sleep problems such as motor movement or sleep apneas. Informant-report sleep questionnaires are likely restricted to use with adults with IDD with overnight support staff or co-residing family, thus not easily generalized to all adults with IDD. Similarly, self-reports of sleep problems by adults with IDD are not easily generalized to all adults with IDD.

Syndrome/Condition Specific Sleep Problems

Angelman Syndrome

Prevalence—Among children with Angelman syndrome (AS), sleep problems have been reported among 20–80% of individuals, with a more recent review citing rates of 48–70% (Tietze et al., 2012; Walz, Beebe, & Byars, 2005; Williams et al., 1995). In AS, sleep problems of specific concern include insomnia (sleep initiation and sleep maintenance), parasomnia, daytime sleepiness, and sleep-related breathing problems, with reported rates varying depending on how sleep was measured (Goldman, Bichell, et al., 2012). A study combining actigraphy, rating scales and PSG reported significant difficulties with sleep latency, frequent and longer night waking, fragmented sleep and shorter sleep duration (Goldman, Bichell, et al., 2012). Using PSG, the sleep architecture for young children with AS includes poor efficiency, a lower percentage of time in REM sleep, and a higher

percentage of time in slow wave sleep (Miano et al., 2004). These sleep problems were replicated on parental rating scales. Parent rating scales also indicated individuals with AS are reliant on sleep facilitators, disoriented when aroused, and awaken easily to loud noises (Walz et al., 2005).

Associated problems—Sleep problems in individuals with AS are associated with age and potentially to seizures (Goldman, Bichell, et al., 2012). Comparisons across research reports suggest a lessening of severity of sleep problems with age, with improvements noted from preschooler to school-age children. However, these conclusions ignore the persistence of sleep problems in individuals with AS. The relationship between sleep problems in individuals with AS and associated seizures has been mixed. Two studies have suggested that seizures were associated with more severe sleep problems (Clayton-Smith, 1993; Didden, Korzilius, Smits, Curfs, & Dykens, 2004). A more recent study using parental reports did not support an association between seizures and sleep problems (Walz et al., 2005).

Sleep problems in children with AS impact their family environment. Shorter sleep duration in children with AS is associated with more frequent night waking in their parents, variable bed time is associated with parent stress, and longer sleep latency is associated with parental insomnia and daytime sleepiness (Goldman, Bichell, et al., 2012).

Treatment—The evaluation of treatments to improve sleep among individuals with AS has generally been limited to case reports and case series (Clayton-Smith, 1993; Summers et al., 1992). These treatment trials have included behavioral and combined behavioral treatment with medication. Some researchers have speculated that the longer sleep latency common in children with AS is associated with poor sleep facilitators or poor sleep hygiene, which would be amenable to behavioral sleep interventions or medications to support sleep initiation (Walz et al., 2005). In one survey, 29% of individuals with AS were taking medications to support sleep (Walz et al., 2005). In a small sample of children with AS, 53% were taking melatonin (Goldman, Bichell, et al., 2012). Medication trials have generally been open-label and uncontrolled, although demonstrating improvements in sleep latency and motor restlessness (Braam, Didden, Smits, & Curfs, 2008a; Zhdanova, Wurtman, & Wagstaff, 1999). Two small studies that specifically assessed the efficacy of medication use of children with AS. The first was an open label trial in 13 children wherein sleep duration increased from baseline levels with melatonin treatment (.3 mg). However, this study also reported lower overall activity levels (as indexed by an actigraphy) which in and of itself may have affected the actigraphy estimates of sleep. A second study, a randomized double-blind placebo-controlled study, of 8 individuals with AS reported several improvements in sleep with melatonin (2.5 – 5 mg), when compared to baseline and placebo, including shorter sleep onset latencies, earlier sleep onset time, fewer night awakenings, and longer sleep durations (Braam et al., 2008a).

Cornelia de Lange Syndrome

Prevalence—According to research estimates, 55% to 72% of children with Cornelia de Lange syndrome (CdLS) have a comorbid sleep problem, the most common of which is

insomnia (bedtime settling, and night awakenings) (Basile, Villa, Selicorni, & Molteni, 2007; Berney, Ireland, & Burn, 1999; Gualtieri, 1991; Hall, Arron, Sloneem, & Oliver, 2008; Tietze et al., 2012). However, when comparable to a case-controlled sample of individuals with IDD matched for age, gender, degree of intellectual disability (ID) and mobility the rates in CdLS do not appear elevated (Hall et al., 2008). Recent studies using rating scales suggest a high rate of sleep disordered breathing, with 35% of children demonstrating moderate to severe symptoms, and additional symptoms of insomnia and circadian rhythm disorder (Stavinoha et al., 2011)

Associated problems—Given the high rate of self-injury present in CdLS, the association between sleep problems and self-injury has been explored, but with no reported relationship (Hall et al., 2008). A higher rate of sleep problems is reported among individuals with CdLS with more severe IDD and among children with CdLS in comparison to adults (Berney et al., 1999; Rajan et al., 2012). Small sample sizes preclude statistical analyses to determine if sleep problems become less prevalent with age, but are suggestive that difficulties with sleep onset decline from childhood into adulthood in individuals with CdLS (Rajan et al., 2012).

Treatment—No research was identified that has evaluated treatments for sleep problems among individuals with CdLS. However, in studies of diverse IDD (like those seen in CdLS), a combination of individualized behavioral therapy and medication has been effective (Didden & Sigafoos, 2001).

Cri du Chat Syndrome

Prevalence—Among children with Cri du Chat syndrome (CDC), sleep problems are reported among 30–50% (Maas et al., 2009; Tietze et al., 2012). Sleep questionnaire reports suggest difficulties with insomnia (bedtime settling, sleep anxiety, night waking, and poor quality sleep) and sleep disordered breathing (Cornish, Oliver, Standen, Bramble, & Collins, 2003; Cornish & Pigram, 1996; Maas et al., 2009). Snoring is also frequently reported (Maas, Didden, Korzilius, & Curfs, 2012). Researchers have suggested that the rate of sleep problems in individuals with CDC is not different from that reported among individuals with IDD or other genetic syndromes (Maas et al., 2009; Maas et al., 2012).

Associated problems—No research was identified that assessed associated problems relative to sleep problems solely among individuals with CDC.

Treatment—In one descriptive survey of a small sample, 13% of individuals with CDC were taking medications to support sleep (Maas et al., 2009). Among those reported to be using medication to support sleep, half of parents indicated that medication was helpful. Parents reporting receiving psychological supports for sleep found it helpful, but those receiving educational or general advice did not find the advice helpful in supporting the sleep problems in their children with CDC (Maas et al., 2009).

Down Syndrome

Prevalence—Among children with Down syndrome (DS), sleep problems are reported among 31–54% (G. Stores & Stores, 2013; Tietze et al., 2012). Measurement of sleep problems has included PSGs, actigraphy, sleep diaries, and sleep questionnaires. Sleep problems of specific concern among children with DS include insomnia, OSA, and daytime sleepiness. With actigraphy and/or PSG, children with DS have more fractured sleep, as measured by longer WASO, longer time in bed, lower sleep efficiency, less time in REM sleep, and more movement during sleep as compared to typically developing children (Ashworth et al., 2013; Ashworth, Hill, Karmiloff-Smith, & Dimitriou, 2015; Harvey & Kennedy, 2002). On rating scales, children with DS were noted to have difficulties with insomnia (sleep onset, bedtime settling, sleep anxiety and bedtime resistance, sleep maintenance and night waking), parasomnias, and sleep-disordered breathing (Ashworth et al., 2013; Breslin, Edgin, Bootzin, Goodwin, & Nadel, 2011; Cotton & Richdale, 2006; R. Stores, Stores, Fellows, & Buckley, 1998). Night waking is reported to be frequent and for long periods of wakefulness, and to also be related to restlessness during sleep.

Obstructive sleep apnea (OSA) is a specific concern in DS, affecting 31–63% of individuals (de Miguel-Diez, Villa-Asensi, & Alvarez-Sala, 2003; Marcus, Keens, Bautista, von Pechman, & Ward, 1991; Stebbens, Dennis, Samuels, Croft, & Southall, 1991). Slightly lower rates of OSA are reported in community samples of individuals with DS, affecting 24–59% of individuals (Dahlqvist, Rask, Rosenqvist, Sahlin, & Franklin, 2003; Stebbens et al., 1991). However, both community and clinic samples report risks significantly higher than the 3–7% reported in the general population (Punjabi, 2008). The increased risk of OSA is likely related to certain facial and physical features present in DS, including mid-facial hypoplasia, mandibular hypoplasia, glossoptosis (retraction or downward displacement of tongue), an abnormally small upper airway, superficially positioned tonsils, relative tonsillar and adenoidal encroachment, and hypotonia of the upper airway (Churchill et al., 2012; Marcus et al., 1991; Roizen & Patterson, 2003).

Associated problems—For individuals with DS, several demographic and daytime behaviors have demonstrated relationships with sleep problems. Older age (within school-age children with DS) is related to a decrease in total sleep time, fewer parasomnias and lower sleep anxiety and bedwetting. Additionally, males have a higher frequency of parent reported sleep problems and daytime sleepiness (Ashworth et al., 2013; Breslin et al., 2011; Maris et al., 2016; R. Stores, 1993). However, age and gender findings have not been replicated in relation to OSA (Maris et al., 2016).

Among children with DS, associations between sleep problems and daytime behaviors and daily life habits are common. Specifically, parent reports of sleep problems were associated with higher scores of irritability, hyperactivity and stereotypies, but surprisingly not to lethargy, on the Aberrant Behavior Checklist (R. Stores, 1993). In addition, parent-reported sleep problems were also associated with parent reports of difficulty across 11 domains of functional outcomes, including mealtimes, fitness, personal care, communication, home life, mobility, responsibilities, relationships, community life, school, and recreation (Churchill, Kieckhefer, Bjornson, & Herting, 2014).

Sleep disordered breathing, specifically OSA, is linked with lower verbal IQ scores and poorer performance on tests of cognitive flexibility among children with DS (Breslin et al., 2014). Further, a higher number of apneas per hour was related to difficulties with visuo-perceptual skills, such as orientation, among young adults with DS, suggesting that the severity of OSA can have a large impact on some cognitive skills (Andreou, Galanopoulou, Gourgoulianis, Karapetsas, & Molyvdas, 2002).

Adults with DS are also at high risk for sleep problems, increased by their additional risk for dementia (Trois et al., 2009). Among adolescents and young adults with DS, several parent-reported sleep problems were associated with poorer executive functioning. Specifically, insomnia was associated with poorer verbal fluency, OSA was associated with poorer verbal fluency and inhibitory control, and daytime sleepiness was associated with poor inhibitory control (Chen, Spanò, & Edgin, 2013). The severity of OSA assessed using PSG was related to younger age and obesity among adults with DS, but findings regarding the relationship to obesity have also been inconsistent (Fitzgerald, Paul, & Richmond, 2007; Shires et al., 2010; Telakivi, Partinen, Salmi, Leinonen, & Härkönen, 1987; Trois et al., 2009).

Treatment—Treatments for sleep problems in individuals with DS are currently tailored to the nature of their sleep problems, particularly OSA. While no clinical trials have been conducted examining the efficacy of positive airway pressure (PAP) or surgical interventions among individuals with DS, case reviews and pre-post designs suggest modest improvements following adenotonsillectomy among children with DS (Shete, Stocks, Sebelik, & Schoumacher, 2010). However, following surgery, many children with DS continue to need other interventions, such as PAP or surgical revisions (Shott et al., 2006). Children with DS are reported to accept and adhere to CPAP treatment (O'Donnell, Bjornson, Bohn, & Kirk, 2006).

Behavioral treatments for sleep have been evaluated using group administered formats and educational pamphlets. Minimal improvements in sleep have been noted in group administered treatment sessions (R. Stores & Stores, 2004). However, children with DS were not screened for sleep problems prior to study entry, likely limiting the impact of treatment. Research evaluation of melatonin are few, although case series suggest improvement with melatonin with young children and adolescents with DS (Jan, Espezel, & Appleion, 1994).

Fragile X Syndrome

Prevalence—The research literature on sleep problems in fragile X syndrome (FXS) is limited, in part because sleep problems generally appear consistent with those experienced by typically developing peers and in part because sleep problems are consistent with their ID and not specifically FXS (Harvey & Kennedy, 2002). However, animal models of FXS document sleep problems with the phenotype and parent reports (although limited) also endorse sleep problems for some individuals with FXS (Zhang et al., 2008). Research studies estimate that 31–77% of individuals with FXS also have a sleep problem, which is consisent with reported rates in other IDD groups (Kronk et al., 2010; Richdale, 2003). Children with FXS do experience shorter sleep duration, difficulties settling to sleep, and increased WASO (Gould et al., 2000; Miano et al., 2008; Richdale, 2003). Children and

young adults with FXS demonstrate a high percentage of stage 1 sleep, and low percentage of REM sleep, and a low number of REM episodes when compared to typically developing children (Miano et al., 2008). Recent findings suggest that despite having similar sleep problems when examining conventional sleep parameters, that individuals with FXS demonstrate disturbances in their sleep microstructure, specifically lower transient slow EEG oscillations within non-REM sleep (Miano et al., 2008).

Associated Problems—According to one FXS study, sleep problems are associated with poor health, listening skills, adaptability, and in those with a higher number of co-occurring conditions (Kronk et al., 2010). Similarly, Richdale (2003) reported an association between sleep problems and child psychopathology. Few studies exist that specifically address FXS outside of its common comorbid conditions (e.g., ASD). For this reason, associated features of sleep problems in FXS may be informed by the ASD and larger IDD literature. For example, sleep problems are associated with more daytime anxiety and sensory sensitivities in children with ASD (Mazurek & Petroski, 2015).

Treatment—Randomized controlled clinical trials of melatonin have been conducted on small samples of children with FXS alone, with the permutation or with comorbid ASD. Although the sample size for the individual subtypes was small, overall melatonin demonstrated improvements in total sleep time, sleep onset latency, and earlier bedtimes overall for this sample of children (Wirojanan et al., 2009). Additionally, behavioral sleep treatments have been demonstrated as effect in a small group (n = 5) of children with FXS (Weiskop, Richdale, & Matthews, 2005).

Prader-Willi Syndrome

Prevalence—Common sleep problems reported among individuals with Prader-Willi syndrome (PWS) include difficulties with insomnia (settling, sleep onset, maintenance), early morning waking, and excessive daytime sleepiness (Cassidy, McKillop, & Morgan, 1990; Cotton & Richdale, 2006; Gibbs, Wiltshire, & Elder, 2013). Among children with PWS, sleep problems are reported among 35–100%, and specifically include excessive daytime sleepiness and snoring (Richdale, Cotton, & Hibbit, 1999; Tietze et al., 2012). In a population study, 20% had a diagnosed sleep disorder (Butler et al., 2002). Using actigraphy, children with PWS are reported to have longer WASO, but shorter sleep latency. These findings persist when controlling for Body Mass Index (BMI) (Gibbs et al., 2013). The frequency of night waking and total time asleep is comparable to typically developing children, but children with PWS remain awake for longer and exhibit more daytime sleepiness (Gibbs et al., 2013).

Using PSG, studies have continually replicated shorter sleep latencies in children with PWS, but demonstrated variable results regarding sleep latency in adults with PWS (Joo et al., 2010; Verrillo et al., 2009; Vgontzas et al., 1996). PSG studies have also replicated findings of a greater frequency and duration of WASO among children with PWS, but not among adults (Verrillo et al., 2009). Abnormal REM sleep patterns have been reported, with individuals with PWS having REM sleep onset earlier during the night (Manni et al., 2001).

Associated problems—In contrast to other syndrome groups where age is often associated with deceasing rates of sleep difficulties, among children with PWS, the reported rate of sleep problems is reported to increase with age (Richdale et al., 1999). Given the high rate of obesity among individuals with PWS, and the association between obesity and OSA, the rate of OSA has been investigated among individuals with PWS. While some individuals do demonstrate OSA, many do not despite marked obesity (Vela-Bueno et al., 1984). More recent case reviews have suggested a link between BMI, sleep arousals, and hypoxemia during sleep (O'Donoghue et al., 2005).

Sleep problems in children with PWS affects the child and their family. In a parent survey, child's sleep problems were detrimental to the child (87%), adversely impacted other family members (44%), and adversely impacted child and family well-being (32%) (Cotton & Richdale, 2006). Another study identified that the severity of OSA is related to higher parental reports of child impulsivity and inactivity (O'Donoghue et al., 2005).

Several studies of individuals with PWS have investigated the possible link between sleep disordered breathing and excessive daytime sleepiness, with mixed findings. Daytime sleepiness is considered to be a central feature of PWS, and not secondary to nighttime sleep disturbances, with hypothalamic dysfunction suspected as contributing to excessive daytime sleepiness (Nixon & Brouillette, 2002).

Treatment—Although sufficient intervention studies have not directly assessed individuals with PWS, treatment recommendations to support the sleep of individuals with PWS have been provided based on treatment recommendations for the general population (Nixon & Brouillette, 2002). Recommended treatments for OSA include adenotonsillectomy, weight loss, PAP, and tracheostomy. The effectiveness of adenotonsillectomy in a small sample suggests that although this surgery improves the obstructive apnea/hypopnea index (AHI) and oxygen desaturations among children with PWS, postoperative complication are also present (Pavone et al., 2006). A review of studies in PWS suggests improvements following adenotonsillectomy, but with residual OSA post-surgery (Sedky, Bennett, & Pumariega, 2014). Recommendations for treating central apnea include nasal mask, tracheostomy, and weight loss. Recommendations for treating excessive daytime sleepiness include behavioral treatments (extending total time asleep, supporting sleep onset) and consideration of medication to support sleep.

Smith-Magenis Syndrome

Prevalence—The physiology of individuals with Smith-Magenis Syndrome (SMS) predisposes them to having disturbed circadian rhythm. Specifically, melatonin rhythms are often inverted, impacting their ability to fall asleep. Among children with SMS, sleep problems are reported in all children (100%) across multiple studies (Tietze et al., 2012). On rating scales, individuals with SMS (infants to adults) demonstrate significant sleep problems, in 65–100% of individuals (Greenberg et al., 1996; Smith, Dykens, & Greenberg, 1998). Common sleep problems included insomnia (bedtime settling, frequent and long sleep waking), parasomnias (bedwetting), excessive daytime sleepiness (daytime napping), short sleep cycles, and snoring (Smith et al., 1998). As expected, reported rates of

bedwetting was most common among younger children (82% under 10 years of age). Using PSG, over half of a small sample demonstrated abnormalities in REM sleep, primarily reduced REM sleep (Greenberg et al., 1996).

Associated problems—Using a rating scale to assess sleep problems, relationships were found with older age and earlier morning waking, more frequent naps, more WASO, and shorter sleep duration (Smith et al., 1998).

Treatment—Responses to a survey suggest that 59% of individuals with SMS use medication to facilitate sleep, with higher use of medication among individuals over the age of 10 years (Smith et al., 1998). Several case studies report effectively treating circadian sleep disturbances using melatonin and β1-adrengic antagonists (Carpizo et al., 2006; Chou, Tsai, Yu, & Tsai, 2002; Van Thillo, Devriendt, & Willekens, 2010). A case series of children with SMS reported a reversal of melatonin patterns with the treatment of β1-adrenergic antagonists during the day and melatonin prior to bedtime (De Leersnyder et al., 2001). In total these studies reflect the treatment of only 13 children with SMS. However, within a rare disorder such as SMS they present growing evidence that melatonin treatment (coupled with a β1-adrenergic antagonist) may help children with SMS who present with inverted or altered circadian patterns.

Williams Syndrome

Prevalence—Sleep problems are reported in 36–57% of children with Williams syndrome (WS) with sleep latency as a common concern (Tietze et al., 2012). On rating scales, children with WS were noted to have difficulties with insomnia (initiating sleep, night waking, and sleep anxiety), parasomnias (bed-wetting, limb movement), daytime drowsiness, and sleep duration (Annaz, Hill, Ashworth, Holley, & Karmiloff-Smith, 2011; Ashworth et al., 2013; Goldman, Malow, Newman, Roof, & Dykens, 2009). Among adolescents and young adults, self-reports indicate a high level of daytime drowsiness, with 34% meeting criteria for excessive daytime sleepiness (Goldman, Malow, et al., 2009). Using actigraphy, studies have replicated that children with WS have long sleep latencies, and that adolescents and young adults with WS have long sleep latencies, poorer sleep efficiency, more WASO, fragmented sleep and increased motor movements (Ashworth et al., 2015; Goldman, Malow, et al., 2009). More recently, studies using PSG in children with WS have been conducted, suggesting additional problems with sleep architecture. Children with WS were reported to have poorer sleep efficiency, decreased REM sleep and increased slow wave sleep (Gombos, Bódizs, & Kovács, 2011; Mason et al., 2011).

Associated problems—The shorter night sleep experienced by children with WS has been associated with lower language development in preschoolers, and possibly with learning performance in school-age children (Axelsson, Hill, Sadeh, & Dimitriou, 2013; Dimitriou, Karmiloff-Smith, Ashworth, & Hill, 2013). In children with WS less total sleep time is associated with older age (Ashworth et al., 2013). Further, sleep duration is associated with cardiac problems in school-age children with WS, and sleep latency associated with asthma and allergies (Annaz et al., 2011). In relation to maternal

characteristics, frequent night wakings among school-age children with WS are reported to be associated with maternal sleep and mood (Axelsson et al., 2013).

Treatment—No research was identified that has evaluated treatments for sleep problems among individuals with WS.

Autism Spectrum Disorders

Prevalence—For individuals with autism spectrum disorder (ASD) parent/caregiver reports of sleep problems are common with estimates of roughly 50 to 80% (Couturier et al., 2005; Goodlin-Jones et al., 2009; Liu, Hubbard, Fabes, & Adam, 2006; Schreck & Mulick, 2000). Sleep problems in ASD are diverse and include insomnias, parasomnias, and sleep disordered breathing (Elrod et al., 2016; Hirata et al., 2016). Sleep problems are among the first concerns parents report in young children later diagnosed with ASD and persist through adolescence and into adulthood (Goldman, Richdale, Clemons, & Malow, 2012; Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005; Ozonoff et al., 2009). Sleep problems are not a constant in the development of individuals with ASD, but are fairly stable with sleep problems noted across the life course (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; Croen et al., 2015). Only a few studies directly assess the stability of sleep problems/behaviors in ASD noting short-term stability for 3–6 months without intervention (Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2011; Goodlin-Jones et al., 2009) but numerous studies document elevated rates of sleep problems in children, adolescents, and adults using cross-sectional designs (Croen et al., 2015). Sleep problems are more common in individuals with ASD who also have comorbid ADHD, anxiety, depression, or gastrointestinal problems (Klukowski, Wasilewska, & Lebensztejn, 2015). Research on comorbid ID and ASD with respect to sleep problems is less consistent. Several studies report elevated rates of sleep problems in individuals with ASD and ID (Giannotti et al., 2008) but at least two studies also demonstrate no significant difference between individuals with ASD without ID and ASD with ID (Johnson, Turner, Foldes, Malow, & Wiggs, 2012; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008).

Insomnia is the most common type of sleep problem in individuals with ASD and may include prolonged sleep onset latency, extended night awakenings, and early morning rise times (Baker, Richdale, Short, & Gradisar, 2013; Richdale & Schreck, 2009; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012; Souders et al., 2009). Less common but documented sleep problems in ASD also include frequent night awakenings, a free-running sleep rhythm, sleep disordered breathing, high sleep variability, and several parasomnias (Giannotti et al., 2008; Goldman, Richdale, et al., 2012; Liu et al., 2006; Richdale & Schreck, 2009; Schreck & Mulick, 2000; Tordjman et al., 2012). Individuals with ASD are a heterogeneous group; similarly, their sleep problems are just as diverse.

The highest rates of sleep problems in ASD come from studies utilizing parent-report indices like the Child Behavior Checklist (CBCL) or the CSHQ. Several studies have substantiated parent-reported sleep problem reports using more objective measures of actigraphy and PSG (Goldman, Richdale, et al., 2012; Malow et al., 2006; Wiggs & Stores, 2004). However, several studies also note parent reports of sleep problems are higher than

those specified using behavioral criteria (Goodlin-Jones et al., 2009; Gringras et al., 2014; Schreck & Mulick, 2000). For example, in a study of young children with ASD, other (non-ASD) developmental disabilities (DD), and no known diagnosis, Goodlin-Jones and colleagues reported elevated sleep problems via parent report in both the DD and ASD groups but only found elevated behaviorally defined sleep problems (e.g., frequent night awakenings) in the DD group (Goodlin-Jones, Tang, Liu, & Anders, 2009). Similarly, Schreck and Mulick (2000) found that parents of children with ASD reported comparable amounts of sleep in their children when compared to controls but endorsed higher rates of sleep problems. Additionally, behaviorally focused treatments have been effective in reducing parent reports of sleep problems but these changes did not always coincide with actigraphically-measured changes in sleep behavior (Johnson et al., 2013).

Associated problems—Sleep problems are associated with elevated rates of daytime behavior problems in children with ASD and with elevated rates of anxiety and depression in older children and adults (Allik, Larsson, & Smedje, 2006; Goldman et al., 2011; J. A. Henderson, Barry, Bader, & Jordan, 2011; Mazurek & Petroski, 2015; Mazurek & Sohl, in press; Sikora, Johnson, Clemons, & Katz, 2012). Across the life course, sleep problems are associated with more severe ASD symptomology (e.g., higher rates of repetitive behaviors) and GI problems (Klukowski et al., 2015; Schreck, Mulick, & Smith, 2004; Tudor, Hoffman, & Sweeney, 2012). Sleep problems are so common in ASD they are, at times, considered a phenotype marker (Cohen et al., 2014; Goldman, Surdyka, et al., 2009). Studies following this approach, report individuals with ASD and sleep problems display more intellectual disabilities, inattention, hyperactivity, and restricted/repetitive behaviors when compared to individuals with ASD without sleep problems (Elrod & Hood, 2015; Goldman, Surdyka, et al., 2009; Mazurek & Sohl, in press).

Treatment—Sleep problems in ASD may be biological, medical, or behavioral in nature and treatment of such problems should start with a detailed medical and family history. The three most common sleep treatment approaches in ASD include parent/caregiver education, behavioral, and pharmacological. Studies of *parent/caregiver education* programs focus on providing parent/caregivers with developmentally appropriate sleep information in various forms along with basic behavioral approaches to support optimal sleep. Information provided via pamphlets did not improve parental perceptions or child sleep behaviors (Adkins et al., 2012) but small group information sessions have been effective in improving parental sleep perceptions and child sleep onset delay (Malow et al., 2014; Reed et al., 2009).

Behavioral treatments in individuals with ASD include several cognitive behavioral approaches, various extinction protocols, sleep hygiene, chronotherapy, scheduled awakenings, sleep restriction, visual supports, stimulus fading, and reward programs. Using the standards put forth by Chambless and Hollon, two systematic reviews of behavioral treatments for sleep problems in ASD have been published (Chambless & Hollon, 1998; Schreck, 2001; Vriend, Corkum, Moon, & Smith, 2011). Within these reviews, empirical support is the strongest for standard extinction approaches when treating night awakenings or cosleeping and scheduled awakenings for reducing night terrors. Several small studies of

individuals with ASD document improvements in sleep with behavioral therapy (Reed et al., 2009) but without adequate replication these treatment approaches do not meet the standard of 'well-established' or ' possibly efficacious' treatments. Given the diverse nature of individuals with ASD and their sleep problems, current field recommendations include using an individualized treatment approach that starts with education and behavioral therapy (Malow et al., 2014). If these approaches are not successful, then a combined behavioral and pharmacological treatment plan are recommended (Cortesi, Giannotti, Sebastiani, Panunzi, & Valente, 2012).

Pharmacological treatments for sleep problems in ASD include most commonly melatonin and may also include α-agonists, anticonvulsants, antidepressants, atypical antipsychotics, and benzodiazepines. In a registry study of 1,518 children with ASD, Malow and colleagues reported that roughly 40% of children with ASD were treated pharmacologically for sleep problems (Malow et al., 2016). A full review of pharmacological treatments in ASD is beyond the scope of this review; however, we have included a section on the most commonly used medication - melatonin.

Several studies have assessed the efficacy of melatonin in treating sleep problems in individuals with ASD and associated genetic conditions (e.g., FXS, tuberous sclerosis). Although the dose, duration, and elements of sleep affected by melatonin vary considerably across studies, the cumulative findings provide support for melatonin treatment (Doyen et al., 2011; Guénolé & Baleyte, 2011; Rossignol & Frye, 2011; Schwichtenberg & Malow, 2015). The most consistent finding across studies of individuals with ASD and associated genetic conditions is decreases in sleep onset latency. Findings regarding other elements of sleep are less consistent. For example, some studies report increased night awakenings (Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt, 2003) and others report decreased night awakenings (Garstang & Wallis, 2006). Although this inconsistency and others like it could stem from differences in dose, melatonin type (fast release or continuous release), administration time, duration of administration, and sleep measurement technique as well as individual differences in melatonin metabolism. Overall, studies of melatonin in individuals with ASD are promising and report relatively few side effects (i.e., headaches, vomiting, upset stomach, dizziness, diarrhea, daytime sleepiness). Considering the relatively low rate(s) of side effects and the consistent findings regarding sleep onset latency, melatonin treatment for individuals with ASD who struggled with sleep onset is supported by the literature. For other types of sleep disturbances (e.g., early morning awakenings, several nighttime awakenings) the research findings are less consistent.

Idiopathic Intellectual Disability

Prevalence

When examining children with idiopathic IDD, rates of sleep problems ranged from 34–86%, with rates varying for specific subtypes of sleep problems (Bartlett et al., 1985; Clements et al., 1986). The variability in reported rates of sleep problems is influenced by how sleep problems are measured in the individual studies, but reported sleep problems persist at the same rate in longitudinal studies (Quine, 1991).

Difficulties with insomnia, both with settling to sleep and staying asleep, in individuals with IDD range from 8–34% based upon a systematic review of the literature from 1990–2011 (van de Wouw et al., 2012). However, when actigraphy was used to evaluate insomnia (sleep latency and WASO), sleep problems were more common, occurring in 72% of a large community sample (van de Wouw, Evenhuis, & Echteld, 2013). The sleep of older adults with IDD is reported to be fragmented, inconsistent with regards to rhythm, and of lower relative amplitude (more fragmented sleep rhythm) (Maaskant, van de Wouw, van Wijck, Evenhuis, & Echteld, 2013).

Associated problems

Sleep problems are an increasingly important area of research among individuals with IDD as researchers and clinicians recognize the bidirectional impact of sleep with comorbid medical and psychological conditions, cognition and executive functioning, mood and maladaptive behavior, and medication use. Some researchers have speculated that difficulties with sleep may contribute to a pattern of poorer cognitive outcomes for individuals with IDD (Harvey & Kennedy, 2002). However, sleep problems, as measured by actigraphy and pulse oximetry, were not related to attention tasks among children with genetic syndromes (Ashworth et al., 2015). In contrast, this study did replicate the relationship between sleep problems and attention among typically developing children. This finding may suggest that sleep does not have a significant impact on cognitive and executive functioning, or it may suggest the need for improved outcome measures of cognitive and executive functioning in order to adequately detect the impact of sleep problems on individuals with IDD (Esbensen et al., under review).

Consistent with typically developing children, a pattern of findings suggest that sleep problems (e.g., insomnia) are more frequent among younger children (Richdale, Francis, Gavidia-Payne, & Cotton, 2000). More severe levels of ID have been associated with more severe sleep problems, less time in REM, lower REM density, and difficulties with sleep onset (Churchill et al., 2012; Didden, Korzilius, Aperlo, Overloop, & Vries, 2002; Harvey & Kennedy, 2002). Among adolescents with IDD, the timing of sleep (late to bed, late to rise) was associated with the adolescent being overweight or obese, controlling for sleep duration (Vanhelst, Bui-Xuan, Fardy, & Mikulovic, 2013).

While directional interpretations cannot be made due to the cross-sectional nature of many studies, sleep problems are associated with maladaptive behaviors, health and mental health conditions, and medication (Didden et al., 2002; Doran et al., 2006). Among children with IDD, parent ratings of sleep problems were correlated with parental ratings of maladaptive behaviors and with the presence of seizures (Didden et al., 2002; Quine, 1991; Richdale et al., 2000). Sleep problems in school-age children with IDD was also related to more frequent and more intense parenting hassles, parental stress and well-being, parenting behaviors, and parental sleep (Quine, 1991, 1992; Richdale et al., 2000). Among adults with IDD, maladaptive behaviors have consistently been related to sleep problems reported on rating scales or observations of sleep (van de Wouw et al., 2012). Caregiver ratings of sleep problems in adults with IDD is also related to respiratory conditions and visual impairment (Boyle et al., 2010).

Several large studies of aging adults with IDD using actigraphy have identified associations between sleep problems and demographic and environmental variables. Females tended to spend more time in bed, but not necessarily asleep (van de Wouw et al., 2013). Older age, severity of ID, motor impairment (bound to wheelchair), living arrangement and mental health (depression) were also associated with more time in bed (Maaskant et al., 2013; van de Wouw et al., 2013). However, it should be noted that some of these relationships may be related more to caregiver/caregiving needs with supporting the physical needs of aging adults with IDD rather than true difficulties with sleep problems (Richdale & Baker, 2014). Overall, some researchers state there is no clear evidence for a relationship between sleep problems and demographic variables of age, gender and level of ID, particularly among adults with IDD (van de Wouw et al., 2012). Fragmented sleep was associated with lower physical activity, dementia, epilepsy, sensory impairments, and spasticity (Maaskant et al., 2013).

Sleep problems do not only impact the individual with IDD, but also others in their household. Parents report that sleep problems in children with IDD disrupt the sleep of other family members (Cotton & Richdale, 2006; Robinson & Richdale, 2004).

Treatment

There is evidence to support that some sleep interventions, that are effective in the general population, are also beneficial for individuals with IDD (Lancioni, O'Reilly, & Basili, 1999). The behavioral sleep treatments of extinction and graduated extinction with preschool and school-age children with IDD have received support, with study designs including case series and randomized control trials (Montgomery, Stores, & Wiggs, 2004; Richdale & Wiggs, 2005; Thackeray & Richdale, 2002). Parental/caregiver instruction has been found to be effective in improving sleep, both with individualized instruction and with educational booklets, with effects being maintained over 6 months (Montgomery et al., 2004). Educational training for staff supporting adults with IDD has demonstrated improvements in sleep efficiency, declines in time in bed and daytime napping (Hylkema, Petitiaux, & Vlaskamp, 2011).

Among adults, the use of CNS medication is reported to be associated with longer sleep duration and better sleep efficiency (van de Wouw et al., 2013). A meta-analysis of the use of melatonin with individuals with IDD demonstrates that it consistently is related to decreases in sleep latency and night wakings, and improvements in total sleep time (Braam et al., 2009).

Future Research

Sleep problems continue to be an area of concern for many individuals with IDD or specific genetic syndromes, with reported rates higher than those in the general population. The literature to date on individuals with IDD, various genetic syndromes or disorders has been able to replicate certain findings relating to associated sleep problems. However, different sleep measurement methodologies contributed to varied prevalence rates and patterns of associated problems. Further, suitable interventions lack strong empirical evidence and implementation in the community. This limited evidence within our research community is

due in part to the challenges of gathering sufficient sample sizes of individuals with specific genetic syndromes. While there does exist some variability in sleep problems across syndrome or disorder subgroups of individuals with IDD, there are some commonalities that can be capitalized on when designing future research studies. There are also promising steps that have been or are being taken to advance our understanding and treatment of sleep problems among individuals with IDD.

Recommendations for Definition and Measurement of Sleep

When studying sleep in individuals with IDD, our field is encouraged to be more consistent in how we define sleep problems and how we measure sleep problems. The definition of sleep problems has varied across studies, in part due to how sleep problems were measured. Given that the DSM-5 criteria for sleep disorders now mirrors more closely the ICSD-3 classification system, the use of common terminology to define sleep in encouraged. Future research needs to be consistent in how sleep problems are defined, in an effort to more clearly evaluate differences across subgroups of individuals with IDD and changes in sleep problems across the age-span (Medicine, 2014).

As evidenced by the above review of the literature, the prevalence of sleep problems can vary tremendously based on the assessment tool. In order to establish an accurate measure of sleep problems, our field is strongly encouraged to use a combination of objective tools to assess sleep. Meritorious research in sleep problems in individuals with IDD should use several different methods for assessing sleep (i.e., PSG and actigraphy). While sleep questionnaires and parental reports of sleep problems have not correlated well with PSG in identifying sleep problems, they continue to warrant evaluation for utility in serving as a valid measure of improving sleep outcomes, or for initially screening for sleep problems. When sleep questionnaires are used, research needs to acknowledge the limitations of this selected tool in measuring different types of sleep problems.

Our field is also encouraged to make use of advances in technology in order to assess sleep in the individual's home environment. Other techniques have been developed to assess sleep. *Portable PSGs* can be used in the individual's own home and technology continues to develop with the use of more subtle wiring (i.e., wires embedded in head gear, or within clothing). This technology has been used to measure sleep and OSA among children with DS (Breslin et al., 2014). *Videosomnography* (VSG) is a sleep recording method that is growing in popularity and includes indexing sleep from video recordings of sleep in the natural sleep environment. The use of VSG is attractive to researchers and clinicians because of (1) the 'stand-off' or no contact requirements, (2) in-home recording options, and (3) its ability to capture dyadic or behavioral sleep associations (Sadeh, 2015). For individuals with ASD or other developmental concerns the electrode or sensor placements associated with PSG or even actigraphy can be distressing and can disrupt sleep. Videosomnography has been used to diagnose sleep problems in a clinical populations (Ipsiroglu et al., 2015) and in numerous research studies of children with ASD, ID, and other developmental concerns (Ariagno et al., 2003; J. M. Henderson, France, Owens, & Blampied, 2010; Schwichtenberg et al., in press; Sitnick et al., 2008). Currently, VSG coding is labor intensive, taking roughly

1 hour to code a single night. Recent advancements in video-processing techniques may reduce this time and make the coding process automated.

Several ‘mass’ market devices now provide estimates of sleep using accelerometers (e.g., Basis, Jawbone, Fitbit, smart watches, smart phone apps), respiration sounds/movement (e.g., S+, Mimo), bioimpedance (e.g., Jawbone), optical (e.g., Basis), ballistocardiography (e.g., Beddit) and other proprietary sensors. The accuracy and clinical utility of these devices is inconclusive at this time (Grifantini, 2014). Overall, it appears that many of these devices over estimate sleep duration in adults and children in the general population (de Zambotti et al., 2016; Evenson, Goto, & Furberg, 2015); however, they may have utility in helping track improvements in sleep or changes in typical sleep patterns. Researchers within the field of IDD are encouraged to monitor research advances on these devices and evaluate their utility in assessing sleep in individuals with IDD.

Recommendations for Research Design

The literature on sleep problems in IDD encompasses a variety of subpopulations, genetic syndromes and disorders. Given the difficulties of collecting sizable samples of specific genetic syndromes and the similarities of sleep problems across several syndrome groups, our field is encouraged to consider both the genetic syndrome and the specific sleep problems in designing future studies. While combining all individuals with IDD may mask sleep problems more common to a specific genetic syndrome, targeting a specific sleep problem across pre-specified syndrome groups may be an appropriate methodology for larger scale studies targeting a treatment for that sleep problem.

The literature on sleep in IDD has generally encompassed a wide age range. This feature of research is not unusual given the rarity of some genetic syndromes (Rosen et al., 2011). However, it is a concern worth noting in future research. There is a need to consider that sleep problems present in early childhood may vary from those present in school-age children, adolescents, and adults, and that variance may depend on the specific genetic syndrome. In SMS sleep changes with age may be less of a concern as sleep problems are related to inverted melatonin rhythm which likely persists across age. In contrast, behavioral sleep problems in younger children with DS will differ from sleep problems associated with the onset of dementia or mental health concerns in adults with DS. Thus, future research is cautioned to consider the age of the individuals sampled for sleep problems. Otherwise, studies that span the lifespan are likely to mask that rates of different sleep problems will vary across the lifespan of individuals with specific genetic syndromes. Surveys of sleep problems in individuals with IDD are also often cross-sectional. Future longitudinal research is warranted to assess changes in sleep problems across specific developmental periods.

There is a need for studies that assess the prevalence of sleep problems to consider treatments already in place. Prevalence estimates can vary based on the above mention syndrome and age heterogeneity in a sample. Prevalence estimates can also vary based on the ongoing use of effective behavioral or pharmaceutical interventions, or previous surgeries that may improve sleep disordered breathing. Discounting this information can contribute to diverse prevalence estimates in the research literature.

While research advances have recently been heavily focused on children with ASD, there continues to be a need to better understand sleep problems in the heterogeneous population of IDD in order to provide tailored interventions specific to the sleep problem. Sleep research on specific syndromes is advancing, but continually plagued by small sample sizes due to the low prevalence rates of the specific syndromes. Advances have also been made in the reporting of specific syndrome groups within larger heterogeneous samples (Braam, Didden, Smits, & Curfs, 2008b; Didden et al., 2002; Montgomery et al., 2004). However, few studies provide data on these smaller syndrome subgroups relative to syndrome-specific treatment outcomes. Presenting findings on small syndrome samples within a larger study of individuals with IDD, with the use of consistent sleep measures, supports the ability to combine findings across studies. Few studies are randomizing smaller syndrome subgroups to intervention groups (Montgomery et al., 2004). Future studies are encouraged to randomly assign genetic syndromes within their sample (block randomization) to treatment groups, in an effort to support the ability to infer treatment effects.

Recommendations to Advance Evidence-Based Treatment

The evidence in support of sleep interventions for individuals with IDD or specific genetic syndromes is limited. Extinction and graduated extinction are probably efficacious for common sleep problems in individuals with IDD (Montgomery et al., 2004; Richdale & Wiggs, 2005; Thackeray & Richdale, 2002). Evidence for these behavioral interventions is consistent with the general population, where extinction, graduated extinction and early intervention/parent education meet criteria for being well-established interventions, and scheduled awakenings for being probably efficacious among children with bedtime refusal and frequent night waking (Kuhn & Elliott, 2003). However, a recent meta-analysis was unable to include many studies evaluating treatment interventions with individuals with IDD due to limited information on outcome measures consistent across studies (Meltzer & Mindell, 2014).

As with any intervention, it is important to ensure that the intervention is tailored to the condition. In the case of sleep problems, while it is beneficial to understand what types of sleep problems are more common in specific genetic syndromes or disorders, it is imperative to match the sleep intervention to the specific sleep problem. In pursuing larger scale randomized control trials of sleep interventions, our field is encouraged to balance the type of sleep problem and underlying contributing cause with the type of genetic syndrome in order to advance our knowledge of evidence-based treatments. Syndrome groups may be worth including within block randomization to ensure that various syndrome groups are equally allocated to treatment or control groups. By expanding the sample eligible to study interventions and including genetic syndromes within the study design, sufficient samples can be collected to assess efficacy of other sleep interventions.

Summary

Our understanding of sleep problems in individuals with IDD and specific disorders or genetic syndromes continues to grow, and our field continues to make use of advances in sleep assessment and diagnostic criteria. Although sleep problems contribute to difficulties with learning, attention, affect regulation, memory, and daytime behavior problems, the use

of individualized treatment plans that build on established behavioral or medical sleep approaches and approved pediatric medications have improved the lives of many individuals and families. The sleep assessment gold standard, PSG, continues to be a challenge for some individuals with IDD but recent advancements in home-based PSG and no-contact or minimal-contact sleep assessment methods are improving clinical diagnostic options. Studies of sleep and IDD include diverse sleep assessment methods, ages, diagnostic criteria, and treatment approaches. Despite this, findings within and across disorders/syndromes are building consensus. Overall, sleep problems are common and treatable in individuals with IDD and for some syndromes/disorders (e.g., SMS, DS) our understanding of the physiological or anatomical mechanisms have greatly improved treatment approaches. The impact of sleep on outcomes for individuals with IDD cannot be understated and continues to warrant ongoing research to improve how we measure sleep problems, how we report findings to improve comparisons across studies, what are the associated risk factors and outcomes of sleep problems, what are developmental changes in sleep problems over the lifespan, and what sleep interventions are effective for what specific sleep problems.

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