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A review of the nature and treatment of sleep disorders in individuals with developmental disabilities

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Abstract

This paper describes research on the prevalence, correlates, and treatment of sleep disorders in individuals with developmental disabilities. A significant number of individuals with developmental disabilities have disordered sleep, although prevalence estimates vary from 13% to 86%. Constitutional variables, including age, presence of physical and sensory impairments, and certain genetic syndromes and medical conditions appear to be related to sleep disturbance, but the evidence is mainly correlational and therefore cannot be said to show a causal relation. While a number of behavioral interventions have proven effective in the treatment of sleep disturbance, and drug therapy involving melatonin appears promising, epidemiologic work on the correlates of sleep disorders appear to have had little impact on treatment. Consideration of the nature of sleep disorders may enhance treatments for individuals with developmental disabilities. © 2001 Elsevier Science Ltd. All rights reserved.

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1. Introduction

During the past 10 years, a growing number of studies have evaluated the prevalence, correlates, and treatment of sleep disorders in individuals with developmental disabilities. This paper reviews this growing literature with the aim to gain a better understanding of the nature of sleep disorders in persons with developmental disabilities. Consideration of the extent and circumstances of sleep disorders that affect individuals with developmental disabilities may lead to new and more effective treatments.

Although the prevalence and characteristics of sleep disorders have been fairly well documented in typically developing children and adults, research on sleep disorders among individuals with developmental disabilities has only recently begun. Given this recent and growing database, a review of the literature seems timely. Accordingly, one aim of the paper is to review the results of epidemiologic surveys that have examined the prevalence and characteristics of sleep disorders among individuals with developmental disabilities. A review of this literature is important for gaining a better understanding of the nature of sleep disorders. A second aim is to review constitutional and environmental variables associated with sleep disorders. A third aim of this paper is to describe treatment strategies that have been used to address sleep disorders in individuals with developmental disabilities. There appears to be only one recent review on the treatment of sleep disorders in individuals with developmental disabilities (Lancioni, O'Reilly & Basili, 1999).

2. Definition and measurement of sleep disorders

Sleep disorders are classified into two major categories: dyssomnias and parasomnias (Stores, 1999). Dyssomnias include a number of primary sleep disorders. Examples of primary sleep disorders include difficulty initiating or maintaining sleep (i.e., delayed settling, frequent night waking), early morning waking, and excessive sleepiness during the day. Dyssomnias can be further classified as either intrinsic or extrinsic depending on the presumed origin of the disorder (Stores, 1999). Intrinsic sleep disorders are presumed to arise from within the body. Some individuals may experience seizures, sleep apnea or involuntary limb movements, for example, that lead to frequent waking at night. Extrinsic disorders are those that can be attributed to external factors, such as a sleeping environment that is too warm, cold, light, or loud. Extrinsic disorders might also be related to a disrupted circadian sleep-wake cycle.

Parasomnias are abnormal events that interrupt sleep. Common types of parasomnias include teeth grinding, enuresis, nightmares, and headbanging. Parasomnias are often further classified depending on when they occur during the sleep process. For example, some parasomnias (e.g., sleepwalking) may arouse the person from deep sleep, whereas other problems are perhaps more likely to

occur during REM-sleep (e.g., nightmares) or during the transition to sleep (e.g., headbanging).

In addition to parasomnias and dyssomnias, numerous associated problems may also emerge when sleep is disturbed. The individual may become excessively tired and irritable during the day. Behavioral manifestations of sleep disorders can include increased aggression and other behavior problems and disruptive behaviors related to going to sleep, such as crying, bedtime tantrums, calling out, screaming, co-sleeping, and leaving the bed (Quine, 1992).

As discussed in more detail below (see Limitations), while some studies have used direct measures such as EEG-recordings or actigraphy and direct observation with time sampling to evaluate sleep disorders, the majority of the studies used indirect measures such as surveys and questionnaires. Moreover, different criteria have been used to identify sleep disorders.

3. Prevalence and characteristics of sleep disorders

3.1. Prevalence

Several studies have investigated the prevalence of sleep disorders in children and adults with developmental disabilities. Sleep disturbance was a major problem for 13% to 86% of the samples. Percentages have varied depending on the individuals' age, living environment, type of sleep disorder, and measures used, as well as caregivers' interpretation of the survey items, their expectations, levels of tolerance, and general perceptions as to what constitutes disordered sleep.

Bartlett, Rooney, and Spedding (1985), for example, studied 214 children with severe mental retardation. All of the children lived at home. Eighty-six percent of children under 6 years of age had a reported sleep disorder. Slightly lower but still fairly high percentages of children from 6 to 11 years old (81%) and from 12 to 16 years (77%) were said to have some type of sleep disorder. The specific types of sleep disorders that were reported by parents included waking at night (56%), problems in settling to sleep (56%), and difficulty in getting the child to go to bed (53%).

Quine (1991) completed a 3-year longitudinal survey of 200 children with severe mental retardation who ranged from 1 to 18 years of age. Sleep disorders were assessed initially, and the assessment was repeated 3 years later. At the initial assessment, 51% of the parents reported settling difficulties in the child, while 67% reported frequent night waking. Fifty percent to 75% of the children who exhibited sleep disorders when first assessed still showed these problems 3 years later.

In one of the few studies to include a control group of typically developing children, Richdale, Gavidia-Payne, Francis, and Cotton (2000) examined scores on several indices of sleep disorders on 52 children with mild to profound mental retardation and compared these scores to those from a control group. Overall, Richdale et al. found sleep disorders in 58.6% of the children, and between-group

analyses revealed that both past (66.7%) and present (57.7%) sleep disorders were more common in children with developmental disabilities than in typically developing children.

Finally, Poindexter and Bihm (1994) used direct observation to assess the prevalence of short sleepers in 103 individuals with profound mental handicap living in an institution. The mean age for the group was 29.8 years with a range of 11 to 58 years. In this study, direct observations were made every 30 min to 60 min over a 21-month period to determine whether an individual was awake and up, awake and in bed, awake and in bed but restless, or asleep. A short sleep night was defined as five or less hours of sleep per night, and individuals were short-sleepers if they showed one or more short-sleep nights per week on average. From these observations, 39% of the individuals were identified as short sleepers.

3.2. Sleep patterns

Sleep patterns refer to a profile of the amount, timing, and quality of sleep that is typical for any given individual over a period of time. Comprehensive analyses of sleep patterns should incorporate polysomnographic recordings (e.g., actigraphy, EEG) during sleep. However, only a few studies have evaluated sleep patterns of individuals with developmental disabilities (cf. Espie et al., 1998). Espie et al. (1998) investigated the sleep patterns of 28 adults with severe to profound mental retardation and seizure disorder. Twelve individuals lived in a family home, three in a community home, and thirteen in a residential facility. Data were collected for seven nights by caregivers who completed sleep diaries. In addition, EEG-recordings were obtained on one night. Overall, these adults spent approximately 42% of each 24-hr period in bed, but the quality of their sleep was questionable. Only 12 individuals (43%) evidenced any REM-sleep, and total REM-sleep was only about 30 min on average. NonREM-sleep was markedly impoverished. While organic factors (e.g., brain damage) could be related to reduced REM-sleep, an alternative explanation is that the frequent use of antiepileptic medications and other drugs (e.g., benzodiazepines) in this population might suppress the duration of REM-sleep.

In a study utilizing direct observation only, Carr and Neumann (1999) conducted hourly checks from 9 p.m. to 7 a.m. for 471 individuals with severe to profound mental retardation. Observers recorded the total number of hours each person spent in bed, as well as the total number of hours of sleep, total amount of interruption (by toileting or position changes), and the total number of nights with insomnia. The main dependent variable was the sleep efficiency index (SEI), which refers to the total number of hours of sleep minus the amount of time spent awake, divided by the number of nights. The higher the SEI, the more efficient the sleep cycle (i.e., lesser fragmentation and shorter sleep latency). They found an average SEI of 7.17 hr (standard deviations were not given) for their sample, a pattern that was almost one hour less than is recommended for adults (Hobson, 1989).

Results from one study suggested that the sleep patterns of children and adults with developmental disabilities may differ from those of age-matched peers (Piazza, Fisher & Kahng, 1996). Over an approximate 3-week period, 51 individuals from 3 to 21 years of age, who had been admitted to an inpatient unit for the treatment of daytime problem behavior, were observed briefly at half-hour intervals to determine whether the person was asleep or awake. Individuals with developmental disabilities had less total sleep and more nighttime disruptive behaviors than did their nonhandicapped peers. Average duration of night waking was 48.3 min, and, on average, these individuals required 69 min to fall asleep. However, total number of hours of sleep per day decreased with age, which was similar to peers.

3.3. Limitations

In considering the general trends from epidemiologic surveys and studies on sleep patterns, several limitations and methodological issues should be highlighted. First, most epidemiologic studies used indirect assessment (i.e., surveys, questionnaires) to collect data. While this approach is legitimate, the resulting data are often of questionable reliability and validity. For example, Bruni, Cortesi, Giannotti, and Curatolo (1995) analyzed parent's logs of sleep behavior of the children and concluded that parents missed frequent, brief seizure-related awakenings. Espie et al. (1998) found that caregivers overestimated sleep quality when their ratings were compared to actual sleep as measured by EEG, and Hering, Epstein, Elrey, Iancu, and Zelnik (1999) found that questionnaire data led to an overdiagnosis of abnormal sleep patterns relative to the use of actigraphy. Unfortunately, only a few studies have used more direct measures such as observations and actigraphy to examine sleep disorders and patterns.

Second, different studies often used different criteria or cut-off points to define a sleep disorder. This makes comparison across studies difficult. For example, in defining early waking, Wiggs and Stores (1996) used a 5 am cut-off, whereas Brylewski and Wiggs (1998) used 6 am. Piazza et al. (1996), in contrast, define early waking as a period in which the individual is awake for at least 1 hr and during which the individual does not re-initiate sleep before scheduled wake-up time. Third, our attempts to synthesize the literature were hindered by the fact that the studies often included individuals with a mix of etiologies. Finally, the relatively low response rates (percentage of returned questionnaires) in most studies also limited the generality of the conclusions.

Despite these methodological limitations, the emerging database clearly indicates that sleep disorder is a major problem for many individuals with developmental disabilities. Good evidence is also emerging on the types of sleep disorders that are common in this population. In the next section, we review studies that provide evidence on variables that appear to be associated with sleep disorders in individuals with developmental disabilities.

4. Variables associated with sleep disorders

In this section, we summarize research findings on the relation between various constitutional (e.g., age, diagnosis, medical conditions) and environmental variables and sleep disturbance in individuals with developmental disabilities.

4.1. Age

Results of studies on the relation between sleep disorders and age are equivocal. Quine (1991) found that a significantly higher proportion of children under age 5 exhibited night waking problems, and Wiggs and Stores (1999) found a significant positive correlation between age and the duration of childhood sleeping problems. Among a sample of adults who lived in a community home, a difference in mean age was found between those with and without sleep disorders (Brylewski & Wiggs, 1999). Those with sleep disorders had a mean age of 49.6 years ($SD = 15.7$), whereas those without a sleep disorder had a mean age of 45.2 years ($SD = 14.6$). On the other hand, Bartlett et al. (1985) found similar high rates of sleep disturbance across a range of age groups. Another study found that age was not significantly related to measures of sleep quantity and sleep quality in developmentally disabled children with and without autism (Schreck & Mulick, 2000). Further complicating the picture are the results of Carr and Neumann (1999) who found no age-related differences in the mean SEI of their sample.

4.2. Disability and Other Diagnoses

4.2.1. Level of mental handicap

The evidence on the relation between level of mental handicap and sleep disturbance also is equivocal. Clements, Wing, and Dunn (1986) and Richdale, Cotton, and Hibbitt (1999) found no association between the presence or severity of sleep problems and level of mental retardation. In contrast, Piazza et al. (1996) found a significant correlation between the amount of appropriate sleep and both IQ and expressive language scores. Similarly, Espie and Tweedie (1991) found that adults with severe/profound mental retardation spent twice as much time awake at night than did adults with mild/moderate mental retardation.

4.2.2. Genetic disorder and other syndromes

Several studies have provided data on the prevalence and characteristics of sleep disorders for specific genetic syndromes and autism associated with developmental disabilities. For example, excessive daytime sleepiness appears to be prevalent among individuals with Prader-Willi syndrome (PWS). Cassidy, McKillop, and Morgan (1990), for example, conducted a study among 25 individuals with PWS who were between 20 months and 42 years of age (mean age = 13 years). Daytime somnolence was reported in 52% of the sample. Other frequently reported problems included daytime napping after age 5 years (89%), snoring (44%), and restless movements during sleep (40%). Another study showed a

higher prevalence of sleep disorders among individuals with PWS than among nonhandicapped peers (Richdale et al., 1999). Helbing-Zwanenburg, Damen, and Kamphuisen (1992) recorded the sleep-wake continuum of 13 individuals with PWS (mean age in years: 31) during 48 hr. Polygraphic measures were compared to those recorded with 19 nondisabled individuals. Significant differences between groups were found for measures of sleep-onset REM, which was five times more common in the PWS group than in the control group. These results suggest a higher prevalence of narcolepsy in individuals with PWS.

Research findings also indicate a high prevalence of sleep disorders among individuals diagnosed with other syndromes such as Angelman, Smith-Magenis, and San Filippo. Genetically, Angelman syndrome (AS) is closely related to PWS. Clarke and Marston (2000) examined sleep disturbance in 73 children and young adults with AS (15 q) who were severely mentally handicapped. Forty-two percent of the sample was reported to experience disordered sleep. Smith, Dykens, and Greenberg (1998) investigated sleep and nighttime behaviors among 39 individuals with Smith-Magenis syndrome who ranged from 1.5 to 32 years of age (mean = 10.5 years). The most commonly reported forms of disruptive nighttime behaviors were settling problems, delayed sleep onset, bedtime refusal, bedwetting, snoring, teeth grinding, and apnea attacks. The level of parental concern about these behaviors seemed significant because 59% of the individuals were given medication to facilitate sleep. Colville, Watters, Yule, and Bax (1996) investigated sleep disorders in 80 children with Sanfilippo syndrome from 4.4 to 25.6 years of age (mean: 10.2 years). Seventy-eight percent had an identified sleep problem, and the problem was considered severe for 46% of these children.

Only a few studies have investigated the prevalence of sleep disorders in children with autism. In a study by Patzold, Richdale, and Tonge (1998), sleep disorders were assessed in 38 children with pervasive developmental disorder (i.e., autism and Asperger's syndrome), most of whom had a mental handicap. It was found that 63.2% and 76.3% had current and past sleep problems, respectively. Frequent night waking, restless sleep, and bedtime problems were reported in 23.3%, 72.2%, and 32.8% of the cases, respectively. Means of sleep latency and duration in min of night waking were 32 min (SD = 24.4) and 30.2 min (SD = 35), respectively. That the sleep-wake pattern of children with autism may be different from the pattern of typically developing children and developmentally disabled children without autism, was suggested in a recent study by Hering et al. (1999). Twenty-two children with autism, aged 3 to 12 years, participated. Sleep questionnaires and actigraphy were used to investigate sleep patterns. Scores on several parameters of sleep (e.g., sleep onset time, sleep duration) were compared with a control group of typically developing children. There were no differences between the groups, except for sleep offset time (early waking). Children with autism woke one hour earlier than the controls. In general, however, sleep patterns of autistic children were similar to those of the typically developing children.

4.2.3. Medical condition

The most frequently reported medical conditions associated with sleep disorders were recurrent ear infections (87%), constipation (53%), allergies/eczema (45%), and other types of infection (45%) (Smith et al., 1998). Quine (1991) and Brylewski and Wiggs (1998) also found an association between epilepsy and sleep problems. Children and adults with sleep disorders were more likely to suffer from epilepsy than children with no sleep disorders. Poindexter and Bihm (1994), however, found that epilepsy was not associated with short-sleep patterns.

4.2.4. Breathing-related problem

Sleep apnea appears to be relatively common in children with developmental disabilities and may be related to seizure disorders. Koh, Ward, Lin, and Chen (2000), for example, found that treatment of sleep apnea led to improved seizure control in five of nine children with neurodevelopmental disorders. Marcus, Keens, Bautista, von Pechmann, and Ward (1991) found evidence for obstructive sleep apnea and other breathing-related sleep disorders in a group of 53 children with Down syndrome. However, Richdale et al. (1999) found no relationships among sleep disorders, snoring, and apnea in 52 children with mental handicap.

Obstructive sleep apnea may be related to excessive daytime sleepiness which is often reported in individuals with PWS, although results are contradictory. For example, contrary to the findings by Cassidy et al. (1990), Vela-Bueno et al. (1984) found no evidence of sleep apnea (central, peripheral, or mixed) among eight individuals with PWS, although severe hypoventilation during sleep was found in one case.

4.2.5. Sensory handicap

Poindexter and Bihm (1994) found that institutionalized adults who were blind or deaf/blind were less likely to be short sleepers than adults without such impairments. This is surprising when one considers the fact that light is a powerful external cue for the sleep-wake cycle, and sleep disturbance is common in blind people (Palm, Blennow & Wetterberg, 1991).

4.2.6. Physical handicap

Quine (1991) found that children with sleep disorders were more likely to have a diagnosis of cerebral palsy than children without sleep problems. In a study by Poindexter and Bihm (1994), cerebral palsy was inversely related to short-sleep patterns in that individuals with cerebral palsy were less likely to be short-sleepers.

4.3. Residential placement

Espie and Tweedie (1991) investigated sleep disorders in 120 adults with mild to profound mental retardation. In this sample, 57% lived in the community and 43% lived in a large residential facility. Mean sleep efficiency scores of these two

groups were 88% and 90%, respectively. While the mean sleep efficiency index did not differ significantly between groups, many individual scores for those living in the large facility indicated inefficient sleep (i.e., $< 85\%$). Espie and Tweedie and Espie et al. (1998) suggested that lowered sleep efficiency may be associated with institutionalized routines that do not coincide with individual sleep needs and preferences.

4.4. Adaptive behavior

Quine (1991) showed that sleep disorders were associated with frequent nighttime incontinence and adaptive skill deficits in a number of areas (i.e., communication, academic, and self-help skills). However, results of Quine (1992) indicated that incontinence was not related to severity of the sleep disorder. Brylewski and Wiggs (1998) concluded that individuals with nocturnal incontinence were likely to show more night waking, parasomnias, snoring, and excessive daytime sleepiness than individuals without incontinence.

4.5. Daytime problem behavior

Daytime problem behaviors may be associated with sleep disorders. Brylewski and Wiggs (1999) explored the association between sleep disorders and daytime problem behaviors in a sample of 205 adults with moderate to profound mental handicap who lived in community-based group homes. Results showed that individuals with sleep disorders scored significantly higher on three of the five subscales of the Aberrant Behavior Checklist (ABC) (Aman, Singh, Steward & Field, 1985) (i.e., Irritability, Stereotypy, and Hyperactivity). Specific topographies of problem behavior, such as self-injury, aggression, and screaming, were more severe in the sleep-disordered group. Wiggs and Stores (1996) also found that children with sleep disorders had higher scores on all factors of the ABC, except Inappropriate Speech.

In some cases, sleep disorders appear to influence self-injurious behavior (SIB). Symons, Davis, and Thompson (2000), for example, observed 30 adults with profound mental retardation with and without SIB. Repeated nighttime observations were used to determine whether an individual was asleep or awake. Adults with a history of SIB slept less than adults in the matched control group. They also found a higher variability in the number of intervals asleep across nights in adults with SIB than in controls. The authors hypothesized that there may be a reciprocal relation between the endogenous opioid system and REM-sleep disturbance, which leads to a cycle of SIB, opioid release, and sleep deprivation. This hypothesized relation between sleep disturbance and SIB is consistent with the fact that individuals who exhibited self-injurious behaviors had lower SEI scores when compared to individuals with mainly destructive and aggressive behaviors (Carr & Neumann, 1999).

Disordered sleep may represent an establishing operation for daytime problem behavior. This hypothesis was supported by a case study with a 31-year-old

severely mentally handicapped man who showed aggressive behavior (O'Reilly, 1995). It was found that daytime aggression was higher when the man was sleep deprived (i.e., < 5 hours) in comparison to when he had slept for more than 5 hr the night before. Similar relations between sleep deprivation and daytime occurrence of self-injury and aggression have been found (O'Reilly & Lancioni, 2000).

4.6. Caffeine intake

Intake of at least four mugs of tea or coffee after 6 p.m. was associated with sleep delays of more than 1 hr and a shorter sleep duration than those who drank less (Brylewski & Wiggs, 1998).

4.7. Maintaining (reinforcing) consequences

Results of several studies suggest that reinforcement contingencies may shape and maintain nighttime disruptive behaviors. For example, Bramble (1997) and Didden, Curfs, Sikkema, and de Moor (1998) showed that withholding parental attention following nighttime disruption resulted in relatively quick and lasting reductions in sleep disorder symptoms of children with developmental disabilities who lived at home. The type of consequence that maintains nighttime disruptive behaviors may vary across individuals and could include access to parental attention or preferred activities such as being allowed to stay up and watch television. In other cases, negative reinforcement may play a role in maintaining disruptive nighttime behaviors if such behaviors result in escape from nonpreferred stimuli, such as a dark bedroom, or avoidance of being placed into bed.

The variables that maintain disruptive nighttime behaviors may not necessarily be responsible for its emergence. Sleep disorders that occur naturally during infant development or that are related to medical problems such as an ear infection may come under operant control. In addition, fear-related sleep disorders may be classically conditioned, such as in cases where the individual experiences a traumatic event that is associated with bedtime or sleeping. For example, in one of our own studies (see Didden et al., 1998), a traumatic experience during the day (i.e., being stuck in an elevator) appeared to influence the emergence of settling problems in a 7-year-old boy with mild mental retardation. In this case, desensitization or gradual distancing (see Treatment section) was effective in normalization of the sleep-wake cycle within three weeks.

To summarize this section, we must conclude that the nature of the associations between the presence of sleep disorders and other variables remains unclear. These studies provide correlational data. Some of these variables may be both a cause and an effect or neither a cause nor effect of sleep disorder. For example, sleep disorders may cause daytime problem behaviors, and vice versa. Furthermore, both variables may be related to an unknown third variable, such as medical problems (e.g., epilepsy). Despite uncertainties about the nature of these

associations, identification of such variables is important in that this may enable one to predict the likelihood of sleep disturbance and to design effective treatments.

5. Treatment

An important consideration in treatment is analysis of the variables that set the occasion for and maintain sleep disturbance and sleep related problems. In terms of general treatment strategies, it is possible that the manipulation of variables associated with sleep disorders may lead to improved sleep. Variables outlined in the previous section that possibly could be manipulated include reducing caffeine intake, treating medical conditions, altering the environment to make it more conducive to sleep, and teaching adaptive behavior (e.g., nighttime continence). However, empirical studies on the effects of such manipulations are almost lacking. In one relevant study, Carr and Neumann (1999) assessed the effects of an individualized sleep monitoring program on the SEI in 471 profoundly to mildly mentally handicapped individuals who lived in a large residential setting. A graphic sleep monitoring system where staff were instructed to modify each individual's bedtime to coincide with his or her apparent sleep preferences resulted in a durable increase in mean SEI-indices for all levels of mental handicap. These types of antecedent manipulations would appear to hold considerable promise for the treatment of sleep disorders in individuals with developmental disabilities, and more research is clearly warranted.

In terms of the existing empirical evidence, several strategies have been shown to be effective in the treatment of sleep disorders in children and adults with developmental disabilities. Most of these strategies can be classified as behavioral, including chronotherapy, bedtime fading, extinction, gradual distancing, sleep-wake scheduling, and light therapy. There have also been a few promising reports on pharmacological intervention involving the use of melatonin.

5.1. Behavioral Procedures

5.1.1. Chronotherapy

Chronotherapy is a treatment designed to synchronize the endogenous circadian cycle with the daily schedule. It involves systematically delaying bedtime until the individual goes to bed and falls asleep at a desirable time. Chronotherapy is hypothesized to be effective because it capitalizes on natural carcadian drift. Research in this area has shown that the length of the human circadian cycle (i.e., the internal clock that regulates the sleep-wake cycle) is approximately 1 hr longer than the 24-hr day (see e.g., Hobson, 1989). The body must therefore reset its internal clock each day in order to maintain a regular sleep-wake cycle. Sleep problems are conceptualized as a failure to synchronize the body's internal clock to external stimuli and in the absence of such cues, sleep onset occurs approx-

imately one hour later each day due to circadian drift. Chronotherapy has been used with success for nonhandicapped adults with delayed sleep phase insomnia. In a case study, Piazza, Hagopian, Hughes, and Fisher (1998) demonstrated the effectiveness of chronotherapy with an 8-year-old girl with severe autism who exhibited irregular sleep onset times, frequent night and early waking, and short total sleep times. Piazza et al. conclude that this type of treatment may be useful for individuals who display irregular sleep-wake patterns for whom other treatments have been unsuccessful. Chronotherapy has also been used successfully with four congenitally blind children aged 4–12 years with moderate to severe intellectual disability (Okawa et al., 1987).

5.1.2. Bedtime fading

With bedtime fading, baseline data are used to determine a time when rapid sleep onset is highly probable. In contrast to chronotherapy, bedtime is made earlier over time. Initial bedtime during treatment is determined by calculating the average sleep onset time during baseline and then adding 30 min whereby the individual is not allowed to fall asleep prior to this time. Piazza and Fisher (1991), for example, used fading with four children with profound intellectual disabilities who showed various types of sleep problems. Fading consisted of adjusting the child's bedtime by 30 min each night based on latency to sleep onset for the previous night. If the child fell asleep within 15 min of bedtime, this time was then 30 min earlier on the next night. However, if the child did not initiate sleep within 15 min, bedtime was made 30 min later on the subsequent night. A response cost component, in which the child was removed from bed and was kept awake for one hour if sleep was not initiated within 15 min, also was used. According to Piazza and Fisher, the mechanisms responsible for this treatment's effectiveness may involve both classic and operant conditioning. From the classic conditioning framework, the unconditioned stimulus was the physiological state associated with sleep deprivation. From the operant side, failure to sleep was punished via the response cost procedure.

5.1.3. Extinction

Results from studies that used ignoring as treatment suggest that nighttime disruptive behaviors may be positively reinforced by caregiver attention (see e.g., Bramble, 1997; Didden et al., 1998). Two types of extinction procedures, nongraduated and graduated extinction, have been used. With nongraduated extinction, parents were instructed to ignore the child's nighttime disruptive behaviors after they had put the child into bed and left the bedroom. One problem with this procedure is that the disruptive behavior may escalate in severity to the point where the child is in danger of doing serious injury or damage that can not be ignored. However, if parents can weather the storm, nongraduated extinction can rapidly eliminate sleep problems in children with developmental disabilities (Bramble, 1997; Didden et al., 1998).

Graduated extinction involves gradually increasing the delay interval before parents (or other caregivers) attend to the child. On each successive night, the

interval is increased a certain number of minutes until the target behaviors are no longer followed by parental attention. This procedure has also proven to be highly effective in eliminating nighttime disruptive behavior (see Durand, Gernert-Dott & Mapstone, 1996). Results from this study suggested that inadvertent reinforcement of lengthier bouts of nighttime disruptive behaviors did not occur.

5.1.4. Gradual distancing

Howlin (1984) described a stimulus fading procedure called gradual distancing with a 5-year old boy with autism. He required a long time to fall asleep, and his mother would often sit with him an hour or more before he fell asleep. He woke up nightly, at varying times and would not settle unless he was taken into his parents' bed. At the time of the initial assessment, his mother slept all night in the boy's bed. The procedure required that the mother gradually increase her sleeping distance from the child. During the first week, she slept on an inflatable mattress next to the child's bed. The distance between the boy's bed and mother's mattress was then increased gradually. Each time the boy woke, mother would comfort him. During the course of treatment, the mattress was placed immediately outside the child's bedroom. At the end of treatment the mother was able to sleep in her own bedroom. During a 3-month follow-up, the number of times the child woke up at night was reduced to only 3–4 times per week. Gradual distancing seems to be indicated in case of fear-related sleep disorders although validation of fear would seem problematic.

5.1.5. Sleep-wake scheduling

Sleep-wake scheduling refers to setting a fixed time schedule for going to bed and a fixed time for waking. With such a procedure, a threshold time (i.e., observed sleep readiness) is calculated taking into account average sleep requirement. Individuals are put to bed if they exhibit evidence of tiredness, and daytime and evening naps are prevented. Morning rising time is set as an anchor each day. Finally, the threshold time is adjusted until 90% sleep efficiency is achieved. In a study by Espie and Wilson (1993), this type of scheduling was effective in reducing sleep problems in five children and adults with mild to profound mental handicap. Effective use of this procedure would seem to depend on being able to find an optimal sleep-wake schedule for the individual.

5.1.6. Light therapy

Only two uncontrolled case studies have been published on the effects of light therapy. Short and Carpenter (1998) described the use of light therapy with a 34-year-old man with profound mental retardation and vision impairment. His sleep disorders had emerged acutely and consisted of difficulties in initiating sleep and frequent night waking. During light therapy, he was awoken at a set time in the morning and exposed to direct natural light for at least 2 hr by taking him outdoors. Daytime sleeping was discouraged. Treatment resulted in a nor-

malized and stable sleep pattern within two weeks of treatment. Smith et al. (1998) also presented case study evidence of the effectiveness of light therapy in the treatment of sleep problems with a 6-year-old child with Smith-Magenis syndrome who had a history of sleep and other behavioral problems during the winter months. During light therapy, the child was exposed to light for 20 min each morning at 6 a.m. The mechanism underlying the effectiveness of this procedure was considered to result from synchronization of the child's internal biological clock by exposure to simulated sunlight. However, it is difficult to isolate the specific effects of light therapy in these case studies because scheduling was an integral part of treatment.

5.2. Pharmacological use of melatonin

In the dark and at night, the pineal gland synthesizes and releases melatonin. The hypothalamic circadian clock, which is located in the suprachiasmatic nuclei (SCN), regulates the pattern of melatonin synthesis in humans. The SCN has been found to be a major sleep regulator. The light-dark cycle is the most powerful synchronizer known to entrain the endogenous circadian pacemaker on a 24-hr cycle (see e.g., Hobson, 1989). Melatonin levels are high in darkness and low in light. It is hypothesized that endogenous melatonin synthesis is dampened in individuals with visual handicaps and in individuals with developmental disabilities or brain damage.

Palm et al. (1991) described a case involving the use of melatonin with a 9-year-old boy who was blind and severely mentally retarded. Despite a structured bedtime routine with a consistent morning wake time, the child showed a disturbed non-24 hr sleep-wake cycle. Treatment began with 0.5 mg oral melatonin given at 6 p.m. The use of oral melatonin resulted in a dramatic improvement to a typical sleep-wake cycle with the boy having generally uninterrupted sleep from 7 p.m. to 6 a.m. The results of a double-blind, cross-over randomized control trial by Jan, Espezel and Appleton (1994) were consistent with those results. They showed that melatonin was highly effective in establishing a normalized sleep-wake rhythm with 15 young children, some of whom were diagnosed with developmental disabilities. Melatonin was given orally at the desired bedtime at dosages between 2.5 mg and 5 mg daily. Palm, Blennow and Wetterberg (1997) obtained similar results in a series of case studies with 8 developmentally disabled children who were blind. In contrast, Camfield, Gordon, Dooley, and Camfield (1996) found no marked improvement for six children with developmental disabilities who were treated for sleep disturbance using melatonin as compared to within-subject placebo control conditions. Three children did, however, show some improvement in certain aspects of sleep (e.g., fewer arousals per night). Thus, while melatonin appears promising, it may not produce dramatic effects for all children.

In considering the current state-of-the-art in treatment, we agree with Lan-

cioni, O'Reilly, and Basili (1999) that systematic replication of studies is needed, especially with regard to new procedures such as chronotherapy and light therapy. Furthermore, no studies have demonstrated a functional relationship between nighttime disruptive behaviors and its negative and/or positive reinforcers. Procedures for the functional analysis of sleep-related problems are needed to identify variables that may shape and maintain sleep disorders. Finally, quantitative data collected during baseline and treatment in published case studies could permit researchers to calculate effect-sizes as part of meta-analyses (see e.g., Didden, Duker & Korzilius, 1997). Results of such analyses might reveal differential effectiveness of procedures for treatment of sleep disorders in our target group.

6. Conclusion

Our review has shown that sleep disorders are highly prevalent and persistent in individuals with developmental disability. Indeed, compared to nonhandicapped persons, individuals with developmental disabilities appear to be at increased risk for sleep disorders. This difference in prevalence and severity may be explained in part by brain damage. In addition, some sleep disorders appear to be associated with certain syndromes, suggesting that sleep problems could be a behavioral phenotype. However, the association between specific genetic disorders and sleep problems is not yet well understood. To assist in developing new and more effective treatments, an understanding of the variables associated with sleep disorders in persons with developmental disabilities may be helpful. The correlational studies reviewed in this paper point to a number of variables that may be associated with sleep problems, but it remains unclear if these factors have predictive validity for treatment selection or treatment outcome.

There is thus a current literature that professionals can consult when providing services to caregivers of individuals with developmental disabilities who have sleep disorders. However, this literature is limited in that most of the treatment studies have been case studies with limited controls for internal validity, and limited data on generalization and maintenance. There is also a need for systematic replication of treatment studies so that the external validity of treatments can be more fully evaluated. Larger-scale controlled studies are certainly warranted, as are studies that examine the long-term effects and side effects of various treatment procedures. In addition, studies that seek to elucidate factors that are necessary for effective implementation are needed to provide evidence-based practice in this area. Finally, while effective implementation of evidence-based treatments not only eliminate sleep disorders, it is plausible that early intervention to establish appropriate sleeping patterns may help to preempt sleep disorders and the many negative consequences associated with the lack of efficient sleep in individuals with developmental disability.

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