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Examining Sleep Problems in Children with Autism Spectrum Disorders

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EXAMINING SLEEP PROBLEMS

IN CHILDREN WITH AUTISM SPECTRUM DISORDERS

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ABSTRACT

EXAMINING SLEEP PROBLEMS IN CHILDREN

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May 2011

Dissertation supervised by Tammy L. Hughes

Sleep problems are prevalent in children with neurodevelopmental disabilities such as autism spectrum disorders. Inadequate sleep can negatively affect the child's daily functioning as well as serve as a source of stress for the child's family. Although there is a growing body of literature aimed at improving sleep difficulties in children with disabilities, there is less known about sleep difficulties in children with autism spectrum disorders and a particular paucity in regard to treatment for sleep problems. The purpose of this paper was to investigate the effectiveness of a behavioral treatment package designed to reduce bedtime resistance and night awakenings in children and apply this technique to children with autism spectrum disorders.

DEDICATION

This is dedicated to all of those who have suffered sleepless nights, and the loved ones who cared for them.

ACKNOWLEDGEMENT

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LIST OF ABBREVIATIONS

American Psychiatric Association (APA)

Asperger's Disorder/Syndrome (AD)

Attention Deficit/Hyperactivity Disorder (ADHD)

Autism Spectrum Disorder (ASD)

Behavior Assessment System for Children – Second Edition (BASC-2)

Behavioral Evaluation of Disorders of Sleep (BEDS)

Childhood Disintegrative Disorder (CDD)

Children's Sleep Habits Questionnaire (CSHQ)

Developmental Delay (DD)

Diagnostic and Statistical Manual of Mental Disorders, Fourth Edition, Text Revision

(DSM-IV-TR)

Food and Drug Administration (FDA)

Intellectual Disabilities (ID)

Mental Retardation (MR)

Neurodevelopmental Disorder (NDD)

Parenting Events Questionnaire (PE)

Pervasive Developmental Delay (PDD)

Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS)

Teacher Report Form (TRF)

CHAPTER I

INTRODUCTION

The mystery of sleep has been pondered for thousands of years. Although

modern sleep medicine has heralded many discoveries and advances, the function of

sleep remains fully unknown. How is sleep necessary for the brain and the body?

Multiple theories propose the possibility of physical and psychological restoration,

energy conservation, physical growth, brain development, particularly during infancy,

memory consolidation, emotional release, and adaptive functions (e.g. maximize safety).

However, no one theory has yet adequately accounted for all of sleep behavior or

functioning.

Although the function of sleep continues to be studied and debated, researchers

have convincingly demonstrated adverse effects of sleep loss. Studies of sleep

deprivation and fragmented sleep document adverse effects on physical health, memory,

cognition, attention, emotions, motivation, and mood (Dahl, 1996a; Gomez, Bootzin, &

Nadel, 2006; Gozal, 1998; National Sleep Foundation, 2009; Touchette, Petit, Seguin,

Boivin, Tremblay, & Montplaisir, 2007; Walker & Stickgold, 2004; Walker & Stickgold,

2006; Walker, Stickgold, Alsop, Gaab, & Schlaug, 2005). Research in children with

obstructive sleep apnea (OSA) most dramatically demonstrates the relationship between

disrupted sleep and daytime impairments (Hansen & Vanderberg, 1997; Owens, Opipari,

Nobile, & Spirito, 1998). Sleep or lack thereof, has a significant impact on quality of

life.

Adequate sleep is essential for both children and adults, however, sleep is

particularly important to development in children. Dahl (1996b) argues sleep to be the

"primary activity of the brain" during the first two years of life and highlights sleep's "relatively central role in early maturational processes (p. 3)." In a longitudinal study of 1492 Canadian children, Touchette et al. (2007) found short sleep duration (e.g. less than ten hours per night), particularly before age 41 months, was associated with hyperactivity/impulsivity and decreased cognitive performance on neurodevelopmental tests. These findings concur with Sadeh, Gruber, and Raviv's 2002 study of 135 Israeli children in 2nd, 4th, and 6th grades. Children who were "poor" sleepers (e.g., awakened at least three times lasting five minutes or longer and/or at least 10% of the night was spent awake after sleep onset) performed more poorly than "good" sleepers on neurobehavioral tests, and had higher rates of behavior problems per parent report. In Tara and Potts-Datema's (2005) review of 20 articles investigating the association between sleep among school-age children and academic outcome, the majority of the studies showed children with sleep disorders or decreased sleep (compared to peers) had lower performance on cognitive tasks, problems with attention and concentration, and lower grades. Taken together there is a growing research base indicating sleep problems are detrimental to typically developing children.

Children with neurodevelopmental disabilities are particularly at risk for sleep problems, and prevalence rates range from 13% to 88% (Didden & Sigafoos, 2001; Jan & Freeman, 2004; Quine, 1991; Wiggs & Stores, 1996), depending upon the study definition. More specifically, children with Pervasive Developmental Disorders (PDD) frequently present with sleep problems with prevalence rates ranging from 44% to 86% (Cotton & Richdale, 2006; Couturier, Speechley, Steele, Norman, Stringer, & Nicolson, 2005; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Liu,

Hubbard, Fabes, & Adam, 2006; Polimeni, Richdale, and Francis, 2005; Richdale & Prior, 1995; Wiggs & Stores, 2004). Reported sleep problems include bedtime resistance, unplanned co-sleeping, delayed sleep onset, decreased sleep duration, night awakenings, sleep-disordered breathing, parasomnias (e.g. nightmares, night terrors, sleep walking, etc.), and daytime sleepiness (Cotton & Richdale, 2006; Courtier et al., 2005; Patzold, Richdale, & Tonge, 1998; Richdale & Prior, 1995; Schrek & Mulick, 2000; Wiggs & Stores, 2004). Similar to typically developing children, problems with initiating and maintaining sleep are among the most common difficulties in children with disabilities (Allik, Larsson, & Smedje, 2006a; reviewed in Stores & Wiggs, 1998; Richdale, 1999, 2001; Wiggs & Stores, 2004). However, in contrast to ratings by parents of typically developing children, parents more frequently categorize sleep problems in children with PDD to be severe (Couturier et al., 2005) and at times persistent into adolescence and adulthood(Oyane & Bjorvatn, 2005; Tani et al., 2003). Similar to typically developing children, there are many different pathways to sleep problems in children with autism. Richdale and Schreck (2009) propose a multifactoral model, hypothesizing sleep problems may occur as a result of: 1) intrinsic biological or genetic abnormalities that alter brain architecture or biochemistry; 2) psychological or behavioral characteristics connected with core or associated features of ASDs; or 3) factors in the family home or environment, including caregiver practices. Treatment for sleep problems also address different etiologies, with pharmacologic intervention affecting biochemistry and behavioral intervention targeting environmental variables which establish and maintain problematic behavior.

Intervention studies demonstrate improved outcomes in children identified with sleep problems. In a review of 52 behaviorally-based treatment studies from 1970 to 2005 (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006), consisting of a sample of over 2,500 children, 80% of the sample demonstrated clinically significant post-intervention decreases in bedtime resistance and night wakings. A smaller number of studies examining over-the-counter (OTC) and prescription medications have also demonstrated reduced sleep onset delay, and increased sleep maintenance and total sleep time, although limited efficacy and safety data exist (Owens and Moturi, 2009). Several behavioral and pharmacological studies in children with neurodevelopmental disabilities have also demonstrated reductions in disruptive bedtime behaviors post-intervention (Didden & Sigafoos, 2001; Lancioni, O'Reilly, and Basili, 1999). Behavioral and pharmacologic intervention studies in children with ASDs have demonstrated reduced sleep latency, a reduction in settling problems, night waking, and unplanned co-sleeping (Christodulu & Durand, 2004; Howlin, 1984; Johnson & Malow, 2008; Piazza, Fisher, & Sherer, 1997; Piazza, Hagopian, Hughes, & Fisher, 1998; Reed et al., 2009; Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005).

<center>Significance of the Problem</center>

Poor sleep patterns are stressful to parents (Goodlin Jones, Tang, Liu, & Anders, 2008; Honomichl, Goodlin Jones, Burnham, Gaylor, & Anders, 2002) and have a pronounced negative impact on family members. Parents of children with disabilities report greater levels of stress (Ha, Hong, Seltzer, & Greenberg, 2008; Webster, Majnemer, Platt, & Shevell, 2008) and higher levels of sleep disturbance than parents of typically developing children (Robinson & Richdale, 2004). Reflective of previous

<center>4</center>

studies, a recent sample of 46 mothers parenting children with developmental disabilities, sleep problems in their children predicted poor maternal sleep quality and was significantly associated with increased maternal depression and stress (Chu & Richdale, 2009).

Moreover, parents of children with autism report higher levels of stress than do parents of children with other psychiatric problems and developmental disabilities (Bouma & Schweitzer, 1990; Duarte, Bordin, Yazigi, & Mooney, 2005; Tomanik, Harrism, & Hawkins, 2004). Polimeni, Richdale, and Francis (2007) found over two-thirds of parents of children with autism or Asperger's Disorder reported their own sleep was disrupted due to their child's sleep difficulties. Meltzer (2008) found parents of children with ASDs reported poor sleep quality as well as an earlier morning wake time and shorter sleep duration. Conversely, improving children's sleep improves family well-being (Eckerberg, 2004). In examining the effects of improving sleep in young children with developmental disabilities, both Robinson and Richdale (2004) and Wiggs and Stores (2001) reported that improvements in children's sleep yielded increased parental satisfaction with their own sleep, their child's sleep, and decreased parental daytime sleepiness. In conclusion, the literature demonstrates typically-developing children, children with neurodevelopmental disabilities, and those with autism are similar in terms of the types of sleep problems encountered.

The most prominent sleep problem is insomnia, characterized by difficulty falling asleep and maintaining sleep, and early morning awakenings. Treatment options for insomnia in children with ASDs are similar to those in typically developing children, and consist of pharmacological, behavioral, or combined management. In their 2006 report,

the American Academy of Sleep Medicine (Morgenthaler et al.) calls for future behavioral treatment research for sleep problems in children with autism. In typically-developing children and children with neurodevelopmental disabilities, behavioral interventions have been proven effective (Morganthler et al., 2006) and are preferred by parents as first-line interventions (Wiggs and Stores, 1996; Williams, Sears, & Allard, 2006). Additionally, safety and efficacy data pertaining to pharmacologic intervention are emerging.

Several small-scale behaviorally-based interventions have been conducted in children with autism to target various sleep problems with promising success. Outcomes include reduced sleep latency, decreased settling problems, night waking, and unplanned co-sleeping, and improved parental satisfaction with the child's bedtime routine (Christodulu & Durand, 2004; Reed et al., 2009; Weiskop et al., 2005) Thus, previous behavioral-based sleep research can serve as a paradigm for future sleep intervention research in children with ASDs. To date, there are no intervention studies investigating the use of a bedtime story paired with positive reinforcement to decrease bedtime resistance and night waking in children with autism spectrum disorders.

Problem Statement

This purpose of this study was to clarify the efficacy and effectiveness of a bedtime story treatment to reduce sleep problems in children with autism. More specifically, this study investigated the impact of this behavioral technique in children with autism who demonstrate bedtime resistance and night awakenings. This study examined the following research questions:

Research Questions and Hypotheses

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1. Will the introduction of a bedtime story treatment package reduce bedtime resistance?

   Hypothesis: Bedtime resistance as reported by parents will decrease as a result of the intervention.

2. Will the introduction of a bedtime story treatment package reduce nighttime awakenings?

   Hypothesis: Nighttime awakenings as reported by parents will decrease as a result of the intervention.

3. Will the introduction of a bedtime story treatment package decrease sleep latency?

   Hypothesis: Sleep latency will decrease as a result of the intervention.

4. Will the introduction of a bedtime story treatment package increase total sleep time?

   Hypothesis: Total sleep time will increase as a result of the intervention.

5. Will improved sleep decrease daytime problem behavior?

   Hypothesis: Daytime behavior problems as measured by parents and teachers will decrease as the result of improved sleep.

6. Will the bedtime story treatment package be acceptable to parents?

   Hypothesis: The intervention will be acceptable to parents.

7. Will levels of behavior at the completion of the intervention be maintained after one month?

   Hypothesis: Levels of behavior at the completion of the intervention will be maintained after one month.

8. Will parents level of perceived stress decrease after the intervention?

   Hypothesis: Perceived stress will decrease after the intervention.

CHAPTER II

LITERATURE REVIEW

Overview of Autism Spectrum Disorders

The term *Pervasive Developmental Disorders* (PDD) refers to a class of neurodevelopmental disorders (NDD) described within the Diagnostic and Statistic Manual of Mental Disorders, Fourth Edition, Text Revision (DSM-IV-TR; APA, 2000). The classification of PDD includes four disorders: Autistic Disorder (autism), Asperger's Disorder (AD), Rett's Disorder, and Childhood Disintegrative Disorder (CDD). PDD-NOS is assigned when autistic symptomology is present, but criteria for autism or AD is not met due to age of onset or subthreshhold levels. Parents and professionals frequently utilize the term Autism Spectrum Disorders (ASD) to refer to the continuum of related symptoms encompassing autism, AD, and PDD-NOS. The boundaries among these disorders is not yet fully clear; however, they share a triad of neurological impairments including impairments in communication, impairments in social interaction, and restricted repetitive and stereotyped patterns of behavior, interests, or activities (APA, 2000). PDDs are considered pervasive as symptoms pervade all areas of development. Table 2.1 compares the developmental and behavioral manifestations and age of onset of symptoms:

Table 2.1

Pervasive Developmental Disorders

Diagnostic Category	Developmental and behavioral manifestations	Symptom onset
Autistic Disorder	• Impaired social interaction • Impaired communication • Restricted repetitive and stereotyped patterns of behavior, interests, and activities	Prior to age 3 years
Asperger's Disorder	• Impairments in social interaction • Restricted, repetitive, and stereotyped patterns of behavior, interests, and activities • No significant delay in language or cognition	Infancy or childhood
Pervasive Developmental Delay, NOS	• Social impairments in either verbal/nonverbal communication or with stereotyped behavior, interests, and activities	Infancy or childhood
Rett's Disorder	• Rapid deterioration of behavior and mental status after 7 months of age • Loss of fine and gross motor skills • Severe language impairment • Primarily in girls	Prior to 18 months
Childhood Disintegrative Disorder	• Loss of previously acquired skills (language, social skills, bowel/bladder control, play, or motor) • Abnormal social interaction, communication, or restricted, repetitive, and stereotyped patterns of behavior, interests, and activities	After 2 years of age

(Source: APA, 2000; Chawarska & Volkmar, 2005)

Etiology and Prevalence

First reported upon by Leo Kanner in 1943 and Hans Asperger in 1944, the cause of ASD continues to be debated among parents, professionals, and scientists. Leo Kanner

(1949) initially suggested autism was a congenital disorder in which children were born lacking the typical motivation for social interaction and emotional engagement. He also suggested the parent-child relationship may contribute to psychopathology. Bruno Bettelheim (1967) promulgated parental emotional unavailability as the pathogenesis of autism. Bernard Rimland (1964) disputed the 'refrigerator mother' theory and proposed neurological impairment as the cause of symptoms of autism. Rimland later asserted vaccines (e.g. thimerosal) to be a direct cause of autism and indeed, this continues to be a heated debate, with the vaccine theory often supported by parents. Within the scientific community, ASD is considered to be a NDD of genetic origin, with symptoms arising from abnormalities in brain structure and functioning. The incidence of autism in siblings is 10-60 times greater than in the general population (Fombonne, 1999) and the high rate of concordance among monozygotic twins suggests a genetic component (Baily, Le Couteur, Gottesman, & Bolton, 1995; Folstein & Rutter, 1977; Steffenburg et al., 1989). No one single gene can account for ASD, with current models suggesting the involvement of multiple genes, as well as a gene-environment interaction (Muhle, Trentacoste, & Rapin, 2004). Prevalence estimates of PDDs from 1997 to 2004 range from 0.7/10,000 to 72.6/10,000 (Fombonne, 2005a) with current rates estimated at 60 per 10,000 (Fombonne, 2005b). Males are diagnosed more frequently than females (mean ratio 4:1) with no differences across ethnicity or social class (Fombonne, 2005b).

Phenotype

Autism is a behaviorally defined disorder characterized by a broad constellation of symptoms. Defined by a pattern of six symptoms across the domains of social interaction, communication, and patterns of behavior (two per domain; APA, 2000), there

is significant heterogeneity in symptoms and functioning among individuals diagnosed with autism. Chawarska and Volkmar (2005) describe symptoms of autism in infancy and early childhood. Children with autism may demonstrate diminished interest in other people, little affection towards familiar caregivers, abnormal eye contact, and a diminished interest in reciprocal interactions. Language or functional communication skills man not develop, in addition to unusual vocalizations, or failure to share interest. Children with autism may excessively mouth items or demonstrate an aversion to touch in infancy, and as they develop, exhibit hand and finger movements, use objects inappropriately (e.g. spinning or nonfunctional uses), demonstrate repetitive and inappropriate interests and play, and demonstrate hyper- or hyposensitivity to sounds, textures, tastes, and visual stimuli.

Deficits in social reciprocity impact the ability to form attachment relationships, imitate another person, share a focus of attention with another person, understand another person's emotions, and engage in pretend play. These impairments in children with autism were once thought to be due to an inability or complete lack of desire to interact with other people. More recent research suggests social deficits may be due to impairments in understanding and responding to social information (Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998). Deficits in the ability to understand that other people have different point of view (e.g. theory of mind) impact the ability to understand the social language and intent of other. Children with autism have varying ability to initiate, respond to, and maintain social interactions.

Communication delays are the second area of core deficit. It often is the first domain of concern identified by families whose children are later diagnosed with autism.

Some children initially develop early language skills, but may lose language milestones between 18 and 24 months (Richler et al., 2006). Others may never develop language, or develop language in which true reciprocal conversational exchanges are impossible. Early language is often characterized by labeling (instead of requesting), echolalia (echoing speech), abnormal prosody or inflection, and improper use of pronouns. In addition to expressive and pragmatic impairments, children with autism often demonstrate deficits in receptive communication.

The third core domain surrounds atypical behavior. Repetitive, perseverative, and stereotyped behaviors are often apparent and disruptive. Children with autism frequently adhere to a strict routine and have difficulty adapting to change. Young children often demonstrate attachments to unusual items or may focus on the part of a toy instead of using the item for its intended purpose. Other behaviors may include lining up objects, staring out of the corners of their eyes, or visually inspecting aspects of objects. Children may not spontaneously develop pretend play, or will play in a rote manner. Stereotyped movements such as pacing, spinning, running in circles, flipping light switches, rocking, hand waving, arm flapping, and toe walking are common. Individuals with autism frequently demonstrate unusual responses to sensory input. Related symptoms include self-injurious behavior and feeding/eating problems. Additionally, although sleep problems are not part of the diagnostic criteria for autism, they are frequently reported clinically and in the literature in persons with ASDs.

Asperger's Disorder and Pervasive Developmental Delay-Not Otherwise Specified

Asperger's Disorder (AD), also called Asperger Syndrome, has only been recognized as a distinct clinical entity in the United States for a brief time. Consistent

diagnostic criteria has not been applied to AD, although in general it is agreed upon that the diagnosis of AD usually involves relatively intact intellectual and language functioning, accompanied by impairments in reciprocal social interaction associated with autism. According to the DSM-IV-TR (APA, 2000), AD is diagnosed if three symptoms, two related to social reciprocity and one to habitual behaviors are present. Language development must be within normal limits (e.g. two-word phrases by 2 years of age; longer phrases by 3 years of age), although pragmatic language impairments are common, and a diagnosis cannot be given if a child was previously diagnosed with autism. In addition to the aforementioned diagnostic criteria, individuals with AD often demonstrate the characteristics originally described by Hans Asperger (1944) including: impairment in nonverbal communication; idiosyncrasies in verbal communication; special interests; intellectualization of affect; clumbsiness and poor body awareness; and conduct problems (e.g. failure at school, noncompliance, etc).

The diagnosis of PDD-NOS is used to describe children who do not have the number or distribution of symptoms for another diagnosis within the ASDs, or have atypical presentation but functional impairment in the relevant areas. There is no minimum number of symptoms necessary to diagnose PDD-NOS, resulting in significant heterogeneity among individuals given his clinical diagnosis. As in children with autism, related symptoms of sensory, feeding, and sleeping problems are commonly reported in children diagnosed with AD and PDD-NOS.

Children with diagnoses of AD and PDD-NOS have been chosen for this study for the following reason. By the definition provided in the DSM-IV-TR, children with AD and PDD-NOS demonstrate fewer autistic symptoms that children diagnosed with

Autistic Disorder. While pragmatic language remains a significant concern in children

with AD, expressive language grossly follows a typical pattern of development.

Additionally, children with autism frequently present with co-morbid mental retardation

(MR), between 40% and 69% (Baird et al., 2000; Chakrabarti & Fombonne, 2001) while

children with AD present with IQs >70. Therefore, clinically and anecdotally, children

with the diagnoses of AD and PDD-NOS are typically higher functioning than children

diagnosed with autism, increasing their ability to learn and generalize the skills taught

within the context of this intervention. Children with Rett Disorder and CDD were not

selected due to significant impairments in functioning as well as low incidence of the

disability.

Sleep Architecture

Sleep is an essential physiological function that begins in utero. Once thought to

be a time of rest, the brain is active during sleep. Sleep alternates or "cycles" among five

phases. The stage termed non-rapid eye movement (NREM) sleep is associated with

reduced neuronal activity. NREM sleep consists of four stages, corresponding to the

depth of our sleep. While time spent in each of the sleep stages varies by age, the stages

are the same for children and adults. Stage 1 consists of the transition between

wakefulness and sleep. During stage 2, breathing and heart rate are regular, body

temperature decreases, and one becomes disengaged from the environment. Termed

"slow wave sleep" (SWS) or delta sleep, stages 3 and 4 are the deepest and thought to be

the most restorative periods. Early in the night, stages 3 and 4 of NREM dominate, while

stage 2 NREM and REM sleep prevail in the latter half of the night. Rapid eye

movement (REM) sleep is associated with dreaming and a high level of brain activity,

although the skeletal musculature is virtually paralyzed. The purpose of REM sleep remains the subject of research and debate, but its importance to development is evidenced by the amount of time neonates spend in REM sleep (approximately 50%) compared to older adults (approximately 13.8%-15%) (Roffwarg, Muzio, & Dement, 1966).

Sleep is controlled by processes and structures within the brain. Neurotransmitters, chemicals in the brain, control whether we are asleep or awake by acting on different groups of neurons. The master circadian pacemaker, the suprachiasmatic nucleus (SCN), is located within the anterior hypothalamus and has been referred to as our "biological clock." Sleep is affected by endogenous and exogenous factors. Autonomic processes exhibit circadian rhythmicity, and include body temperature, cardiovascular function, melatonin and cortisol secretion, metabolism, and sleep. Most important of the exogenous factors is the daily environmental cycle alteration of light and darkness. Biological processes impacted by external factors, or circadian rhythms, are also affected by locomotor activity, feeding, excretion, sensory processing, social cues, and learning capability.

The amount of sleep a child needs changes with developmental stages. As children age, there is a shift from time spent asleep to wakefulness during the day over the first five years of life (National Sleep Foundation, 2009). The average amount of sleep needed steadily declines throughout childhood and adolescence. Additionally, as children enter adolescence there is an increasingly large discrepancy between school-night and weekend sleep patterns. Dahl (1996a) defines sufficient sleep as the amount necessary to permit optimal daytime functioning.

Table 2.2

Sleep Needs

Age	Sleep Needs
Newborn (0-2 months)	10.5-18 hours
Infants (3-11 months)	9–15 hours
Toddlers (1-3 years)	12–14 hours
Preschoolers (3-5 years)	11–13 hours
School-aged Children (5-12 years)	10- 11 hours
Teens (11-17)	8.5-9.25 hours
Adults	7-9 hours
Older Adults	7-9 hours

(Source: National Sleep Foundation, 2009)

In addition to biological factors, sleep patterns are influenced by psychological, cultural, social, and family factors. Sleep and patterns also differ among individuals. Although a variety of physiological and environmental factors influence sleep-wake rhythms, individual differences (e.g. 'morning lark' or 'night owl') are prominent regarding peak alertness, and may be evident from an early age.

Sleep Disorders in Typically Developing Children

Parental concerns regarding sleep are common. Approximately two-thirds of adults (n = 1,473) surveyed in the 2004 *Sleep in America* poll, conducted by the National Sleep Foundation, indicated they would like to change something about their child's sleep. Owens (2001) reports sleep problems to be one of the top five parental concerns reported to pediatricians. Approximately 20% to 30% of all children experience some type of sleep disturbance during childhood (Gaylor, Burnham, Goodlin-Jones, and

Anders, 2005; Jenni, Fuhrer Zinggeler, Iglowstein, Molinari, & Largo., 2005; Lui, Lui, Owens, & Kaplan, 2005; Sadeh, Raviv, & Gruber, 2000; Touchette et al., 2005). Younger children experience more sleep problems; however, sleep problems may be persistent into adolescence (Sheldon, 1998). Additionally, only one in five of the adolescents surveyed indicated they get an optimal nine hours sleep on school nights, with 45% getting less than eight hours sleep per night during the week (National Sleep Foundation, 2006). Thus, sleep problems are salient to caregivers, more prevalent in early childhood, but are also evident in adolescence.

The variables of developmental changes (e.g. changes in sleep need; shift to daytime wakefulness) and individual differences in sleep can make it difficult to define when sleep is a problem and when a sleep problem becomes a clinical disorder. Durand (1998) believes sleep problems may be measured in terms of how well the child functions during the day, and how disruptive sleep difficulties are to a child and his or her family. Ferber (1996) describes sleep to be problematic if it disturbs the child, the child's family, or both. Assessing daytime functioning requires special consideration in children. In addition to problematic nighttime behaviors, during the daytime children may manifest adult symptoms of sleepiness such as yawning or drowsiness. They may also manifest other symptoms including irritability, low frustration tolerance, and a short attention span (Dahl, 1996a), or paradoxical symptoms such as hyperactivity and restlessness. Ring et al. (1998) describes associations between insufficient sleep and ADHD symptomology. Stores & Wiggs (2001, p. 17) highlight how pediatric sleep problems differ from adult sleep problems:

- The effects of sleep disturbance may be pervasive and affect cognition and behavior.

- Sleep disturbance may result in overactivity, irritability, or other symptoms similar to ADHD, in contrast to adults, who are typically sleepy or underactive.

- Children's sleep disorders are very treatable.

- Parenting factors are prominent to the causation, maintenance, and/or treatment of children's sleep disorders.

In assessing sleep problems, there are three basic types: difficulty getting to sleep or staying asleep; sleeping too much; and, disturbed episodes that interfere with sleep (Stores & Wiggs, 2001, p. 15). Further, the authors distinguish between the sleep problem (e.g. complaint) and underlying cause (e.g. sleep disorder). Each type of problem may be caused by a variety of sleep disorders.

Classification of Sleep Disorders

Lack of a common definition in identifying sleep disturbance is problematic. For childhood sleep disorders, there appears to be neither an appropriate, nor common classification system. The American Psychological Association (2000) divides sleep disorders into four categories according to etiology and describes 13 different sleep disorders including: Primary Sleep Disorders; Sleep Disorder Due to Another Mental Disorder; Sleep Disorder Due to a General Medical Condition; and Substance-Induced Sleep Disorder. Primary Sleep Disorders exclude the etiology of the other three categories, and are thought to arise from endogenous factors, further complicated by exogenous factors (APA, 2000). The category of Primary Sleep Disorders is further subdivided into Dyssomnias and Parasomnias. Dyssomnia is an expansive term used to

describe difficulty initiating or maintaining sleep, or excessive sleepiness. They are

characterized by a disturbance in the amount, quality, or timing of sleep. According to the

DSM-IV-TR (2000), "Parasomnias are disorders characterized by abnormal behavioral or

physiological events occurring in association with sleep, specific sleep stages, or sleep-

wake transitions (p. 630)." The problems represent the activation of physiological

systems at inappropriate times during the sleep-wake cycle, and include nightmares, sleep

terrors, sleepwalking, and parasomnia not otherwise specified.

Like the DSM-IV-TR (2000), the *International Classification of Sleep Disorders,*

Revised, Diagnostic and Coding Manual (ICSD-DCM; American Academy of Seep

Medicine, 2001) divides sleep into four, albeit different, categories: Dyssomnias,

Parasomnias; Sleep Disorders Associated with Mental, Neurological, or Other Medical

Disorders; and Proposed Sleep Disorders. This nosology is more comprehensive than the

DSM-IV-TR, and includes over 80 different sleep disorders. Unlike the previous two

classification systems, the *Diagnostic Classification of Mental Health and Developmental*

Disorders of Infancy and Early Childhood: Revised Edition (DC 0-3R; Zero to Three,

2005) focuses on children from ages birth to three. This system is the least

comprehensive of the three, and only names Sleep Behavior Disorder, with two types of

conditions, Sleep-Onset Disorder and Night-Waking Disorder, delineated.

The literature reports problems with all three diagnostic systems.   Robinson &

Waters (2008) indicate utilizing strict DSM-IV-TR criteria may fail to account for factors

unique to pediatric sleep problems (e.g. developmental changes in sleep habits, parent-

child interactions, and behavioral disorders). Regarding the ICSD-DCM (American

Academy of Sleep Medicine, 2000), Gaylor, Goodlin-Jones, and Anders (2001) report,

"the labels are cumbersome and the criteria neither empirically nor developmentally determined (p. 61)." They also report the DC 0-3 (Zero to Three, 2005) to lack an empirically derived, quantitative metrics to assist in the classification of sleep problems (Gaylor et al., 2001), and instead have proposed a developmental sleep disorders classification system for children ages birth to three years which classifies sleep problems by frequency and severity (Gaylor et al., 2005).

In 2006, a definition of pediatric insomnia was endorsed by a consortium of experts assembled by the National Sleep Foundation, in conjunction with Best Practice Project, Management, Inc (Mindell et al., 2006). This group concurred with criticisms surrounding current nosologies, and believed a need was apparent to create a definition which reflected differences between pediatric and adult sleep disorders. Therefore, the following definition, based on the International Classification of Sleep Disorders-2, was created. Pediatric Insomnia is defined as, "Repeated difficulty with sleep initiation, duration, consolidation, or quality that occurs despite age-appropriate time and opportunity for sleep and results in daytime functional impairment for the child and/or family (Mindell et al., 2006, p. e1225). This definition intentionally includes behavioral insomnia, as well as insomnia which may result from multiple etiologies, including behavioral, environmental, psychiatric, medical, and psychosocial.

Behaviorally-Based Sleep Problems

The literature commonly discusses childhood sleep problems in terms of those which are physiologically-based (e.g. obstructive sleep apnea, restless leg syndrome, etc.), and those that are behavioral-based. Behavioral-based complaints typically include complaints of bedtime problems (e.g. bedtime resistance or refusals; nighttime fears), or

night awakenings (Mindell & Meltzer, 2008), and are among the most commonly reported types of problems. While the DSM-IV-TR (APA, 2000) describes problems which may be behaviorally-based under the label Primary Insomnia (307.42), it does not make any reference to children, or the effect of parent behaviors on the development or maintenance of these problems. The ICSD-DCM (Zero to Three, 2005) includes behaviorally-based sleep problems under Extrinsic Sleep Disorders, beneath the broader category of Dyssomnias. As described below, parents are particularly influential in contributing to the following disorders: Inadequate Sleep Hygiene, (307.41-1), Limit-setting Sleep Disorder (LSSD; 307.49-4), and Sleep-onset Association Disorder (SOAD; 307.42-5) (ICSD-DCM; 2001).

While children may be vulnerable to sleep problems due to physiology, parental behaviors play an integral role in the development of behaviorally-based problems. As Blampied and France (1993) state, children must adjust patterns of sleeping and waking to sleeping practice familiarly and culturally determined. Behaviorally-based sleep problems result when a child is reinforced (e.g. rewarded) for a behavior which results in a delay in going to bed, or when the onset only occurs when particular conditions are present. For example, in LSSD, a child may stall going to sleep by running from the room at bedtime, repeatedly calling out for a parent, or making multiple requests (e.g. another story, water, bathroom). The parent may fail to set limits or may occasionally "give in" to the child's requests, resulting in a delayed bedtime. Hence, the child has inadvertently learned behaviors which result in bedtime resistance or settling problems. A similar pattern may be seen in SOAD. For example, a mother may nurse or bottle-feed a child until they fall asleep, then place the child in his or her crib. The mother may also

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feed and hold the infant each time it cries throughout the night, until the infant has resumed sleep. Thus, the mother has taught the child to associate being held and fed as the conditions for falling asleep.

Many families and practitioners may believe physiologically-based sleep disorders to be more problematic; however, behaviorally-based problems may be as significant. Owen et al. (1998) compared 152 children ages 2 to 12 with Obstructive Sleep Apnea and behaviorally-based sleep problems. Results indicated the latter can be associated with more significant behavior problems at bedtime and during the day. Due to the impact of caregivers on sleep, when assessing childhood sleep problems, it is essential parents or caregivers report on all aspects including parent-child interactions.

Assessment of Sleep Disorders

In addition to various diagnostic classification systems, several subjective and objective tools are utilized to measure sleep problems in children. It is first recommended that an extensive sleep history be obtained, focusing on the sleep schedule, including the child's 24 hour sleep-wake cycle during the week and on weekends (Mindell & Meltzer, 2008; Stores & Wiggs, 2001). Clinical interview may be supplemented with screening tools such as the Epworth Sleepiness Scale (ESS; Johns, 1991), the Children's Sleep Habits Questionnaire (CSHQ; Owens, 2001), or the BEARS (Owens & Dalzell, 2005) depending upon the nature of the sleep problem. Clinical interview and screening tools may also be paired with a sleep diary, to gather information regarding sleep/wake times, bedtime routines, night waking, etc.

Objective measures provide the most accurate information about sleep. Audiovisual recordings, even with the family's own video equipment, may be used to

corroborate subjective input. Standard sleep recording devices include actigraphy and polysomnography. An actigraph is a watch-like device which may be worn on the wrist or ankle, or sewn into a child's clothing. The device monitors body movement via sensors and data is downloaded into a computer program which applies validated algorithms to the data to score sleep and wake states. While actigraphy does not measure stages of sleep, it is particularly useful in obtaining objective data in children who are not able to tolerate the more invasive polysomnography, or to measure sleep/wake cycles over a period of time.

Polysomnography (PSG) is considered the "gold standard" in sleep assessment. Known as a "sleep study," it measures sleep cycles and stages by recording brain waves (Electroencephalography; EEG), electrical activity of muscles, eye movement, breathing rate, blood pressure, blood oxygen saturation, and heart rhythm. Standard PSG involves recording EEGs, EOGs (Electrooculogram), and EMGs (electromyogram), to measure NREM, REM sleep, and sleep continuity. However, one limitation is that it is labor intensive and expensive and is often not well tolerated by young children or children with disabilities. Thus, it generally cannot be used for large-scale population studies in children.

Sleep in Children with Intellectual Disabilities

Multiple studies have evaluated sleep in children with varying levels of intellectual disability (ID). These studies generally focus on sleep physiology, or on descriptions of sleep problems. Several studies utilize heterogeneous samples of children with varying levels of ID. More recent research has become increasingly focused on sleep problems in specific populations, such as children with autism, Down syndrome,

ADHD, or children with physical disabilities. Studying homogenous groups (e.g. by disorder) may assist in understanding the origins of sleep problems associated with conditions as well as assist in delineating treatment effects (Wiggs and Stores, 2001). Of the studies which include heterogeneous samples (e.g. children with various disabilities and varying levels of ID), parent survey is the most common methodology. As children with ASD and sleep problems are discussed in extensive detail in the next section of the paper, studies which utilized heterogeneous groups of children with ID will be examined in this section.

Early studies of sleep in developmental disabilities indicated high prevalence rates of sleep disturbance. In 1985, Bartlett, Rooney and Spedding studied 214 children with severe ID. Prevalence rates for sleep problems ranged from 86% in children under 6 years of age, 81% in children 6 to 11, and 77% in children 12 to 16. Most common sleep problems included night waking (56%), settling problems (e.g. falling asleep) (56%), and bedtime resistance (53%). Quine (1991) also studied children with severe ID; however, she conducted a three year longitudinal study of 200 children with severe ID between the ages of 2 and 18 years. Similar to Bartlett et al.'s (1995) results, half (51%) of parents indicated settling difficulties, and two-thirds (67%), frequent night waking. Notably, over half of this group reported these same problems three years later, indicating sleep problems may be persistent in children with ID.

Richdale, Francis, Gavidia-Payne, & Cotton (2000) conducted a study which compared the sleep of children with ID to TD controls. The authors surveyed 52 children with ID ages 2-19 years, in comparison with 25 age-matched TD peers. Notably, 57.7% of the children with ID presented with a current or past (66.7%) sleep problem, compared

with 16% (current sleep problem) and 33.3% (past) of the control group, respectively. In over half the children with ID and sleep problems, they had been present for over two years. Further, the presence of sleep problems was significantly associated with behavioral problems and parental perception of hassle frequency and intensity.

Quine's 2001 study reiterated the high prevalence of sleep problems in children with ID compared to TD peers. In a comparison of 182 children ages 4 to 12 years of age receiving special education services (e.g. in a special school setting), to 576 age-matched typically developing peers (mainstream setting), children with disabilities had significantly higher rates of sleep problems compared to children in mainstream schools. Like her 1991 study, children with disabilities had significantly higher rates of sleep problems; however, the three most frequently reported types of problems (e.g. settling problems, night waking problems, and sleeping in parent's bed) were the same in both groups. Maternal stress was associated with sleep problems in both the children in mainstream schools and those in special schools. Additionally, epilepsy, which is significantly more common in children with NDD, was associated with sleep problems.

In 2001, Didden, Korzilius, van Aperlo, van Overloop, and de Vries surveyed parents of 286 children between ages 1 and 19 years of age with mild to profound ID regarding sleep problems in relation to behavior. Almost all the children in the sample (99.4%) reported one type of sleep behavior or mild sleep problem, while 16.1% exhibited a severe sleep problem, with settling problems (4.2%), night waking (10.8%), and early waking (4.2%) the most common type of problem. Children with more severe levels of ID were more likely to have a sleep problem, and children with sleep problems

demonstrated higher levels of daytime behavior problems (e.g. hyperactivity, aggression, and non-compliance).

Two studies by Robinson and Richdale (2004) endorsed the chronic nature of sleep problems in children with ID. The first survey consisted of families who believed their child had a past or current sleep problem, while the second survey first asked families to describe sleep problems, and then indicate if they believed a sleep problem to be present. In the first survey of 149 of parents with mild to profound ID, 36.2% of the children experienced a past sleep problem lasting an average duration reported for 6.3 years. 25.5% of the sample had a current sleep problem, with the average duration lasting 8.8 years. Like Didden et al. (2002), children with more severe levels of ID experienced the highest frequency of past and current sleep problems. In the second study, 243 families of children with various levels of ID were also surveyed, with 62.8% of the sample reporting some form of problematic sleep behavior. Early waking, settling, and night waking were the most commonly reported problems. Where parents believed a sleep problem was present, the average duration was reported to be 7.1 years. Between the two studies, approximately one-third to three-fourths of parents with past and current sleep problems, sought treatment.

Prevalence rates of sleep problems in children with NDD vary; however, as a whole, the literature suggests sleep problems are more prevalent in children with NDD than in TD children. Like the etiologies of various NDDs, the etiologies of sleep problems in this population are diverse. In their discussion of sleep hygiene and children with NDDs, Jan et al. (2008) suggest a variety of disease-related factors contribute to sleep problems, including, "location of brain abnormalities, the severity of developmental

delay, associated sensory loss, health problems, and pain (p.1344)." Mindell and Owens (2003) suggest increased sleep problems may be related to existing behavioral problems, neurological problems, medical problems, and psychiatric conditions. Didden et al. (2002) propose sleep problems as a behavioral phenotype given the associations among genetic disorders and sleep problems (p. 538). As with TD children, environmental and caregiver factors, as well as sleep practices (bedtime routine, scheduling) and physiologic factors (e.g. intake of caffeine, exercise) contribute to quality of sleep. In addition to physiological factors, Wiggs and Stores (2001, p. 50) suggest other factors which may predispose children with intellectual impairments to sleep problems, including:

- Difficulty learning good sleep hygiene.

- Decreased limit-setting by parents due to feelings of guilt or compassion leading to overdependence of children during settling or night waking.

- Difficulty establishing good sleep habits due to co-existing emotional or behavioral problems.

- Parents minimizing sleep problems or believing them to be "untreatable."

While sleep problems are more prevalent in children with NDDs, the nature of sleep disturbance is similar to that seen in TD children and includes settling problems, night waking, and co-sleeping (Polimeni et al., 2007, p.77; Taira, Takase, & Sasaki, 1998; Wiggs & Stores, 2004).

<div align="center">Sleep in Children with Autism Spectrum Disorders</div>

Theoretical Basis of Sleep Problems

The etiology of sleep problems in children with ASDs is unknown. Richdale and Schreck (2009, p. 4) propose a biopsychosocial model, hypothesizing sleep problems

may occur as a result of: 1) intrinsic biological or genetic abnormalities that alter brain architecture or biochemistry; 2) psychological or behavioral characteristics connected with core or associated features of ASDs; or 3) factors in the family home or environment, including caregiver practices.

Biological and physiological research has suggested clock gene abnormalities may be a causative factor in autism, and results in temporal abnormalities with circadian/sleep architecture and circadian hormone abnormalities including altered serotonin and melatonin. Nicholas, Rudrasinghma, Nash, Kirov, Owen, and Wimpory's (2007) study of clock genes (genes which encodes proteins regulating circadian rhythm) suggested the problems in timing, memory, and sleep characteristic of individuals with autism are consistent with their positive findings associating clock genes with autism. Bourgeron (2007) suggested clock genes in connection with low melatonin may result in abnormal circadian rhythms in ASD. Three studies have endorsed decreased melatonin in children with ASD. In 2000, Kulman, Lissoni, Rovelli, Brivio, and Sequeri found abnormal circadian rhythms and decreased melatonin levels in all of 14 children with autism, compared with 20 age-matched controls. In their analysis of 50 children and adolescents with autism in compared with 88 age- and gender-matched controls, Tordjman, Anderson, Pichard, Charbuy, and Touitou (2005) found the autism sample demonstrated significantly lower rates of nocturnal melatonin, which the authors hypothesized may be related to a dysregulation in neuroendocrine functions involved in circadian rhythms or may be related to altered serotonin neurobiology. Both findings (e.g. decreased melatonin levels) was supported more recently by Melke et al. (2008) in a multi-center international study of 250 children and adults with autism and AD compared

with controls. The authors posited the low melatonin concentration was caused by a primary deficit in the ASMT gene, which encodes the last enzyme of melatonin synthesis, and is deleted in many individuals with ASD.

Researchers studying sleep neurophysiology have speculated central nervous system involvement in sleep problems in children with ASD. Elia et al. (2000) used polysomnography to evaluate the sleep patterns in 17 male children and adolescents with autism, seven children with mental retardation (MR) and fragile X syndrome, and five age- and gender-matched controls. Based on results, the authors suggested differences in sleep patterns in individuals with autism (in comparison with the MR and TD groups) and hypothesized monoaminergic abnormalities in the central nervous system in individuals with autism. Limoges, Mottron, Bolduc, Berthiaume, and Godbout's (2005) study utilizing polysomnography also endorsed differences in sleep patterns and neurophysiology in 27 adults with ASD compared to 78 age- and gender-matched controls, and hypothesized this may be associated with atypical cortical organization. In 16 children with autism and intellectual disability (ID), Miano et al., (2007) found changes in sleep microstructure in Stages 3 and 4 of NREM sleep, compared to controls. Bruni et al. (2007) also used polysomnography to compare the sleep patterns of 8 children with AD, 10 children with autism, and 12 age-matched controls. Similar to Miano et al.'s study (2007), significant differences were found in sleep microarchitecture among the three groups. Thus, genetic and physiological studies indicate a biological basis for sleep problems in children with ASD.

Sleep problems in children with ASD may also be related to psychological or behavioral characteristics connected with core or associated features of the disorders,

although few studies to date have investigated this relationship. (Richdale and Schreck, 2009). Children with ASDs are vulnerable to co-morbid conditions associated with sleep problems, including anxiety, depression, and ADHD (Leyfer et al., 2006). Co-morbid mental retardation may also be a risk factor in sleep problems; however, the impact is not clear as most studies have examined children with classic autism, many of whom demonstrate coexisting intellectual deficits (Johnson & Malow, 2008). Richdale and Prior (1995) theorized the inability to use social cues in children with autism results in an inability to synchronize the sleep/wake cycle. The relationship between severity of symptoms of autism and sleep problems was examined by Schreck, Mulick, and Smith in 2004, and by Hoffman, Sweeney, Gilliam, Apodaca, Lopez-Wagner, and Catillo in 2005. In their retrospective review of 55 children ages 5 to 12 years of age (M = 8.2) diagnosed with autism (inclusion for study based on a Gilliam Autism Rating Scale GARS; Gilliam, 1995) score of ≥ 80), Schreck et al. (2004) examined demographics information, the GARS and the BEDS to understand if the fundamental characteristics of autism could be exacerbated by sleep problems. Results indicated communication problems were significantly related to increased sensitivity to stimuli in the sleeping environment and by periods of screaming during the night. These two characteristics (sensitivity and periods of screaming) were related to increased symptoms of autism. Screaming and fewer hours of sleep per night predicted increased stereotypic behavior, and fewer hours slept predicted difficulties with social interaction and overall diagnostic characteristics of autism. Hoffman et al. (2006) extended this study with a larger sample (N = 72 (with $\geq$ 80 on the GARS; ages 4 to 15 years; M = 8.2 years) and use of the Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, McGuinn, & Nobile, 2000) to measure

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sleep problems. Results indicated sleep-disordered breathing and parasomnias predicted the severity of symptoms. While a clear relationship between the core and associated symptoms of ASD and sleep problems remains unclear, further research is needed understand these associations.

The third component of Richdale and Schreck's (2009) biopsychosocial model consists of factors in the family home or environment, including parenting practice. Owens (2008) suggests several child, parent, and environmental variables impacting sleep and the development of sleep problems.

Table 2.3

Variables impacting sleep

Child	Parent	Environment
Temperament	Parenting style	Physical sleep environment
Behavioral Style	Discipline style	Family composition
Cognitive abilities	Maternal depression	Lifestyle issues
Language abilities	Family stress	Socioeconomic status
Circadian preference	Parent education level	
Comorbid conditions	Knowledge of child development	
Medical conditions	Parent sleep	
Television viewing habits		

(Source: Owens, 2008)

The proposed biopsychosocial model highlights the complex, multi-factorial nature of the etiology of sleep problems in children with ASD. It also underscores the need for further research to clarify the relationship among variables. While the etiology

is not fully known, a larger body of literature (e.g. over 15 studies) examining the prevalence and nature unequivocally demonstrates children and adolescents with ASDs are particularly vulnerable to problems with sleep.

Prevalence and Nature of Sleep Problems

Similar to studies of children with other NDDs, parents of children with ASD frequently report sleep to be problematic. Over 15 studies since 1995 have examined the nature and prevalence of sleep problems in ASD, and consist of studies which exclusively rely upon parent report, and those which combine parent report with objective measures (e.g. actigraphy or videosomnography). Of the 10 studies exclusively relying upon parent report, three of the studies lacked a comparison group. This is problematic as the validity of parent report measures may be compromised as parents may have difficulty reporting on sleep due to their own sleep habits. Secondly, it may be difficult to discriminate among certain types of problematic sleep behavior due to delays in communication and behavior inherent to ASDs. Nonetheless, the following three studies are consistent in endorsing the high prevalence of sleep problems in this population (54% - 86%).

Utilizing sleep diaries, the CSHQ, and the Parenting Events Questionnaire (PE), Honomichl et al. (2002) gathered sleep data on 100 children with ASD (including four with Rett's and Angelman's syndrome, or Childhood Disintegrative Disorder) over 12-weeks. 54% of parents reported sleep problems including delayed sleep onsets and fragmented sleep. Lui et al. (2006) found 86% of their 167 sample of children with ASD had at least one sleep problem almost every day with the most frequently reported problems being bedtime resistance (54%), insomnia (56%), parasomnias (53%), morning

rise problems (45%), daytime sleepiness (25%), and sleep disordered breathing (25%). Williams, Sears, & Allard (2004) reported similar types and rates of sleep problems in their sample of 210 children with autism with over half the sample (53.3%) reporting difficulty falling asleep, restless sleep (40%) unwillingness to sleep in own bed (39.5%) and frequent awakenings (32%). Unlike the previous studies, the authors divided the sample into those with autism and co-existing MR, and those without. The group with MR experienced significantly higher rates of nighttime wakenings. Similar patterns of the prevalence and nature of sleep problems in children with ASD are endorsed in the studies which utilized a comparison group.

Richdale and Prior (1995) also divided their sample of 39 children with autism, although they utilized criteria of an IQ < 55 to classify 12 of the children as low functioning, and an IQ > than 55 to classify high functioning. These two groups were then compared with age-matched typically-developing (TD) controls (in two separate studies reported within the same article). The low-functioning autism group napped more, went to bed earlier, and demonstrated significantly longer sleep latency, night awakenings, and earlier wake time compared to controls. The high-functioning autism group also took a significantly longer time to fall asleep, stayed awake for longer periods of time at night, and woke earlier than the controls. Age-related changes in sleep patterns were also reported in all three groups and children under the age of 5 years had the highest incidence of sleep problems. Patzold et al. extended this study in 1998 by adding an age, gender, and IQ-matched control group. Significantly more children in the ASD (n=38) than in the control group (n=36) were reported as having sleep problems, both currently and in the past, with problems settling, and night waking, and when awake,

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children in the ASD group were more disruptive (e.g. talking, singing, grunting, laughing, making noises, banging on walls, playing, and running around). Notably, children in the ASD group required additional parental conditions to fall asleep, and failure to meet these conditions resulted in the child refusing to get into bed, tantruming, etc). There were no differences among age or IQ (e.g. high versus low functioning) in the ASD group.

In 2000, Schreck and Mulick examined parental reports of sleep problems in children ages 5 years to 12 years 11 months with autism (GARS score ≥80; n = 38), compared to those with PDD (not specified as PDD-NOS or a PDD; n = 17); MR alone (n = 22), children attending special education classes without MR or autism (n = 49) and an age-matched control group (n = 43), to understand the presentation of sleep problems in children with autism and PDD, parents perceptions, and amount of sleep. Parents completed demographic information, the Gilliam Autism Rating Scale (GARS: Gilliam, 1995), and the Behavioral Evaluation of Disorders of Sleep (BEDS) (Schreck, Mulick, & Rojan, 2003). Despite age differences, there were no significant differences in the average number of hours slept per night, by age or group. Parent of children with autism reported more sleep problems than the other groups as well as a greater need to facilitate sleep such as the use of medication or a pacifier, although in actuality to autism group did not have higher rates of medication usage than the other groups. Further analysis conformed the children with autism exhibited significantly more dyssomnias and parasomnias including more nightmare behavior such as screaming (with and without waking), sleep walking, and acting out dreams.

One study compared sleep problems in children with autism, Down syndrome (DS), and Prader-Willi syndrome (PWS) to TD controls (Cotton and Richdale, 2006).

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The prevalence of sleep problems in the disabilities group was at least four times higher than in TD children, with problems in children with autism the most prevalent. Settling difficulties and co-sleeping were more prevalent in autism whereas sleep maintenance problems were common in autism, DS, and PWS, than in controls. Thus, the literature which compares children with co-existing MR and autism generally did not demonstrate differences based on intellectual functioning. This group of studies is in contrast to those describing sleep problems with other NDDs. These differences may be due to co-morbid conditions (epilepsy, genetic conditions, etc.) in the other NDD sample. Based on the results in the preceding studies, several authors have hypothesized that sleep problems may be related to the core or related deficits inherent in autism, as opposed to intellectual functioning.

Unlike the previously reported control group studies, Courturier et al. (2005) individually matched 23 children with an ASD between the ages of 5 and 12 years with IQs >70 to peers matched by age, gender, and geographic region. Although the sample size was small due to the desired homogeneity of the matched groups, a significantly higher proportion of the children with ASD were reported to have a sleep problem based on a CSHQ score of 41 or higher (78%) compared to controls (26%) with significantly higher problems for the ASD group in sleep onset delay, sleep duration, sleep anxiety, and daytime sleepiness.

In their 2006 study, Hoffman et al. attempted to correct methological errors from previous studies (e.g. small sample sizes, particularly with the autism groups, limited information regarding demographics, unclear inclusion/exclusion criteria, lack of control groups, varied definitions of autism, nonstandardized measures of autism, and possible

bias due to recruiting methods). Hoffman et al.'s sample consisted of 106 parent

volunteers from a university-based behavioral intervention and patent education program,

and the community sample (N = 168) from local community organizations (n=14),

students in university psychology courses (n = 69), and from data collected by graduate

students (n = 85) as part of an assignment. With the exception of a higher level of

education in the community groups, the demographics (race and family income), did not

differ. The authors used the CSHQ subscales to measure sleep problems and the GARS

to verify a diagnosis of autism. Results indicated children with autism experienced

significantly more sleep problems than the community sample on seven of the eight

subscales, including Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep

Anxiety, Night Wakings, Parasomnias, and Sleep Disordered Breather, with no difference

for the Daytime Sleepiness Subscale. Additionally, bedtime resistance and sleep anxiety

decreased with age in both the autism and community sample. The authors noted their

autism sample may not have been representative and additional measurement techniques

should be utilized in the future, such as sleep diaries, actigraphy, and videosomnography.

More recently, Krakowiak et al. (2008) studied a large sample consisting

exclusively of young children (ages 2 to 5 years; M=3.6 years) with ASD (n=303),

Developmental Delay (DD; n=63), and 163 population-based TD controls. Overall, 53%

of the children with ASD and 46% of the children with DD had at least one sleep

problem occurring 'frequently' or 'always' compared with 32% of the TD sample.

Notably, only 1.2% of the TD reported the sleep problem affected daily functioning of

the child, in contrast with 21% and 20% of the ASD and DD groups, respectively. THE

ASD and DD groups also reported significantly higher impact on family functioning

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(23% and 21%) compared with the TD group (2.5%). Both the ASD and DD group demonstrated significantly more problems with sleep onset and night awakenings than the DD group, with the ASD group demonstrating the highest report of problems.

A smaller body of literature used objective measures in conjunction with subjective measures to describe sleep in children with ASD. Hering, Epstein, Elroy, Iancu, and Zelnik (1999) conducted the first study to utilize actigraphy to measure sleep patterns in autism. Twenty-two randomly selected children with autism (3 – 12 years) were compared with controls via questionnaire. From this group, eight children with autism, with reported sleep problems, and eight controls were monitored via actigraph for 72 hours. Although the parents in the sleep problem/autism group reported earlier waking and multiple night arousals, actigraph data demonstrated only differences in waking times (e.g. children with autism woke approximately one hour earlier than controls). The authors hypothesized parents of children with autism may be more sensitive to nighttime behavior, or night waking may not be longer than in typical children, but behavior is disruptive and wakes other household member. Wiggs and Stores (2004) attempted to address this discrepancy in their study of 69 children with ASDs using sleep questionnaires, diaries, and actigraph data for five nights, although this study did not offer a control group of TD children. Eighty-three percent of the sample was reported as having a past sleep problems and 67% a current sleep problem, with a repetition of previously described problems most commonly occurring (e.g. difficulty falling and staying asleep). Based on ICSD-R criteria, the most common underlying sleep disorder was behavioral (e.g. problems related to limit-setting and sleep conditions required for the child to fall asleep). Actigraph data was available for 62 children and

data was compared to norms for 46 children ages 6 to 12 years. Unlike Hering et al.'s study (1999), when compared with the norms, the autism sample demonstrated sleep abnormalities (e.g. very early or late onset and waking times, increased sleep latency, and frequent night wakings); however, the group with reported sleep problems did not differ from the group without sleep problems, indicated the role of parent perception of sleep problems.

Goodlin-Jones et al. (2008) also utilized a multi-method approach to examine how sleep problems differ among TD children, children with autism, and those with developmental delay (DD).   The authors improved upon previous studies by controlling for age- and diagnosis-related effects, comparing 68 preschool children with autism were compared with 57 children with DD, and 69 TD controls that were developmentally-matched. Additionally, multimethod data collection occurred, including actigraphy, sleep diary, and the CSHQ. Like Hering et al.'s study (1999) there was incongruence among the objective and subjective report measures; however, children in the autism sample slept significantly less than the other two groups, while the DD sample demonstrated significantly more and longer night awakenings. Regardless, parent perception of a sleep problems was associated increased self-reports of stress.

Lastly, four studies examined sleep in children with AD; two exclusively utilized parent report and three incorporated actigraphy. In 2005, Polimeni et al., compared sleep problems and treatment outcomes of children with autism and AD to TD controls. The authors surveyed 66 parents of TD children (ages 2-11), 53 parents of children with autism (ages 2-16), and 52 parents of children with AD (ages 4-17) on demographics, sleep patterns, the presence and nature of sleep problems, treatment, and success of the

treatment, in addition to a sleep survey. No significant differences were found between

groups on hours of sleep per night; however, significantly fewer sleep problems were

reported for the TD group than the autism group (73%) and the AD group (73%).

Interestingly, 50% of the TYP group reported sleep problems. There were no significant

differences in severity or type of sleep problems among the groups, although the AD

group demonstrated significantly higher symptoms of sleep disturbance as measured by

the BEDS total score, than the other groups. Allik, Larsson, & Smedje (2006a and

2006b) reported on the sleep patterns in 32 children (8-12 years) with AD or high-

functioning autism (HFA) compared to age-matched controls utilizing questionnaires,

sleep diaries, and actigraphy for seven days. Results revealed children with AS/HFA

went to bed and woke approximately 40 minutes earlier than controls, although both

groups went to bed and awoke significantly later on the weekends. Parent and actigraph

data confirmed significantly longer sleep latency for the AD/HFA group. Allik et al.

(2006b) found almost a third of their AD/HFA sample (10/32) met criteria for pediatric

insomnia and parents and teachers reported more autistic, emotional, and hyperactivity

symptoms than in the group without insomnia.

Most recently, Paavonen et al. (2008) also demonstrated a higher prevalence of

sleep disturbance in children with AD than in TD controls. The authors compared the

prevalence of sleep disturbance in 52 (4.8-17 years; M=10.1) children with AD compared

to 61 TD controls, but uniquely, using children in addition to parents, as informants.

Both parents and children (58.3%) of the AD group reported significantly more sleep

problems, and the majority (68.5%) of controls slept over 9 hours per night, compared to

41.2% of the AD group. Parents in the AS group reported significantly more sleep-

related fears, while the children's self-report endorsed significantly more insomnia, night wakings, difficulties falling back asleep, too short sleep duration, and tiredness. Children with AS also reported significantly more negative attitudes towards sleep and sleep-related fears.

In conclusion, recent studies of sleep in children with ASD utilized more rigorous methodologies than previously reported. Improvements included comprehensive diagnostic evaluations of participants, excluding children with defined neurologic diseases (e.g. Fragile X syndrome or Rett syndrome), and controlling for confounding variables such as co-morbid conditions or medication use. Attempts have been made to control for the confounding variable of MR, and it appears that ASD may be an independent risk factor for sleep problems. Additionally, more recent studies utilized multiple-method data collection techniques including objective and subjective measures, and attempted to provide matched controls through the use of a narrow age range, matched neurodevelopmental comparison groups. Regardless of the limitations, this body of literature overwhelmingly describes a high prevalence of sleep problems in children with ASD, particularly insomnia. Problems with initiating and maintaining sleep may often have an underlying behavioral basis. The literature focusing on children with AD highlights the pervasive nature of sleep problems in this population. Further research is required to understand the interaction of AD and age (e.g. changes in circadian rhythms inherent to puberty), and other variables such as co-morbid anxiety in sleep problems and children and adolescents with AD. Despite many unanswered questions, it is clear that intervention is warranted for children with ASDs and sleep problems.

Treatment of Sleep Problems

Discussions of treatment for childhood sleep problems are described in the literature by the etiology of the problem, symptoms, or the population being studied. As the scope of this paper highlights sleep in children with ASD and behavioral intervention, discussions of treatment will exclude those for sleep disorders caused by a specific physiologically-based conditions (e.g. surgery for obstructive sleep apnea or medication for narcolepsy) typically not associated with ASD.

Treatment of Sleep Problems in Typically Developing Children

Ward, Rankin, and Lee (2007) describe commonly reported sleep problems. Of infants, toddlers, and preschool children, approximately 15% to 40% experience difficulty with settling problems and night waking (Goodlin-Jones, Burnham, Gaylor, & Anders, 2001; Thunstrom, 1999, 2002). Bedtime resistance occurs in approximately 10% to 20% of school-aged children (Archbold, Pituch, Panabi, & Chervin, 2002; Owens et al., 2000; Smedje, Broman, & Hetta, 2001). Obstructive sleep apnea occurs in approximately 1% to 3% of children (Owen, Canter, & Robinson, 1995; Hultcranz, Lofstrand-Tidestrom, & Ahlquist-Rastad, 1995; Ferreira et al., 2000). Owens, Rosen, and Mindell (2003) also found sleep problems to be more common in infants and toddlers, decrease with age, but remain consistent in children ages six through adolescence. Thus, discussions of treatment in the literature focus on these commonly reported problems and often are targeted towards young children.

Improved sleep hygiene is the first-line treatment intervention in childhood sleep disturbance (Jan et al., 2008; Mindell & Meltzer, 2008). Sleep hygiene is described by Meltzer and Mindell (2004) as, "a set of sleep-related behaviors that expose persons to activities and cues that prepare them for and promote appropriate times and effective

sleep (Jan et al., 2008, p. 1344)." Table 2.4 indicates targeted areas for "good sleep hygiene."

Table 2.4

Sleep Hygiene

Environmental	Scheduling	Sleep Practices	Physiologic
Sleep position	Sleep/wake times	Bedtime routines	Medical conditions
Bedding	Daytime naps	Bedtime associations	Mealtimes
Sleep Space Physical Environment		Ability to self-sooth	Exercise

(Source: Based on Jan et al., 2008)

While it is not fully understood why sleep hygiene promotes sleep, Jan et al. (2008) propose it may:

- Entrain intrinsic circadian rhythms to the external environment.

- Condition behavior to associate certain activities and environments with sleep.

- Promote sleep by reducing arousal and promoting relaxation.

- Decrease anxiety in some children with neurodevelopmental disabilities.

The authors acknowledge recommendations for sleep hygiene are based on clinical expertise due to limited empirical evidence. After sleep hygiene, treatment is likely to employ a pharmacological or behavioral intervention.

Pharmacologic Sleep Intervention in Typically Developing Children

Several factors may potentially impact the decision by physicians and parents to utilize medication for the treatment of sleep problems. Variables may include the presence of a co-morbid condition (e.g. psychiatric, neurodevelopmental disability),

parent stress, acceptability to parents, previous treatment with behavioral intervention, physician characteristics, and severity of the sleep problems/impact on child and parent functioning (Owens et al., 2003). There is currently no Food and Drug Administration (FDA) approved medication for pediatric insomnia.

Despite this, pharmacological intervention for childhood sleep problems is a common practice among community-based pediatricians. Schnoes, Kuhn, Workman, and Ellis (2006) surveyed 800 pediatricians in four states to assess prescribing practices specific to sleep disturbance. Of the 222 returned surveys, 96% of the respondents indicated they treated sleep disturbance. Behavioral intervention (alone) was utilized by 92.8% of the sample, medication by 39.6% (alone), and a combination of the two, by 55%. Behavioral intervention was most frequently utilized in infants and preschool-aged children, with medication and combined treatment most frequently used with adolescents. The most commonly prescribed medications were antihistimines (e.g. diphenhydramine), clonidine, melatonin, and imipramine (tricyclic antidespressent), and other antidepressants (e.g. trazadone).

The findings of this study are similar to Owens et al.'s 2003 survey of 671 pediatricians regarding the use of medication to treat pediatric insomnia. This study found the same most commonly prescribed medications, with 77% of respondents having recommended over-the-counter medications and 58% prescribing medications for sleep in the past six months. Use of pharmacological treatment for sleep problems occurred typically for short-term, situational use (e.g. travel, acute pain, or acute stress) with nonprescription medications, and longer-term use of prescription medications for children with NDDs, psychiatric conditions, or ADHD (p. e632). Both studies concluded

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pediatricians often prescribe medications for pediatric sleep problems; however, there is a paucity of research in which the efficacy, tolerability, and safety profiles are evaluated.

In response to concerns regarding the common use of pharmacological intervention for pediatric sleep problems (e.g. insomnia) despite limited safety and effectiveness studies, a consensus statement, was developed by a consortium sponsored by the National Sleep Foundation and Best Practice Project Management, Inc. (Mindell et al., 2006). Recommendations were made regarding study design, participants, assessment, standard of care treatment (e.g. sleep hygiene and behavior intervention), and ethics. See Pelayo and Dubik (2008) for a more thorough discussion of medications used to treat pediatric sleep disturbance.

Behavioral-Based Sleep Intervention in Typically Developing Children

After sleep hygiene, behaviorally-based intervention is the first-line intervention. These types of sleep interventions are based on the theory of behaviorism, which originated in the early 1900s with Ivan Pavlov's theory of Classical Conditioning. Pavlov's supposition was later extended by the work of John Watson, Edwin Guthrie, Clark Hull, and notably, B.F. Skinner's theory of Operant Conditioning. While classical conditioning is characterized by a biological, or uncontrollable response to a conditioned stimulus, operant conditioning involves a voluntary response of an organism, which occurs when the organism is reinforced (e.g. rewarded). For example, when a child is given praise by a parent for a certain behavior, that behavior is likely to reoccur, in order to elicit the parent response.

Skinner then applied these basic principles to describe how to shape or change behavior, teach complex series of behavior (chaining), how to extinguish behaviors, and

differentiate between reinforcement and punishment. Skinner's theory of operant conditioning provided the foundation for teaching methodologies such as behavior modification, Applied Behavior Analysis, Mastery Learning, Direct Instruction, and has had significant influences on both regular and special education practices. Ormrod (2004) summarizes several basic assumptions of learning based on behaviorism including:

- Principles of learning apply equally to different behaviors and species.

- Learning can be studied objectively via focusing on a stimulus and response.

- Learning can only be describes as a measurable change in behavior (internal processes are exluded).

- Organisms are not predisposed to behave in certain ways.

Additionally, in the behaviorist literature, learning is referred to as conditioning. Behaviorism has also provided the foundation for the treatment of sleep problems which are considered to have an environmental (e.g. influenced by parent of caregiver behavior) component or etiology. Moore, Meltzer, and Mindell (2008) describe the primary goal of behavioral sleep interventions, "…is for children to develop positive sleep-related associations and self-soothing skills to fall asleep at bedtime and return to sleep following night waking independently (p. 572)."

In 2006, the American Academy of Sleep Medicine published practice parameters for bedtime problems and night awakenings in typically developing children ages 0 to 4-11 years of age (Morgenthaler et al.), based on a review of 52 individual treatment studies from 1970 to 2005 (Mindell et al., 2006). Collectively, the sample included over 2500 children. The review indicated 94% of the behavioral interventions produced

clinically significant improvements in bedtime resistance and night wakings, with over 80% of the children demonstrating clinically significant improvement that was maintained for three to six months. Nine of the 52 studies (17%) were randomized treatment control trials, 4 studies (8%) were randomized trials with high alpha and beta errors. The balance of the studies consisted of nonrandomized trials or case studies.

Extinction and parent education/prevention were supported by the highest levels of evidence. Extinction was examined in 23 studies, 21 of which were effective. Extinction consists of putting a child to bed and ignoring negative behaviors such as yelling or crying. This approach can be quite stressful for parents, and often results in an "extinction burst," in which behaviors may worsen before improvement is evidenced. Consistency is essential to extinction as intermittently attending to the child will result in intermittent reinforcement, which only serves to strengthen the negative behaviors. Graduated extinction, in which parents ignore behavior, but briefly check on the child at gradually longer intervals, has greater parental acceptance and was recommended by the American Academy of Sleep Medicine based on three randomized trials.

Parent education and prevention is designed to prevent sleep problems. It consists of written materials or in-person education, focusing on teaching children to self-sooth and develop positive sleep habits through a consistent sleep schedule, and bedtime routine. This treatment was found to be effective according the highest standard based on four randomized trials. Delayed bedtime as well as scheduled awakenings both were supported by one randomized trial; however, the latter was suggested to have diminished acceptability to parents and less utility in very young children. Insufficient evidence was

available to recommend any single therapy over another or any multi-faceted intervention over a single therapy.

Several methological issues were identified by the authors. First, lack of standard definitions and criteria for sleep problems, as well as outcome measures, were noted. Secondly, much of the literature was based on single-case design studies, which may limit external validity; however, the authors recognized experimental single-case research is stronger at isolating the mechanism of change than large group designs. Another problem with the literature is lack of studies which utilize objective assessment methods (e.g. actigraphy, polysomnography, etc). Still, parental subjective experience is clearly meaningful regarding the impact of the intervention on caregivers. Another concern is the lack of comparison regarding delivery methods of the intervention (e.g. one-on-one training, written materials, etc.). Finally, the body of behavior intervention sleep research typically included a short follow-up period of six months or less.

Treatment of Sleep Problems in Children with Neurodevelopmental Disabilities

Treatment options for insomnia in children with NDDs are similar to those in typically developing children, and usually consist of pharmacological, behavioral, or combined management. However, treatment of pediatric sleep problems in children with disabilities is complicated by additional physiologic, medical, or psychiatric factors. For example, pain is the strongest contributing factor in sleep problems among children with physical disabilities (Hemmingsson, Stenhammar, & Paulsson, 2008). Wiggs and Stores (2001) suggest certain treatment options (e.g. extinction) may be inappropriate in children with coexisting medical problems. In addition to these variables, Quine's (1991) path model of sleep problems in children with disabilities highlights the complex

interaction of child and family variables (communication skills, behavior problems, quality of marriage, maternal stress, etc.) which may impact treatment, including the ability of caregivers to manage treatment procedures.

This is further complicated by caregiver beliefs, which influence the decision to seek treatment, treatment choice, and acceptability. Robinson and Richdale (2004) found parents' most common reason not to seek treatment was the attribution of the sleep problem to the child's disability, or medical condition. Keenan, Wild, McArthur, and Espie (2006) also found parents most frequently attributed the cause of sleep problems to the child's medical problem or disability, although overall, parents believed the sleep problems to be controllable or curable. Despite attributing sleep problems to their child's disability or medical condition, approximately one-third (33.3%) of parents in study 1 (Robinson & Richdale, 2004) sought treatment for a past sleep problems, and over one-half (53.1%) sought treatment for a current sleep problem. In study 2 (described within the same article), 76.2% of parents had sought treatment for their child's sleep problem. In Keenen et al.'s study (2007), 60% of parents sought treatment. Results from the aforementioned studies are in contrast with Didden et al. (2002), in which only 19% of parents sought treatment. These results indicate significant variability in parents' willingness to seek treatment for their child's sleep problems.

There is also great variability in parent perception in effectiveness and satisfaction with treatment. Robinson and Richdale (2004) also surveyed parents regarding effectiveness and satisfaction regarding treatment, with no significant difference among the groups. However, behavior intervention was rated in the most effective for past sleep problems, but was rated the same effectiveness as drugs alone, for current sleep

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problems. In study 2, medical treatment 64.6% of the participants utilized a medical treatment and 50% a behavioral treatment. Although there were no significant differences among treatment effectiveness ratings, interestingly, behavioral interventions were rated the lowest. Given a forced choice, participant's in Keenen et al.'s study (2007), parents rated both behavioral treatment and melatonin as acceptable; however, over half (53.5%) preferred behavioral treatment to pharmacological (34.5%). This study was limited in that it compared behavior intervention to melatonin, which may be perceived as a rather innocuous drug.

In their 1999 review, Lancioni et al. found 22 studies from 1984 to 1998 which examined sleep interventions in children with severe or profound intellectual impairment with concurrent sleep problems. Five behavioral intervention strategies (in 14 studies) were examined (e.g. sleep scheduling, bedtime routine plus gradual extinction; extinction; bedtime fading with or without response cost, chronotherapy), and eight studies examined the use of melatonin. Over three-fourths of the participants were believed to have demonstrated some improvement; however, methodological problems somewhat limited the impact of the results. For example, in only two of the studies reviewed, the investigators reported explicitly on their methods to assess the sleep problems. In other studies, confounding variables were not taken into account.

Didden & Sigafoos' (2001) review of treatment of sleep disorders in developmental disabilities (both children and adults) reflects the need for larger-scale studies, comparison control groups, examination of long-term effects of sleep intervention, and studies that seek to elucidate factors necessary for effective implementation. As a whole, this body of literature demonstrated behavioral and

pharmacologic treatment (e.g. melatonin) was promising, but needed further evaluation. Additionally, this literature provided a foundation for improving the design of current intervention studies and areas of future research.

In the past decade, several small-scale studies examined treatment of sleep problems in children with NDDs, with the literature increasingly focusing on treatment of sleep problems in specific populations, such as children with autism or ADHD. Despite the prevalence of sleep problems in children with NDDs and the fairly substantial body of literature describing the efficacy of behavioral interventions in typically developing children, a small body of intervention research applies to children with developmental disabilities.

Treatment of Sleep Problems in Children with Autism Spectrum Disorders

Quality sleep promotes optimal emotional, behavioral, and cognitive functioning. In addition, treating sleep problems may also decrease parental stress and improve family functioning. The characteristics associated with autism have been found to be especially challenging, with parents of children with autism reporting higher levels of stress than parents of children with Down syndrome, other psychiatric problems, or developmental disabilities (Hoffman et al., 2008). Further, sleep problems in children with autism negatively impacts caregiver sleep (Lopez-Wagner et al., 2008) and stress is higher in parents of children with ASDs and sleep problems than in children with ASD without sleep problems (Doo & Wing, 2006). Thus, effective intervention is essential to child and family well-being. Despite this, less than half of parents of developmentally disabled children with sleep problems receive help (Wiggs & Stores, 1996).

Wiggs and Stores (1996) found parental preference for behavioral intervention, despite medications being the most commonly prescribed treatments. In their survey of 202 families of children with ASDs, Williams et al. (2006) found behavioral interventions (92%) were tried with much greater frequency than medications (35%) and behavioral interventions were more frequently perceived as helpful. As the scope of this paper highlights sleep in children with ASD, discussions of treatment will exclude those for sleep disorders caused by a specific physiologically-based conditions (e.g. surgery for obstructive sleep apnea or medication for narcolepsy) not frequently associated with ASD.

Pharmacologic Sleep Intervention in Children with Autism Spectrum Disorders

As previously noted, there are no FDA approved medications for pediatric insomnia. Despite this, there is some research to support the use of pharmacologic interventions, particularly melatonin, in children with ASD. Several trials in children with other NDDs and exclusively in children with NDDs have demonstrated favorable results. Recently, Wirojana et al. (2009) employed a four-week, randomized, double-blind, placebo-controlled crossover design in 12 children between the ages of 2 years and 15.3 years with autism, FXS, or both. Results indicated statically significant differences in increased total night sleep duration in 10 participants, decreased sleep latency time for all participants, and earlier sleep-onset time in 9 participants during melatonin treatment as compared with placebo. Night awakenings were decreased in 7 participants, but not significantly. No significant side effects were reported which is consistent of most other studies examining melatonin. Three trials examined melatonin exclusively in children with ASD. Anderson, Kaczmarska, McGrew, and Malow (2008) conducted a larger

retrospective chart review of 107 children ages 2 to 18 years of age with a diagnosis of an autism spectrum disorder. The majority of parents reported sleep was no longer a concern (25%) or improved sleep (60%) after treatment. In an open-label trial of combined controlled-release and fast-release melatonin in 25 children with ASDs (Giannott, Cortesi, Cerquiglini, & Bernabei, 2006). All children demonstrated improvement in sleep diaries and questionnaires. One month after melatonin was discontinued, 16 children returned to pretreatment sleep scores. As melatonin has a relatively short half-life, extended-release melatonin may be helpful for children with problems with sleep-maintenance. Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt (2003) found decreases in sleep latency as well as improvement in daytime behaviors as measured by the CBCL in a sample of 15 children with AD. Limited side-effects (e.g. 3 children) were reported from the aforementioned studies, but included mild tiredness, difficulty awakening, dizziness, and diarrhea.

Other studies investigating medication to treat sleep in children with ASD and co-morbid conditions examined the effectiveness of ramelteon (Stigler, Posey, & McDougle, 2006), clonidine (Ming, Gordon, Kang, & Wagner, 2008), risperidone (Capone, Goyal, Grados, Smith, & Kammann, 2008), tryptophan (Doan, 1998), iron supplements (Dosman et al., 2007), and secretin infusions (Honomichl, Goodlin-Jones, Burnham, Hansen, & Anders, 2002). Despite favorable results, these studies consisted of single-subjects or small sample and require more research to measure effectiveness and tolerability.

Behaviorally-Based Sleep Intervention in Children with Autism Spectrum Disorders

Behavioral techniques utilized in treating TD children, have also been used to treat children with ASDs. Further, behavioral techniques (e.g. Applied Behavior

Analysis; ABA) are widely for teaching and managing daytime behavior problems in children with autism, making it an intuitive approach to treating sleep problems in this population. The first study examining behavior intervention for sleep problems in children with autism was published in 1964. Wolfe, Risley, and Mees applied extinction and mild punishment (door closure) to sleep problems in a 2.5 year-old hospitalized child with autism. Howlin (1984) reduced bedtime resistance and night waking in a 6 year-old boy with autism by gradually increasing the mother's sleeping distance from the child. Durand, Gernert-Dott, & Mapstone (1996) included a 12 year-old boy with autism in a multiple baseline study where sleep problems were treated with bedtime routines and graduated extinction. Following treatment, the child with autism demonstrated a decrease in bedtime resistance and sleep onset. Piazza, Fisher, and Sherer's (1997) study of 14 children with severe MR, sleep problems, and daytime behavior problems included three children with autism. Their study compared the use of delayed bedtime and removal from the bed if sleep onset was more than 15 minutes compared to bedtime fading (e.g. scheduling bedtime when sleep onset was likely to occur) alone. Interestingly, one child with autism demonstrated the most improvement, while another child, the least improvement. Schreck's 2001 review article revealed four basic themes in behavioral techniques from the preceding body of literature: bedtime routines, extinction, stimulus fading, and faded bedtimes, with only extinction providing sufficient evidence for possibility of an efficacious intervention for sleep problems in ASD. The literature in the past decade builds upon these techniques, but includes other components such as parent education, sleep hygiene, positive reinforcement, and use of social stories and bibliotherapy.

Although almost all behavioral techniques (e.g. studies of TD children and those with NDDs) recommend the establishment of a bedtime routine and good sleep hygiene; however few studies examine these components. While the mechanism of sleep hygiene is not fully understood, it may work by entraining intrinsic circadian rhythms to the external environment. Additionally, behavioral conditioning results in the association of certain activities and environments to promote sleep. Recently, Reed et al. (2009) piloted a study in 20 families to examine the impact of a parental behavioral sleep education workshop on insomnia in children with ASD. A series of three workshops focused on: 1) establishing effective daytime and nighttime habits and a bedtime routine; 2) minimizing night waking(s) and early morning awakenings (graduated extinction and positive reinforcement), and 3) individualized sleep concerns and medical causes of sleep disorders. Researchers utilized several subjective and objective measures (e.g. actigraphy) to baseline and post-treatment data. Results indicated CSHQ scores on the insomnia subscales improved significantly including sleep onset delay, bedtime resistance, sleep duration, and sleep anxiety, while night wakings did not improve significantly and persisted in the majority of the children. Actigraphy data (for 12 out of the 20 children) supported CSHQ results, and improvements were seen in daytime behavior (e.g. hyperactivity, self-stimulatory behavior, sleep disturbance, and restricted behavior). Parental stress did not change, but 77% believed their children's sleep habits improved. This study uniquely contributed to the literature by demonstrating parent education and sleep hygiene as effective strategies for improving sleep with ASDs. Several strengths of the study included multi-method data collection techniques, and a sample which included several children on medication and diverse cognitive functioning,

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to allow for generalization to a diverse population of children with ASDs. Limitations included small sample size, lack of a control group, and reliance solely upon parent report to measure daytime behavior improvements.

Like Reed et al.'s (2009) study, Christodulu and Durand implemented positive bedtime routines. They paired this with sleep restriction and utilized a multiple baseline design to treat chronic bedtime resistance and night awakenings in four children (ages 2-5) with physical and developmental disabilities (2 of the 4 participants were diagnosed with an ASD). Sleep restriction, based on the premise that excess time in bed exacerbates insomnia (Spielman, Saskin, & Thorpy, 1987), involves reducing time spent awake in bed as well as the time spent asleep, until the desired sleep schedule is achieved. This may be considered to be advantageous as it prevents behaviors frequently associated with extinction. Actigraph data was collected on one child to determine reliability of parent reporting with sleep diaries (the other three children were resistant to the device). A reduction in total sleep time was reported in three out of the four participants. Despite this, bedtime disturbance and night awakenings reduced in all participants, and all parents reported an increase in parent satisfaction prior to intervention and at one month follow-up.

Weiskop et al. (2005) also evaluated the effectiveness of a parent training program as described by Reed et al. (2009); however this program targeted the use of behavioral principles (less focus on sleep hygiene), including the techniques of bedtime routines, reinforcement, effective instructions, partner support, and extinction. Weiskop et al. initially presented on case in a 5 year-old boy with autism in 2001, which was later included in their 2005 study. Utilizing a concurrent multiple baseline design, participants

included five children with autism, one with AD, and seven children with Fragile X

syndrome (FXS). Only baseline data was available for the child with ASD as he was

withdrawn from the study due to illness. Children ranged in age from 1.1 years to 9.1

years (mean age 5.1 years). None of the children with an ASD were taking medication.

Data was collected by parents via a sleep diary, the Goal Achievement Scale (GAS;

Hudson, Wilken, Jauernig, & Raddler, 1995), and a social validity questionnaire. After

obtaining a detailed sleep history and conducting a functional behavior assessment,

parents participated in five training sessions designed to teach behaviorally-based

principles of learning. Sessions focused on establishing a bedtime routine, reinforcement,

visual supports, partner-support skills, and extinction (for resisting bedtime and/or getting

out of bed). All of the autism participants demonstrated an extinction burst in pre-sleep

disturbance, followed by a dramatic decrease. Number of nights per week the child fell

asleep alone and in their own bed dramatically increased during the intervention, and was

mostly maintained at the 12 month follow-up. Night wakings improved in three of the

five participants with autism and one did not decrease as there were no night wakings at

baseline. One child patterns of night wakings remained variable. Lastly, in the children

with autism, co-sleeping dramatically decreased, and the trend persisted at the 12 month

follow-up. Of the five children with FXS, two experienced an extinction burst, but

overall, all showed decreased pre-sleep problems, increased ability to fall asleep, and

decreased co-sleeping. Interestingly, for all the participants, improvement in pre-sleep

disturbance only occurred after extinction procedures were implemented, with a rapid

effect. Improvement in night waking was variable in the children with FXS. Most

improvements were maintained at the three month follow-up (no 12 month follow-up was

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conducted for this group). GAS results indicated most parents perceived an improvement in their child's sleep. Regarding social validity, reported program strengths included outcomes, support, and instruction. The greatest reported weakness by parents was time commitment; however, 100% of respondents concluded they would recommend the program to a friend and 70% gave the program the maximum rating.

The authors acknowledged several methological problems, including, participant retention (two participants who dropped out of the program had the highest level of behavioral problems), and lack of objective data collection methods. Limited ability to generalize results (e.g. due to participants limited to two disability groups) was also cited as a limitation; however, this could also be cited as a strength as homogeneity of sample may assist in determining what interventions are most beneficial for these populations. Additionally, the intervention was limited by its intensive nature, requiring a high level of therapist time and support, and time of parental involvement. Despite these weaknesses, several strengths were also apparent. First, this study included follow-up data collection at three months as well as a year, unlike most other studies reviewed. Secondly, parents choose sleep-related goals and determined what constituted success, as opposed to predetermined scored chosen by clinicians. Lastly, the treatment is accompanied by an extensive manual for replication.

One single subject case study examined the use of a personalized social story in conjunction with behavioral strategies (e.g. positive reinforcement, visual supports, bedtime routine, and graduated extinction) to reduce co-sleeping, bedtime resistance, and night awakenings in a four-year-old boy diagnosed with severe learning disabilities, ASD, and speech and language delay (Moore, 2004). Although reportedly successful,

this study was greatly limited by lack of results data and the implementation (specifically therapist-parent interactions) was not specifically described. However, the use of a children's story to treat sleep problems is an interesting and understudied intervention.

Only one other study (Burke, Kuhn, & Peterson, 2004) examined the use of a story and behavioral strategies to reduce sleep problems, albeit in TD children with daytime behavior problems (no diagnoses reported). Utilizing a multiple baseline single case design, disruptive bedtime behaviors and night awakenings decreased in four children (ages 2 to 7). This study had a stronger methodology as data regarding sleep problems was obtained via the CSHQ and sleep diary, detailed intervention and results data were provided, data was collected on daytime behavior via the CBCL, and children were followed-up after three months. Like most others, the study was limited by its small sample and subjective data collection techniques; however, this study makes a unique contribution as the intervention involved minimal cost, time, or therapist support, and utilized an activity (e.g. reading a bedtime story) already part of most children's bedtime routine.

Thus, it is proposed that the study by Burke et al., (2004) serve as the foundation for an intervention treatment package designed to reduce bedtime resistance and night waking(s) in children with ASDs. While the aforementioned study was conducted in a small sample of TD children, the intervention was highly effective. Next, the activity is one which is developmentally-appropriate for children ages 3 to 10 years and most families typically read books as part of their bedtime routine. While the study conducted by Williams et al. (2006) indicated reading a story before bed and rewards for falling asleep were perceived among the least helpful behavioral intervention, this may not be

valid for two reasons. First, it was unlikely stories were read as methods of teaching and reinforcing appropriate bedtime expectations. Secondly, reward selection would likely impact effectiveness as children with autism often have unique reinforcers than TD children. These two problems will be addressed by the nature of the story (which conveys parental and child expectations) as well as support from the researcher (e.g. selecting highly reinforcing rewards). Perhaps most attractive, the proposed invention involves very little cost, time, or therapist intervention. Moreover, it is delivered by parents in the comfort of their home. The treatment will also be enhanced by including written parent information on sleep hygiene and principles of extinction and reinforcement which may be kept and utilized for reference.

In conclusion, treatment for sleep problems in children with ASDs is likely multi-factorial and relates to atypical development found in the ASDs within the context of intrinsic biological or genetic abnormalities, psychological or behavioral characteristics associated with the core features of ASDs, and factors in the family home or environment. Behavioral intervention has been and is currently utilized to treat a variety of learning and behavioral problems in children whose deficits originate from an underlying genetic or biological basis. In the literature, behavioral techniques have been applied to the treatment of sleep problems, in TD children, those with NDDs, and specifically, in children with ASDs. Moreover, the behavioral techniques of ABA are widely accepted for teaching and managing behavior problems in children with autism, have been applied to sleep problems in children with ASDs, and served as the basis for the proposed intervention.

CHAPTER III

METHODS

Introduction

The effectiveness of the intervention was evaluated utilizing a single-case, nonconcurrent multiple-baseline across subjects research design. This method was chosen for the following reasons: First, historically derived from laboratory research on operant conditioning (Kazdin, 1982), the single-case design allowed for continuous assessment of the targeted behavior over time, compatible with data collection methods utilizing direct observation. Secondly, the multiple baseline design allowed participants to be treated without the need for withholding or withdrawal of the treatment, or reversal of the design. Lastly, single-case design was an appropriate method for treatment package evaluation (Kazdin, 1982) on a single subject or small group of subjects to determine possible effectiveness, feasibility, and to resolve methodological problems before attempting to implement the intervention in a larger sample.

Participants

A clinical sample was recruited via an electronic notification to families who participated in the voluntary list serve managed by The Autism Center, located in Pittsburgh, PA. Inclusion criteria included children with a diagnosis of Asperger's Disorder or Pervasive Developmental Disorder-NOS (American Psychological Association, 2000), as measured by the Gilliam Autism Rating Scale or Gilliam Asperger's Disorder Scale (Gilliam, 1995) in addition to a diagnosis by a licensed psychologist. Children were within the age ranges of 3 years to 10 years of age as the story may not be developmentally appropriate for children younger or older. Participants

also met the following criteria established by Mindell and Durand (1993; cited in Burke et al., 2004, p. 390):

1. Medical etiologies were not believed to contribute to the sleep disturbance.

2. For a minimum of three nights per week, the child resisted going to bed, fell asleep in a location other than his or her bed, or required parental intervention or presence to return to sleep.

3. The parents indicated to the clinician a desire for the child to fall asleep independently and sleep in his or her own bed throughout the night.

4. The sleep problems had been occurring for a minimum duration of 4 weeks.

Measures

Parents completed four questionnaires including a Background Information Sheet, the Children's Sleep Habits Questionnaire (CSHQ), the Child Behavior Checklist (CBCL), and the Treatment Evaluation Inventory (TEI). In addition, they were asked to complete a brief entry in a sleep diary each morning and night. All forms were completed by the caregivers at home. The Background Information Sheet was completed prior to the start of the study. The CBCL and CSHQ was administered pre- and post-intervention, as well as at the one-month follow-up. Sleep diaries were completed for the duration of the baseline and intervention phases, as well as for a period of one week at the one month follow-up.

Background Information Sheet

The Background Information Sheet was developed for this project and consisted of questions regarding demographics of the caregiver(s) including education, employment, income, and geographic location.

The Children's Sleep Habits Questionnaire, Abbreviated Version (CSHQ)

The Children's Sleep Habits Questionnaire (CSHQ) is a retrospective, 45 item parent checklist designed to screen common sleep problems in children ages 4 through 12 years of age (Owens, Spirito, & McGuinn, 2000) based on the International Classification of Sleep Disorders (ICSD-DCM; American Academy of Seep Medicine, 2001) diagnoses. Items are rated by parents or caregivers on a three-point scale, indicating behaviors related to sleep which *typically*, *usually*, and *sometimes* occur over a "typical" recent week. The abbreviated version, utilized for this study, consists of 33 items and contains all of the subscales and items which are scored on the original assessment. The CSHQ items reflect eight key domains reflected in clinical sleep complaints, including: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness. An overall score of 41 is suggested by the authors as a cut-off score for referral for a sleep problem as it correctly identified 80% of the clinical sample in the study. However, as there are no established norms for the total and subscale scores, the authors recommend the instrument is most useful in comparing samples or assessing sleep pre- and post-intervention. The CSHQ is made available by the authors free of charge at http://www.kidzzzsleep.org/researchinstruments.htm.

Referring to the dependability or trustworthiness of an instrument, Urbina (2004, p. 117) describes reliability in measurement to be "a quality of test scores that suggests they are sufficiently consistent and free from measurement error to be useful." To assess reliability and validity, Owens et al. (2000) surveyed parents of 469 school-aged children, aged 4 through 10 years (community sample) and parents of 154 patients diagnosed with

sleep disorders in a pediatric sleep clinic. Children aged eleven on above were removed from the sample to minimize possible pubertal influences on sleep. The closer a reliability coefficient is to 1.0, the more reliable the test. Two reliabilities were reported on the CSHQ, internal consistency and test-retest reliability. Internal consistency was measured using Cronbach's alpha (α) coefficient and test-retest reliability was calculated utilizing Pearson's correlation coefficients for the subscale scores and Spearman Brown's correlation coefficient for the item scores. Internal consistency for the entire CSHQ was .68 for the community sample and .78 for the clinical sample. Alphas for individual subscales ranged from .36 to .70 in the community sample and .44 to .83 in the clinical sample with Sleep Duration, Bedtime Resistance, Daytime Sleepiness, and Sleep Anxiety having the highest internal consistency. Test-retest reliability was assessed in a volunteer sample of 60 parents from the community sample after a two-week interval, and with the exception of Sleep Duration (.40) ranged from .62 to .79 on the individual scales of the test.

Test validity, as defined by Anastasi & Urbina, (1997), "concerns what the test measures and how well it does so (p. 113)," and should be shown to correlate with various criteria used in making decisions or predictions (Urbina, 2004). Validity was investigated by comparing the clinical sample to the community sample for each item and subscale. Difference in 30 out of 33 items were statistically significant at the p<.001, with three items on the Daytime Sleepiness subscale not significant. The clinical sample had significantly higher scores (p<.001) than the community sample on all subscales as well on total score (after controlling for age and SES), indicating higher levels of sleep problems in the clinical sample. Lastly, sensitivity and specificity were examined using

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the Receiver Operator Characteristic (ROC) curve with sensitivity calculated at .80 and specificity at .72, indicating 80% of the clinical group was correctly identified.

A new questionnaire, the Family Inventory of Sleep Habits (Malow et al., 2009) was recently created based on the CSHQ, to measure sleep hygiene in children with autism. Due to the fact this assessment is focused exclusively on this aspect (e.g. sleep hygiene), it was not considered for use in the study.

Sleep Diary

A sleep diary was developed for the project based on the current research questions and a sample from the American Academy of Sleep Medicine. The structured diary form was created to assist parents in recoding the behaviors targeted for intervention, including the frequency of disruptive bedtime behaviors, night awakenings, and sleep events, such as the time the child went to bed, sleep latency, as well as medication usage.

The Child Behavior Checklist (CBCL)

The Child Behavior Checklist (CBCL 4-18) (Achenbach, 1991) is a widely used, standardized instrument to measure behavioral and emotional problems in children 4 to 18 years of age (Achenbach, 1991). Raters may include parents/caregivers, teachers, or self-report, and consists of a 113-item three-point scale (0 to 2) combined with a seven-part social competency checklist. The instrument yields scores on internalizing and externalizing behaviors, a total score, and scores on DSM-IV-related (APA, 2000) scales. The CBCL yields T scores (Mean = 50; Standard Deviation = 10), with a score below 65 falling in the normal range, between 65 and 70 falling within the borderline range, and a score of 70 or above within the clinical range.   A preschool version of the CBCL was

created for children ages 2 to 3 years, and it is composed of a 100-item behavior problems checklist (Achenbach, 1992). The younger version of the CBCL contains a Sleep Problems subscale, in addition to six other subscales.

The CBCL is considered to be a highly reliable and valid instrument. Reliability was measured using Person's r (test-retest), Cronbach's alpha (internal consistency) and Pearson's r (inter-rater between parents). Across ages and groups, including the preschool version, the internal consistencies of the Externalizing and Total Problem scores are in the .92 to .96 range, and in the .88 to .92 range for the Internalizing Scale (Doll, Furlong, & Wood , 1998; Furlong & Wood, 1998). Regarding validity, multiple indices demonstrate high concurrent correlations with similar correlations on the Connors (1973) Parent Questionnaire (.59 to .86) and the Quay-Peterson (1983) Revised Behavior Problem Checklist (.59 to .88). Strong discriminant validity has also been evaluated (Furlong & Wood, 1998).

Treatment Evaluation Inventory - Short Form

The Treatment Evaluation Inventory – Short Form (TEI-SF; Kelley, Heffer, Gresham, & Elliott, 1989) is a shortened and simplified version of the Treatment Evaluation Inventory (TEI; Kazdin, 1980). It consists of nine items on a 5-point scale, and is used to assess parental judgments of treatment acceptability and perceived efficacy. Coefficient alpha estimate of internal consistency for the TEI-SF is .85, and did not differ significantly from internal consistency for the original TEI (.89) (Kelley et al., 1989). Additionally, the TEI-SF was found to discriminate among three different treatments and loaded on two factors as did the original TEI.

Research Design

The study utilized a single-case, nonconcurrent multiple baseline design across two children and families. In multiple baseline designs the performance is examined across more than one baseline. To effectively show a change in behavior, the behavior only changes when the intervention is applied. When the intervention is introduced, a test is made between the level of performance and the projected level of the baseline. In instances in which data is gathered on two or more persons, the design begins with the observations of baseline performance of the same behavior for each person. After the behavior has reached a stable rate, either by staying the same or becoming worse, the intervention is applied to one individual at a time while the other participants do not receive the intervention and baseline data is still collected. The participant who is exposed to the intervention is expected to have a change in behavior, while the others are expected to remain stable. The procedure continues until all participants receive the intervention (Kazdin, 1982).

A minimum of two baselines is needed to display that the intervention is effective. The effectiveness of the intervention is clearer when more participants or baselines are used. The use of more baselines ensures that the intervention was responsible for the behavior change, and outside factors did not affect the results. If the behavior of all participants does not change, the researcher may be able to reason why the intervention worked only under certain conditions (Kazdin, 1982).

The design was chosen as it allowed for continuous assessment of the targeted behavior over time, participants could be treated without the need for withholding or withdrawal of the treatment, and it provide the opportunity to evaluate efficacy, feasibility, and to resolve methodological problems before attempting to implement the intervention in a larger sample. Additionally, the effect of the intervention may be

cumulative, and thus, difficult to reverse. While the nonconcurrent design incorporates features of the traditional concurrent multiple baseline design, measurements and manipulations across data series are not temporally aligned. The baseline phase is applied for differing lengths of time, allowing the researcher to apply the intervention when a stable baseline trend is revealed. This also provides for more flexibility in subject recruitment, as opposed to starting all subjects at the same time with the concurrent design.

Major threats to internal validity for this study included history, maturation, testing, instrumentation, selection biases, attrition, and diffusion of treatment (Kazdin, 1982). The use of a multiple baseline design directly addressed issues of history: the possible impact of common events will be controlled by different baseline and starting points for intervention. There was no change in instrumentation over the course of intervention and no attrition of participants. Selection biases and diffusion of treatment were not factors as all participants will receive the same intervention. Maturation was unlikely a threat to internal validity as sleep patterns do change with age; however, this change occurred in terms of years, not over the brief time span of the study. Measurement via the sleep diary could be problematic if caregivers did not complete the diary on a daily basis, and then sought to record data at one episode. The impact of this problem was minimized by weekly phone calls or emails from the investigator reminding caregivers to record the data on a daily basis. Fidelity to intervention was measured via use of the sleep diary.

Major threats to external validity for this study included generality across subjects, generality across times, generality across behavior change agents, reactive

experimental arrangements, reactive assessment, and multiple treatment interference (Kazdin, 1982). Threats regarding generality were addressed by use of the multiple baseline design with more than one participant. Follow-up data at one month was collected to address threats to generality across time, specifically, maintenance. Regarding generality across behavior change agents, the intervention is easily replicated as the storybook provided guidelines for its use within the text. Reactive experimental arrangements and assessment was not an issue as the reading of a bedtime story was a developmentally appropriate activity which children participated in prior to the intervention. Additionally, parents collected the data, as opposed to an unfamiliar third-party. Multiple-treatment interference was a threat to external validity as the intervention consisted of a treatment package presented simultaneously.

The independent variable for this study was the bedtime story intervention package. The dependent variables for the study were as follows: The Children's Sleep Habits Questionnaire, Abbreviated Form, sleep diary, Child Behavior Checklist, and Treatment Evaluation Inventory, Short Form. Dependent variables measured by the sleep diary were based upon Burke et al.'s 2004 study, and included:

Disruptive bedtime behaviors – Including, but not limited to stalling, noncompliance, vocal protests, calling out for parents, crying, screaming, tantrums, complaining, demanding, and aggression. To minimize caregiver burden while accurately assessing disruptive behaviors, caregivers were asked to count each episode of an overt behavior (e.g. hitting, yelling, etc). For each minute an extended behavior lasted, one instance of behavior was recorded.

Night wakings – Anytime a child arouses a parent/caregiver and requires him or her to do something to settle the child. If the child sleeps in a caregiver's bed, the number of instances and duration of each instance was recorded.

Sleep onset time – The time from when the child is in bed with the lights out until the time the parent observed the child asleep.

Total Sleep time – The time from which the child went to sleep and woke in the morning, minus total time of night waking(s).

Procedures

Participants were obtained via a study notice sent via the computerized list serve from the Autism Center, located in Pittsburgh, Pennsylvania. The list serve was a voluntary email list managed by the Autism Center. Participants were required to meet the criteria reported in the *Participants* section described above. Following Institutional Review Board approval from the University, informed consent was obtained from each parent/caregiver participating in the intervention. Consent for participation was reviewed with all caregivers with clearly outlined benefits and risks to participation.

Once recruited and informed consent was signed, caregivers were given the option of reviewing study materials in person with the investigator at a location of their choice, or independently. *The Sleep Fairy* story was sent via mail, and caregivers had the option of receiving hardcopy of the study materials, or materials (excluding the book) via email. Parents were provided with a self-addressed stamped envelope to return study forms. No compensation was provided for the study; however, families were allowed to keep *The Sleep Fairy* book, regardless of length of participation.

Baseline

Caregivers completed the Background Information Sheet, CBCL, and the CSHQ upon receiving study materials. They also completed sleep diary entries until stability of targeted behaviors was determined (Kazdin, 1982). Baseline data was then sent to the investigator via mail.

Intervention

The intervention consisted of a bedtime story treatment package including: two parent handouts regarding sleep hygiene and behavioral principles, *The Sleep Fairy* book, and the use of positive reinforcement and extinction. At the beginning of the study, parents were provided information regarding good sleep hygiene, and were asked to follow the principles outlined in the handout, for the duration of the study. Caregivers were then be asked to read *The Sleep Fairy* (Peterson & Peterson, 2003) at the conclusion of the child's nightly routine, and follow the principles of extinction and reinforcement outlined on the introductory two pages of the text and in the informational handout. The text consisted of 14 pages of text and illustrations, which described how two children overcame their bedtime problems and learned how to get ready for bed, and stay in bed without difficulty. A poem embedded in the story specified appropriate bedtime behaviors and described how the sleep fairy left a treat (positive reinforcement) for children who followed bedtime routines and fell asleep without problems. The two introductory pages provided parents with brief instructions on how to set clear bedtime expectations, use reinforcement contingent on appropriate bedtime behavior, and select reinforcers. Please note, although previously referred to as a social story by Burke et al. (2004), *The Sleep Fairy* does not meet criteria as described by Gray and Garand (1993) and was referred to as a bedtime story throughout the study.

The story was read each night for two weeks, and then faded by the parents as determined by the procedures within the text. Data obtained via the sleep diary was collected for a period of 30 days. Participants were contacted via phone or email, depending upon their preference, one time per week to answer questions or concerns. At the end of the 30-day period, caregivers again completed the CSHQ, the CBCL, and the Treatment Evaluation Inventory – Short Form. Data was sent to the investigator via mail.

One month Follow-Up

One month from the completion of the 30-day intervention, caregivers were asked to collect one week of sleep diary data. At the end of the week, they again completed the CSHQ and the CBCL. Forms were completed via hard sent to the investigator via mail.

Data Analysis

The parent/caregiver of each subject completed the Children's Sleep Habits Questionnaire and the Child Behavior Checklist at the start of the baseline phase, completion of the intervention phase, and at the one month follow-up. Sleep diaries were completed one per day for the duration of the baseline, treatment, and one-week follow-up phase. The Treatment Evaluation Inventory – Short form was completed at the end of the 30 day intervention. The collected data was analyzed by use of visual analysis (Kazdin, 1982), and effect size using Cohen's d (Allison & Gorman, 1993).

Visual Analysis of the Graphed Data

Five criteria were employed in the visual analysis of graphed data (Kazdin, 1982): (a) changes in mean level of performance across phases, (b) changes in level of performance from the end of one phase to the beginning of the next phase, (c) changes in

trend or slope from one phase to the next, (d) the latency of behavior change across phase, and (e) percentage of nonoverlapping data.

Effect Size Calculation

Along with visual analysis, effect size was calculated to provide a measure of the magnitude of treatment impact. Effect size was calculated using Cohen's *d* (Allison & Gorman, 1993). Cohen's d is the standardized difference between the means of two groups. The formula is $d = (M_{treatment} - M_{control})/s$, where $M_{treatment}$ is the mean of the treatment, $M_{control}$ is the mean of the control, and s is the standard deviation.

Children's Sleep Habits Questionnaire and Child Behavior Checklist

The results of the CSHQ and CBCL scales completed prior to baseline and at the termination of the intervention, as well as the CSHQ and CBCL completed at the one month follow-up by the parent/guardian were analyzed descriptively to examine changes over time.

Treatment Evaluation Inventory, Short Form

The results of the TEI-SF completed at the end of the intervention phase was analyzed descriptively.

Research Questions and Hypotheses

1. Will the introduction of a bedtime story treatment package reduce bedtime resistance?

   Hypothesis: Bedtime resistance as reported by parents will decrease as a result of the intervention.

2. Will the introduction of a bedtime story treatment package reduce nighttime awakenings?

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Hypothesis: Nighttime awakenings as reported by parents will decrease as a result of the intervention.

3. Will the introduction of a bedtime story treatment package decrease sleep latency?

    Hypothesis: Sleep latency will decrease as a result of the intervention.

4. Will the introduction of a bedtime story treatment package increase total sleep time?

    Hypothesis: Total sleep time will increase as a result of the intervention.

5. Will improved sleep decrease daytime problem behavior?

    Hypothesis: Daytime behavior problems as measured by parents and teachers will decrease as the result of improved sleep.

6. Will the bedtime story treatment package be acceptable to parents?

    Hypothesis: The intervention will be acceptable to parents.

7. Will levels of behavior at the completion of the intervention be maintained after one month?

    Hypothesis: Levels of behavior at the completion of the intervention be maintained after one month.

8. Will parents level of perceived stress decrease after the intervention?

    Hypothesis: Perceived stress will decrease after the intervention.

CHAPTER IV

RESULTS

Single Subject Analysis of the Research Questions

Baseline data was collected in the form of the Children's Sleep Habits

Questionnaire, Abbreviated Form (CSHQ; Owens et al., 2000), the Child Behavior

Checklist (CBCL; Achenbach, 1991), and a 7 to 10-day sleep diary. During the 30-day

intervention, data was collected in the form of a sleep diary, measuring items including

sleep latency, disruptive bedtime behaviors, night waking, and perceived parent stress.

At the end of the 30-day intervention, parents again completed the CSHQ and the CBCL.

Additionally, parents or caregivers completed the Treatment Evaluation Inventory –

Short Form (TEI-SF; Kelley et al., 1989) to measure treatment acceptability and

perceived efficacy. Lastly, one month following the completion of the intervention,

another 7 to 10-day sleep diary was completed, in addition to an additional CSHQ and

CBCL. Data was collected by the participants in their homes, and occurred from May

2010 to August 2010. Data from Participant 1, for days seven and eight obtained during

the baseline phase, was omitted as it was collected on a holiday and not indicative of the

child's typical daytime or bedtime routine. Additionally, data was not collected on days

24 and 25 during intervention for the same participant, thus, data is provided for 28 days.

Data was analyzed using visual analysis (Kazdin, 1982), percentage of nonoverlapping

data points (Scruggs, Mastropieri, & Castro, 1987), and effect size (Allison & Gorman,

1993).

Research Question 1

Does the introduction of a bedtime story treatment package reduce bedtime resistance?

Hypothesis: Bedtime resistance as reported by parents will decrease as a result of the intervention.

CSHQ results, Bedtime Resistance subscale.

Changes in scores on the *Bedtime Resistance* subscale of the CSHQ were examined from the baseline, intervention, and follow-up phases for each participant (See Table 4.1). Consisting of six items rated from one to three, a score of six indicates the lowest level of problems related to bedtime resistance, while a score of 18 indicates the highest level of concerns. As rated by his mother, Participant 1 demonstrated a decline across phases of the study, with his raw score starting at an 11 in the baseline phase, a seven at the end of the intervention phase, and a six at the 30-day follow up, indicating minimal concerns in regard to bedtime resistance. Participant 2, also rated by his mother, demonstrated variability in symptoms, with a baseline score of ten, an intervention score of eight, and a follow-up phase score of 11.

Table 4.1

CSHQ Bedtime Resistance Subscale

CSHQ Item	Subject 1			Subject 2		
	Baseline	Intervention	Follow-up	Baseline	Intervention	Follow-up
Goes to bed at same time	1	1	1	2	1	1
Falls asleep in own bed	2	1	1	2	1	2
Falls asleep in other's bed	2	1	1	2	2	2
Parent in room to sleep	2	1	1	1	1	2
Struggles at bedtime	2	2	1	1	2	2
Afraid of sleeping alone	2	1	1	2	1	2
Subscale Total	11	7	6	10	8	11

Visual Analysis of the Graphed Data

Five criteria were employed by the experimenter to analyze the collected data (Kazdin, 1982): (a) changes in the mean level of performance across phases, (b) changes in the level of performance from the end of one phase to the beginning of the next phase, changes in the trend or slope from one phase to the next, (c) changes in trend or slope from one phase to the next, and (d) the latency of behavior changes across phases, and (e) percentage of nonoverlapping data. To address this research question, results from the sleep diaries from baseline through the one month follow-up period were analyzed for each participant.

Changes in sleep diary means.

Mean bedtime resistance for Participant 1 at baseline was 6.16 incidents of disruptive bedtime behaviors, decreasing to 3.25 mean episodes during intervention phase, with a final decline to 0.57 mean incidents at the 30-day follow-up. Participant 2 also evidenced a similar decline in mean episodes of bedtime resistance, with a baseline mean of 4.88 occurrences, declining to 1.53 mean episodes during the intervention phase, and a mean of 0.00 at the week-long 30-day follow-up.

Changes in level.

Drastic change from the end of baseline to the start of treatment was not observed for Participant 1 (See Figure 4.1), with a total of eight episodes of bedtime resistance at the end of the baseline phase, to a total of five instances of bedtime resistance on the first night of intervention. Conversely, a vivid change was observed in Participant 2, with three episodes of bedtime resistance on the final night of baseline data collection, to an increase of seventeen instances of bedtime resistance on the first night of intervention.
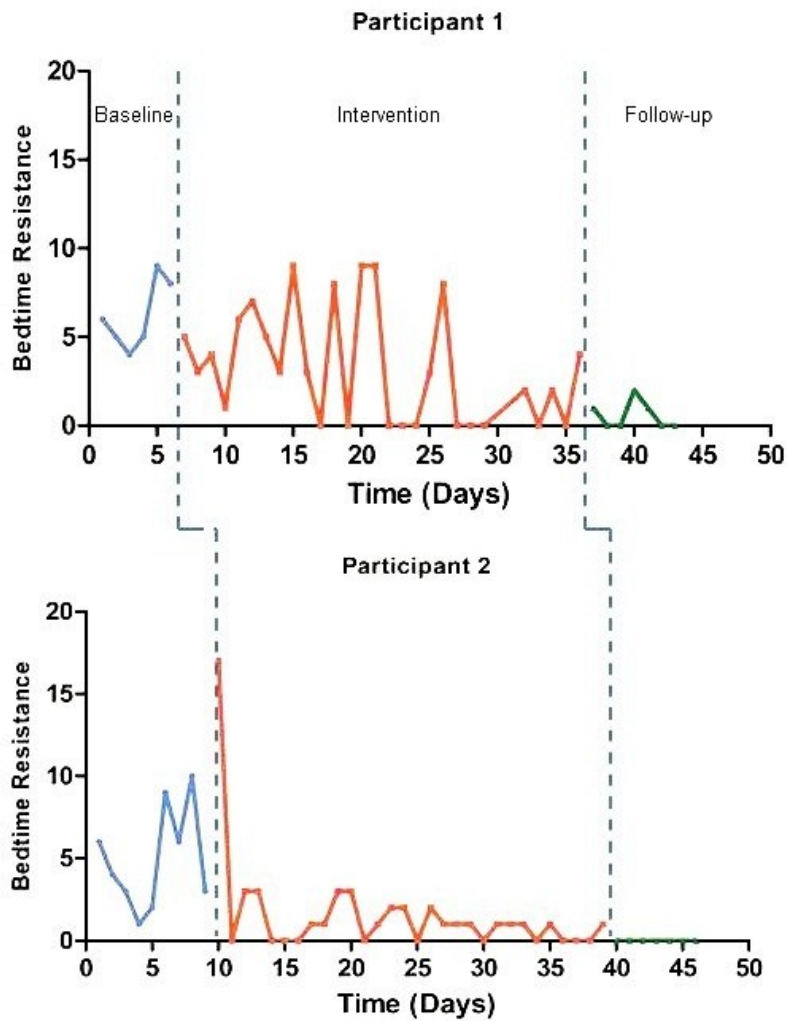
Figure 4.1 Occurances of disruptive bedtime behaviors by intervention phase

Changes in trend.

Examination of the linear regression trend line for each participant indicated a

decline of instances of disruptive bedtime behaviors over the course of treatment.

Participant 1 evidenced an overall increase in disruptive bedtime behaviors during the

baseline phase, but evidenced a steady decline in the intervention as well as follow-up

phases. Participant 2 also evidenced a declining trend within the intervention phase, and

a less significant decline during the follow-up phase; however, no instances of disruptive

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bedtime behaviors were recorded during the week-long data collection which occurred 30 days post intervention.

Latency of change.

Visual inspection of data indicated that change in behavior occurred immediately after the beginning of treatment for each participant based on sleep diary data. Participant one evidenced a small decrease in instances of bedtime resistance, while participant two experienced a significant extinction burst, with a change from three instances of bedtime resistance on the final night of baseline, to 17 instances on the first night of intervention.

Percentage of nonoverlapping data

Percentage of nonoverlapping data points was employed to further insure careful visual analysis. The less overlap found between data points, the more effective and reliable the intervention (Scruggs, Mastropieri, and Castro, 1987). From the results of the sleep diary, visual inspection revealed that 63.75% of points were nonoverlapping between the baseline and intervention phase for Participant 1 (11 data points overlapped). For Participant 2, 43.34% of the data points (17 data points overlapped) in the intervention phase were nonoverlapping with the baseline phase; however, a wider range of data points (nine) was noted for the second participant. The overall total for this study indicated the 51.72% of the data was nonoverlapping between the baseline and intervention phases.

Effect Size

Along with visual analysis, effect size was calculated to provide a measure of the magnitude of treatment impact, and compared baseline to overall intervention. Effect

size was calculated using Cohen's *d* (Allison & Gorman, 1993). The effect size of the sleep diary results measuring occurrences of bedtime resistance from the baseline phase to the end of the intervention phase was large for both participants with .89 for Participant 1 and .99 for Participant 2. It was also strong from the intervention phase to the 30-day follow-up phase with an effect size of .85 for Participant 1, but medium (.53) for Participant 2.

Research Question 2

Will the introduction of a bedtime story treatment package reduce nighttime awakenings? Hypothesis: Nighttime awakenings as reported by parents will decrease as a result of the intervention.

CSHQ results, Night Wakings subscale

Three items on the CSHQ measured night wakings on the CSHQ, with a score of nine signifying the highest level of difficulties and a score of three indicating minimal concerns (See Table 4.2). Participant 1's score was a six at baseline, and at the end of the intervention, with a decline to four at the 30-day follow-up. Participant 2's score was a seven in the baseline phase, with a decrease to four in the intervention and an increase to 6 at the follow-up portion of the study.

Table 4.2

CSHQ Night Wakings Subscale

CSHQ Item	Subject 1			Subject 2		
	Baseline	Intervention	Follow-up	Baseline	Intervention	Follow-up
Moves to other's bed in night	3	2	1	3	2	2
Awakes once during night	1	2	2	3	1	2
Awakes more than once	2	2	1	1	1	2
Subscale Total	6	6	4	7	4	6

Visual Analysis of the Graphed Data

   Changes in sleep diary means.

   Night wakings were reported infrequently in the sleep diary during all phases of data collection. Participant 1 demonstrated a mean of .833 night wakings during the baseline phase, with a decrease to .75 at the end of the intervention phase, and a slight increase to .786 at the 30-day follow-up. Participant 2 also demonstrated infrequent night wakings. A mean occurrence of .222 wakings occurred during the baseline phase, .1 at the end of the intervention, and 0.00 at the 30-day follow-up.

   Changes in level.

   Along with the low level of incidence in night wakings across participants and phases of the study, negligible changes in the level of night wakings were observed for both participants from the final night of baseline to the first night of intervention (See Figure 4.2). Participant one demonstrated no incidents on the last night of baseline, and one incident on the first night of the intervention phase. Participant two demonstrated no

change in night wakings from the baseline to intervention phase, with three episodes
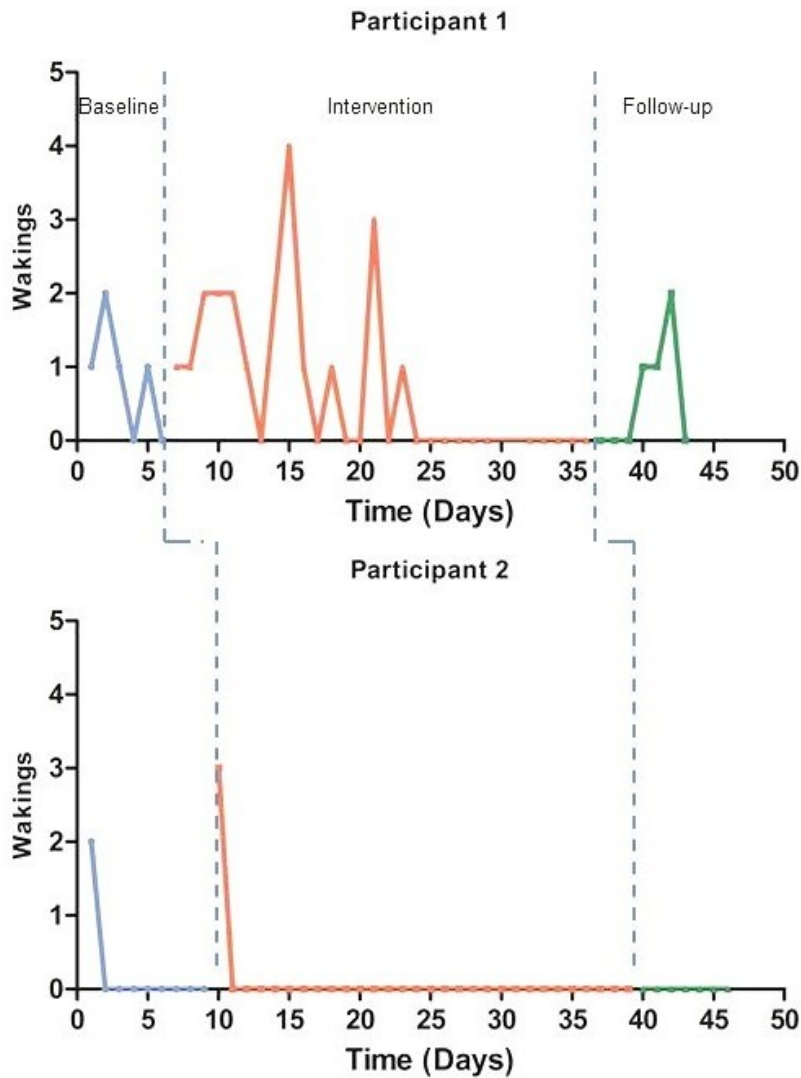
recorded both nights.

Figure 4.2 Occurances of night wakings by intervention phase

Changes in trend.

Examination of the linear regression trend line for each participant evidenced a

decline in the trend of night wakings for both participants from the baseline to

intervention phases; however, the decrease was negligible. Participant 1 evidenced a

minor increase in the trend line from the intervention phase compared to the follow-up, while Participant 2 evidenced a minor decline across phases.

Latency of change.

Visual inspection of data revealed an inconsistent pattern of change in night wakings for Participant 1, with a trend of two or less night wakings occurring for three days or more at the 21st night of intervention. A low frequency of night wakings was observed for Participant 2, with a small extinction burst occurring on the first night of the intervention phase, but subsequently decreasing to zero incidents per night for the balance of the intervention and follow-up phases.

Percentage of Nonoverlapping data

Visual inspection of the sleep diary revealed overlapping data points in all but two nights of the intervention phase for Participant 1, resulting in a low percentage of nonoverlapping data (7.15%).Participant 2 also evidenced a similar trend, with all but one data point overlapping, resulting in a 3.34% of nonoverlapping data.

Effect Size

The effect size based on sleep diary results measuring occurrences of bedtime resistance from the baseline phase to the end of the intervention phase for Participant 1 was extremely low ($d = .08$) as was the effect size from the intervention to the follow up phase ($d = .17$). Negligible effects were also observed for Participant 2 with an effect size of .212 from baseline to intervention, and .202 from intervention to the 30-day follow-up.

Research Question 3

Will the introduction of a bedtime story treatment package decrease sleep latency?

Hypothesis: Sleep latency will decrease as a result of the intervention.

CSHQ results, Sleep Onset Delay subscale

One item on the CSHQ measured sleep latency, the time it takes to fall asleep, with a score of three signifying the greatest level of concern and a one representing the least difficulty with sleep onset. Participant 1 was rated as a one across phases, signifying the child usually (e.g. 5 – 7 times per week) falls asleep in 20 minutes. Participant 2 was rated a one at baseline and intervention, and a two (e.g. 4 – 5 times per week falls asleep within 20 minutes) at the 30-day follow-up.

Visual Analysis of the Graphed Data

Changes in sleep diary means.

Mean sleep latency for Participant 1 at baseline was 37.5 minutes, decreasing to 24.82 mean minutes during the intervention phase, with a final decline to 13.57 minutes at the 30-day follow-up. Participant 2 also evidenced a decline in sleep latency from baseline to the intervention phase, with a sleep latency mean of 13.22 minutes, declining to 10.65 during the intervention phase. An increase to 14.71 mean minutes was reported at the 30-day follow-up, which began on September 1, 2010.

Changes in level.

A significant increase in sleep latency was noted for Participant 1 from the baseline to the intervention phase (See Figure 4.3), with an increase in sleep latency from 30 minutes the last night of baseline, to a sleep onset of 80 minutes on the first night of intervention. A less dramatic change was noted for Participant 2, with a ten minute increase in sleep latency on the first night of intervention.
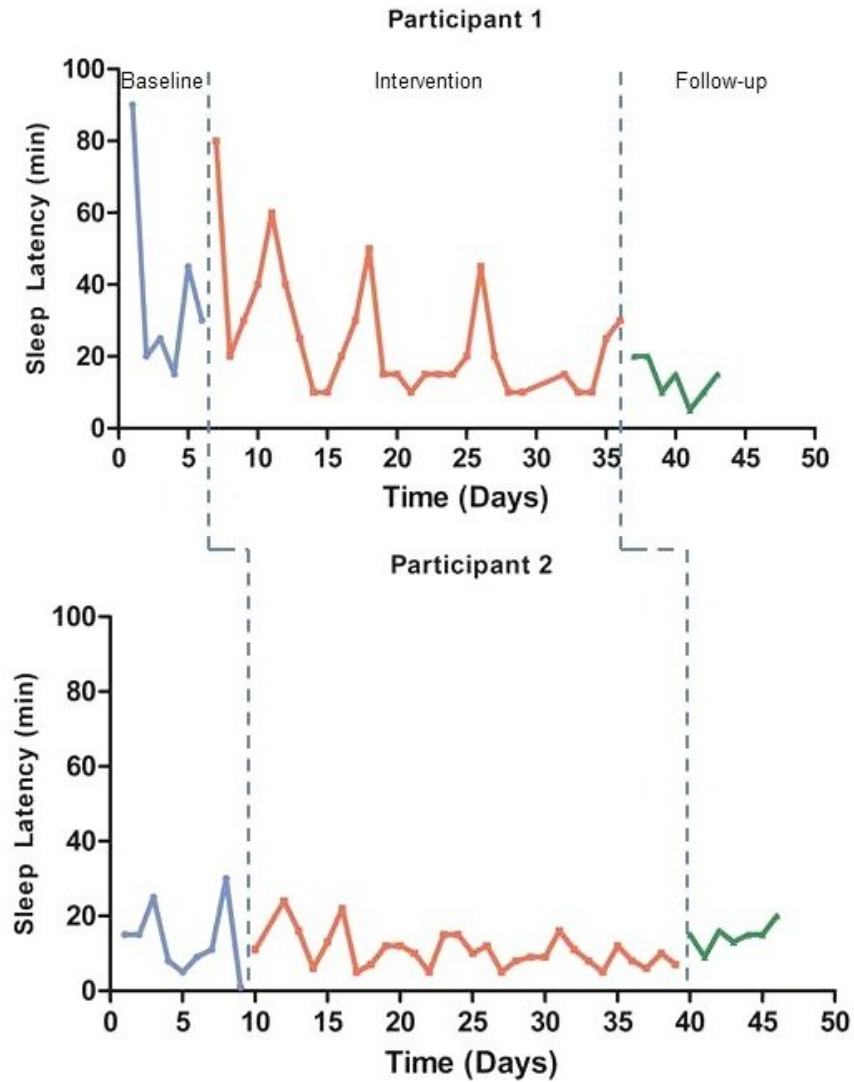
Figure 4.3 Sleep latency in minutes by intervention phase

Changes in trend.

Examination of the linear regression trend line indicated a decline in sleep latency

for Participant 1 across phases. Participant 2 also evidenced a declining trend within the

intervention phase, but demonstrated an increasing trend within the 30-day follow-up

phase.

Latency of change.

Visual inspection of data indicated change in sleep latency occurred immediately after the beginning of treatment for Participant 1. Participant 2 experienced a less dramatic increase; however, the last night of the baseline phase, sleep latency was noted to be one minute, the shortest sleep onset throughout data collection for this participant.

Percentage of nonoverlapping data

Visual inspection of sleep diary data revealed that only 3.58% of the intervention data was nonoverlapping between the baseline and intervention phase for Participant 1 (27 out of 28 data points overlapped); however, a wide range in sleep latency time (105 minutes) was observed within the baseline phase. For Participant 2, a similar trend was noted, with only one data point out of 28 nonoverlapping, resulting in the same percentage (3.58%) of nonoverlapping data. Again, a large range (115 minutes) was noted for sleep latency for Participant 2. The overall total for this study indicated that 3.58% of the data was nonoverlapping between the baseline and intervention phases for sleep latency.

Effect Size

The effect size of the sleep diary results measuring sleep latency for between both phases was moderate for Participant 1, with $d = .646$ from baseline to intervention, and $d = .692$ from intervention to the 30-day follow-up. It was less strong from the baseline to the intervention phase for Participant 2 ($d = .420$), but stronger from intervention to the follow-up ($d = .849$); however, this signified an increase in sleep latency time from intervention to follow-up for Participant 2.

Research Question 4

Will the introduction of a bedtime story treatment package increase total sleep time?

Hypothesis: Total sleep time will increase as a result of the intervention.

CSHQ results, Sleep Duration subscale

Changes in scores on the *Sleep Duration* subscale of the CSHQ, consisting of three items, were examined from the baseline, intervention, and follow-up phases for each participant (See Table 4.4). As rated by his mother, Participant 1 demonstrated a decline from the baseline to intervention, maintained at the 30-day follow-up, with raw scores of five, three, and three, respectively. Participant 2 demonstrated variability and was rated a seven at baseline, a three at intervention, and a six at the 30-day follow-up.

Table 4.3

CSHQ Sleep Duration Subscale

CSHQ Item	Subject 1			Subject 2		
	Baseline	Intervention	Follow-up	Baseline	Intervention	Follow-up
Sleep too little	1	1	1	3	1	2
Sleeps the right amount	2	1	1	3	1	2
Sleeps same amount each night	2	1	1	1	1	2
Subscale Total	5	3	3	7	3	6

Visual Analysis of the Graphed Data

Changes in sleep diary means.

Mean total sleep time for Participant 1 at baseline was 553 minutes, decreasing to 543 minutes during intervention phase, with an increase to 595 minutes at the 30-day follow-up. Participant 2 demonstrated a decrease in total sleep time throughout the study, with a mean of 524 minutes at baseline, 520 during the intervention, and 518 at the 30-day follow-up.

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Changes in level.

No change in total sleep time (10 hours) from the end of baseline to the start of intervention was noted for Participant 1 (See Figure 4.4). A change in 25 minutes was observed in Participant 2, with a total sleep time of 514 minutes (8 hours and 34 minutes) the last night of baseline to 489 minutes (8 hours, 9 minutes) the first night of intervention.
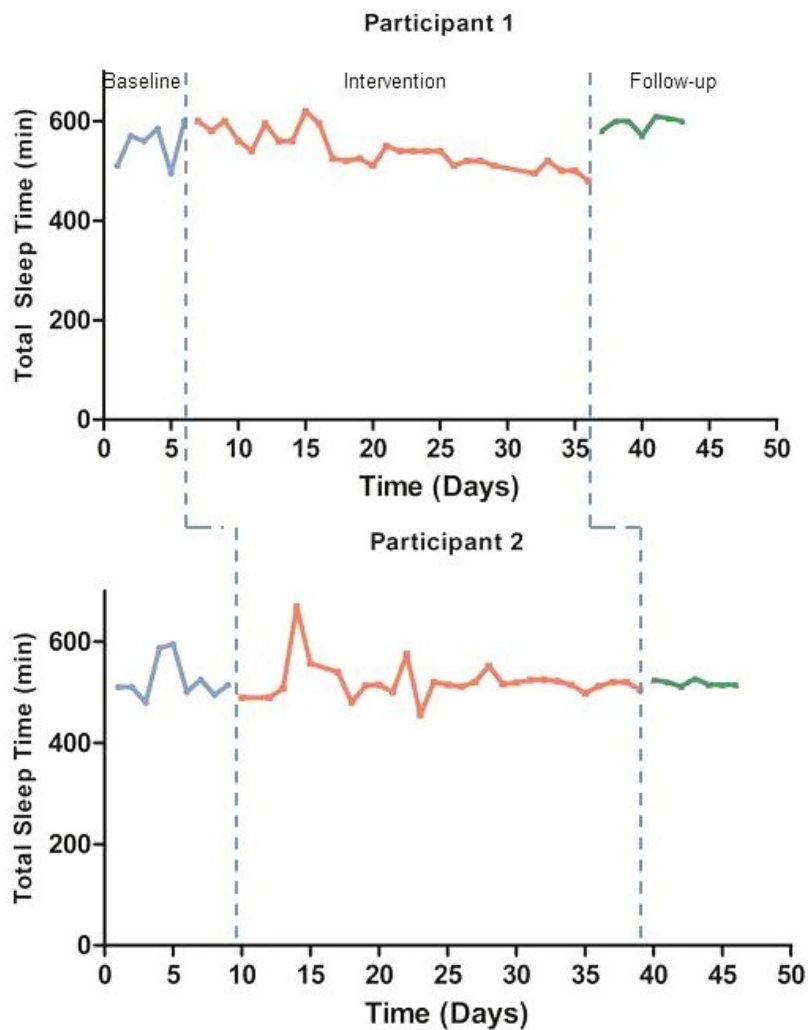

Figure 4.4 Total sleep time in minutes by intervention phase

Changes in trend.

Examination of the linear regression trend line for each participant indicated variability in trend over the phases. Participant 1 evidenced an overall increase in the total sleep time during the baseline phase, a small decreasing trend during the intervention phase, but an increasing trend within the 30-day follow-up phase. Participant 2 evidenced a decreasing trend throughout all phases, indicating an overall decrease in total sleep time.

Latency of change.

Visual inspection of data indicated no apparent pattern in regard to the latency of change due to the variability of the data for both participants.

Percentage of nonoverlapping data

From the results of the sleep diary, visual inspection revealed that 100% of the data points were overlapping between the baseline and intervention phase for Participant 1, with a range in total sleep time of 105 minutes (495 – 600 minutes) observed within the baseline phase. For Participant 2, a wide range of total sleep time was also evidenced within the baseline phase (100 minutes) with a range from 495 minutes to 595 minutes. Six data points out of a total of 28 data points in the intervention phase were noted, resulting in 21.47% nonoverlapping data for Participant 2. The overall total of nonoverlapping data in regard to total sleep time for this study was 10.72% between the baseline and intervention phases.

Effect Size

The effect size of the sleep diary results measuring total sleep time was initially small from baseline to intervention for Participant 1 ($d = .267$), but large from the intervention phase to the 30-day follow-up ($d = 1.30$). It was negligible for Participant 2

with $d = .08$ from baseline to intervention, and $d = .07$ from intervention to the 30-day follow-up.

<center>*Research Question 5*</center>

Will improved sleep decrease daytime problem behavior?

Hypothesis: Daytime behavior problems as measured by parents and teachers will decrease as the result of improved sleep.

Parent endorsements on the CBCL indicated total behavioral concerns within the Clinically Significant range for both participants at baseline, with a score of 65 for Participant 1 and a score of 69 for Participant 2. Participant 1 was again rated with clinically significant concerns at the end of the intervention phase, which subsequently declined to concerns within the Borderline range at the 30-day follow-up. Participant 2 also evidenced a decline in behavioral symptoms, with a rating of 65 at the end of the intervention phase, and a 61 (Borderline range) at the 30-day follow-up.

<center>*Research Question 6*</center>

Will the bedtime story treatment package be acceptable to parents?

Hypothesis: The intervention will be acceptable to parents.

Parent satisfaction was rated via the TEI-SF, a nine item, five-point Lickert scale with responses ranging from *Strongly Disagree* to *Strongly Agree*, with a higher score (possible 45) indicating the greatest satisfaction. The parent of Participant 1 rated all items as *Strongly Agree*, with a mean score of 45. The parent of Participant 2 rated all items as *Agree*, with a mean score of 36

<center>*Research Question 7*</center>

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Will levels of behavior at the completion of the intervention be maintained after one month?

Hypothesis: Levels of behavior at the completion of the intervention be maintained after one month.

As this question was addressed by questions one through four, please refer to the data reported in regard to bedtime resistance, night wakings, sleep latency, and total sleep time.

Research Question 8

Will parents level of perceived stress decrease after the intervention?

Hypothesis: Perceived stress will decrease after the intervention.

Parent stress was measured via a four-point scale, with parents recording *No Stress*, *Low Stress*, *Moderate Stress*, or *High Stress* in regard to their child's bedtime routine. Results were then converted to a four point scale, with one indicating the lowest level of stress and four indicating the highest level of stress. The parent of Participant 1 evidenced a slight, but decreasing trend in perceived stress in regard to her child's sleep with a mean of 1.83 at baseline, 1.60 at intervention, and 1.42 at the 30 day follow-up. The parent of Participant 2 demonstrated a slight decrease in perceived stress from a mean of 1.77 at baseline to a mean of 1.2 during the intervention phase. *Low Stress* was reported throughout the 30-day follow-up phase, resulting in a mean of 2.

CHAPTER V

DISCUSSION

The current study examined the impact of a bedtime story treatment package on disruptive bedtime behaviors in children with ASDs. The treatment consisted of a bedtime story treatment package including: two parent handouts regarding sleep hygiene and behavioral principles, *The Sleep Fairy* book (Peterson & Peterson, 2003), and the use of positive reinforcement and extinction. At the beginning of the study, parents were provided information regarding good sleep hygiene, and were asked to follow the principles outlined in the handout, for the duration of the study. Caregivers read *The Sleep Fairy* at the conclusion of the child's nightly routine, and followed the principles of extinction and reinforcement outlined on the introductory two pages of the text and in the informational handout. The first hypothesis was that bedtime resistance would decrease, based on parent report, following the treatment applied by parents.

Analysis of the data for the first research question indicated that both of the participants experienced decreases in disruptive bedtime behaviors (e.g., verbal protests, calling out for a parent, stalling, etc.) post intervention. That is, descriptive analysis from the Children's Sleep Habits Questionnaire (CSHQ), visual analysis, and effect size calculations from sleep diary data all showed a decrease. CSHQ means and sleep diary data demonstrated a consistently decreasing trend in symptoms for Participant 1. In contrast, CSHQ rating for Participant 2 indicated variability in disruptive bedtime behaviors while sleep diary data indicated a consistent downward trend in bedtime resistance over the course of treatment for both subjects. Notably, a strong extinction burst was evidenced for Participant 2 on the first night of intervention. That is, prior to

the treatment, Participant 2 demonstrated an average of less than five incidents of disruptive bedtime behaviors throughout the baseline phase, with three incidents occurring the night prior to the first night of intervention. Upon treatment, disruptive bedtime behaviors raised to 17 incidents, clearly demonstrating an extinction burst, a sudden and temporary increase in the response's frequency. Effect sizes for Participant 1 and Participant 2 were stronger from baseline to the intervention phase (.89 and .85, respectively), but still robust from baseline to the 30-day follow-up (.85 and .53). Effect size, recommended in the *Publication Manuel of the APA* (American Psychological Association, 2001), was calculated to provide further evidence of the treatment's success beyond the use of visual analysis and nonoverlapping data points. The effect size data show a strong intervention effect (e.g., the intervention worked) while parents were applying the bedtime routines. The effect of this intervention remained at a similar (strong) level for participant one. However, the strength of the effect decreased to a moderate, yet still effective level for participant two. Effect sizes from the sleep diary results of this study provided strong evidence of the bedtime story treatment package as a valid treatment for certain aspects of sleep problems.

The second questioned examined the effect of the bedtime story treatment package on night wakings. The question hypothesized that the intervention would decrease night wakings based on descriptive analysis of CSHQ scores, and visual analysis and effect size calculation of sleep diary data. Examination of this question indicated the treatment had a negligible effect on night wakings; however, wakings occurred infrequently for both participants in the baseline phase. Results of the CSHQ indicated a two point decline in CSHQ scores from baseline to the end of intervention

(six to a four) for Participant 1, while Participant 2 demonstrated a one-point decline across phases (seven to six). Based on CSHQ scores, Participant 1 demonstrated the most significant change in the symptom of *Moving to other's bed in night*, with this reportedly occurring five to seven times per week in the baseline phase, to zero to one times per week in the follow-up phase.

Sleep diary data also demonstrated a negligible change in night wakings with a decline from .833 incidents during the baseline phase to .786 incidents at the 30-day follow-up. Participant 2 also demonstrated minimal changes from baseline (.222) to post intervention (0.00) with only one night in which wakings occurred in the baseline phase, and again on the first night of intervention. Again, a small extinction burst was noted for Participant 2 on the first night of intervention, with an increase from zero incidents of night wakings for eight nights prior to the intervention, and three incidents occurring on the first night of treatment. As would be expected based on the limited number of night wakings evidenced for both participants across treatment phases, effect size was extremely low, with an effect size of .17 from baseline to the 30-day follow-up for Participant 1, and .20 for Participant 2.

The third research question examined the impact of the intervention on sleep latency, the time it took the child to fall asleep after getting into bed with the lights out. The question hypothesized that sleep latency would decrease based on descriptive CSHQ data, visual analysis of the sleep diaries, and effect size. Participant 1 was rated as a one across phases, signifying the child usually (e.g. 5 – 7 times per week) fell asleep in 20 minutes. Participant 2 was rated a one at baseline and intervention, and a two (e.g. 4 – 5 times per week falls asleep within 20 minutes) at the 30-day follow-up.

Interestingly, while Participant 2 was observed to have extinction burst at the beginning of intervention in regard to bedtime resistance and wakings, Participant 1 was observed to have a significant increase in latency from the last night of baseline (30 minutes) to a latency of 80 minutes on the first night of intervention. An overall declining trend in sleep latency was noted from baseline to the 30-day follow-up for Participant 1 with a 23.93 minute decline in sleep latency from baseline to post intervention. A small decline was initially noted for Participant 2 (2.57 minutes) from baseline to intervention, but an increase of 4.06 minutes was noted from the intervention phase to the 30-day follow-up. It should be noted that 30-day follow-up data collection occurred the first week in September, also coinciding with the start of school for Participant 2. Effect sizes were moderate for Participant 1 with .64 from baseline to intervention and .69 from intervention to the 30-day follow up. Conversely, the strongest effect size for Participant 2, .84 from intervention to follow-up, signified an undesired increase in sleep latency.

The fourth research question examined total sleep time, and hypothesized an increase in total sleep time as a result of the intervention as measured by descriptive CSHQ analysis, and sleep diary data analyzed visually and via effect size calculation. Three questions on the *Sleep Duration* subscale of the CSHQ examined total sleep time. Participant 1 demonstrated a decline from the baseline to intervention, maintained at the 30-day follow-up, with raw scores of five, three, and three, respectively while Participant 2 demonstrated variability and was rated a seven at baseline, a three at intervention, and a six at the 30-day follow-up. Visual analysis of sleep diary data indicated variability in total sleep time for Participant 1, with an unwanted, declining trend throughout the

intervention for Participant 2. Mean total sleep time for Participant 1 initially decreased

by 10 minutes from baseline to intervention, but increased by 52 minutes from

intervention to the follow-up phase, signifying an overall increase from baseline to post

treatment of 42 minutes. While a small difference, Participant 2 slept a mean of six

minutes less post treatment than during the baseline phase. A small effect size of .26 was

initially noted for Participant 1, but a large effect size of 1.30 was noted from baseline to

the 30-day follow-up phase. Conversely, a negligible effect size of .08 and .07 was noted

across phases for Participant 2.

The fifth research question examined problematic daytime behavior, and

hypothesized a decrease in total behavioral concerns as measured by the CBCL, with data

analyzed descriptively. As the intervention was conducted predominantly over the

summer break, the Teacher Report Form (TRF) was not utilized for the study. Both

participants were initially rated with behavioral concerns in the Clinically Significant

range (Participant 1, 65 and Participant 2, 69) on the *Total Problems* subscale of the

CBCL, with a decline in symptoms to concerns within the Borderline range (62 and 61,

respectively) at the end of the 30-day follow-up.

The sixth research question examined treatment acceptability and hypothesized

the intervention would be acceptable to parents based on ratings analyzed descriptively

on the Treatment Evaluation Inventory, Short Form (TEI-SF). The parents of both

participants positively endorsed the bedtime story intervention package with all nine

items on the TEI-SF rated as *Strongly Agree* resulting in a mean score of 45 for Parent 1

and all items rated as *Agree,* with a mean score of 36 for Parent 2.

The seventh research question, which evaluated the levels of behavior at the completion of the intervention as composed to behavior at the end of the 3-day follow up, was addressed by research questions one thru four. Please refer to the aforementioned data.

Research question eight examined perceived parent stress regarding their child's sleep and hypothesized perceived stress would decrease as a result of the intervention. Parent stress was measured on a four-point Lickert scale, with parents recording *No Stress (1)*, *Low Stress (2)*, *Moderate Stress (3)*, or *High Stress (4)* in regard to their child's bedtime routine. Parent 1 evidenced a slight, but decreasing trend in perceived stress in regard to her child's sleep with a mean of 1.83 at baseline, 1.60 at intervention, and 1.42 at the 30 day follow-up while the parent of participant 2 demonstrated a slight decrease in perceived stress from a mean of 1.77 at baseline to a mean of 1.2 during the intervention phase. *Low Stress* was reported by Parent 2 throughout the 30-day follow-up phase, resulting in an increased mean of 2.

Conclusions

Relevant Literature

Findings from this study found that the bedtime story intervention package was effective in reducing bedtime resistance, nighttime wakings, sleep latency, and symptoms of daytime behavior problems. Results were largely congruent with Burke et al.'s 2004 study upon which the current intervention was based, although overall levels of problematic sleep behaviors in the current study were significantly less at baseline than reported in the previous study. Research was also consistent with previous behavioral interventions (Christodulu & Durand, 2004; Moore, 2004; Reed et al., 2009; Weiskop et

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al., 2005) addressing sleep problems in children with ASDs which utilized behavioral techniques shared by the current study (e.g. bedtime routines, sleep hygiene, extinction, and reinforcement).

Relevant Theory

Based on B.F. Skinner's theory of Operant Conditioning, behavioral theory has provided the foundation for the treatment of sleep problems considered to have an environmental (e.g. influenced by the parent of caregiver) component or etiology. Behaviorism assumes learning occurs via focusing on a stimulus and response, manipulating setting events, antecedents and consequences, and is measured by describing an observable, quantifiable change in behavior. Preferred by parents for treatment for sleep problems (Wiggs and Stores, 1996; Williams et al., 2006), the American Academy of Sleep Medicine published practice parameters for bedtime problems and night awakenings (Morgenthaler et al.) based on 52 individual treatment studies from 1970 to 2500 (Mindell at al., 2006) with extinction and parent education focusing on sleep hygiene and behavioral principles supported by the highest levels of evidence.

Based on sleep hygiene and behavioral principles, results of the current study provided confirmation that the intervention effectively reduced bedtime resistance. Results indicated a negligible reduction in wakings, although participants demonstrated low levels of wakings at baseline and across phrases. Sleep latency decreased for both participants, while the effect on total sleep time was variable. Although high levels of disruptive bedtime behavior and night wakings were not evidenced in the current study, the participants demonstrated clinically significant sleep problems as measured by the

CSHQ (e.g. total score of 41 or greater; Owens et al., 2000), and parents perceived a need for intervention.

The current study is particularly relevant as it provides evidence of treatment effectiveness in a small sample of children with ASDs. Given the high prevalence rate of spectrum disorders with 44% to 86% of this population presenting with sleep problems (Cotton & Richdale, 2006; Couturier et al., 2005; Krakowiak et al., 2008), intervention options are needed. Thus, results indicate that this bedtime story intervention package can, therefore, successfully enable parents to reduce sleep problems in their child diagnosed with an ASD.

Limitations

While this study was implemented according to the methodological design, some limitations did exist. Due to the study occurring within the summer months, teacher ratings of daytime behavior were not completed as originally planned. Next, while reliability checks were requested by the investigator, neither family participated in random reliability checks due to the reported unavailability or burden of utilizing a second data collector. Fidelity to intervention was less of a risk of the steps to be completed for the intervention (e.g. read the story nightly and deliver the intervention based on child behaviors) as a reminder was provided in the front of the *Sleep Fairy* text, in the sleep diary, and via the weekly email reminders provided by the investigator. However, parents completed the treatment unsupervised in the privacy of their own homes. Thus, true fidelity to treatment is unknown.

Data collection methods and measures posed some limitations to the current study. As noted in Chapter 3, measurement via the sleep diary was a threat to the study

as caregivers completed the sleep diary independently with the exception of a weekly reminder to record data on a daily basis, via email based upon parent preference. For example, parents may have recorded data for several nights at one time, instead of on a daily basis, resulting in inaccurate data. This risk to internal validity was further complicated by the aforementioned lack of reliability checks. Results were also complicated by discordance among data collections methods, specifically, between the CSHQ and the sleep diary. For example, as measured by the sleep diary, a fairly significant decrease was noted for Participant 2 in disruptive bedtime behaviors at baseline (m = 4.88) with a decline to a mean of 0.00 at the follow-up phase. Despite this, results of the CSHQ reflected a slight increase in bedtime resistance concerns from baseline to the follow-up phase.

In regard to measurement of daytime behavior problems, the CBCL is considered to be a reliable and valid instrument used to measure a child's change in behavior over time or following a treatment; however, raters provided data on behavior or emotional problems during the past 6 months, thus making it less sensitive to short-term change. Additionally, parent stress was measured via an investigator conceived four-point rating scale, as opposed to a measure with documented reliability and validity ratings. Lastly, the follow-up occurred 30 days post-intervention. As sleep problems are often persistent in children with ASDs (Oyane & Bjorvatn, 2005; Tani et al., 2003), follow-up should occur at later time points, such as six months or one year.

Although the results were promising, they represent an initial evaluation of two subjects. Further, the two participants demonstrated low levels of nighttime awakenings throughout the baseline phase, resulting in minimal levels of change during the

intervention or the 30-day follow-up phase. Lastly, a final limitation is data collection via subjective methods (e.g. parent ratings and sleep diaries) as opposed to objective measures of sleep problems such as actigraphy or video recording.

Recommendations for Future Research

While findings from this study were conclusive and provided empirical support for the bedtime story treatment package, further research of this treatment remains a need. Future investigations should represent a larger sample of children and include children experiencing higher level of sleep problems, particularly nighttime wakings. To obtain higher recruitment and study retention rates, investigators may consider implementing the intervention during the school year, in order to minimize confounding variables such as extended vacations and atypical daytime and nighttime schedules. Given the high number of children diagnosed with Autistic Disorder who present with sleep problems (Krakowiak et al., 2008), future investigation should also include this population and may include expanded ages to allow participation of children older than age ten whose cognitive abilities reflect functioning of a younger child. Further, prospective research should also seek to identify which components of the intervention (e.g. establishing a bedtime routine, attention from parents, reading the story, or positive reinforcement) contribute to the overall positive effect of *The Sleep Fairy*.

Due to the discrepancy between sleep diary data and caregiver report on the CSHQ, it is recommended that future study may consider the use of more objective measures, including actigraphy or video recordings. In addition, fidelity checks should be completed on a regular basis to ensure accurate reporting. Fidelity checks may be completed via videotaping one random night per week, to ensure the treatment is

delivered as prescribed, parents respond to bedtime resistance or night wakings correctly (e.g. they don't inadvertently provide intermittent reinforcement), data is counted accurately, and the reinforcer is only delivered when children demonstrate the expected behavior (e.g. no incidents of bedtime resistance and they independently fall asleep in their own beds if they awaken through the night).

Replication of this study may also include may include a parent training on what constitutes normal sleep patterns and the amount of total sleep time recommended for children by age. Additionally, a parent component to the intervention in which the parents are given the opportunity to review the graphed sleep diary data in order to compare their perception of their child's sleep in comparison to the actual data. In the current study, one parent consistently reported an increase in their child's sleep problems, despite the data indicating a decreasing trend. This may be a reflection of the parental sleep difficulty, as opposed to actual sleep problems in the child. As Rinsley (1994) notes, a parent may seek treatment for their child, as opposed to seeking treatment for themselves. Thus, the intervention may help the parent to accurately evaluate their child's sleep, as well as address adult sleep hygiene and sleep patterns.

Results of this study provided evidence of a bedtime story treatment package as an effective intervention to reduce sleep problems in children with ASDs. The parents of participants successfully utilized sleep hygiene and behavioral principles guided by a bedtime story to reduce bedtime resistance and night wakings. The developmentally appropriate intervention was delivered with minimal supervision in the child's home environment by the parent or caregiver. These results contribute to the knowledge base

of effective behavioral interventions in children with an ASD and provide sleep problem

treatment options for psychologists and parents of children with spectrum disorders.

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