

EVALUATION OF SOCIAL STORIES IN SLEEP TREATMENT FOR CHILDREN WITH  
AUTISM SPECTRUM DISORDER: A FEASIBILITY STUDY

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## DEDICATION

In memory of my mother, Linda K. Fisher,  
who is the foundation of my life, my work, and all that I am.

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Lora Israelsen, UTSW 2020

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by

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A majority of children with autism spectrum disorders (ASD) experience sleep disturbances which are apt to negatively impact cognition, behavioral functioning, and the general trajectory of psychosocial development. Additionally, pediatric sleep disorders may contribute to disordered sleep in parents and reduced quality of life for family members. While children with ASD often respond positively to general standard of care in pediatric sleep medicine, there are few sleep treatments that address ASD-specific factors that contribute to sleep disturbances. This limitation may be addressed through the development of a treatment that incorporates sleep behavioral strategies in a format developed for children with ASD, such as a social story. This study evaluated the feasibility and acceptability of using a social story on bedtime routine developed for children as a complementary intervention for standard of care in a pediatric sleep clinic. Results from this study indicated social stories are acceptable as a complementary tool to sleep treatment. All participants approached for the study gave consent. Of those enrolled in the treatment group, all participants randomized to the treatment group read the story for at least one week as part of the bedtime routine (100%) with a mean utilization of 19.4 days during the month. Participants reported the social story was easy to implement and well received by the children, though feedback indicated the need for personalization and flexibility in the

implementation of social stories. There were no significant differences in pediatric sleep outcomes between treatment groups. There were significant challenges related to the feasibility of the study in terms of gathering follow-up data, partially due to the impact of the COVID-19 worldwide pandemic that occurred during the study. Secondary analyses found no significant differences in parental sleep outcomes or psychosocial functioning between treatment groups. These findings suggest that social stories may be a promising tool in pediatric sleep treatment for children with ASD, but additional research is warranted to clarify its efficacy.



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## LIST OF ABBREVIATIONS

ADHD	Attention-deficit/Hyperactivity Disorder
ADOS	Autism Diagnostic Observation Schedule
ASD	Autism Spectrum Disorders
CADD	Center for Autism and Developmental Disorders
CMC	Children's Medical Center
COVID-19	Coronavirus Disease 2019
CSHQ	Children's Sleep Habit Questionnaire
GABA	Gamma-aminobutyric acid
PedsQL	Pediatric Quality of Life Questionnaire – Family Impact Module
PSQI	Pittsburgh Sleep Quality Index
TAU	Treatment-As-Usual
TAU+SS	Treatment-As-Usual + Social Story

## **CHAPTER ONE**

### **Introduction**

#### **STATEMENT OF THE PROBLEM**

A majority of children with autism spectrum disorders (ASD) experience sleep disturbances, particularly delayed sleep onset and frequent nighttime awakenings. Due to the critical role sleep plays in a host of cognitive skills, emotional functioning, and behavioral management, these disturbances are apt to have immediate and long-term impacts on the child's cognitive development and psychosocial functioning.

Additionally, since pediatric sleep disorders often lead to disrupted sleep for caregivers, sleep disorders in children with ASD may impact the quality of life for parents, caregivers, and family members. By treating pediatric sleep disturbances, clinicians may improve the child's general functioning and impact the trajectory of their development, while also improving quality of life and general functioning for the entire family.

It is worth noting that the treatment of pediatric sleep disorders among children with ASD may not be a straightforward process for many families. Parents of children with an ASD diagnosis and significant sleep disturbances could pursue treatment through a variety of clinical avenues. For example, they may first report sleep difficulties to their pediatrician, therapist, or other providers who specialize in autism and developmental differences. These clinicians often provide basic psychoeducation on general sleep hygiene or bedtime strategies but may not have the training needed to address patient-specific issues with sleep.

Alternatively, the parents may seek treatment from a pediatric sleep clinic, where the child's specific sleep challenges may be treated more effectively but the sleep psychologist, nurse practitioner, or medical doctor may not have the requisite skills or expertise for supporting learning and behavioral change in children with ASD. With these considerations in mind, a

simple treatment tool that can transition between clinical settings to complement various clinical specialties would be ideal for addressing sleep disorders in children with ASD. In order to transition easily between treatment settings, the tool should be low cost and easy to implement. A social story (i.e., a narrative handout written in accordance with general guidelines developed by Carol Gray) on bedtime routines may prove to be the ideal tool since social stories are frequently used to help children with ASD learn new skills, flexible to personalized treatment, low cost, and easily implemented by various types of clinicians.

The present study evaluated the feasibility and acceptability of using a social story on bedtime routines as a complement to standard treatment for patients with ASD in an outpatient pediatric sleep disorders clinic. Specifically, the study used quantitative data and qualitative feedback to gain a better understanding of the parents' and child's utilization, satisfaction, and ease of implementing the social story bedtime routines. Analyses were conducted to assess the efficacy of the social story by comparing sleep outcomes, quality of life factors, and treatment satisfaction for parents and children in the control and treatment groups. Results from this study will support further research and development of ASD-specific treatment tools for pediatric sleep disorders.

## **CHAPTER TWO**

### **Review of the Literature**

### **SLEEP AND AUTISM SPECTRUM DISORDER**

#### **Pediatric Sleep**

While sleep medicine still has many unanswered questions about sleep, it is clear from decades of research that sleep plays a crucial role in human life and impacts the cognitive, emotional, and physical development of children (Stores, 1999). Researchers have consistently highlighted the relationship between sleep and critical cognitive functions such as learning capacity, memory consolidation, working memory, executive function associated with the prefrontal cortex, and general neural plasticity (Curcio, Ferrara, & De Gennaro, 2006; Dewald, Meijer, Oort, Kerkhof, & Bogels, 2010; Kopasz et al., 2010).

Researchers note disruptions in sleep and poor sleep quality in children are also associated with poor academic performance and higher rates of behavioral problems (Curcio et al., 2006; Meijer, Habekoth, & van den Wittenboer, 2001; Sadeh, Gruber, & Raviv, 2003). One study found that when children's sleep is reduced by an hour for four consecutive nights, their functioning is significantly impaired across several critical domains of development including positive affective response, emotional regulation, short-term memory, working memory, and attention (J. L. Vriend et al., 2013).

The effects of sleep disruptions on cognitive performance may be especially pronounced for more vulnerable subgroups of children, including African American children and children from families with lower socioeconomic status (Buckhalt, El-Sheikh, & Keller, 2007). Follow-up studies indicated that when researchers control for SES, lower cognitive performances for African American boys (compared to European American boys) are associated with poor sleep efficiency, which can persist for two years after the period of poor sleep efficiency ((Philbrook,

Hinnant, Elmore-Staton, Buckhalt, & El-Sheikh, 2017). Researchers indicate that certain environmental factors such as management of family adversity, discrimination, and race-based stressors may exacerbate the negative outcome of poor sleep on cognitive development for these vulnerable populations.

Researchers have found that some of the outcomes associated with inadequate or poor sleep such as impaired attention, impulsivity, and difficulty modulating emotions can resemble symptoms of developmental disorders such as Attention-deficit/Hyperactivity disorder (ADHD) (Dahl, 1996). Indeed, disrupted sleep is a common symptom for a variety of mental health concerns and developmental disorders, including autism spectrum disorders.

### **Autism and Sleep Disorders**

Autism spectrum disorders are developmental disorders that can range widely in terms of general presentation but are broadly characterized by two constellations of functional differences: the presence of restricted or stereotyped behavioral patterns or interests and the absence of typical patterns of social communication, interpersonal interaction, and/or language development (American Psychiatric Association, 2013a). Due to these differences in behavior and social functioning, along with differences in their overall neurological development, children with ASD often function differently from their typically developing peers across a host of activities, including sleep.

Decades of research have shown that sleep disturbances are a common concern for the majority of children with an ASD diagnosis (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Maxwell-Horn & Malow, 2017; Richdale & Schreck, 2009). While roughly one third of typically developing children have sleep disturbances, researchers estimate that 50% to 80% of children with an ASD



diagnosis experience significant difficulties with sleep (Krakowiak et al., 2008; Reynolds & Malow, 2011; Veatch, Maxwell-Horn, & Malow, 2015). In particular, children with ASD often demonstrate delayed onset of sleep, an increase in the number of nighttime awakenings, and poor sleep efficiency (Kotagal & Broomall, 2012; Krakowiak et al., 2008).

The consequences of sleep disruptions are not limited to difficulties at night. Rather, these sleep disturbances are associated with a host of potential problems for the children, including impacting their daytime behavior and neurocognitive development, particularly related to memory and learning (Kotagal & Broomall, 2012). This may be especially concerning given that many children with ASD already learn in a manner that differs from many of their peers and often require alternative educational supports due to differences in communication skills. By effectively treating the sleep disruptions, clinicians have the potential to improve the child's overall functioning, including progress in memory, learning, emotional regulation, and general behavioral management.

### **Physiological Factors**

There are a variety of hypothesized physiological causes for sleep disturbances in children with ASD. These hypotheses include disruptions in the production, activity, and maintenance of sleep-critical neurotransmitters and hormones, including gamma-aminobutyric acid (GABA), serotonin, and melatonin (Reynolds & Malow, 2011). The possibility of poor melatonin development in particular may have a direct impact on the quantity and quality of sleep in children with ASD, since melatonin is critical for the onset and maintenance of sleep (Reynolds & Malow, 2011).

Additionally, low iron levels in children with ASD may also contribute to restless sleep (Veatch et al., 2015) due to the relationship between iron deficiency, specifically ferritin levels

of less than 50 ng/ml, and restless leg syndrome (Sun, Chen, Ho, Earley, & Allen, 1998). Iron deficiency in children with ASD may be tied to dietary and eating concerns since roughly 70% to 90% of children with ASD have atypical feeding behaviors (Reynolds & Malow, 2011), which often includes restrictions in the variety of food these children are willing to eat and ritualized behaviors around eating which can impact the amount of food they consume.

### **Behavioral Factors**

In addition to hypothesized physiological causes, many children with ASD have behavioral patterns or communication deficits which may exacerbate the impact of the physiological causes of sleep disturbances. Indeed, though the etiology of a sleep problem is often multifaceted, behavioral insomnia is commonly a critical factor that causes or intensifies sleep challenges in children with ASD (Johnson & Malow, 2008). Behavioral factors that are most likely to contribute to insomnia for children with ASD and other neurodevelopmental disorders include poor sleep hygiene, maladaptive sleep associations, and challenges with limit-setting at bedtime (Grigg-Damberger & Falls, 2013). More specifically, most behavioral symptoms that impede sleep in children are connected to both the behavior of the child and the behavior of the parent such as poor or inconsistent bedtime limits or reliance on the parent (or another external support) to help them fall asleep (Meltzer, 2010).

Additionally, given that communication difficulties are present for children with ASD, these children may not fully understand their parents' expectations around bedtime routines and sleep (Reynolds & Malow, 2011). Given the significant role behavioral factors play in the sleep onset and maintenance for many children with ASD, once concerns for other significant sleep disorders have been eliminated, the recommended treatment for sleep disorders in children with ASD (as with neurotypical children) is behavioral therapy with guidance on improving sleep

hygiene and bedtime routines, combined with melatonin as needed (Miano, Giannotti, & Cortesi, 2016)

Beyond the impact these sleep disturbances may have on the child's personal development, the sleep disruptions may also contribute to significant stress in caretakers and family members (Kotagal & Broomall, 2012; Stores, 1999). Pediatric sleep disorders often lead to disruptions in sleep for parents and caregivers due to a range of sleep-related challenges. For example, parents often report difficulties due to undesired co-sleeping with their child, having to get out of bed multiple times to care for the child during the night, or experiencing a sense of constant worry about sleep. Even after controlling for other relevant demographic and clinical factors, studies have found bedtime resistance and daytime sleepiness in children is significantly correlated with parenting stress (Byars, Yeomans-Maldonado, & Noll, 2011). The concerns and challenges associated with their child's sleep indirectly impact the parents' sleep, making parents more vulnerable to their own difficulties with memory, emotional regulation, and behavioral management (Reynolds & Malow, 2011).

### **Treatment for Sleep Disorders**

The question then is what contributes to efficacious sleep treatments for patients with ASD? In general, practitioners of sleep medicine use a variety of physiological, behavioral, and educational strategies to improve sleep behaviors for pediatric patients. Physiological treatments such as taking iron supplements to combat restless leg syndrome or using melatonin supplements are routinely used in pediatric sleep populations, generally with good effect in both typically developing children and children with atypical developmental patterns. For example, when compared against a placebo, melatonin was found to advance sleep onset and increased total sleep time among children with ADHD (Van der Heijden, Smits, Van Someren, Ridderinkhof, &

Gunning, 2007). Recent studies have shown promising results in the use of melatonin for children with ASD and sleep issues, including significant improvements in daytime behavior for the child and quality of life for parents (Schroder et al., 2019) while also increasing sleep duration by an average of 57.5 minutes per night after the use of prolonged release melatonin (Gringras, Nir, Breddy, Frydman-Marom, & Findling, 2017).

Educational approaches and materials are also routinely used as part of sleep treatment for pediatric patients. Many of these educational approaches specifically target problematic sleep-related behaviors such as outlining a visual bedtime routine for children or instituting behavioral reward charts. These approaches have the potential to be a helpful tool in the successful implementation of treatment strategies, since much of pediatric sleep medicine relies on the parents' ability to successfully implement behavioral strategies at home. In general, the most important factor in sleep treatment for all ages is education and support for effective sleep hygiene which includes a regular bedtime routine, consistent sleep schedule, and developing a calming environment that promotes sleep (J. Vriend & Corkum, 2011).

Providing strategic support and education on sleep hygiene is particularly important since many parents may attempt to implement measures that seem correct but may worsen sleep (e.g., instituting a bedtime that is too early for their child's natural circadian rhythm and therefore counterproductive). Education regarding sleep hygiene often includes developing strategies with parents to ensure the room is quiet, dark, and cool; electronic devices are turned off and ideally outside the bedroom; regular use of a calm, rewarding bedtime routine (brush teeth, choose pajamas, and read or have special time with parent); and a consistent sleep schedule that starts at the same time each night (J. Vriend & Corkum, 2011). The assumption among many treatment

providers is that educational materials that support effective sleep hygiene helps parents better understand and adhere to recommended treatments.

### *Sleep Treatment for Children with ASD*

Due to the combination of known physiological and behavioral factors associated with sleep disorders in children with ASD, sleep treatment for patients often includes a combination of physiological treatments, behavioral treatments, and educational approaches for both the parental and child education, in alignment with what is used in standard sleep treatment. However, within the field of ASD research, a handful of researchers have begun to examine what types of educational approaches or materials are most effective for the ASD population.

One randomized study found that the use of an educational pamphlet developed to target insomnia in children with ASD was not found to significantly improve sleep latency for children (Adkins et al., 2012). In the qualitative findings for this study, parents noted that the information in the pamphlet was helpful but reported it would have been more useful to have specific examples for how to implement the information. Another study examined a more extensive parent-based sleep education in both individual and group sessions presented in one, two, or three educational workshops. These researchers found that while outcomes did not differ by educational format, overall the parent-based sleep education was associated with improvements in the children's sleep onset delay (Malow et al., 2014).

While parent-based educational workshops are effective for the ASD population, it is important for clinicians to have alternate options for supporting the educational needs of these families. Not all parents will be able to consult a pediatric sleep expert for individual sessions or participate in educational groups. To support the challenge of limited resources and expertise in this field, the Sleep Committee of the Autism Treatment Network has developed guidelines to

diagnose and treat insomnia in children with ASD which includes: screening all children with ASD for insomnia, screen for potential medical issues that contribute to insomnia, determine and develop therapeutic interventions for the child that include parent education and behavioral approaches as a first-line treatment, consider the use of pharmacologic therapies, and follow-up with patients (Malow et al., 2012).

In general, educational approaches for sleep treatment in an ASD population should be delivered and implemented in a manner that works for the parents' and children's needs. Parent-based educational programs or targeted educational materials that help parents understand how to implement strategies at home are likely to be effective elements of sleep treatment for children with ASD. By developing effective handouts or pamphlets, clinicians will have low-cost resources that help parents successfully address sleep disorders in children with ASD at home. One example of an educational handout that might be effective in sleep treatment would be a social story, a type of educational tool routinely used for children with an ASD diagnosis.

#### *Social Stories as a Treatment with the ASD Population*

Social stories were developed by Carol Gray in the 1970s as a targeted treatment for helping children with ASD understand and implement new behavior. Social stories are meant to be simple, concrete stories that help children learn new behavior or routines through verbal repetition (Gray, 2014). The theory behind social stories is that a child with ASD often does not have a clear understanding of the parents' (or others') expectations for their behavior. By reading the same story over and over, the social story theoretically acts as a learning device to help the children internalize the social expectations.

While social stories have been used as a therapeutic treatment for children with ASD for decades, the research supporting their use in sleep treatment is sparse and inconsistent. One study

examined what they called a social story, which included a description of parental expectations for appropriate bedtime behavior and outlined rewards for meeting those expectations. They paired this story with positive reinforcement program in a small group of pediatric patients (n=4) with disruptive bedtime behavior. The researchers found that consistent use of the story and rewards was associated with a decrease in the frequency of disruptive bedtime behaviors. They also found fewer problems with sleep maintenance following the implementation of the story. Parents in this study reported improvements in daytime functioning and a high sense of self-efficacy in implementing the treatment at home (Burke, Kuhn, & Peterson, 2004).

Though the results of the study are promising, the story in this study does not follow standard guidelines for social stories typically used for children with ASD (Gray, 2014), the very small sample size impacts generalizability, and decision to directly pair use of the story with a positive reinforcement program limits the ability to interpret the efficacy of the story alone. To build from this study, we developed a social story that focuses on sleep treatment that we hope may be helpful both in sleep medicine settings, where clinicians specialize in sleep treatments rather than treatments for children with ASD, as well as in clinics where clinicians focus on treatments for ASD but may also field questions about improving sleep. In the current study, we evaluated the feasibility and acceptability of using social stories to augment treatment of patients with ASD in a pediatric sleep clinic and conducted preliminary analyses of efficacy.

## SUMMARY

Disordered sleep is a clinical concern for the majority of children with ASD, with the potential for indirect negative consequences on the sleep and general quality of life for their families. Though there are many standard pediatric sleep treatments that can be implemented to address disordered sleep, there are few treatment tools developed to address both sleep concerns and behavioral factors specifically associated with ASD. Additionally, in standard sleep treatment, patients receive extensive education on sleep to guide their implementation of new behavioral strategies. However, research indicates that while parents appreciate the knowledge, they experience difficulties when attempting to implement the new knowledge at home.

The social story, a common clinical or educational tool used in a variety of settings, may aid children with ASD in understanding expectations around bedtime routines and the development of new behaviors. Though social stories are unlikely to be useful as a standalone treatment, they may serve an essential role in supporting the efficacy of standard sleep treatment by providing a tangible tool for parents to implement at home. In this study, we explored the feasibility and acceptability of social stories as a complementary clinical tool for sleep treatment for children with ASD. We also conducted analyses on changes in pediatric sleep, parental psychosocial functioning, parental sleep, family functioning, and general treatment satisfaction.



## AIMS AND HYPOTHESES

### Primary Aim and Hypothesis

Aim 1. To examine the feasibility and acceptability of a social story on bedtime routines as a complementary treatment tool for behavioral insomnia in children with ASD and determine if social stories enhance treatment outcomes for children assigned to TAU+SS.

*Hypothesis 1:* We expect that social stories will be both feasible and acceptable as a complementary treatment tool as measured by participants' reported adherence to treatment, comfort with intervention, improvements in pediatric sleep, and satisfaction questions at follow up.

### Secondary Aim

Aim 2: To evaluate the efficacy of social stories for parents of participants assigned to TAU+SS, as measured by results of questionnaires on parental sleep, psychosocial functioning of parents, and general family function.

*Hypothesis 2.* By treating the child's sleep disorder, we hypothesize that we may also observe a secondary improvement in the parents' quality and duration of sleep, parental psychosocial functioning, and overall family function.

## **CHAPTER THREE**

### **Methodology**

### **METHODS**

#### **Patient Recruitment**

Study participants were recruited from Children's Health Sleep Disorders Center at Children's Medical Center (CMC) in Dallas, TX. Participants were pediatric patients (4 to 10 years of age) with an Autism Spectrum Disorder diagnosis who required treatment for behavioral insomnia from a sleep psychologist (W. David Brown, Ph.D., DABSM, DBSM). All study participants had a complaint of behavioral-based sleep insomnia at the time of recruitment with parental confirmation that the child struggled to fall asleep within 30 minutes of being put in their bed at least three nights per week, per the diagnostic standard for insomnia (American Psychiatric Association, 2013b).

The Children's Health Sleep Disorders Center regularly provides treatment to children with an ASD diagnosis who have been referred by other providers, including the Center for Autism and Developmental Disorders (CADD). To aid in recruitment, the sleep psychologist at the Sleep Center reserved certain appointments times for new patients referred from CADD during the study. This allowed CADD-referred patients to bypass the wait list for treatment at the Sleep Center. The study also included current patients of Dr. Brown at Children's Health Sleep Disorders Center who met study criteria with ongoing need for treatment of behavioral insomnia.

The age range was limited from 4 to 10 years of age due to psychometric properties of sleep questionnaires (i.e., the *Children's Sleep Habit Questionnaire* was normed on children 4 years of age and older) and developmental considerations (i.e., the text of the social story is most appropriate for children rather than adolescents).

The current study did not have the requisite resources needed to confirm the accuracy of an ASD diagnosis with the diagnostic standard, the Autism Diagnostic Observation Schedule (ADOS). Consequently, we relied on provider-based documentation of the ASD diagnosis in EPIC, the electronic medical record system used by Children's Medical Center. The majority of participants 88.2% (n=15) received therapeutic care at CADD or additional clinics at CMC that specialize in the support of autism and other developmental disorders. However, despite the majority of participants receiving care at CADD, a review of medical records indicated that most participants did not receive their initial evaluation at CADD which nearly always includes an ADOS as part of diagnostic practice. For the majority of participants, it is unknown if an ADOS was utilized in the diagnostic process as only 17.6% (n=3) of participant medical records included results of an ADOS.

Co-morbid developmental disorders or medical conditions were not included in the exclusion criteria, therefore participants remained eligible for the study even if they had Attention-deficit/Hyperactivity Disorder (ADHD), epilepsy, apnea, or another significant pediatric disorder so long as behavioral factors also appeared to be a significant factor in their sleep difficulties. Exclusion criteria included parents or children who were not conversant in the English language or children who were nonverbal, due to the English language format of the social story and questionnaires.

Between August 2019 and February 2020, all patients were recruited in-person at the Sleep Disorders Center at Children's Medical Center in Dallas, TX. In March 2020, the Sleep Center was temporarily closed due to the worldwide pandemic from Coronavirus Disease 2019 (COVID-19) and recruitment for the current study was temporarily halted. Following IRB approval for study modifications related to COVID-19 factors, recruitment for the current study

resumed in July 2020. From July 2020 to September 2020, participants were recruited primarily via virtual telehealth visits, with questionnaires completed over video platforms whenever possible. When connectivity issues or other technical issues impacted audio/visual reception, questionnaires were completed over the telephone with the researcher reading questions to the parents. Social stories were mailed via the United State Postal Service to participants as needed. Approved consent and assent practices were continued across recruitment formats.

Table 1: Inclusion and Exclusion Criteria

	<b><u>Criteria</u></b>	<b><u>Rationale</u></b>
<b><u>Inclusion</u></b>	Age of child is between 4 years, 0 months and 10 years, 11 months	The social story text was developed for young children and the <i>Children's Sleep Habit Questionnaire</i> was normed for children older than 4 years of age.
	Parent speaks English proficiently.	Follow-up phone calls and surveys were conducted in English.
	Parent is able to provide informed consent and child is able to provide assent. Assent to be gathered verbally from the child in the presence of the parent by asking if the child is comfortable with the researcher asking about the child's sleep and function. Assent was gathered prior to the distribution of questionnaires.	Ethical need for awareness and selection of possible treatment choices.
	An ASD diagnosis recorded in EPIC for the child.	The pilot study focused on children diagnosed with ASD.
	Child consistently (i.e. at least 3 nights per week) demonstrated difficulty falling asleep (i.e. awake for more than 30 minutes after being placed in bed).	The social story was developed to complement behavioral treatments for disturbances in sleep onset and sleep maintenance.
<b><u>Exclusion</u></b>	Child's sleep disturbances are suspected to be caused primarily by medical issues (e.g., apnea, narcolepsy).	The treatment recommendations in this study would not be the primary treatment for medical sleep disturbances.
	Parent lacks English literacy.	The social story and questionnaires were written in English.
	Child is nonverbal or does not use productive language (i.e., language is primarily echolalia or scripted speech).	The social story treatment relies on comprehension of language, which may differ for a nonverbal child's and impact their ability to understand and implement the treatment.

## Study Procedures

We identified patients who appeared eligible for the study based on basic data available in the medical chart. As part of this process, we prescreened all patients on Dr. Brown's clinical schedule from August 2019 to September 2020. We also reviewed the waitlist for the Sleep Disorders Clinic in July 2020 to identify possible participants who had been referred to the clinic since the start of the study (August 2019 – July 2020) but had not yet scheduled an initial appointment. Any potential participants on the waitlist were contacted by clinical staff to try to schedule an appointment. While researchers did not formally recruit from CADD at CMC, CADD providers were informed of the study. If a CADD provider had a patient who required support for disordered sleep, they reviewed inclusion criteria and encouraged their patients to contact the Sleep Disorders Center regarding the study.

The researcher determined whether patients met inclusion/exclusion criteria (as outlined in Table 1) in the initial interview with parents which was conducted in a private office in the Sleep Center or as part of the virtual visit. The researcher reviewed study procedures with the parents to ensure that they were interested in the project and willing to participate (e.g., respond to follow-up phone calls and emails, comply with treatment). If the family was interested, the research staff presented the study and obtained consent and assent either prior to or during their clinical appointment. Patients who are ineligible for participation in the study received standard treatment.

From August 2019 to February 2020, participants were enrolled immediately prior to their appointment with the sleep psychologist (Dr. W. David Brown) at Children's Health Sleep Disorders Center. From July 2020 to September 2020, participants were enrolled during their telehealth visit with Dr. Brown, with consent and assent gathered and questionnaires completed

at the end of the virtual visit. All families (100%) broached in person or during a virtual visit expressed an interest and willingness to participate in the study.

One parent asked to be included in the study who reported her child had been diagnosed with ASD. However, she had never seen a provider for ASD-related treatment, did not have ASD listed as a diagnosis in their child's medical record, and could not recall details of the diagnostic process. We were uncertain if she met inclusion criteria so encouraged her to bring school records for our review at a follow-up appointment, but because of transportation and technological limitations, the parent was unable to attend follow-up appointments. She never officially consented or enrolled in the study and thus was not counted in our attrition rate.

Another parent called the clinic to inquire about the study (after being referred by their CADD provider) but expressed hesitation to participate upon learning that the clinical visit associated with the study was generally charged to the patients' insurance. As a note, all patients participating in the study were referred for sleep concerns and billed accordingly. No additional visits or charges were ordered solely for the study. Research and clinical staff followed routine clinical protocols and standards for all study participants. Although the researcher offered flexibility around the financial aspect of the clinical visit for this family, the mother requested time to think about it. The research and clinical staff were unsuccessful in later attempts to contact the family. They never consented or enrolled in the study and thus were not included in our attrition rate, but her hesitation to participate due to insurance concerns is worth noting as a possible concern for families. It is unknown if other families hesitated to participate for financial concerns or other reasons after receiving a referral from CADD as no other families contacted research or clinical staff at the Sleep Center and expressed their concerns.

Upon completion of the baseline measures, all participants who met inclusion criteria for the study were randomized at a 1:1 ratio into one of two conditions: Treatment-As-Usual (TAU) and Treatment-As-Usual with a social story handout (TAU+SS). The randomization schedule was produced using a random number program (e.g. Excel software, RAND function). The sleep psychologist providing TAU in this study (Dr. W. David Brown) was blinded to the assigned randomization during the initial evaluation to avoid unintentional changes to TAU provided to each patient.

### **TAU condition**

Treatment as usual was administered by Dr. W. David Brown either in-person or virtually through the outpatient pediatric sleep clinic at CMC. Information gathered from the baseline research measures was incorporated into clinical treatment planning whenever relevant. Treatment for participants in this group often included a diagnostic interview, patient education on sleep, adjustments to bedtime routines and sleep schedules, behavioral treatments for sleep, supplements to promote healthy sleep such as melatonin or iron, positive reinforcement strategies, and other supports as needed to promote positive psychosocial functioning both during the day and at night.

While patients in this group received verbal education on sleep and bedtime routines, they did not receive any tangible handouts to guide the implementation of the bedtime routine each night. The TAU condition included the obtaining of baseline measures. Information gathered from the baseline measures was incorporated into clinical treatment planning when relevant. As part of the study design, participants in the TAU group received follow-up calls at two weeks when possible and were scheduled to complete follow-up measures at four-weeks.



### *TAU + SS condition*

The TAU+SS group also received standard care that included sleep education, adjustments to bedtime and sleep schedule, behavioral treatments and strategies, recommendations for supplements, etc. In addition, this group received a social story that included text and images which outlined a healthy bedtime routine (see Appendix A). The social story was minimally personalized to the child's bedtime routine, including the addition of their name, specific bedtime, and name of the caregiver that helped with bedtime. Parents were instructed to read the social story nightly for one month as part of the child's bedtime routine to promote positive sleep-related behaviors. As a note, the parents were asked to read the same social story every night as part of the guiding theory behind social stories that the repetition of the stories acts as a learning process to help children with ASD internalize social or behavioral expectations.

The parents were encouraged to track adherence by recording their frequency of reading the social story via an adherence tracker attached to the back of the social story and through verbal report at the 4-week follow-up appointment. They were also asked to record any difficulties they experienced while using it. This adherence tracking was intended to guide the researcher in the assessment of how “dosing” or frequency might impact outcomes (i.e., differences in outcomes for child whose parents read the social story nightly versus those who read it only a few times in a month). It also documented experiential challenges associated with TAU+SS. Parents were instructed to cease utilization of the social story if it became too challenging for the child or the family to implement.

### **Sample Size**

The sample size for this study was designed to establish a proof of concept for the feasibility and acceptability of social stories as a complementary treatment to standard pediatric sleep treatment. While the originally planned analytic sample size was 40, due to a delay in IRB approval, challenges experienced by the research staff while the primary researcher was on maternity leave, and the global COVID-19 pandemic, the sample size was reduced.

A total of 19 participants (4 to 10 years of age) enrolled and completed baseline data for the study as part of their visit to Children's Health Sleep Disorders Center in Dallas, Texas. Following a randomization schedule, enrollment was evenly split with 9 participants in the TAU + SS arm and 10 participants in the TAU arm. We removed two participants from the sample, one due to age (the child was recruited at 3 years, 11 months) and one due to significant missing data (questionnaires were administered but not recovered from the parent and we were unable to contact the family after the appointment). Of the remaining 17 participants, 47.1% (n=8) were in the TAU+SS group and 52.9% (n=9) were in the TAU group. Baseline data included all data on the child's sleep as well as accompanying data from their parents. With an attrition rate of 29%, we were able to collect follow-up data on a final sample size of n=12.

### **Data Collection & Analysis**

Data collection began at the initial visit and continued through a follow-up session. The follow-up session was always scheduled to be 4 weeks after the initial visit, but the majority of participants cancelled or rescheduled their follow-up appointments (often multiple times). It should be noted that the rate of cancellations and reschedules during this study was similar to cancellation rates generally seen at the Sleep Center. Due to these challenges with scheduling and communication, the majority of patients enrolled in this study were not seen 4 weeks after the initial visit but instead had a delayed follow-up visit. One outlier participant was seen 197

days after baseline (the significant delay was due to clinic closure and participant-specific challenges related to the COVID-19 pandemic and insurance). Of the remaining participants, the mean number of days from baseline to follow-up was  $38 \pm 9.2$  days with a range of 25 to 53 days.

From August 2019 to February 2020, parent-report measures were administered in a private consult room prior to the start of their appointment. The researcher sat with the parents and child to provide in-person support and clarify any questions while parents completed the questionnaires with pen and paper. From July 2020 to September 2020, participants were enrolled at the end of their telehealth visit with Dr. Brown, with questionnaires completed verbally over a secure virtual health platform (the researcher read questions and parents provided their response). The questionnaires were unable to be emailed to parents due to privacy concerns related to personal health information. Data collection was managed by manually entering data into a secure spreadsheet from paper questionnaires. The researcher called study participants to follow-up on any missing data as needed.

We assessed outcomes on pediatric sleep (as measured through parent-report measures), treatment compliance, parental sleep, parental psychosocial functioning, and overall satisfaction. We collected data on parental psychosocial functioning from the parent most involved or impacted by the bedtime routine and nighttime behavioral management. When parents felt they were equally involved in bedtime and behavioral management, we asked the parents to choose which parent would prefer to answer the questions or would be most likely to attend the follow-up session.

## MEASURES

### Demographic Data

Baseline demographic data for participants was gathered from EPIC, the electronic medical record software used by CMC.

- EPIC Data: Age, legal sex, diagnostic data related to the ASD diagnosis, clinical care for ASD-related and sleep treatments, and time between baseline and follow-up visits.
- Data related to rates of reschedule attempts and cancellations were tracked by the research and clinical staff

### Aim 1

The primary outcome measures to assess feasibility, acceptability, and effect on pediatric outcomes, including adherence rates, attrition rates, and subjective pediatric sleep data (i.e., sleep diaries and sleep measures).

- *Children's Sleep Habit Questionnaire (CSHQ)*: The CSHQ is a parent-completed questionnaire developed to evaluate common sleep problems in children based on their behavior and sleep-related functioning. It includes 33 items across eight subscales (bedtime resistance, sleep-onset delay, sleep duration, sleep anxiety, night awakenings, parasomnias, sleep-disordered breathing, and daytime sleepiness). All subscales and total CSHQ scores were calculated for this study, however analyses focused on the three subscales most closely tied to sleep onset disorder (bedtime resistance, sleep-onset delay, and sleep duration). Most items are scored on a Likert scale from 1 (usually experiences symptoms of disordered) to 3 (rarely experiences symptoms of disordered), with two items scored on a

Likert scale from 0 to 2. Research suggests that the reliability of the subscales range from 0.62 to 0.79 with internal consistency of subscales that ranged from 0.56 to 0.93 for clinical groups (Owens, Spirito, & McGuinn, 2000). One study has found that the CSHQ identified proportion of sleep disturbances among children with ASD at a similar rate to use of actigraphy ((Souders et al., 2009).

- *Sleep Diary*: The diary is a parent-completed measure of the child's nightly sleep schedule for a one-week time period. All prescreened patients who appear to be eligible for the study, based on their EPIC profile, received a sleep diary as a baseline measure. Although it was originally planned that sleep diaries would be mailed to patients, due to the frequently shifting clinical schedule and high reschedule rate of appointments with potential participants, the clinical staff often did not have sufficient warning to mail diaries prior to the appointment. Parents were asked to fill the diaries to the best of their recollection at the start of the appointment. Parents were asked to complete a second diary following treatment, which was collected at the follow-up appointment. The majority of parents forgot to track the sleep schedule with the diary, so follow-up data related to the sleep diary was also based on parent recollection. Though imprecise, the sleep diary allowed us to examine changes in perception of the quantity of pediatric sleep.
- *Recruitment, Enrollment, Completion, and Drop Out Rates*: Recruitment, enrollment, completion, and dropout numbers were captured to help establish proof of concept for the feasibility and acceptability of the proposed study.

**Aim 2:**

Secondary outcomes measures included an adherence questionnaire based on the original treatment recommendations, a quality of life measure looking at overall family functioning, and patient satisfaction. Outcome measures are outlined below:

- *Pittsburgh Sleep Quality Index (PSQI)*: The PSQI is a parent-completed measure that measures sleep quality over a one-month time period. It was developed by researchers at the University of Pittsburgh and has been used extensively for both clinical and research purposes. It consists of 10 items which contribute to seven sleep-related components that produce a global score of sleep quality. Each item is scored on a Likert scale from 0 to 3 based on frequency of symptoms. Research suggests that the reliability of the PSQI global score is 0.87 for primary insomnia patients with a sensitivity of 98.7 and specificity of 84.4. with a global PSQI >5. Validity analyses indicated high correlations with sleep log data, though lower correlations with polysomnography data (Backhaus, Junghanns, Broocks, Riemann, & Hohagen, 2002). We collected the PSQI for the parent at baseline and at the follow-up appointment.
- *Pediatric Quality of Life Inventory – Family Impact Module (PedQL )*: A parent-report measure used to assess the impact of the child’s health condition on the parents’ functioning (i.e., physical, emotional, social, cognitive, communication, and worry) and the overall family experience (i.e., daily activities and family relationships). This study utilized the portion of the questionnaire (36 items) focusing on function and experience of the parents/family over a 7-day period. Each item is scored on a Likert scale from 0 (Never) to 4 (Almost Always). The overall PedsQL questionnaire has been found to have excellent reliability (0.92)

for parent report (Varni, Burwinkle, Seid, & Skarr, 2003) and the Family Impact Module has additionally demonstrated internal consistency and reliability in a community sample (Medrano, Berlin, & Davies, 2013).

- *Patient satisfaction*: A brief questionnaire developed for this study to assess the parent's level of satisfaction with the treatment received in treatment at the sleep clinic.
- *Adherence*: The adherence tracker is a chart developed for this study (attached to the social story) and is also embedded in the follow-up sleep diary for TAU+SS participants. Adherence was also collected verbally during the follow-up session by self-report. At the follow-up session, parents were asked to recall initial treatment recommendations, ability to maintain adherence to the recommendations, and any challenges that impacted adherence.

Table 2. Timeline for Outcome Measures

	<u>Measure</u>	<u>Time Points</u>	<u>Data for Child or Parent?</u>
<u><b>Aim 1</b></u>	Children's Sleep Habits Questionnaire (CSHQ)	Baseline, Follow-up	Child
	1-week sleep diary	1 weeks prior to evaluation, 1 weeks prior to follow-up	Child
	Patient satisfaction	Baseline, Follow-up	Parent
	Adherence	Follow-up	Child, Parent
<u><b>Aim 2</b></u>	Pittsburgh Sleep Quality Index (PSQI)	Baseline, Follow-up	Parent
	Pediatric Quality of Life Inventory (PedsQL) – Family Impact Module	Baseline, Follow-up	Parent, Family



## STATISTICAL ANALYSES

We utilized data from the final randomized sample of patients and their parents for all statistical analyses. Analyses were carried out using IBM SPSS Statistics version 27.0. The level of significance was set at  $\alpha = 0.05$  (two-tailed) and, to address multiple testing (where applicable), p-values will be adjusted using the False Discovery Rate (Benjamini & Hochberg, 1995).

### **Baseline Demographics and Characteristics**

We used descriptive statistics, Fisher's Exact test, chi-square tests, and independent sample t-tests to examine the baseline data of the total sample and compare demographic characteristics between TAU and TAU+SS groups.

### **Primary Aim Analyses**

#### *Feasibility and Acceptability of Social Stories*

The assessment of feasibility and acceptability of social stories as a complementary treatment tool for behavioral insomnia aids in the development of sleep treatments designed to address specific needs of children with ASD. It may also inform the feasibility and design of future controlled studies in larger populations in related topics.

The primary analyses focused on a core question: does data from parents and children indicate that social stories are acceptable and implementable as a complementary treatment tool for behavioral insomnia? To address this, we analyzed trends in the implementation of the treatment, adherence rates, and overall satisfaction with social stories when implemented at home. Satisfaction was collected via the satisfaction questionnaire and follow-up questions. We summarized qualitative findings derived from parents' verbal responses for why ease of implementation and interest in social stories differed between families.

We assessed descriptive statistics to assess initial indicators of the efficacy of social stories in treatment. The social story was developed to target behaviors around sleep onset delay, so changes related to CSHQ bedtime resistance and sleep-onset delay subscales were of particular interest in these analyses. We also analyzed changes in the CSHQ sleep duration subscale, paired with changes in the sleep diary, to examine overall changes in the quantity of the child's sleep and assess changes in total CSHQ sleep score. We utilized paired t-tests and analysis of covariance (ANCOVA) with the level of significance set at  $\alpha=.05$  (two-tailed) to analyze data in the primary analyses.

### **Secondary Aim**

#### *Aim 2: TAU+SS vs TAU on Parental Sleep, Psychosocial Functioning, and Family Function*

The second set of analyses explored trends in parental sleep, psychosocial functioning, and quality of life for family, both for the total group to assess overall treatment as well as a comparison of between control and treatment groups. Sleep outcome data were derived from the PSQI and psychosocial functioning from the PedsQL. To examine overall changes in parental sleep, we compared changes in the global PSQI score as well as reported sleep quality and sleep efficiency at baseline and follow-up. We examined changes in parental function and family experience by analyzing changes core PedsQL domains by from baseline to follow-up. Satisfaction was collected via the satisfaction questionnaire and follow-up questions. We utilized paired t-test and ANCOVA to analyze data in the secondary analyses. The level of significance was set at  $\alpha=.05$  (two-tailed) for all analyses.

### **Missing Data**

The researcher reviewed all questionnaires prior to the patient's dismissal from the appointments to identify missing fields in the questionnaire to limit the prevalence of missing

data. However, some parents were unable to complete the questionnaires before returning home due to behavioral challenges with their children. In those instances, the researcher followed-up by phone call to capture the missing data and at times referenced the original clinical report with the consent of the parent to aid in parent recollection.

A few parents marked questions as N/A when they had no evidence either way (e.g., claimed they never listened to their child at night so was unsure if they snored or gasped while breathing). For scoring purposes, N/A responses on the CSHQ were scored with a '1' which aligns with the symptoms rarely or never occur. This is a conservative scoring approach and felt appropriate since the symptoms likely would have been noticeable if they occurred regularly, even if parents or caregivers were not specifically watching for them.

One parent chose to complete follow-up data for their child but not for their own sleep and functional status. Consequently, the sample size for the analyses associated with the sample size for the primary research aim (which relies solely on pediatric outcomes) is larger than the sample size for the secondary research aim, which focuses on parental outcomes.

## **CHAPTER FOUR**

### **Results**

### **RESULTS OF ANALYSIS**

#### **Total Sample**

##### *Baseline Characteristics*

At baseline, the sample consisted of 17 pediatric participants plus their parent/caregiver, 47.1% (n=8) of whom were randomized into the TAU+SS group and 52.9% (n=9) into the TAU group. The sex ratio of the pediatric sample (based on legal sex documented in the medical record) was 76.5% (n=13) male and 23.5% (n=4) female. Through chance, all females in the sample were randomized to the treatment (TAU+SS) group. Consequently, the control group was 100% (n=9) male; the treatment group was 50% (n=4) male and 50% (n=4) female. Overall, the mean age of the pediatric sample was  $6.7 \pm 2.1$  years. The mean age for male participants overall  $6.6 \pm 2.1$  years, with a mean age of  $5.6 \pm 2.4$  years in TAU+SS and  $7.3 \pm 1.8$  years in TAU. The mean age for female participants (all in TAU+SS) was  $6.7 \pm 2.4$  years. Demographic information (i.e., parental age, parental sex) was not collected on parents of study participants.

Following attrition, our final analytic sample was comprised of 12 participants and their parent(s) who participated in data collected at baseline and final follow-up. Within the final analysis group, the sample size for primary analyses differs from the secondary analyses sample due to missing data from one parent who completed questionnaires for their child but not for themselves. Additionally, although we collected data at a 2-week check-in call, we experienced challenges with cancellation and failures to respond when attempting to reach participants by telephone. Consequently, we did not include data collected from the brief telephone call in the

final analysis since not all participants who attended their final follow-up session responded to the 2-week call.

### *Findings from Qualitative Report at Baseline*

Although this study did not include a formal qualitative component, the verbal comments parents made while completing the questionnaires further illustrated the broad impact sleep deprivation has on them and their children. Many parents talked for extended periods of time about their worries for their children and their own well-being while completing the questionnaires, with at least seven parents taking an hour or more to complete the questionnaires with the researcher, a task that takes approximately 15 minutes if the parent is focused solely on responding to the questionnaires. One parent resisted filling out the sleep diary, saying they could only describe the nights of the previous week as “hell.”

Several parents began to cry or tear up while working through the questions on the *PedsQL: Family Impact Module* questionnaire, particularly during the sections on emotional functioning, social support, and worry about their children. Some parents said they felt like they were “drowning” and that years of sleep deprivation contributed to a belief that their lives were forever changed. One woman lamented, “I’m not the same person I used to be [because of] no sleep,” and described her preference for pursuing personal goals in the early morning hours and the sense of loss she felt from not being able to engage in those activities for many years. Parents described their sense of desperation to find a solution to the sleep deprivation, noting they had used various behavioral strategies, several types of weighted blankets, different versions of melatonin (brands, pills, etc.), curtains and light-blockers, lavender lotions, and nightlights all to no avail.

In many ways the comments made by parents while completing the questionnaires highlighted the desperation and frustration that has crept into their lives following years of sleep deprivation and challenges with nighttime behaviors. These concerns were not limited to their concerns for their child with ASD and their own wellbeing, but extended to other family members as well, particularly other children in the home. Several parents described that one child's sleep difficulties constantly interrupted the sleep of their siblings. One parent, after describing her deep love for all her children, stated she wondered at times if she had, "messed up my older son's life" due to the challenges the family faced from his younger sibling and general lack of sleep in the home. The comments underscored the deeply felt concerns these parents carried for all members of the family and spoke to their fears of how sleep deprivation may impact their children's functioning and long-term development.

#### *Demographic Characteristics for Analytic Sample*

In the final analytic sample (n=12), 58.3% (n=7) were randomized into the TAU group and 41.7% (n=5) were randomized into the TAU+SS group. Demographics characteristics of the analytic group are presented in Table 3. There were differences in the sex distribution between groups. Similar to the overall sample described above, the control group was 100% male whereas the treatment group was 60% male; these differences were not statistically significant. There were not significant differences in age between the TAU and TAU+SS groups. Although females were on average older than male participants (7.8 vs 6.5 years), this difference in age by sex was not statistically significant.

Clinical characteristics of the sample are also presented in Table 3. The average total CSHQ sleep score was above the proposed cutoff of 41 (Owens et al., 2000), indicating participants as a whole likely met clinical criteria for disordered sleep. Study participants

reported that on average they experienced behavioral resistance at bedtime, delayed sleep onset, and limited sleep duration the majority of evenings in a week. Differences between bedtime resistance and delayed sleep onset were not statistically significant between TAU and TAU+SS. Differences in sleep duration based on CSHQ responses were statistically significant ( $p=0.01$ ) but differences in sleep duration per diary report were not significant between TAU and TAU+SS. Differences in clinical characteristics of the sample were not significant for age nor sex.

Parents reported that on average their sleep fell in the Fairly Bad to Very Bad range with 71 percent sleep efficiency, which indicates clinical concern for disordered sleep for parents at baseline. There were no significant differences in physical functioning, emotional functioning, cognitive functioning, overall total sleep, and family function between TAU and TAU+SS.

Table 3: Baseline Demographics and Clinical Characteristics of Analytic Sample

Variables	Total (n=12)	TAU+SS (n = 5)	TAU (n = 7)	p value
<b>Demographics</b>				
Pediatric Age: Years (M (SD))	6.72 (2.24)	5.67 (2.34)	7.78 (1.71)	0.29
Male (n (%) )	10 (83.3%)	3 (60.0%)	7 (100%)	0.23
<b>Measures (M (SD))</b>				
CSHQ Total	54.75 (7.5)	56.00 (3.35)	53.50 (10.46)	0.59
CSHQ Bedtime Resistance	10.33 (2.4)	10.50 (2.59)	10.17 (2.48)	0.83
CSHQ Sleep Onset Delay	2.00 (0.85)	2.00 (0.89)	2.00 (0.89)	1.00
CSHQ Sleep Duration	6.83 (2.21)	8.33 (0.52)	5.33(2.25)	0.01
Sleep Diary	7.68 (1.54)	7.65 (1.79)	7.72 (1.42)	0.95
Parental Sleep Quality	2.25 (0.87)	2.50 (0.84)	2.00 (0.89)	0.34
Parental Sleep Efficiency	0.72 (0.20)	0.67 (0.19)	0.78 (0.21)	0.34
Parental Total PSQI	12.25 (4.45)	12.50 (4.76)	12.00 (4.56)	0.86
Parental Physical Function	4.26 (2.69)	3.28 (1.88)	5.25 (3.18)	0.22
Parental Emotional Function	2.18 (1.20)	2.47 (1.27)	1.90 (1.16)	0.44
Parental Cognitive Function	2.22 (0.92)	2.07 (0.83)	2.37 (1.05)	0.60
Family Function	1.60 (1.17)	1.17 (0.65)	2.03 (1.46)	0.21

Note: Differences between most groups were not significant when running a Chi Square Independence Test, Fisher's Exact Test (1-sided, for sex), and independent sample t-tests.



### *Analytic Findings - Overall Treatment Outcomes*

Although follow-up data did not provide evidence of efficacy specific to the social stories, it did indicate that in general sleep treatment yielded positive outcomes for both children and parents. This indicates that targeted sleep interventions can directly improve sleep for the child and indirectly improve parental sleep and functioning. Although parents continued to report symptoms of disordered sleep and challenges associated with sleep deprivation for their child and themselves, findings from a paired samples t-test demonstrated an overall reduction in frequency of symptoms and improvements in functional challenges associated with sleep deprivation.

In particular, the results of a paired samples t-test indicated that participants in this study experienced a significant improvement in sleep duration based on CSHQ parent report ( $p = .006$ ,  $SD = 1.4$ , Cohen's  $d = 2.7$ ) and sleep diaries ( $p = .001$ ,  $SD = 1.0$ , Cohen's  $d = 1.0$ ) following treatment at the sleep clinic (for both TAU and TAU+SS groups). Parents also reported a significant improvement in their own physical functioning analysis ( $p = 0.02$ ,  $SD = 2.4$ , Cohen's  $d = 2.44$ ). Additionally, while other findings were not significant, the overall trend in the data indicated clinical improvements for both children and parents following treatment of pediatric sleep disorders. Results for outcomes for total sample are presented in Table 4.

Table 4: Treatment Outcomes for Total Sample (TAU and TAU+SS)  
*Results of Paired Samples T-Test*

	Baseline M (SD)	Follow-Up M (SD)	p value
Pediatric Outcomes			
CSHQ Bedtime Resistance	10.33 (2.43)	10.08 (2.71)	0.75
CSHQ Sleep Onset Delay	2.00 (0.85)	1.50 (0.67)	0.11
CSHQ Sleep Duration	6.83 (2.21)	5.42 (2.27)	0.006
Sleep Diary	7.68 (1.54)	8.55 (1.29)	0.01
CSHQ Total	54.75 (7.52)	50.83 (10.45)	0.07
Parental Outcomes			
Sleep Quality	2.40 (0.84)	2.10 (0.88)	0.19
Sleep Efficiency	0.71 (0.20)	0.71 (0.21)	0.89
PSQI Total	12.45 (4.61)	11.82 (4.90)	0.45
Physical	4.26 (2.69)	2.26 (1.01)	0.02
Emotional	2.18 (1.20)	2.01 (1.11)	0.41
Cognitive	2.22 (0.92)	2.10 (1.08)	0.71
Family Functioning	1.60 (1.17)	1.17 (0.88)	0.16

## Primary Aims

- Aim 1. To examine the feasibility and acceptability of a social story on bedtime routines as a complementary treatment tool for behavioral insomnia in children with ASD and determine if social stories enhance treatment outcomes for children assigned to TAU+SS.

### *Feasibility of the Research Study – Participation Factors*

Regarding the feasibility of this study, our core questions included: Would families be interested in participating? Would we be able to collect enough data to assess strengths, weaknesses, and efficacy of the social story? In terms of willingness to participate, all parent – children dyads (100%, N=19) approached prior to or during their appointment with Dr. Brown consented to participate in the study, suggesting that the feasibility of this study was not hampered by patient concerns regarding the risks or expectations of the study. It should be noted that one mother called to request information on the study and expressed hesitation to participate upon learning the study was conducted as part of a standard clinical visit that was typically charged to insurance; she was not included in the attrition rate since she never officially consented or enrolled in the study.

However, beyond the initial willingness to participate, we experienced moderate challenges with no-shows, cancellations, and call backs to gather data over the phone. This impacted the study both with initial recruitment (i.e., through screening we identified potential participants who later canceled or failed to attend their initial appointment) as well as with ongoing retention efforts following enrollment. Of those who enrolled in the study, more than two thirds of the participants (70.6%) in the study either rescheduled, failed to show, or canceled their follow-up appointments at least once.

Within this group, the number of cancellations or no-shows ranged from 1 to 5 appointments per patient with a mean number of 1.58 of cancellations or failures to show within this group. Although clinical staff did not consistently track all attempts to reschedule appointments with patients, based on the known number of rescheduling attempts, members in this group were on average called 3.33 times (range of 1 to 10 callbacks per patient) by clinical or research staff to reschedule their follow-up appointment before follow-up data were successfully collected or rescheduling attempts were ceased. These challenges with retention severely impacted our data collection and undermined the overall feasibility of the study, particularly in relation to exploring efficacy of social stories.

#### *Acceptability of Social Stories as a Treatment Tool – Adherence and Implementation*

Regarding the acceptability of social stories as a treatment tool in sleep medicine, our core questions included: Would families adhere to treatment and be able to implement it easily? Would the social story impact satisfaction?

Notwithstanding the notable challenges with retention, feedback on the implementation and utilization of the social story was generally positive and promising in terms of the acceptability of social stories as a treatment tool. Of the 5 participants enrolled in the TAU + SS group, all parents (100%) reported using the social story at least once and said it was easy to implement into their bedtime routine. Although the majority of parents failed to consistently use the adherence tracker, they verbally reported utilizing the social story at bedtime an average of  $19.4 \pm 10.2$  days, with a range of 7 days to more than 30 days ( $M=19.4$ ,  $SD=10.2$ , range: 7-30). It should be noted that although we capped frequency of utilization at 30 days to align with the original study design, several parents reported using the social story after follow-up.

As part of our evaluation of acceptability, we assessed if social stories impacted satisfaction at the clinic but found no difference in satisfaction with Dr. Brown and treatment at the Sleep Center between TAU and TAU+SS. All participants (100%) reported they would recommend treatment with Dr. Brown at the Sleep Center for other children with similar concerns at both baseline and follow-up. As part of the satisfaction survey, parents were asked to identify which aspects of the visit they found most helpful and only one parent noted the social story as a driver for their satisfaction. In general, most parents stated their satisfaction was tied to increased education about sleep, guidance on bedtime routines, and the opportunity to discuss their concerns. While this does not support the idea that social stories may be more acceptable than other sleep treatments, it does speak to the need for support around bedtime routines.

#### *Acceptability of Social Stories as a Treatment Tool – Qualitative Report*

Verbal feedback on the social story indicated that for nearly all parents assigned to the TAU+SS group, it was easy to integrate the social story into their child's bedtime routine. Several parents reported their child attached to the social story quickly and was eager to read it at night. One parent reported that the social story was such a positive experience for her child at bedtime that her child requested additional social stories for her morning routine. It is worth noting that within that parent-child dyad, the parent also reported that the child preferred to read the social story to her mother rather than have the mother read it to her.

One parent reported they stopped at 25 days due to illness, which disrupted the bedtime routine but returned to using the social story after the illness, which occurred after the 30-day mark. A subset of the parents (40%, n=2) indicated that they stopped using the story at some point in the trial and did not intend to return to the story. One parent said they stopped using it after 10 days because too many features of the bedtime routine had been changed as a result of

the total treatment plan (i.e., altered bedtime, restriction of television shows before bed, etc.) and she felt it was overwhelming for her child. She later noted that she believed the social story would be improved with greater personalization to the child. In a similar vein, another parent stopped using the social story after 7 days because she felt her child did not understand or connect to the story.

*Analytic Findings – Pediatric Treatment Outcomes, TAU and TAU+SS*

The analysis of changes in pediatric sleep from baseline to follow-up focused on three CSHQ subdomains (Bedtime Resistance, Sleep Onset Delay, and Sleep Duration) as well as the total change in sleep. Sleep duration was also assessed by analyzing baseline to follow-up of data from sleep diaries. Symptoms of disordered sleep were rated by frequency per week (3 = Rarely or 0-1 times per week, 2 = Sometimes or 2-4 times per week, 1 = Usually or 5-7 times per week), so a higher score indicates a child frequently experiences a variety of symptoms of disordered sleep. One-way ANCOVA analyses were conducted to compare treatment outcomes at follow-up for TAU and TAU+SS with sex and respective baseline (Time 1) included as covariates and follow-up pediatric sleep outcomes (bedtime resistance, sleep duration, and CSHQ total; Time 2) as outcomes. Group Least squares Means (adjusted for the covariates in the model) were included in analyses.

The difference between the two intervention groups on change in sleep onset delay as measured by CSHQ2 was trending toward significance,  $F(1,8) = 4.87, p = 0.06$ , Cohen's  $d = 1.27$ ). There was no significant difference between the two intervention groups in bedtime resistance as measured by CSHQ1,  $F(1,8) = 0.28, p = 0.61$ , Cohen's  $d = 0.30$ ). There was no significant difference between the two intervention groups in sleep duration as measured by CSHQ3,  $F(1,8) = 0.49, p = 0.51$ , Cohen's  $d = 0.40$ ), or sleep diary,  $F(1,8) = 0.86, p =$

0.38, Cohen's  $d = 0.54$ ). There was no significant difference between the two intervention groups in total CSHQ sleep score,  $F(1,8) = 0.81, p = 0.40$ , Cohen's  $d = 0.52$ ). Results for sleep outcomes of pediatric participants in the sample are presented in Table 5.

Effect size, as interpreted by Cohen's  $d$ , are described as small at 0.2, medium at 0.5, and large at 0.8 (CohenJ., 2013). The study intervention, though insignificant, was associated with a large effect size on sleep onset delay, a medium effect size on sleep duration and total pediatric sleep, and a small effect size on sleep duration from baseline to follow-up.

### Secondary Aims

- Aim 2: To evaluate the efficacy of social stories for parents of participants assigned to TAU+SS, as measured by results of questionnaires on parental sleep, psychosocial functioning of parents, and general quality of life for the family.

#### *Analytic Findings – Changes in Parental Sleep, TAU+SS vs. TAU*

The analysis of changes in parental sleep from baseline to follow-up focused on two PSQI subdomains (Sleep Quality and Sleep Efficiency) as well as total sleep score. As with the CSHQ, the PSQI rates symptoms of disordered sleep were rated by frequency (3+ time a week, Once or twice a week, Less than once a week, Not during the past month) so a higher score indicates a parent frequently experiences a variety of symptoms of disordered sleep.

One-way ANCOVA analyses were conducted to compare treatment outcomes at follow-up for TAU and TAU+SS with sex and respective baseline (Time 1) included as covariates and follow-up parental sleep outcomes (Sleep Quality, Sleep Efficiency, and PSQI total; Time 2) as outcomes. Group Least squares Means (adjusted for the covariates in the model) were included in analyses. The difference between the two intervention groups in total PSQI score was trending toward significance,  $F(1,7) = 3.55, p = 0.10$ , Cohen's  $d = 1.14$ ). There was no significant

difference between the two intervention groups in parental sleep quality,  $F(F(1,6) = 0.28, p = 0.61, \text{Cohen's } d = 0.34)$ . There was no significant difference between the two intervention groups in parental sleep efficiency,  $F(F(1,7) = 0.01, p = 0.92, \text{Cohen's } d = 0.06)$ . The insignificant study intervention was associated with a large effect size on total parental sleep and a small effect size with parental sleep quality. Results for sleep outcomes of parental participants in the sample are presented in Table 6.

#### *Analytic Findings – Changes in Parental Psychosocial Functioning, TAU and TAU+SS*

The analysis of changes in parental psychosocial functioning from baseline to follow-up focused on three PedsQL subdomains (Physical Functioning, Cognitive Functioning, and Emotional Functioning) as well as total Family Functioning. Symptoms of distress were rated by frequency of challenges in a given category in the 7 days immediately prior to completing the questionnaire (4 = Almost Always, 3 = Often, 2 = Sometimes, 1 = Almost Never, 0 = Never), so a higher score indicates the parent or family frequently experiences distress or challenges.

One-way ANCOVA analyses were conducted to compare treatment outcomes at follow-up for TAU and TAU+SS with sex and respective baseline (Time 1) included as covariates and follow-up parental psychosocial outcomes (Time 2) as outcomes. Group Least squares Means (adjusted for the covariates in the model) were included in analyses. The differences between the two intervention groups in reported cognitive functioning was trending toward significance,  $F(F(1,8) = 3.14, p = 0.12, \text{Cohen's } d = 1.02)$ . There was no significant difference between the two intervention groups in reported physical functioning,  $F(F(1,8) = 2.58, p = 0.15, \text{Cohen's } d = 0.93)$ . There was no significant difference between the two intervention groups in reported emotional functioning,  $F(F(1,8) = 0.04, p = 0.84, \text{Cohen's } d = 0.12)$ . There was no significant difference between the two intervention groups in total score for Family Functioning,  $F(F(1,8)$



= 0.008,  $p = 0.93$ , Cohen's  $d = 0.05$ ). The insignificant study intervention was associated with a large effect size on total parental cognitive and physical function. Results for psychosocial outcomes of parental participants in the sample are also presented in Table 6.

## **COVID-19**

In June 2020, we developed a brief questionnaire to assess the potential impact of COVID-19 on this study and sleep outcomes in general due to heightened anxiety within families, disrupted schedules, reduction in other services/therapies for the children, and other challenges that accompanied the pandemic. We administered this questionnaire over the phone to participants enrolled prior to the pandemic shutdown and during virtual visits to participants enrolled after the pandemic.

Parent responses to these questions did not indicate an obvious trend in sleep challenges associated with the pandemic. The majority of parents reported no change in sleep following the pandemic, though some parents noted their child's sleep appeared to improve following the shutdown (which the parents attributed to lower stress from not attending school or maintaining a busy schedule) and others reported worsened sleep after the pandemic (which the parents attributed to stress and worry about parents' jobs or the health of elderly loved ones).

Table 5: Analysis of Covariance in Pediatric Sleep  
Equation Predicting Effect of TAU+SS on Change, Time 1 to Time 2

	LSM (SE)	95% CI	LSM Group Difference (SE)	95% CI for Group Difference	F Statistic (df)	p-value	<i>d</i>
Sleep Onset Delay			0.89 (0.40)	-0.04 to 1.81	4.87 (1, 8)	0.06	1.27
TAU + SS	1.94 (0.27)	1.33 to 2.56					
TAU	1.06 (0.27)	0.44 to 1.68					
Bedtime Resist			0.89 (1.69)	-2.99 to 4.78	0.28 (1, 8)	0.61	0.30
TAU + SS	10.53 (1.11)	7.97 to 13.09					
TAU	9.64 (1.11)	7.08 to 12.20					
Sleep Duration			-0.94 (1.34)	-4.03 to 2.16	0.49 (1, 8)	0.51	0.40
TAU + SS	4.95 (0.80)	3.11 to 6.79					
TAU	5.89 (0.80)	4.04 to 7.73					
Sleep Diary			-0.55 (0.60)	-1.93 to 0.82	0.86 (1, 8)	0.38	0.54
TAU + SS	8.27 (0.40)	7.35 to 9.19					
TAU	8.83 (0.40)	7.90 to 9.75					
CSHQ Total			4.26 (4.74)	-6.68 to 15.20	0.81 (1, 8)	0.40	0.52
TAU + SS	52.96 (3.16)	45.67 to 60.25					
TAU	48.70 (3.16)	41.41 to 55.99					

Note: Covariates included Sex and respective baseline (Time 1) measure; Cohen's *d* interpretation: .2 (small), .5 (medium), .8 (large) (Cohen, 2013)

Table 6: Analysis of Covariance on Change in Parental Sleep & Psychosocial Function  
*Equation Predicting Effect of TAU+SS on Change, Time 1 to Time 2*

	LSM (SE)	95% CI	LSM Group Difference (SE)	95% CI for Group Difference	F Statistic (df)	p-value	<i>d</i>
PSQI Total			2.65 (1.40)	-0.67 to 5.97	3.55 (1, 7)	0.10	1.14
TAU + SS	13.02 (0.89)	10.92 to 15.13					
TAU	10.38 (0.99)	8.05 to 12.71					
Sleep Quality			-0.25 (0.47)	-1.40 to 0.90	0.28 (1, 6)	0.61	0.34
TAU + SS	2.00 (0.28)	1.31 to 2.69					
TAU	2.25 (0.35)	1.39 to 3.11					
Sleep Efficiency			-0.008 (0.08)	-0.19 to 0.17	0.01 (1, 7)	0.92	0.06
TAU + SS	0.70 (0.05)	0.59 to 0.81					
TAU	0.71 (0.05)	0.59 to 0.83					
Cognitive Function			1.07 (0.60)	-0.32 to 2.46	3.14 (1, 8)	0.12	1.02
TAU + SS	2.63 (0.40)	1.70 to 3.57					
TAU	1.57 (0.40)	0.63 to 2.50					
Physical Function			1.00 (0.62)	-0.44 to 2.44	2.58 (1, 8)	0.15	0.93
TAU + SS	2.77 (0.41)	1.82 to 3.72					
TAU	1.76 (0.41)	0.81 to 2.71					
Emotional Function			-0.11 (0.55)	-1.38 to 1.15	0.04 (1, 8)	0.84	0.12
TAU + SS	1.95 (0.35)	1.15 to 2.78					
TAU	2.06 (0.35)	1.26 to 2.87					
Family Function			0.51 (0.56)	-1.25 to 1.35	0.008 (1, 8)	0.93	0.05
TAU + SS	1.19 (0.37)	0.34 to 2.04					
TAU	1.14 (0.37)	0.29 to 1.99					

Note: Covariates included Sex and respective baseline (Time 1) measure; Cohen's *d* interpretation: .2 (small), .5 (medium), .8 (large) (Cohen, 2013)

## **CHAPTER FIVE**

### **Discussion**

#### **SUMMARY AND RELEVANCE**

This study was developed as a feasibility study for utilizing social stories in the treatment of behavioral insomnia for children with ASD. The social stories provide a narrative framework for supporting the implementation of a consistent, positive bedtime routine. All pediatric participants had received a diagnosis for ASD that had been documented by medical providers in their electronic medical record, though it is unknown what percentage of participants received an ADOS during their diagnostic assessment. All adult respondents were primary caregivers for the child diagnosed with ASD. The majority of patients (88%) received specialized care at CADD or other clinics at Children's Medical Center in Dallas, TX to support their therapeutic needs associated with ASD.

Additionally, all participants sought specialized care at Children's Health Sleep Disorders Center to manage concerns related to behavioral insomnia and disordered sleep. Pediatric participants presented with insomnia with particular challenges associated with bedtime resistance as well as other features of disordered sleep (i.e., frequent/prolonged nighttime awakenings). The majority of pediatric participants within the analyzed sample were male, which is common for any sample of children with ASD since boys are significantly more likely to be diagnosed with ASD. The average age in our sample was 6.7 years. Participants were randomized into control (TAU) and treatment (TAU+SS) groups upon enrollment.

In general, parental participants also presented with symptoms of disordered sleep as well as challenges in psychosocial functioning per their report on the PSQI and PedsQL. For this study, we focused on physical, emotional, and cognitive functioning for parents, but parents reported challenges in nearly all domains of psychosocial functioning. Levels of disordered

sleep, as measured by CSHQ and PSQI, and psychosocial functioning, as measured by PedsQL-Family Impact, was not significantly different between groups. Analyses indicated that treatment in general at the Sleep Center was associated with a trend of an overall improvement in sleep, with significant improvements in pediatric sleep duration and parental physical functioning.

Qualitative findings from this study highlighted the anxiety, concern, and sense of loss many parents feel related to ongoing sleep deprivation, indicating the importance of addressing sleep concerns for children when working to support the child, parent, and entire family unit. Based on these findings, it may be critical for clinicians who specialize in ASD but not sleep medicine to screen for sleep challenges routinely as it may be underreported in ASD clinics. By screening for disordered sleep more regularly, clinicians may become more practiced in providing support and education for healthy bedtime routines and behavioral strategies, refer patients to sleep providers more frequently, and be more likely to utilize treatment tools like the social story or other tools that effectively address disordered sleep.

Regarding study feasibility, results from this study indicated significant limitations that negatively affected the execution of the study design. More specifically, interest in the study and willingness to participate was high at recruitment, suggesting social stories may be a feasible complementary treatment for behavioral insomnia among children with ASD. However, we also experienced a high rate of canceled, rescheduled, or missed appointments throughout the study. Although rate of missed appointments during this study aligned with general trends of cancellation and reschedule rates at the Sleep Center, it still impacted the study. These issues with retention limited our ability to collect follow-up data and undermined the feasibility of the study design. While many of these challenges were connected to issues that arose with the global COVID-19 pandemic, these difficulties were present in recruitment efforts prior to the pandemic

and may highlight standard challenges this patient population faces due to their significant load of therapeutic treatments.

Children with ASD frequently have a complex calendar of therapeutic routines, medical appointments and educational responsibilities that complicate their (and their parents') ability to consistently engage in important but nonessential activities, whether that be a research study or other activities they would like to pursue. In our study, parents referenced the challenges they experience trying to maintain a complex schedule of appointments, with parents explaining that they felt like they were in "survival mode all the time" and on the "go, go, go... [dealing with] insurance, therapy, school" for their children. Indeed, some participants noted the COVID-19 pandemic facilitated an easier research follow-up as it allowed them to complete the questionnaires over the phone with the researcher when they were unable to keep an original appointment. In sum, the challenges we faced in retention limited the feasibility of effectively assessing the efficacy of social stories. Yet these challenges also highlight the importance of a research design that allows for flexibility, sensitivity, and responsiveness when providing clinical care and conducting research with children with ASD as scheduling challenges appear to be a common feature in the lives of many families.

Despite challenges with the feasibility of executing the study in alignment with original protocols, results from the study are promising in terms of the acceptability of social stories as a treatment. All participants in the treatment group utilized the social story for at least one week, with an average utilization rate of 19 days (out of 30 possible days). Several children continued to use the social story after 30 days and one requested a social story for other routines in her day. Qualitative feedback from the treatment group indicated that the social story was easy to implement and generally well received by the children, while also emphasizing the importance of

personalization and flexibility to ensure ease of implementation for all children and family situations. Indeed, results of this study indicate that more liberality in personalization of the social story and flexibility in implementation guidelines may have increased adherence for families in this study. Though social stories were well received by participants, quantitative results did not reveal significant differences in results in pediatric sleep, parental sleep, parental psychosocial functioning, family function, and general satisfaction between groups. There was no indication based on the data in this study that it is preferable to standard sleep treatment.

## COMPARISON OF FINDINGS TO THE LITERATURE

The results of the current study indicate that social stories are acceptable as a complementary tool in pediatric sleep treatment for children with ASD and a useful aid for parents to implement treatment strategies at home. This study contributes to the small literature on the use of social stories as a therapeutic treatment for sleep challenges in children with ASD. The findings from the current study align with the results of an earlier study which found a decrease in the frequency of disruptive bedtime behaviors, improved sleep maintenance, and improvements in daytime functioning following use of positive reinforcement strategies paired with social stories (Burke et al., 2004) . The current study extended this line of research and adds to it with a larger sample size, a randomized control design, and an intervention tool that was standardized but also minimally personalized to the child. In the current study, parents reportedly appreciated the ease of implementation of the social story at home.

The qualitative feedback we received in this study suggested that adherence may have been higher if the social stories has been adapted to more closely align with children's developmental level or tailored to the family situation. Personalization is a key recommendation for social stories, according to the guidelines set forth by Gray (Gray, 2014)). While personalization of the social stories may be inherently limited in a randomized control study due to the nature of the design, it may prove to be a critical feature of successful utilization of social stories in a clinical environment.

The findings from this study also connect to literature on the role of educational handouts in sleep treatment. Researchers have previously found that while parents found the information within educational pamphlets on sleep to be helpful, they also noted it was challenging to implement at home without targeted guidance which may have contributed to a lack of significant results in improving sleep latency for children (Adkins et al., 2012). The results of the



current study indicate that parents and children found social stories to be an acceptable bridge between education from a clinical provider and implementation of strategies at home. More broadly, researchers have examined the efficacy of certain educational strategies for parents within the ASD population. Intensive in-person sleep education, whether delivered in group or individual sessions was associated with improvements in children's sleep onset delay (Malow et al., 2014), which complements the overall treatment findings of the current study that the pediatric participants experienced improvements in sleep duration and the trend toward significant differences in sleep onset delay between treatment groups.

Additionally, the findings from this study on parental functioning add further support to previous findings which found that pediatric sleep disorder are associated with significant stress in family members (Kotagal & Broomall, 2012; Stores, 1999) and may influence parents' cognitive and psychosocial functioning (Reynolds & Malow, 2011). Although the current study has limited results on the efficacy of social stories and their impact on parental functioning, the qualitative findings suggest that overall the social stories helped parents maintain a positive bedtime routine with their child.

## **LIMITATIONS**

This study has several limitations which may impact the generalizability and interpretation of the data. One primary limitation is the diagnostic data for our evaluable sample. Recruitment was restricted to children with ASD listed in their electronic medical chart. However, most participants had limited information regarding the original diagnostic criteria, particularly whether an ADOS (the gold standard for ASD diagnosis) was utilized in the diagnostic process. Due to limitations of resources, we were unable to confirm symptoms of ASD in a systemic manner via the administration of an ADOS. While the majority of our sample was also seen at clinics dedicated to supporting ASD treatments and therefore had likely received an ADOS in the past, we cannot conclusively say that their ASD diagnoses were consistent with ADOS criteria.

There are additional limitations related to the demographics of our sample. The majority of our sample was male (83%) with an average age of 6.7 years. This may limit the generalizability to the population at large because although historically it appeared the ASD diagnostic ratio for boys to girls was 4:1, a recent meta-analysis found that the true ratio is likely 3:1 (Loomes, Hull, & Polmear Locke Mandy, 2017). Our sample may slightly overrepresent boys. Additionally, due to psychometric constraints for the primary pediatric sleep questionnaire (CSHQ), we limited the age to 4 to 10 years of age. This impacts the generalizability of the study to younger children with an ASD diagnosis, who may theoretically respond well to additional structure and guidance during their bedtime routine.

Additionally, while all participants in the study exhibited behavioral insomnia at the time of enrollment, the severity of the behavioral factors varied by child. Some parents described mild bedtime resistance while other parents described significant bedtime resistance as well as a host of other behavioral concerns. This study did not distinguish between severe vs moderate vs mild

behavioral concerns. The intensity of the standard treatment and number of changes recommended by Dr. Brown was likely to differ based on the intensity of the child's behavioral presentation. This variability may confound our findings as the social story may be more or less appropriate and effective depending on the number of interventions the parents are attempting to implement.

Additionally, we recruited our sample from a pediatric sleep clinic, suggesting a population of children with acute sleep disorders who have resources and ability to access specialized care. Many children with ASD and disordered sleep may not live in a geographic area with pediatric sleep specialists and may instead seek treatment for sleep from general community providers. Although the social story was developed specifically as a tool that could be implemented by a variety of providers with a range of expertise, this study did not evaluate social stories as a standalone treatment. This limits the ability to generalize the findings beyond the use of social stories when used as a complementary tool for targeted treatments for sleep.

The study itself was temporarily halted and hampered by the COVID-19 pandemic. Approximately half of the participants were recruited prior to March 2020, when Children's Medical Center in Dallas, TX had to cancel/reschedule hundreds of patients to develop and implement policies and procedures for social distancing, telehealth practices, and additional factors that had to be adjusted to keep patients safe in a global pandemic. The remaining half of participants enrolled in the study in the summer of 2020, when policies and procedures continued to undergo changes and adaptations to the pandemic. Although all participants were called after COVID-19 and asked to identify changes in sleep specifically related to COVID-19, this study did not include an objective assessment of the impact of COVID-19 on their sleep.

The small sample size in this study limits the ability to look at efficacy outcome data. While a sample size of 12 is appropriate for a proof of concept study particularly with a pediatric ASD population, the small sample size reduced statistical power to effectively examine the efficacy of social stories. In addition, the efficacy outcome data were based on self-report and would have been strengthened with an objective measure of sleep such as actigraphy. While this study did not have the resources necessary to support the use of actigraphy and thus relied on sleep diaries, the lack of objective outcome data limits interpretation of the findings.

Additionally, the sleep diary and the adherence tracker both required parents to remember to track data at home and bring the data to their appointment. The vast majority of parents forgot to do this, so their responses were based on their recall of how the child slept the previous week and how often they read the social story. Although the researcher worked to help the parents recollect as accurately as possible, biased recall likely impacted the accuracy of the data on those measures. Finally, the outcome data were also gathered at different points across participants due to significant challenges with cancellations, rescheduling, failing to show, and not answering the phone. The varied timeline for collection of follow-up data limits the ability to interpret the findings.

In addition to limitations with our sample and challenges that impacted data collection, there were also limitations with the social story intervention. One potential limitation is that it is unclear whether the child's positive experience with the social story was directly connected to the social story or if it was due to the one-on-one time they were afforded by sitting with their parents to read a special book every night. The interpersonal effects of the parent-child time are unknown and may interact with any positive effect directly associated with the social story. Additionally, while the social stories were minimally personalized to each child (including

adding a name, a personal bedtime, and the name of the caregiver that most often helps them get ready for bed), they were still in a standardized format. Per report by some parents, the lack of personalization impacted the child and parent's connection with the social story. Consequently, the results of this study may not adequately represent the clinical efficacy of social stories since clinicians are able to personalize and modify stories to more directly match needs and preferences of their patients.

## **FUTURE RESEARCH**

Future research should seek to better understand efficacy of the social story when used in sleep treatment. Part of this should focus on the design and development of the social story, including best approaches for personalizing the social story. Some examples of personalization which may improve efficacy include adjusting aspects of the social story bedtime routine to more closely map with a family's routine, using visual images that more closely reflect the child's race and family's cultural heritage, or modifying the language based on age.

Another focus could be factors that contribute to successful implementation and utilization of the social story. This may include assessing the efficacy when one caregiver reads the story each night versus switching between caregivers, when a child (who is literate) read the social story to themselves versus has their parent read it to them, and timing of the social story as part of the bedtime routine. It may also be worthwhile to examine the comparison of a social story to pre-determined bedtime book that ensures focused attention between the parent and child to examine interactions effect between parental attention and reading the social story.

As part of examining the efficacy, further research may include examining the relative impact of social stories on different components of behavioral insomnia. In addition, research should be conducted to better understand which parents and children are most likely to utilize the social story and how factors of sleep treatment (i.e., number of treatment recommendations, severity of sleep concerns) impact utilization. While all of the participants in the current study utilized social stories at least once, continued usage varied widely amongst participants. Further examination of strengths and limitations of social stories when implemented will be beneficial. A qualitative study may be particularly well suited to examine the factors that impact successful implementation of social stories.

As part of this, the TAU+SS group was too small to effectively test the impact of “dosing” on outcomes, but future examinations should examine whether more repetitions of the social story are associated with stronger outcomes. Conversely, a parent choosing to continue repetitions may indicate greater severity of behavioral resistance at bedtime and/or cognitive delays that impact comprehension of the social story and the ability to quickly learn new tasks. A more expansive study on dosing efficacy would more effectively tease out the impact of repetitions and when more sustained use of the story would be vital for a successful outcome.

In addition to examining and modifying the design, development, and implementation strategies for social stories, there are extrinsic areas of follow-up research that may prove beneficial. For example, it may be critical to examine the efficacy of the social story when used with complementary treatment in a specialized sleep center as well as when it is used as a solo treatment or in an environment where sleep concerns may be addressed even if specialized sleep treatment is not available (e.g., pediatricians’ office, ABA therapist, school). The utilization and efficacy of social stories may differ across settings. The data and findings derived from these alternative settings are apt to provide clarity around the role of social stories in treating sleep disorders among children with ASD.

Future research should also include objective measures of sleep (i.e., actigraphy) and adherence to counter the challenges associated with self-reported data based on recall. Additionally, given the concerns related to high rates of appointment cancellations, rescheduling, and failure to show, future research may benefit from streamlining the study with other services that patients use more frequently (i.e., school, ABA therapy, etc.).

## **Conclusion**

Overall, social stories were well liked and utilized in our sample, though no significant differences were found on outcome measures of disordered sleep between the TAU+SS group and the TAU group. The findings from this study expand on previous research on sleep treatment for children for ASD, indicating that parents and children found social stories to be an acceptable tool to support sleep treatment. Future research should examine how features of the design and development of social stories, as well as factors of implementing social stories at home, affect the efficacy of social stories in the treatment of sleep disorders for children with ASD.



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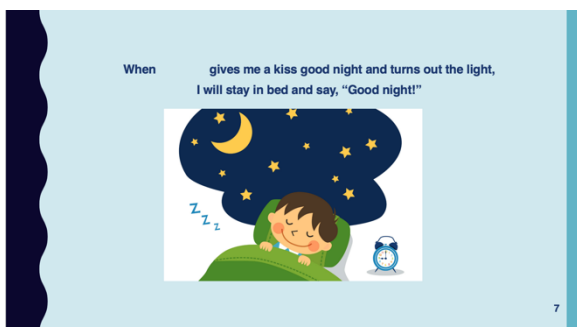
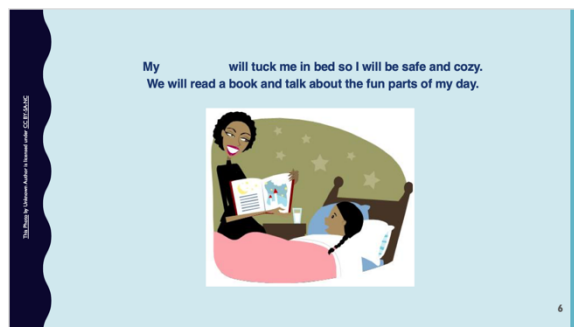
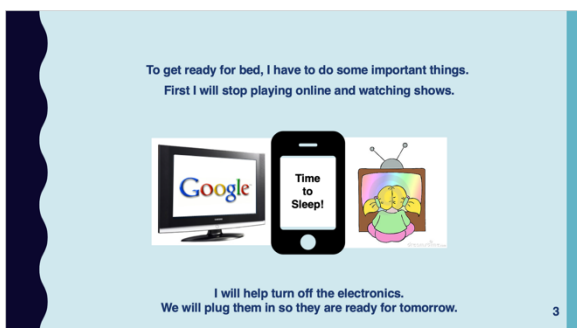
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# APPENDIX A

## Social Story

### Appendix A. 1 Social Story



## APPENDIX B

### Questionnaires

#### Appendix B. 1 Child's Sleep Habits Questionnaire

##### Child's Sleep Habits Questionnaire (pre-school and school-aged children)

The following statements are about your child's sleep habits and possible difficulties with sleep. Think about the past week in your child's life when answering the questions. If last week was unusual for a specific reason (such as your child had an ear infection and did not sleep well or the TV set was broken) choose the most recent typical week.

Answer USUALLY if something occurs **5 or more times** in a week.

Answer SOMETIMES if it occurs **2-4 times** in a week.

Answer RARELY if something occurs **never or 1 time** during a week.

Indicate whether or not the sleep habit is a problem by circling "Yes", "No," or "not applicable (N/A)".

Write in child's bedtime: \_\_\_\_\_ Write in child's usual wake time: \_\_\_\_\_

Child's usual amount of sleep each night (no naps): \_\_\_\_\_ hours and \_\_\_\_\_ minutes

Child's usual amount of sleep each day (naps): \_\_\_\_\_ hours and \_\_\_\_\_ minutes

	1 Usually (5-7)	2 Sometimes (2-4)	3 Rarely (0-1)	Problem?		
1. Child goes to bed at the same time at night	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
2. Child falls asleep alone in own bed	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
3. Child falls asleep within 20 minutes after going to bed	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
4. Child sleeps the right amount	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
5. Child sleeps about the same amount each day	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
6. Child wakes up by him/herself	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A

Child has appeared very sleepy or fallen asleep during the following (check all that apply):

	0 Not Sleepy	1 Very Sleepy	2 Falls Asleep
7. Watching TV	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Riding in a car	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

**PLEASE TURN OVER AND COMPLETE OTHER SIDE!!!**

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	3 Usually (5-7)	2 Sometimes (2-4)	1 Rarely (0-1)	Problem?		
9. Child falls asleep in parent's or sibling's bed	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
10. Child struggles at bedtime (cries, refuses to stay in bed, etc.)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
11. Child needs parent in the room to fall asleep	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
12. Child is afraid of sleeping alone	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
13. Child sleeps too little	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
14. Child is afraid of sleeping in the dark	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
15. Child has trouble sleeping away from home (visiting relatives, vacation)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
16. Child moves to someone else's bed during the night (parent, sibling, etc.)						
17. Child awakens once during the night	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
18. Child awakens more than once during the night	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
Write the number of minutes a night waking usually lasts: _____						
19. Child talks during sleep	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
20. Child is restless and moves a lot during sleep	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
21. Child sleepwalks during the night	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
22. Child wets the bed at night	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
23. Child grind teeth during sleep (your dentist may have told you this)	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
24. Child awakens alarmed by a frightening dream	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
25. Child awakens during night screaming, sweating, and inconsolable	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
26. Child snores loudly	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
27. Child seems to stop breathing during sleep	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
28. Child snorts and/or gasps during sleep	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
29. Child wakes up in a negative mood	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
30. Adults or siblings wake up child	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
31. Child has difficulty getting out of bed in the morning	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
32. Child takes a long time to become alert in the morning	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A
33. Child seems tired in the morning	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<u>Yes</u>	No	N/A

**PLEASE TURN OVER AND COMPLETE OTHER SIDE!!!**

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## Appendix B. 2 Pediatric Quality of Life Inventory – Family Impact Module

*In the past 7 days, as a result of your child's health, how much of a problem have you had with...*

<b>PHYSICAL FUNCTIONING (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some- times</b>	<b>Often</b>	<b>Almost Always</b>
1. I feel tired during the day	0	1	2	3	4
2. I feel tired when I wake up in the morning	0	1	2	3	4
3. I feel too tired to do the things I like to do	0	1	2	3	4
4. I get headaches	0	1	2	3	4
5. I feel physically weak	0	1	2	3	4
6. I feel sick to my stomach	0	1	2	3	4

<b>EMOTIONAL FUNCTIONING (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some- times</b>	<b>Often</b>	<b>Almost Always</b>
1. I feel anxious	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I feel frustrated	0	1	2	3	4
5. I feel helpless or hopeless	0	1	2	3	4

<b>SOCIAL FUNCTIONING (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some- times</b>	<b>Often</b>	<b>Almost Always</b>
1. I feel isolated from others	0	1	2	3	4
2. I have trouble getting support from others	0	1	2	3	4
3. It is hard to find time for social activities	0	1	2	3	4
4. I do not have enough energy for social activities	0	1	2	3	4

<b>COGNITIVE FUNCTIONING (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some- times</b>	<b>Often</b>	<b>Almost Always</b>
1. It is hard for me to keep my attention on things	0	1	2	3	4
2. It is hard for me to remember what people tell me	0	1	2	3	4
3. It is hard for me to remember what I just heard	0	1	2	3	4
4. It is hard for me to think quickly	0	1	2	3	4
5. I have trouble remembering what I was just thinking	0	1	2	3	4

<b>COMMUNICATION (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some- times</b>	<b>Often</b>	<b>Almost Always</b>
1. I feel that others do not understand my family's situation	0	1	2	3	4
2. It is hard for me to talk about my child's health with others	0	1	2	3	4
3. It is hard for me to tell doctors and nurses how I feel	0	1	2	3	4

*In the past 7 days, as a result of your child's health, how much of a problem have **you** had with...*

<b>WORRY (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. I worry about whether or not my child's medical treatments are working	0	1	2	3	4
2. I worry about the side effects of my child's medications/medical treatments	0	1	2	3	4
3. I worry about how others will react to my child's condition	0	1	2	3	4
4. I worry about how my child's illness is affecting other family members	0	1	2	3	4
5. I worry about my child's future	0	1	2	3	4

### DIRECTIONS

Below is a list of things that might be a problem for **your family**. Please tell us **how much of a problem** each one has been for **your family** during the **past 7 days**.

*In the past 7 days, as a result of your child's health, how much of a problem has **your family** had with...*

<b>DAILY ACTIVITIES (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. Family activities taking more time and effort	0	1	2	3	4
2. Difficulty finding time to finish household tasks	0	1	2	3	4
3. Feeling too tired to finish household tasks	0	1	2	3	4

<b>FAMILY RELATIONSHIPS (problems with...)</b>	<b>Never</b>	<b>Almost Never</b>	<b>Some-times</b>	<b>Often</b>	<b>Almost Always</b>
1. Lack of communication between family members	0	1	2	3	4
2. Conflicts between family members	0	1	2	3	4
3. Difficulty making decisions together as a family	0	1	2	3	4
4. Difficulty solving family problems together	0	1	2	3	4
5. Stress or tension between family members	0	1	2	3	4

## Appendix B. 3 Pittsburgh Sleep Quality Index

### Pittsburgh Sleep Quality Index (PSQI)

Instructions: The following questions relate to your usual sleep habits during the past month only. Your answers should indicate the most accurate reply for the majority of days and nights in the past month. **Please answer all questions.**

1. During the past month, what time have you usually gone to bed at night? \_\_\_\_\_
2. During the past month, how long (in minutes) has it usually taken you to fall asleep each night? \_\_\_\_\_
3. During the past month, what time have you usually gotten up in the morning? \_\_\_\_\_
4. During the past month, how many hours of actual sleep did you get at night? (This may be different than the number of hours you spent in bed.) \_\_\_\_\_

5. During the <u>past month</u> , how often have you had trouble sleeping because you...	Not during the past month	Less than once a week	Once or twice a week	Three or more times a week
a. Cannot get to sleep within 30 minutes				
b. Wake up in the middle of the night or early morning				
c. Have to get up to use the bathroom				
d. Cannot breathe comfortably				
e. Cough or snore loudly				
f. Feel too cold				
g. Feel too hot				
h. Have bad dreams				
i. Have pain				
j. Other reason(s), please describe:				
6. During the past month, how often have you taken medicine to help you sleep (prescribed or "over the counter")?				
7. During the past month, how often have you had trouble staying awake while driving, eating meals, or engaging in social activity?				
	No problem at all	Only a very slight problem	Somewhat of a problem	A very big problem
8. During the past month, how much of a problem has it been for you to keep up enough enthusiasm to get things done?				
	Very good	Fairly good	Fairly bad	Very bad
9. During the past month, how would you rate your sleep quality overall?				

	No bed partner or room mate	Partner/room mate in other room	Partner in same room but not same bed	Partner in same bed
10. Do you have a bed partner or room mate?				
	Not during the past month	Less than once a week	Once or twice a week	Three or more times a week
If you have a room mate or bed partner, ask him/her how often in the past month you have had:				
a. Loud snoring				
b. Long pauses between breaths while asleep				
c. Legs twitching or jerking while you sleep				
d. Episodes of disorientation or confusion during sleep				
e. Other restlessness while you sleep, please describe:				

### Appendix B. 4 Sleep Diary

## Sleep Diary

Every morning when you get up complete the sleep diary for the previous night. For example, on Monday morning fill in the information for Sunday night.

[illegible]