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Bedtime Fading with Response Cost for Treatment of Sleep Disturbances in Children with Autism Spectrum Disorder

by

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A DISSERTATION

Presented to the Faculty of the University of Nebraska Medical Center Graduate College in Partial Fulfillment of the Requirements for the Degree of Doctor of Philosophy

Medical Sciences Interdepartmental Area Graduate Program Psychology Department (Applied Behavior Analysis)

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Supervisory Committee:

Keith Allen, Ph.D. Holly Roberts, Ph.D. William Warzak, Ph.D.
As many as 82% of children with Autism Spectrum Disorders (ASD) experience numerous chronic sleep-related problems and at a much higher frequency and severity than their typically developing peers. Behavioral treatments are considered best practice and first-line treatment to address sleep problems. These treatments tend to address one specific sleep-related behavior at a time. Bedtime Fading with Response Cost (BFRC) is a promising intervention that targets a multitude of sleep problems concurrently and has yet to be replicated by more than one investigative team in the home setting with children on the autism spectrum. This study evaluated the effectiveness of BFRC in decreasing sleep disturbances in children with ASD using parents as change agents by implementing treatment in the home environment. A non-concurrent multiple baseline design across three participants was used. Results indicate that BFRC was effective in eliminating unwanted co-sleeping, frequent night awakenings, and dependent sleep onset and reducing sleep onset latency, bedtime resistance, and disruptive sleep-related behaviors. Follow-up data demonstrate that gains were maintained. Parents reported high satisfaction with BFRC and sleep outcomes for their children. This study extends both the
practice and science of parent-implemented behavioral interventions as treatment options for children with ASD and co-occurring sleep problems.

Keywords: autism, sleep, children, parents, behavioral intervention
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Introduction

Background

Autism Spectrum Disorder (ASD) is a neurodevelopmental disorder affecting one out of 68 children in the United States (Center for Disease Control and Prevention [CDC], 2016). ASD is characterized by core deficits in social communication and social interactions and atypical repetitive behaviors and restricted interests (American Psychiatric Association [APA], 2013; CDC, 2016; Reynolds & Malow, 2011). Sleep disturbance is one of the most common and challenging behaviors experienced by children with ASD (Dominick, Ornstein Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Levin & Scher, 2016; Reynolds & Malow, 2011; Wiggs & Stores, 2004) and is frequently associated with co-occurring behavioral difficulties (Adams, Matson, & Jang, 2014; Mazurek & Sohl, 2016). Prevalence rates of chronic sleep-related problems in children with ASD range from approximately 52% to 82% compared with 32% to 50% in children with typical development (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008).

Sleep disturbance is an umbrella term for a range of sleep-related problems presumably lacking an underlying medical basis, as opposed to sleep disorders. In the pediatric sleep literature, the sleep disturbance and sleep problems terms are often used interchangeably (Turner & Johnson, 2012) to refer to common parent-reported sleep concerns such as bedtime refusal, delayed sleep onset, frequent night awakenings, and undesired co-sleeping (Blampied & France, 1993). Parents of children with ASD report a
variety of disturbed sleep behaviors, such as reduced sleep duration, delayed sleep onset, early rising, excessive daytime sleep, lack of independent sleep initiation skills, frequent night waking, difficulty following bedtime routines and undesired co-sleeping (Christodulu & Durand, 2004; Durand, 2002; Knight & Johnson, 2014; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997; Weiskop, Richdale, & Matthews, 2005). Notably, these particular sleep problems are not uniquely different than those reported by parents of typically developing children. It is clear, however, that children with ASD are affected by disrupted sleep at a much higher frequency and with a much greater severity than their typically developing peers (Anders, Iosif, Schwichtenberg, Tang & Goodlin-Jones, 2011; Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Owens & Whitmans, 2004).

Parents’ report that their child’s sleep problems exacerbate difficulties with social functioning, mood, and fatigue (Lopez-Wagner, Hoffman, Sweeney, Hodge, & Gilliam, 2008), which may, in turn, decrease the child’s quality of life and limit independent functioning. Sleep problems in children with ASD can also have detrimental effects on the entire household (Patzold, Richdale, & Tonge, 1998; Tilford, et al., 2015). When children do not sleep, it often means that their siblings and parents do not sleep, either. Parents of children with ASD who sleep poorly during the previous night are likely to have an increased negative affect/decreased positive affect the following day (Mihaila & Hartley, 2016). Additionally, mothers who experience chronic poor sleep perceive their child’s behavior problems as occurring at a higher frequency and severity and/or their own parenting skills as less effective (Mihaila & Hartley, 2016).
Parents of children with ASD experience higher levels of stress and depression and lower levels of marital intimacy than parents of children with and without other disabilities (Fisman, Wolf, & Noh, 1989; Levin & Scher, 2016; Tilford, et al., 2015). Parents of children with ASD report a lack of time for family activities, recreation, and for each other, which can negatively impact the quality of marriage (Fisman, Wolf, & Noh, 1989). This may explain why sleep disturbances experienced by children with ASD often result in increased family stress, marital discord, and decreased parental intimacy (Goodlin-Jones, Tang, Liu, & Anders, 2008; Fisman, Wolf, & Noh, 1989; Hoffman, et. al., 2008; Mihaila & Hartley, 2016; Meltzer, 2011; Patzold, Richdale, & Tonge, 1998; Piazza & Fisher, 1991b; Tilford, et al., 2015).

**Treatment**

Pharmacological agents in conjunction with behavioral intervention are often recommended for the treatment of sleep disturbance in children with ASD (Johnson & Malow, 2008). Medications prescribed to children with ASD to treat sleep disturbance include synthetic melatonin, benzodiazepines, clonidine, beta-blockers, as well as antipsychotics as a means to counter undesired motor effects (Murray, et. al., 2014). Melatonin is a naturally occurring hormone released by the pineal gland to help promote and maintain the circadian rhythm. Synthetically derived melatonin is widely available in the United States as an over-the-counter supplement (Bramble & Feehan, 2005) and is used as a first-line treatment for promoting sleep in children on the autism spectrum (Malow & McGrew, 2008).

To date, the use of behavioral principles (Jin, Hanley, & Beauliue, 2013; Knight & Johnson, 2014; Turner & Johnson, 2012; Vriend, Corkum, Moon, & Smith, 2011) or
melatonin (Malow et al., 2012; Rossignol & Frye, 2011) to address behavioral sleep disturbances in children with ASD has been explored in 16 and 10 studies, respectively. Of the melatonin studies, two represent retrospective reviews of records (Andersen, Kaczmarska, McGrew, & Malow, 2008; Gupta & Hutchins, 2005) and six combine the implementation of behavioral principles with the delivery of melatonin (Garstang & Wallis, 2006; Giannotti, Cortesi, Cerquiglini, & Bernabei, 2006; Malow et al., 2012; Paavonen, Nieminen-von Wendt, Vanhala, Aronen, & von Wendt, 2003; Wasdell et al., 2008; Wright et al., 2010). Only two studies have evaluated melatonin administration alone without concomitant use of behavioral methods, and one of them is a brief case study (Jan, Freeman, Wasdell, & Bomben, 2004; Wirojanan et al., 2009).

Melatonin has been shown to improve sleep outcomes in children with ASD by reducing sleep onset latency (Dodge & Wilson, 2001), and increasing total sleep time (Wirojanan et al., 2009). The sustainability of such benefits, once the medication is withdrawn, has yet to be evaluated. Research results indicate that melatonin does not have an effect on the parental concerns of frequency of night awakenings (Dodge & Wilson, 2001; Wirojanan et al., 2009). Moreover, lacking is the evaluation of melatonin as a treatment option for other disruptive sleep behaviors that parents have identified as their primary concern (e.g., bedtime resistance and undesired co-sleeping) (Christodulu & Durand, 2004; Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005).

At this time, there is no established dosage or recommended length of use of melatonin in addressing sleep problems in children with ASD (Kennaway, 2015; Malow et al., 2012; Rossignol & Frye, 2011). Some children with ASD may experience poor
metabolism and disappearing effects of melatonin, leading to potential dose escalation (Braam, et al., 2012). Similar to other pharmacological agents, melatonin falls short for many parents because taking a medication does not teach healthy sleep behaviors to ensure lasting change (Malow & McGrew, 2008). Therefore, research on the exclusive use of melatonin to improve sleep disturbances in children, with and without ASD, remains limited and needs to be more rigorously evaluated (Kennaway, 2015).

In the field of behavioral sleep medicine, interventions are based on the principles of learning that employ methods of reinforcement, contingency management, shaping, and extinction to teach healthy and appropriate sleep behaviors that last a lifetime. Interventions with a validated body of research to support their utility, referred to as empirically supported treatments (ESTs), are considered first-line treatments in behavioral sleep medicine. The ‘gold standard’ criteria for determining the validity of such interventions were first established by members of the American Psychological Association Division 12 task force (Chambless & Ollendick, 2001). These criteria include definitions of “well established”, “probably efficacious”, and “promising” interventions, which function as guidelines for determining and choosing the best available ESTs (Chambless, Baker, Baucom, Beutler, Calhoun, Crits-Christoph, et al. 1998; Chambless & Hollon, 1998; Powers, 1999).

Behavior-based treatments are considered best practice in addressing sleep problems in typically developing children due to significant improvements experienced by 80% of children treated (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). At this time, research indicates that behavioral interventions for the treatment of sleep problems in children with ASD have yet to be classified as well established or probably efficacious.
(Schreck, 2001; Vriend, Corkum, Moon, & Smith, 2011). This may be due to insufficient methodological rigor, lack of significant or equivalent outcomes, and/or the absence of duplicated studies by more than one investigator or research group that is needed to meet classification requirements.

Behavioral interventions addressing sleep problems in children with ASD include bedtime routine (Piazza, Fisher, & Sherer, 1997; Weiskop, Richdale, & Matthews, 2005); extinction (Durand, Gernert-Dott, & Mapstone, 1996; Weiskop, Matthews, & Richdale, 2001; Wolf, Risley, & Mees, 1964); scheduled awakenings (Durand, 2002); sleep restriction (Christodulu & Durand, 2004; Durand & Christodulu, 2004); stimulus fading (Christodulu & Durand, 2004; Howlin, 1984); chronotherapy (Piazza, Hagopian, Hughes, & Fisher, 1998); parent education (Malow, et al., 2014; Moon, Corkum, & Smith, 2010; Weiskop, Matthews, & Richdale, 2001); bedtime fading with (Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997) and without response cost (DeLeon, Fisher, & Marhefka, 2004); as well as multi-component treatment packages (Jin, Hanley, & Beaulieu, 2013; Knight & Johnson, 2014; Montgomery, Stores, & Wiggs, 2004; Reed, et al., 2009). Among behavioral interventions, most tend to address one specific sleep-related behavior at a time (Piazza & Fisher, 1991b), such as night terrors (Durand, 2002); disrupted sleep-wake cycle (Piazza, Hagopian, Hughes, & Fisher, 1998), and excessive daytime sleepiness (Friedman & Luiselli, 2008). In reality, parents of children with ASD tend to report a multitude of co-occurring sleep problems (e.g., bedtime resistance, delayed sleep onset, frequent night waking, and undesired co-sleeping) (Christodulu & Durand, 2004; Durand & Christodulu, 2004; Durand, Gernert-Dott, & Mapstone, 1996; Reed, et. al., 2009; Weiskop, Matthews, & Richdale, 2001).
Parents of children with ASD report that some sleep disturbances evolve from their own attempts to address other long-standing sleep problems, such as engaging in undesired co-sleeping to reduce night awakenings (Reed, et al., 2009). Undesired co-sleeping can be a part of the initial bedtime routine (Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005) or a response to difficulty falling back asleep and disruptive behaviors (i.e., tantrums, refusal to sleep in own bed, or escaping from the bed and bedroom) following night awakenings (Christodulu & Durand, 2004; Durand & Christodulu, 2004; Durand, Gernert-Dott, & Mapstone, 1996; Howlin, 1984). Therefore, it is not surprising that undesired co-sleeping experienced by children with ASD often coincides with a wide variety of sleep problems, including frequent night awakenings, bedtime disturbances, difficulty falling asleep and reinitiating sleep independently, decreased duration of sleep, early morning awakenings, and refusal to sleep in one’s own bed (Christodulu & Durand, 2004; Durand & Christodulu, 2004; Durand, Gernert-Dott, & Mapstone, 1996; Howlin, 1984; Reed, et. al., 2009; Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005).

Bedtime Fading with Response Cost (BFRC) is a promising behavior-based intervention that targets a multitude of sleep problems concurrently (Piazza, Fisher, & Sherer, 1997). Specifically, BFRC has been demonstrated to decrease disruptive sleep-related behaviors, reduce sleep latency, reduce night awakenings, and increase sleep duration in children with ASD (DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Moser, 1991; Piazza, Fisher, & Sherer, 1997). BFRC has also been effective in reducing (Piazza & Fisher, 1991b; Piazza, Fisher, & Moser, 1991) and even eliminating (Ashbaugh & Peck, 1998) undesired co-sleeping in children without
ASD. Thus far, undesired co-sleeping has not been the primary sleep variable in studies evaluating BFRC with children on the autism spectrum. To date, undesired co-sleeping has been used as the primary dependent variable in only two studies (Weiskopf, Matthews, & Richdale, 2001; Weiskopf, Richdale, & Matthews, 2005) in children with ASD. Both studies focused on parent-defined sleep disturbances and treatment goals.

**Original BFRC Protocol**

BFRC involves temporarily delaying the child’s typical bedtime, establishing a set wake-time (regardless of the amount of sleep acquired the night before), and eliminating access to daytime sleep (for non-napping children ages 4 and older) (Piazza & Fisher, 1991b). Developmental sleep norms in conjunction with parent sleep goals are used to set the appropriate bedtime and wake-time based on the amount of sleep required for each child. Parents are informed of the average hours of sleep per night needed by their child according to his or her chronological age and asked to indicate their preferred time of sleep onset, awakening, and nap (if applicable). The original BFRC protocol began with collecting baseline data on whether the child was asleep or awake every 30 minutes, throughout a 24-hour period, until data were determined stable. Once baseline data are stable, the child’s mean sleep onset time was calculated and the child’s bedtime was temporarily delayed by adding 30 minutes to the average time the child typically fell asleep (i.e., faded bedtime) (Piazza & Fisher, 1991b). For example, if the child’s average time of sleep onset was 10:45 p.m. during baseline, the initial bedtime was set to 11:15pm.

The caregivers placed the child in bed at the scheduled bedtime and observed the child 15 minutes later to determine whether the child was asleep or awake. A caregiver
stood within one foot of the child and whispered his or her name. If the child did not respond to his or her name (i.e., the absence of motor or verbal response), then the child was considered to be asleep, and the caregiver recorded the time of sleep onset. If the child was not asleep, he or she was removed from his or her bed, and restrict access to the bed, bedroom, and sleep for 45 minutes (i.e., response cost). During that time, the child was allowed to partake in regular nighttime activities (e.g., play with toys, watch TV) not related to sleep. At the end of the 45 minutes the child was returned to bed. If the child did not fall asleep within 15 minutes of being returned to bed, the caregivers repeated the response cost procedure by removing the child from bed for 45 minutes and allowed the child to engage in non-sleep related activities. This procedure was repeated until the child fell asleep within 15 minutes of being placed in bed (Piazza & Fisher, 1991b).

Each subsequent night the child’s bedtime was adjusted by 30 minutes based on the time of sleep onset during the previous night. For example, if bedtime was 10:30 p.m., and the child did not fall asleep until 12:00 a.m., the following night the bedtime would be 11:00 p.m.; similarly, if the child fell asleep at 10:30 p.m., the following night the bedtime would be adjusted to 10:00 p.m. For instances of nighttime awakenings, caregivers were encouraged to prompt the child to return to his or her bed every 30 minutes. Caregivers allowed the child to engage in regular nighttime activities. The predetermined wake-time was strongly enforced by the caregivers every day, regardless of the amount and quality of the child’s sleep during the previous night. Caregivers eliminated access to daytime sleep, unless naps were appropriate for the age and developmental needs of the child (Piazza & Fisher, 1991b).

BFRC Procedural Variants
Modifications to the original BFRC protocol have been reported in subsequent studies (Ashbaugh & Peck, 1998; DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997). Variations include the amount of time the bedtime is faded, the duration of time allowed to achieve sleep before response cost is implemented, the duration of the response cost procedure itself, and the duration of faded bedtime. The amount of time added to the faded bedtime and the duration of response cost, i.e., time spent out of bed when not achieving sleep onset, have been evaluated using both 30- and 60-minute intervals. The amount of time allotted between placing a child in bed and checking sleep onset has included both 15- and 20-minute intervals. Fading the bedtime once sleep onset is achieved has been evaluated with both 15- and 30-minute intervals and after either one or two consecutive nights. In addition, the original authors of BFRC suggest fading the bedtime by 15 minutes, rather than 30 minutes, if the child’s bedtime fading plateaus (does not progress) before reaching the bedtime goal (Piazza & Fisher, 1991b). With and without modifications to the original protocol, BFRC has been shown to be effective in addressing various co-occurring sleep disturbances with a typically developing child (Ashbaugh & Peck, 1998), children with intellectual disability (ID) (Piazza & Fisher, 1991b), children with autism (DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997) and related disorders (Piazza, Fisher, & Moser, 1991).

BFRC and Children with Non-ASD Neurodevelopmental Disorders

The bedtime fading procedure without the response cost component was first developed by Piazza and Fisher (1991a) to increase the duration of appropriate nighttime sleep of two children with severe sleep problems. Treatment included utilizing baseline
data of each participant’s average time (i.e., within 15 minutes) of sleep onset that was most likely to occur. Faded bedtime was then determined by calculating the baseline average for sleep onset and adding one half hour. For example, if the average sleep onset time is 10:00 p.m. during baseline, then the initial bedtime during the treatment phase is 10:30 p.m. Treatment required the child to remain awake until the faded bedtime and access to the bed was not permitted until the designated bedtime. Additionally, a predetermined wake time was established and the participant was not allowed to sleep past this time (Piazza & Fisher, 1991a). Based on the previous night’s time of sleep onset, the bedtime was adjusted accordingly, such that if the child fell asleep within 15 minutes of the designated bedtime, the bedtime was adjusted 15 minutes earlier. Correspondingly, if the child did not fall asleep within the allotted 15 minutes, the bedtime was pushed later an additional 15 minutes. Bedtime adjustment required two consecutive nights of the same established sleep onset time before fading the bedtime on the third night.

Two participants, a 6-year-old female diagnosed with attention deficit-hyperactivity disorder (ADHD) and a 4-year-old female diagnosed with profound ID and comorbid tuberous sclerosis were included in the study (Piazza & Fisher, 1991a). Both participants experienced multiple sleep problems, including bedtime refusal, bedtime disruption, and a lack of independent sleep initiation. The 6-year-old participant experienced frequent night awakenings and obtained only four to six hours of sleep per night. Treatment was initiated while the participant lived at home with her grandmother. The 4-year-old participant experienced excessive daytime sleepiness and fell asleep multiple times during the day. Treatment was conducted while the participant resided at an inpatient facility for children with developmental disabilities. A momentary time
sampling procedure (every 30 minutes during a 24-hour cycle) was utilized to record whether the child was in or out of bed and awake or asleep. Trained observers collected data on the participant in the inpatient unit. The grandmother of the in-home participant collected data until she went to bed, with the addition of two scheduled checks at 2:00 a.m. and 4:00 a.m. (Piazza & Fisher, 1991a).

Results indicate that bedtime fading procedure increased the percentage of appropriate sleep (i.e., hours of sleep occurring within the determined timeframe) and increased the duration of sleep for both participants. In addition, bedtime fading eliminated the problem sleep behaviors of night awakening and bedtime refusal for the 6-year-old participant. The authors suggested that bedtime fading is potentially useful for combating multiple sleep problem concurrently as it may help to institute a consistent sleep cycle and establish the bed as a conditioned reinforcer for sleep (Piazza & Fisher, 1991a). One limitation of the study was that the authors did not use an objective measure (e.g., actigraphy) of the participants’ target behaviors. Skill acquisition and the elimination of problem sleep behaviors were based on observers’ and caregiver reports.

The same authors were the first to evaluate bedtime fading with the added component of response cost (BFRC) (Piazza & Fisher, 1991b). The additional treatment component included removing the child from bed and keeping him or her awake for one hour if sleep was not initiated within 15 minutes of the initial bedtime. During the hour of response cost, the child was permitted access to the same items and attention as found during baseline. At the end of the one-hour interval, the child was returned to bed and allowed 15 minutes to fall asleep. If the child did not fall asleep within the allotted amount of time, he or she was removed from the bed again (i.e., response cost) and
required to remain awake for an additional hour. The outlined procedure was repeated until sleep onset was achieved within 15 minutes of being placed in bed. The investigators hypothesized that the response cost component would “enhance treatment efficacy” by allowing for “a more rapid fading of the bedtime”, thus increasing the reinforcing value of sleep (Piazza & Fisher, 1991b).

Participants were four nonverbal children diagnosed with profound ID and living in an inpatient unit for the treatment of ID and self-injurious behaviors. All participants experienced co-occurring sleep problems such as delayed sleep onset, inconsistent sleep duration, night awakenings, early morning awakenings, disruptive bedtime behaviors, and excessive daytime sleep, whereas one participant experienced undesired co-sleeping. Similarly to the first bedtime fading study (Piazza & Fisher, 1991a), the investigators employed a momentary time-sampling (e.g., each half hour) method conducted by trained observers to record whether or not the participants were in or out of bed and awake or asleep. The effectiveness of the procedure was evaluated using a non-concurrent multiple baseline across subjects. The parameters for the bedtime fading component as found in the preliminary study were used in the same manner, with the response cost component previously described (Piazza & Fisher, 1991b).

Two different observers scored the child’s behavior (i.e., awake or asleep, in or out of bed) to assess inter-observer agreement (IOA). Results of the study indicated that all participants either improved in the percentage of intervals of appropriate sleep or decreased in the percentage of intervals of inappropriate sleep. Three out of four participants demonstrated a decrease in the frequency of night awakenings. Furthermore, disruptive bedtime behaviors decreased from 100% to 16% for one participant and
undesired co-sleeping decreased by nearly 50% for another. The authors concluded that each participant benefited from the BFRC procedure and that both classical and operant conditioning may have facilitated treatment efficacy (Piazza & Fisher, 1991b).

A third study, coauthored by Piazza, Fisher, & Moser (1991), sought to evaluate whether BFRC would be effective in improving the aberrant sleep patterns of three girls with Rett Syndrome (RS). Standard BFRC procedures were used for all three participants who were participating in inpatient \(N=2\) or outpatient \(N=1\) services for the treatment of behavior problems. Sleep problems included night awakenings, unwanted co-sleeping, delayed sleep onset, and excessive daytime sleep. Data recording methods and IOA employed by the trained observers and parent participants remained consistent with methods outlined in previous studies (Piazza & Fisher, 1991a; Piazza & Fisher, 1991b).

Results demonstrated an increase in the percentage of appropriate sleep for all participants, although clinical significance was not determined (Piazza, Fisher, & Moser, 1991). In addition, time of sleep onset decreased for one participant, and the duration of night awakenings, as well as the percentage of daytime sleep, decreased for two participants. Outcomes for undesired co-sleeping were not directly reported, although the authors did indicate that the parents of one participant were engaging in co-sleeping less frequently and spending less time soothing their child back to sleep. Similarly to the previous bedtime fading and BFRC studies (Piazza & Fisher, 1991a; Piazza & Fisher, 1991b), no information was provided on how the parent participants were trained to implement the BFRC procedure or collect data on the observable sleep variables (Piazza, Fisher, & Moser, 1991). In addition, sleep variables were not measured objectively.

**BFRC and Typically Developing Children**
To date, only one study replicated the BFRC procedures with a typically developing child. Ashbaugh and Peck (1998) employed an ABAB experimental design with a typically developing 2-year-old female experiencing frequent night awakenings, disrupted sleep, and undesired co-sleeping. The participant lived at home with her biological father and mother, the latter being a member of the investigative team. Ashbaugh and Peck (1998) followed the BFRC protocol and data collection procedures as presented in the Piazza and Fisher (1991b) study, with two minor modifications: 1) The procedure included returning the child to her own bed if she awoke during the night, as requested by her parents; and 2) The response cost component was reduced from 1 hour to 30 minutes. Bedtime for the subsequent night was adjusted earlier by 30 minutes if the child was able to fall asleep within the allotted 15 minutes or later by 30 minutes if the child was not able to fall asleep within 15 minutes of bedtime (Ashbaugh & Peck, 1998). Parent and teacher observations of napping behavior were conducted and compared for IOA. Results of the study indicated that the participant was able to establish a more regular sleep schedule and undesirable co-sleeping was eliminated. The authors noted that slight modifications to the BFRC might not diminish the effectiveness of the intervention (Ashbaugh & Peck, 1998).

**BFRC and Children with ASD**

Three studies have evaluated the effects of BFRC in children with autism and sleep disturbance (DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997). All three studies concluded that faded bedtime, with or without response cost, was successful in improving sleep problems in children with autism. Piazza, Fisher, and Sherer (1997) compared the relative effectiveness of BFRC to
simple bedtime scheduling, which involved establishing a set bedtime and wake time (naps if age appropriate) and permitting sleep only during designated times. Bedtime scheduling also included implementing a consistent bedtime routine and placing the child in bed at the designated times (Piazza, Fisher, & Sherer, 1997). Five of the fourteen participants, ages 5 to 8 years, were diagnosed with ASD. Participants were randomly assigned to either the BFRC or the bedtime scheduling group. Treatment took place in an institutionalized setting as all 14 participants were being treated for severe behaviors in an inpatient unit (Piazza, Fisher, & Sherer, 1997).

Sleep problems experienced by the sample population included early awakenings, difficulties initiating sleep independently, delayed sleep onset, and nighttime awakenings. BFRC procedures, data collections methods, and IOA techniques were adopted from previously described studies (Piazza & Fisher, 1991b; Piazza, Fisher, & Moser, 1991). Result indicated that for all participants the BFRC intervention, compared with bedtime scheduling, produced a greater reduction of sleep problems (Piazza, Fisher, & Sherer, 1997). Of the two children with ASD in the bedtime scheduling group, one showed slight improvement with night waking and falling asleep, while the other child’s sleep problems (i.e., night awakening and early awakening) maintained at baseline levels. All three of the participants with ASD in the BFRC group demonstrated improvements with the near elimination of early awakenings and a decrease in the duration and frequency of night awakenings. Outcomes were maintained at a 4-month follow-up. Strength of this study was the inclusion of a comparison group to demonstrate the effectiveness of BFRC.

A subsequent study evaluated the effectiveness of the faded bedtime component without response cost in solving sleep problems and preventing self-injurious behaviors
(SIBs) in a 4-year-old male with ASD (DeLeon, Fisher, & Marhefka, 2004). The participant awoke frequently throughout the night and engaged in head banging, flopping on the floor, and body hitting, until receiving contingent reinforcement of attention in the form of hugs and comfort. Treatment was conducted in an inpatient unit for the treatment of severe SIBs. BFRC treatment procedures, data collection methods, and IOA criteria were replicated from previous studies (Piazza & Fisher, 1991b; Piazza, Fisher, & Moser, 1991; Piazza, Fisher, & Sherer, 1997). The investigators employed an AB design without a reversal condition due to the severe nature of the participants’ SIBs. Results demonstrated a decrease in the total number of night awakenings, which, in turn, lead to the reduction in the frequency and rate of the wake-related SIBs displayed by the participant (DeLeon, Fisher, & Marhefka, 2004).

The most recent BFRC study examined the effectiveness of a manualized BFRC program in the reduction of problem sleep behaviors in children with ASD (Moon, Corkum, & Smith, 2010). All participants (N=3) had a diagnosis of ASD, lived at home with their biological families, and experienced delayed sleep onset and difficulties with initiating sleep. The parents of all participants were trained to provide the BFRC procedure at home. Each parent received a handbook on the treatment of sleep difficulties, including information on the principles of sleep hygiene, importance of consistent bedtime routines, and behavioral strategies for managing child’s daytime and sleep-related behaviors. Parents met with a therapist for 5 consecutive weeks to review the information provided in the handbook. In addition, at the end of each week, the therapist contacted parents by phone to review the content of the materials learned and to monitor progress (Moon, Corkum, & Smith, 2010).
The BFRC procedure, data collection method, and IOA criteria remained the same as described in preceding studies (Piazza & Fisher, 1991b; Piazza, Fisher, & Moser, 1991; Piazza, Fisher, & Sherer, 1997) with minor exceptions: The duration of time allowed to attain sleep onset was increased from 15 minutes to 20 minutes (Moon, Corkum, & Smith, 2010). Sleep diaries and actigraphy were used to record sleep outcomes. Results indicate a reduction in sleep onset latency for all three participants. Outcomes were maintained at a 12-week follow-up, and parents reported satisfaction with the intervention. Strengths of this study include objective recording of sleep variables (i.e., actigraphy), clear use of behavioral definitions needed to replicate the study, and engaging parents to implement the BFRC protocol in the home environment (Moon, Corkum, & Smith, 2010). A weakness of the study is that it is impossible to isolate BFRC as the only independent variable when the intervention was presented in combination of other treatment strategies, such as the didactic sessions and handbook on behavioral strategies. As a result, the study makes it difficult to determine if treatment outcomes are the result of the BFRC intervention itself.

**Conclusion and Future Directions**

In summary, children with ASD experience various co-occurring sleep problems and disruptive sleep-related behaviors at a greater frequency and severity than typically developing children. Consequently, in children with ASD and their families quality of life and day-to-day functioning suffer. Parents of children with ASD who also experience sleep disturbances are especially at risk for mood disorders and marital dissatisfaction. Effective behavioral treatments that parents can reliably implement at home are limited.
BFRC is a promising intervention that successfully addresses sleep problems and sleep-related problem behaviors in children with and without ASD.

At this time, BFRC has been restricted to controlled studies by several groups of investigators implemented by trained professionals in institution settings. Consideration has yet to be given to how parents uniquely define sleep disturbance within the context of their own home and family. Primary sleep variables tend to be researcher defined, which may not capture the most salient aspects of a child’s sleep disturbances from the parent’s perspective. For example, undesired co-sleeping is a commonly occurring sleep disturbance reported by parents of children with ASD. However, parent-defined primary dependent variables have not been considered in any of the existing BFRC studies. Only one BFRC study engaged parents in implementing treatment of sleep problems in children with ASD in the home setting. However, none of these studies utilized parent-derived sleep goals to determine the onset of treatment or evaluate the effectiveness of BFRC for the reduction of sleep disturbance in children with ASD. Therefore, the primary purpose of this study was to evaluate the effectiveness of BFRC as a parent-implemented home-based treatment for parent-defined sleep disturbances and related sleep problems experienced by their child with ASD.

Method

Participants

Children between ages 4 and 8 years with a clinical diagnosis of autism and sleep disturbance were recruited through a day treatment program for children with ASDs and an outpatient behavioral health clinic located in the Midwest. Participant recruitment continued until a sufficient number of eligible participants (N=3) needed for a single
subject research design was recruited. Participant selection criteria were adapted from other studies in the area of behavioral treatment of sleep disturbance in children (Mindell & Durand, 1993; Weiskop, Richdale, & Matthews, 2005). Recruited participants had a confirmed diagnosis of autism based on the *Autism Diagnostic Observation Schedule, Second Edition (ADOS-2)* (Lord, et al. 2012) and the *Childhood Autism Rating Scale, Second Edition (CARS-2)* (Schopler, Van Bourgondien, Wellman, & Love, 2010), which is standard practice in diagnostics. For the purposes of this study, individuals with a classification of *Autism* or *Autism Spectrum* on the ADOS-2, in combination with a classification of either *Severe* or *Mild-to-Moderate* on the CARS-2, met the criteria for a diagnosis of autism and were deemed eligible for participation in the study. Individuals classified as *Non-Spectrum* on the ADOS-2 and/or *Minimal-to-No-Symptoms* on the CARS-2 were excluded from the study. Eligible participants were required to be living at home with at least one parent at the time of the study and present with a minimum of four-week history of childhood sleep disturbances as denoted by

- resistance to going to bed, defined as one or more verbal protests and/or physical protests a night, for a minimum of three nights per week; or
- delayed sleep onset, defined as more than 20 minutes between bedtime and falling asleep, for a minimum of three nights per week; or
- any of the following situations at bedtime for a minimum of three nights per week:
  - undesired co-sleeping with a parent or sibling, being rocked or held to fall asleep, falling asleep in a location other than his/her own bed; or
  - falling asleep with a bottle or pacifier; or
• spontaneous awakening that requires parental intervention to return to sleep at least once per night, at least three times per week.

Children using pharmacological agents to regulate sleep and/or other behaviors were not necessarily excluded. Unless medically necessary, parents were asked to attempt to maintain their child’s medication regimen throughout the duration of the study. Excluded from the study were families who had received professionally derived information directly from a psychologist, therapist, and/or physician and/or used a behavior-based intervention to address their child’s sleep problems within the past 6 months (Moon, Corkum, & Smith, 2010).

Three potential participants were screened and met criteria for participation. All three parents-child dyads enrolled and completed the study. All three children had been diagnosed with ASD prior to enrolling in the study. Each participant met the classification of Autism on the ADOS-2 and the Severe Symptoms of Autism Spectrum Disorder group on the CARS-2 measures. All names were changed to protect participants’ right to confidentiality. Participant 1, Bill, was a 7-year-old Caucasian male attending the 1st grade. Bill’s parents reported that he exhibited bedtime resistance, refused to sleep in his own bed, and took longer than 20 minutes to fall asleep each night. He woke repeatedly throughout the night and each time sought parental presence to reinitiate sleep by crawling into his parents’ bed. His parents often engaged in undesired co-sleeping with Bill in their bed, as they were asleep when he crawled into their bed and remained asleep with Bill in bed. Participant 2, Holly, was an 8-year-old Caucasian female attending the 3rd grade. Her parents reported that she refused to sleep in her own bed and never fell asleep without parental presence. A parent had to remain next to Holly
each night in their bed and engaged in undesired co-sleeping to get her to fall asleep. Participant 3, Keith, was a 4-year-old Caucasian male attending preschool. His parents reported that he resisted bedtime, had difficulty falling asleep, and experienced frequent awakenings throughout the night. Keith required parental presence to fall asleep at bedtime and following any night awakening, which resulted in undesired co-sleeping in either his bed or his parents’ bed. His parents reported that Keith sometimes stayed awake throughout the entire night.

All children were living with their biological parents and siblings. Each parent reported participating in implementing the BFRC protocol, whereas one designated parent from each dyad was responsible for collecting data and taking telephone calls from a member of the investigative team. During the study, all children continued their previously prescribed regimen of medication (Bill: dexamethasone [Focalin], guanfacine, and melatonin; Holly: risperidone and melatonin; Keith: cetirizine [Zyrtec]).

Table 1 provides a summary of participant characteristics including chronological age, gender, ADOS-2 scores and classification, CARS-2 scores and severity group, and sleep problems meeting selection criteria.

**Materials**

A trained clinician administered the *ADOS-2* and *CARS-2* to evaluate each child’s verbal, social, and cognitive abilities (Gotham, Risi, Pickles, & Lord, 2007). The *ADOS-2* is a semi-structured, standardized assessment for children ages 12 months through adulthood that allowed the examiner to observe and rate behaviors related to communication and social interaction and play skills identified as important to the diagnosis of ASD. Scores based on the examiner’s observations were calculated through
the use of a diagnostic algorithm to yield one of three classifications (i.e., *Autism, Autism Spectrum*, and *Non-spectrum*) (Lord, et al., 2012). The use of the ADOS-2 alone does not provide justification for a diagnosis of ASD; therefore, the CARS-2 was used as a screening tool to compliment ADOS-2 outcomes.

The CARS-2 is a valid and reliable clinician- and parent-completed rating scale for children ages 2-13 years (Schopler, Van Bourgondien, Wellman, & Love, 2010). The CARS-2 is comprised of descriptors of childhood social, developmental, and motor abilities related to ASD symptoms. A total score classifies children into one of three severity groups of ASD symptoms (i.e., *Severe, Mild-to-Moderate*, and *Minimal-to-No-Symptoms*).

Dependent measures of sleep included the following: (a) *Sleep Diary*; (b) *Actigraphy*; (c) *Behavior Diary*; and (d) the *Children’s Sleep Habit Questionnaire* (CSHQ). All measures were collected during baseline (4 to 14 days), end of treatment (11 to 14 days), and at one month post-treatment (4 to 7 days). A *Sleep Diary* is a standardized parent-completed measure in the field of behavioral interventions and sleep medicine with a reasonable validity and high internal consistency (Corkum, Tannock, Moldofsky, Hogg-Johnson, & Humphries, 2001; Mindell & Durand, 1993).

A traditional sleep diary, based on considerations from the literature (Patzold, Richdale, & Tonge, 1998; Weiskop, Matthews, & Richdale, 2001), was developed for this study. One parent per family recorded nightly information regarding their child’s sleep in the sleep diary throughout the duration of the study (see Appendix A). Outcomes recorded in the diary specific to sleep included (a) frequency and duration of night awakenings after sleep onset (FNA; WASO); (b) sleep onset latency (SOL); (c) total
sleep time (TST); (d) time in bed (TIB); and (e) independent versus dependent sleep initiation (ISO; DSO). Sleep efficiency (SE) outcomes were extrapolated from the sleep diary data \([\text{TST/TIB} \times 100]\) and expressed as a percentage.

A slightly modified sleep-related behavior diary (Allen, Kuhn, DeHaai, & Wallace, 2013; Burke, Kuhn, & Peterson, 2004) was added to this study. The same parent recorded nightly information regarding their child’s sleep-related behaviors in the behavior diary throughout the duration of the study (see Appendix B). Diary outcomes specific to sleep-related behavior included (a) frequency and duration of undesired co-sleeping (UC-S) with a parent or a sibling; (b) frequency and duration of bedtime resistance (BR; DBR); (c) frequency of sleeping in a non-designated area; and (d) the location of non-designated sleep (refer to Table 2 for a list of all dependent variables on the sleep and sleep related behavior diaries). Bedtime resistance (BR) frequencies were the sum total of the number of disruptive behaviors that occurred during the bedtime routine once the child was placed in bed and the number of times the child got out of the bedroom after being put in bed for the night. Disruptive Behavior Composite (DBC) mean frequency was determined by dividing the sum total of BR per data collection phase by the number of days during that period. Duration of bedtime resistance (DBR) outcomes were obtained by calculating the duration of disruptive behaviors during bedtime routine and once the child was placed in bed with the duration of time the child was out of the bedroom after being put in bed for the night.

*Treatment integrity* is a measure of the extent to which treatment procedures are implemented as intended (Vollmer & Sloman, 2008). An integrity-monitoring sheet on the BFRC protocol (see Appendix C) was created for this study based on clinical
considerations and the literature (Burke, Kuhn, & Peterson, 2004; Durand & Mindell, 199b; Vollmer & Sloman, 2008). Telephone calls were made to the designated parent responsible for data collection at least three times a week during the treatment phase (Burke, Kuhn, & Peterson, 2004; Durand & Mindell, 1999b). On randomly selected nights, parents answered a series of questions related to components of the BFRC procedure they implemented. Parent responses were recorded on the integrity-monitoring sheet, and a random sample of data collected, ranging from 43% to 71% of participant treatment data, was scored for fidelity. Treatment integrity outcomes were calculated by dividing the sum of correct parent responses by the total number of possible correct responses and multiplying the result by 100 (Vollmer & Sloman, 2008). The bedtime fading components were implemented with 96% integrity for Holly, 97% integrity for Bill, and 100% integrity for Keith. The response cost components were implemented with 100% integrity for all three participants.

To assess the reliability of data collection, periodic telephone checks were used to compare the level of agreement between parent-reported data and actual recorded data for the previous one to three nights during the treatment phase (Burke, Kuhn, & Peterson, 2004; Durand & Mindell, 1999b). The researcher recorded parent verbal reports during each telephone check using a blank sleep diary and sleep-related behavior diary (see Appendix A and B). Random samplings of these telephone checks (i.e., 43% of data for Keith, 50% for Bill, 71% for Holly) were later matched and compared with the data forms returned by parents. Data points were considered reliable if the difference between the reported and recorded durations varied by no more than 15 minutes and observed frequencies varied by no more than one point between data sets (Burke, Kuhn, Peterson,
Reliability was calculated by dividing the sum of agreements by the sum total of agreements plus disagreements and multiplying the result by 100 \([\frac{\sum \text{of agreements}}{\sum \text{of agreements} + \sum \text{of disagreements}} \times 100]\). The result was expressed as a percentage (Durand & Mindell, 1999b). Level of agreement between the telephone checks and parent-recorded sleep diaries was 95% for Holly, and 97% for Bill and Keith each. The level of agreement between parent-recorded sleep-related behaviors was 94% for Holly, 98% for Keith, and 100% for Bill.

*Actigraphy* is an objective measure of motor activity, which, in turn, provides reliable estimates of sleep-wake patterns in children (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). Actigraphy is considered a relatively unobtrusive method for recording sleep patterns in children. Children with autism and/or sensory issues may not tolerate wrist placement of the actigraph device; therefore, ankle placement is recommended (Ancoli-Israel, et al. 2015). The MicroMini Motionlogger actigraph (Ambulatory Monitoring, Inc.), a small sensor about the size of a wristwatch, was secured with a hospital band to each child’s ankle to continuously record movement. Parents were asked to have their child constantly wear the small actigraph on the same ankle, except during bathing or swimming. Zero Crossing Mode (ZCM) was used to count frequencies of signal-based movement for each epoch (e.g., 30 seconds or one minute). Activity data collected were downloaded from the sensor using Act Millennium (version 3.68.0.1) software. Data were scored using Action W (version 2) software and the Sadeh scoring algorithm (Sadeh, Sharkey, & Carskadon, 1994) to make inferences about the child’s sleep-wake cycles. Primary outcome variables included SOL, TST, SE, WASO, and TIB.
The *Children’s Sleep Habits Questionnaire (CSHQ)* (Owens, Spirito, & McGuinn, 2000) is a widely used multidimensional measure to determine the frequency and topography of sleep behaviors for school-aged children between 4-10 years. The parent-completed questionnaire yields reliable psychometric properties with typically and non-typically developing children (Lewandowski, Toliver-Sokol, & Palermo, 2011). CSHQ abbreviated version (CSHQ-A) is composed of 33 items regarding bedtime, sleep behaviors, waking during the night, and morning waking/daytime sleepiness (Owens, Spirito, & McGuinn, 2000). Parents retrospectively reported their child’s sleep for the past week using a Likert scale indicating if the problem occurred “usually” (five to seven nights per week), “sometimes” (two to four nights per week), or “rarely” (zero to one night per week). Parents also indicated if the sleep habit was a problem by circling “Yes”, “No”, or “Not applicable (N/A)”. Items were scored within 8 subscales (i.e., bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night awakenings, parasomnias, sleep disordered breathing, and daytime sleepiness). Total sum of each subscale yielded a total sleep disturbance score (range of 33 to 87), with a proposed cut-off score of 41; higher scores indicate more sleep problems.

Three measures were selected to capture the impact of and satisfaction with treatment for the family. The *Goal Achievement Scale (GAS)* quantifies progress on parent-identified treatment goals and assesses the clinical significance of change in sleep behavior (Hudson, Wilken, Jauering, & Raddler, 1995). For each identified treatment goal (i.e., sleep related behavior), 0% success (i.e., no improvement) was the set baseline rate. Before treatment, parents (with guidance from a member of the research team on developmentally appropriate behaviors) decided what would constitute 100% (total)
success for each goal. Success was dependent on the parents’ level of desired improvement, not on the total elimination of a problem behavior. Change in each goal was expressed as a percentage of success over baseline. For example, parents reported wanting to reduce the frequency of co-sleeping. Parents’ goals represented a personally acceptable level of co-sleeping (e.g., 2 out of 7 nights per week). Baseline rate of undesired co-sleeping (e.g., occurring 7 out of 7 nights per week) was set at 0% success, whereas the goal for 100% total success depended on parents’ preference (e.g., eliminating all but 2 of the 7 nights of co-sleeping per week).

The Parent’s Customer Satisfaction Questionnaire (PCSQ) is a 42-item questionnaire to evaluate treatment satisfaction (McMahon & Forehand, 2003). For the purposes of this study, only 16 questions specific to treatment satisfaction were used; 11 items corresponded to a Likert scale (e.g., 1 (considerably worse) to 7 (greatly improved)). Wording of each response option varied according to the question being asked (e.g., 1 = Very dissatisfied to 7 = Very satisfied; 1 = Strongly not recommended to 7 = Strongly recommended; 1 = Very inappropriate to 7 = Very appropriate; 1 = Very pessimistic to 7 = Very optimistic). Parents evaluated the overall treatment protocol by rating the degree to which they endorsed each statement and answered 5 open-ended questions related to usefulness of the treatment.

The Kansas Marital Satisfaction Scale (KMSS) is a brief 3-item measure designed to rapidly assess relationship satisfaction of married individuals or couples (Crane, Middleton, & Bean, 2000). The KMSS is described as a “reliable and economical” tool for measuring satisfaction with marriage, partners, and marital relationships. Parents separately completed the KMSS by reading each item and marking on the corresponding
scale of 1 (extremely dissatisfied) to 7 (extremely satisfied) the degree to which they agreed with each statement (Schumm, Nichols, Schectman, & Grigsby, 1983). A possible score for each item ranges from 1 to 7; a cut-off score of 17 distinguishes between distressed and non-distressed couples, with a total individual score of 16 or lower indicating marital distress (Crane, Middleton, & Bean, 2000).

**Procedure**

*Initial Interview*

Parents of prospective participants completed a 15-minute telephone screening followed by a face-to-face interview to determine eligibility. During this interview, parents and children met with a member of the investigated team to complete the CARS-2, baseline CSHQ-A, and a demographic questionnaire. A classification of either *Severe* or *Mild-to-Moderate* ASD resulted in the administration of the ADOS-2. Depending on parents’ availability, the ADOS-2 was completed either during the initial session or scheduled for another time that week. Parents were invited to participate in the study once a diagnosis of autism was confirmed and all other eligibility criteria were met. The consent form was reviewed and parents completed the KMSS.

*Pre-Baseline*

Prior to baseline data collection, parents meet with a member of the investigative team for a second session to identify their specific treatment goals. Parents were instructed in the recording of data in the sleep and behavior diaries. They received feedback from a trained clinician as they completed sample diaries for two vignettes describing detailed sleep patterns and sleep-related behaviors (see Appendix D). Once parent dyads correctly completed the sample diaries, they were provided blank diaries.
and an actigraph to begin collection of data on their child’s sleep patterns and sleep-related behaviors (Christodulu & Durand, 2004; Reed et al, 2009).

**Baseline**

Parents met with a member of the investigative team once a week during the baseline phase of the study for approximately 30 minutes to review the sleep and behavior diaries and exchange actigraphs. Once baseline data were determined stable, or 14 consecutive days of data was collected, the treatment phase was introduced, and parents were taught to implement the BFRC protocol. Parents were instructed in the implementation of a modified BFRC protocol (i.e., bedtime procedures, response cost, and bedtime fading) and received an outline of procedures (see Appendix E).

Bedtime fading instructions included:

(i) Adjust bedtime for the following night according to child’s previous time of sleep onset by 15 minutes. For example, if child’s initial bedtime was 10:30 p.m., and he/she fell asleep within 15 minutes of being placed in bed, fade the bedtime to 10:15 p.m.; and

(ii) If child did not fall asleep within 15 minutes of the initial bedtime, push back the bedtime to 10:45 p.m.

Bedtime procedures included:

(i) Follow regular bedtime routine and place child in his/her bed at the initial adjusted bedtime;

(ii) After 15 minutes, check to see if child is awake by standing within one foot of the bed and whispering his/her name. If there are no observable motor or
verbal responses (i.e., opening eyes, talking, etc.) it is safe to assume child is asleep; and

(iii) If determined that child is awake (i.e., he/she responded to whisper), implement response cost.

Response cost procedures included:

(i) Remove child from the bed and bedroom and allow him/her to partake in activities not related to sleep for 30 minutes, such as playing with toys or watching TV; and

(ii) At the end of 30 minutes, return child to his/her bed;

(iii) Repeat the procedure if your child does not fall asleep within 15 minutes of being returned to bed; and

(iv) Address nighttime awakenings by returning child to his/her bed with minimal parental attention and interaction.

Parents received feedback from a trained clinician as they verbally described how to implement BFRC with their child. Once parent dyads correctly reported how to implement each component of BFRC, they were provided blank diaries and an actigraph to begin treatment phase data collection. Finally, each parent completed the KMSS independently, once weekly for the duration of the study.

Treatment

A member of the investigative team assisted parents in determining parent-preferred bedtime goals. A consistent wake time and naptime (if applicable) were derived using developmental norms (Iglowstein, Jenni, Molinari, & Largo, 2003) and parent sleep goals. Baseline data were used to determine the initial bedtime by calculating the average
sleep onset time and adding 30 minutes. Parents were asked to enforce a consistent wake
time every morning regardless of their child’s sleep behaviors the night before. Parents
were also asked to eliminate access to non-established daytime sleep, such as naps, unless
appropriate for the age and developmental needs of their child. Parents were required to
regulate the timing and duration for any established naps, regardless of the amount or
quality of sleep the child had the night before, and to not permit access to sleep outside of
the designated times. If the child eloped from the bedroom, parents were asked to return
him or her to bed and ignore all disruptive behaviors as long as they were not harmful to
the child or others.

Parents were taught a modified BFRC protocol and received an outline of the
following procedures: (a) Bedtime protocol: (i) Follow the regular bedtime routine and
place child in his/her bed at the initial adjusted bedtime. (ii) After 15 minutes, check to
see if child is awake by standing within one foot of the bed and whispering his/her name.
If there are no observable motor or verbal responses (i.e., opening eyes, talking, etc.), it is
safe to assume the child is asleep. (iii) If the child is awake (i.e., he/she responded to your
whisper), implement response cost. (b) Response cost: (i) Remove child from the bed and
bedroom and allow him/her to partake in activities not related to sleep for 30 minutes,
such as playing with toys or watching TV. (ii) At the end of 30 minutes, return child to
his/her bed. (c) Repeat the procedure if child does not fall asleep within 15 minutes of
being returned to bed. (d) Address nighttime awakenings by returning child to his/her bed
with minimal parental attention and interaction. (e) Bedtime fading: (i) Adjust the
bedtime for the following night according to child’s previous time of sleep onset by 15
minutes. For example, if child’s initial bedtime was 10:30 p.m., and he/she fell asleep
within 15 minutes of being placed in bed, fade the bedtime earlier by 15 minutes to 10:15 p.m.. If the child did not fall asleep within 15 minutes of the initial bedtime, push the bedtime later by 15 minutes to 10:45 p.m.

After being trained to implement the BFRC protocol, parents continued to meet with a member of the investigative team once a week for approximately 30 minutes to review sleep and behavior diaries and exchange actigraphs. During the treatment phase, a researcher contacted the designated parent responsible for data collection by telephone approximately every other day to check on “data collection efforts” (Durand & Mindell, 1999). Parents answered a series of questions related to the BFRC procedures and data collected. Information provided was recorded in a modified diary and compared with parent completed sleep and behavior diaries at the end of the study. Parents were asked to complete the CSHQ-A at the end of the treatment phase.

Follow-up

Approximately four weeks after the end of treatment, parents met with a member of the investigative team for approximately 30 minutes to receive data sheets and an actigraph. Parents collected sleep and behavior data for a minimum of 4 and maximum of 7 consecutive days. At the end of the follow-up data collection phase, parents attended a closing session to complete the CSHQ-A and PCSQ and return data and the actigraph. Parents attended between five and eight sessions to complete the study, depending on outcomes (see Table 3). For example, if a stable trend in the baseline and treatment data was observed, then sessions two, three, four, and five were consolidated into two sessions.

Research Design
A non-concurrent multiple baseline design across participants was used (Harvey, May, & Kennedy, 2004; Watson & Workman, 1981). Baseline lengths were staggered across participants and information was collected until data stability and trend were observed or for a maximum of 14 days if stability was not observed, followed by a maximum of 14 days for the treatment phase. It was expected that changes may occur in each participant’s sleep behavior under the treatment condition, while participant’s sleep behavior during baseline may remain unchanged. Follow-up measures were collected one month following the conclusion of treatment, for a maximum duration of one week. Visual inspection of baseline data was utilized to assess stability and trends in the data and determine the introduction of the treatment phase for each individual participant (Harvey, May, & Kennedy, 2004; Watson & Workman, 1981). For example, once the frequency of Bill’s undesired co-sleeping was stable and the trend was not decreasing (after 6 days), the treatment phase was initiated. Similarly, when the frequency of Keith’s undesired co-sleeping was not stable or the trend was decreasing, treatment was delayed until baseline data demonstrated a stable trend in the desired direction (or for a total of 14 days). This time-lagged approach demonstrates experimental control through the sequential introduction of treatment once stable baseline levels in the dependent variables were observed (Harvey, May, & Kennedy, 2004).

**Data analysis**

Means and standard deviations were calculated for WASO, SOL, TST, and SE from data recorded by actigraphy and in sleep diaries (Moon, Corkum, & Smith, 2010). Mean frequencies and standard deviations for FNA, DSO, and non-designated sleep were calculated from data recorded in the sleep and behavior diary. Means and standard
deviations for undesired co-sleeping were also calculated using data recorded in behavior and sleep diaries (Weiskop, Matthews, & Richdale, 2001). Raw scores on the CSHQ-A subscales of bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night awakenings, parasomnias, sleep disordered breathing, and daytime sleepiness were reported (see table; Moon, Corkum, & Smith, 2010). Daily means for the frequency and duration of disruptive behaviors were calculated using data from the sleep and behavior diaries (Weiskop, Richdale, & Matthews, 2005). Level of success of parents’ treatment sleep goals was calculated using the GAS at the end of baseline, intervention, and follow-up conditions and expressed as % (Weiskop, Matthews, & Richdale, 2001; Weiskop, Richdale, & Matthews, 2005). Mean level of satisfaction (%) was also calculated from KMSS raw scores for each parent during all three phases.

**Results**

Data from each child were analyzed individually because a non-concurrent multiple baseline design across participants was used in this study. Changes in trends and patterns for the primary dependent variables are provided below. Each of the three parents dyads generated sleep-related goals for their own child and worked with a member of the investigative team to refine and determine the levels of success for each goal (see Table 4).

**Undesired co-sleeping**

Undesired co-sleeping was the only sleep problem consistently reported by the parents of all three participants. Parents generated a goal using the GAS to reduce undesired co-sleeping in the parent’s bed (see GAS section for details). Frequency and
duration of undesired co-sleeping with parents reduced after the onset of BFRC for all three children as measured by the sleep-related behavior diaries (see Figures 1 and 2). For Holly, undesired co-sleeping occurred once per night during baseline, averaging 653 minutes per night. This decreased to 0 minutes during treatment and was maintained at that level at follow-up. Bill’s frequency of undesired co-sleeping was once per night throughout baseline, with a mean duration of 230 minutes per night, which also decreased to 0 minutes during treatment and was maintained at that level at follow-up. It should be noted that on the third night of treatment, Bill’s parent chose to co-sleep with him in the child’s bed for 90 minutes after an instance of bedwetting. This occasion did not meet the definition of undesired co-sleeping; therefore, the frequency and duration were not included in the final data set. Alternatively, had this one-time occurrence been included, undesired co-sleeping decreased from baseline to 0.1 occurrences and an average of 6 minutes per night at the end of treatment. Keith demonstrated a dramatic decrease from the mean frequency of 2 occurrences per night (mean duration of 57 minutes per occurrence) during baseline to 0.1 occurrences and an average of 1 minute per night during treatment. One episode of undesired co-sleeping occurred on the last night of the treatment phase in the parents’ bed for 15 minutes. Undesired co-sleeping for Keith slightly increased to 0.3 occurrences, with a mean duration of 9 minutes per night, throughout follow-up. These instances occurred on separate days in the parent’s bed, lasting less than 45 and 15 minutes each.

**Non-designated sleep**

Frequency and location of sleeping in a non-designated area were reported for Bill and Keith, whereas Holly maintained zero occurrences during all three phases of the
study. Both Bill and Keith showed reduction in the frequency of sleeping in a non-designated area. For Bill, mean frequencies of sleep in a non-designated area (location not reported) decreased from 0.16 occurrences during baseline to zero occurrences throughout treatment. For Keith, mean frequencies of sleep in a non-designated area decreased by half from baseline (0.13) (located in the car and on the couch) to treatment (0.06) (located in the car). This single episode of sleeping in a non-designated area occurred during a change in routine. Keith’s parents reported they had traveled by car out of town for the day, and the return trip coincided with Keith’s bedtime, at which time he fell asleep and remained asleep for the duration of the car ride (2.5 hours). Follow-up data demonstrated a total elimination of sleeping in a non-designated area for Bill and Keith.

Bedtime resistance

Bedtime resistance (BR) was a reported concern for Bill’s parents only, although all three children engaged in disruptive behaviors during the bedtime routine and after being placed in bed, as recorded in the sleep-related behavior diaries (see Figure 3). For Holly, mean duration of BR per night decreased from 15 minutes during baseline to 0.4 minutes at the end of treatment and further decreased to 0.3 minutes during follow-up. Night-by-night inspection of Holly’s sleep-related behavior diary indicated a one-time change in her bedtime routine (i.e., attending a party for 30 minutes past bedtime), which may have contributed to the duration of BR during follow-up. Both Keith and Bill experienced extinction bursts (increase in mean duration of BR) during the treatment phase, with a sharp decrease beyond baseline levels during follow-up. Bill’s mean duration of BR per night greatly increased from baseline (12.4 minutes per night) to
treatment (55 minutes per night) and then significantly decreased throughout follow-up (4 minutes per night). For Keith, BR mean duration per night increased from baseline (1.3 minutes per night) to treatment (6.3 minutes per night), and decreased to near 0 minutes during follow-up (0.1 minutes per night). Overall, Keith’s BR duration increased by only an average of 5 minutes during treatment and reduced by 6.2 minutes at follow-up.

Parents in this study reported no occurrences of aggressive behavior when returning the child to their own bed following elopement. Night-by-night sleep-related behavior diary for Bill indicated a spike in BR duration by 150 minutes the same day he experienced a significant deviation in routine (i.e., attended only ½ day of school and no swim class), which was preceded and succeeded by nights with BR rates of zero. Bill’s parents noted this change in schedule might have greatly contributed to his resistance to bedtime. Bill’s CSHQ-A bedtime resistance subscale scores decreased from baseline (9) to treatment (6) and were maintained during follow-up (6). Likewise, Keith’s CSHQ-A bedtime resistance subscale scores decreased from baseline (12) to treatment (7) and were maintained during follow-up (6). Holly’s CSHQ-A bedtime resistance baseline subscale score of 14 decreased to 6 during follow-up.

Disruptive behavior composite

Mean scores of Disruptive Behavior Composite (DBC) were extrapolated from the sleep-related behavior diaries and patterns similarly to the mean durations and frequencies of BR for all three children (see Figure 4). Holly’s DBC mean scores decreased slightly from the baseline (1.3 per night) to the end of treatment (1.2 per night) and displayed a downward trend. Her DBC further decreased during follow-up (0.5 per
Night-by-night inspection indicated that disruptive behaviors during her bedtime routine or after she was placed in her bed for the night were eliminated, and only the number of times she escaped from her bedroom after being put in bed for the night contributed to her DBC mean scores. Holly’s nightly DBC scores varied throughout treatment with a subtle downward trend that remained below baseline at the end of follow-up. A change in Holly’s routine may have contributed to one of two instances of BR during follow-up, thus increasing her DBC. Bill’s DBC increased slightly from baseline (5.3 per night) to treatment (5.7 per night).

Similarly to Holly’s, Bill’s night-by-night data indicated that disruptive behaviors during his bedtime routine decreased to almost none. Furthermore, Bill’s disruptive behaviors occurring after he was placed in his bed for the night decreased, while the number of times he came out of his bedroom after being placed in bed for the night increased during treatment. Visual analysis of the plotted data showed an unexpected uptick in the frequency of BR on the same day Bill experienced a significant deviation from his daily routine. Such deviation may have negatively impacted his DBC. His DBC decreased throughout follow-up to 2 per night. As for Keith, his DBC doubled from baseline (1.1 per night) at the end of treatment (2.2 per night) and decreased to near 0 minutes during follow-up (0.1 per night) with no clinically relevant differences across topography.

Night awakenings

Frequent night awakenings (FNA) were a parent-reported sleep problem for Bill and Keith. Both children showed some decrease in the number of night awakenings
during the implementation of BFRC as measured by sleep diaries (see Figure 5). Bill’s FNA slightly decreased from 1.2 occurrences per night at baseline to an average of 1 per night throughout treatment, whereas Keith’s FNA per night decreased from 0.6 per night at baseline to 0.3 per night during treatment. Night-by-night sleep diary data for Bill indicated a progressive decrease in FNA during treatment with four consecutive nights of zero night awakenings nearing the end of treatment with an increase in FNA the following night. Bill used the bathroom independently once that night, which accounts for one FNA. Bill’s parents indicated a deviation from his typical daytime routine resulting in a lack of exercise, which might have had a negative impact on his sleep and contributed to the unexpected increase of FNA. Additionally, at least one night awakening on each of the first, third, and fourth nights of the treatment phase were the result of bedwetting and required parental assistance in changing bedding and pajamas. These instances were not recorded as FNAs. Follow-up data demonstrated a total elimination of FNA for Bill and Keith.

Holly experienced an increase and fluctuation in the frequency of night awakenings from baseline (0 per night) to a mean frequency of 0.7 per night during treatment, which returned to near baseline levels during follow-up (0.5 per night). Prior to BFRC, Holly slept through the night in her parents’ bed with at least one parent in the room at all times. This standard might have been greatly reducing the likelihood of FNA. Therefore, the adjustment to sleeping alone in her own bed may have produced FNA during treatment. Visual analysis of the data showed FNA decreased to zero the last night of treatment, maintained the first three nights of follow-up, and increased by 2 instances the final night of follow-up due to a change in routine (i.e., party attendance and delayed
bedtime). Bill’s CSHQ-A *night wakings* subscale scores decreased from baseline (7) to treatment (6), and further decreased during follow-up (4), whereas Holly’s CSHQ-A bedtime resistance subscale score remained stable from baseline (5) throughout follow-up. Keith’s CSHQ-A *night wakings* subscale scores decreased from baseline (7) to treatment (4) and then slightly increased during follow-up (5).

*Dependent sleep onset*

Dependent sleep onset (DSO) was a parent-reported problem for Holly and Keith as both children required a parent in the room to fall asleep. DSO, as measured by the sleep diaries, was eliminated for all three children (see Figure 6). Holly’s DSO occurred steadily once per night during baseline, whereas Keith’s baseline DSO varied between 1 and 3 times per night with a mean frequency of 1.7 per night. Bill’s baseline DSO generally occurred once a night with a mean frequency of 1.2 occurrences per night. Each child’s DSO decreased to zero after the onset of BFRC and remained at the 0-per-night level at follow-up.

*Sleep onset latency*

Night-by-night inspection of mean sleep onset latency (SOL), as measured by sleep diaries, revealed an overall reduction in SOL at during follow-up for Bill and Keith, whereas Holly’s mean SOL increased above baseline (see Table 5). Bill’s baseline mean SOL (57.5 minutes) decreased by over 50% by end of treatment (25.7 minutes), and again at follow-up (11.3 minutes). This clinically meaningful improvement highlights the effectiveness of BFRC in the reduction of SOL for a child with ASD that experienced significant sleep onset delays. Visual analysis of Bill’s night-by-night sleep diary data
showed an increase in SOL the same day he experienced a change in routine, suggesting a more consistent schedule may have resulted in greater improvement of SOL. Keith’s mean SOL baseline (9.6 minutes) – already an acceptable level of latency – greatly increased during treatment (33.2 minutes). This abrupt increase in SOL occurred the first night of treatment; SOL progressively decreased and remained at a lower level during follow-up (4.3 minutes).

Mean SOL recorded by actigraphy decreased from baseline to the end of treatment for Holly and Bill (see Table 5). Actigraphy treatment phase data for Keith were lost due to a software malfunction. Actigraphy data on Holly’s mean SOL indicated a decrease of 29.7 minutes from baseline (43.5 minutes) to treatment (13.8 minutes) with a return toward baseline levels (32 minutes) at the end of follow-up. Bill’s mean SOL decreased by 50 minutes from baseline (74 minutes) to treatment (24 minutes) and increased by 7.3 minutes at follow-up (31 minutes). Keith’s remaining actigraphy data indicated his mean SOL decreased from baseline (13 minutes) to follow-up (5.6 minutes) by 7.4 minutes. Bill’s CSHQ-A sleep onset delay subscale scores decreased from baseline (3 minutes) to treatment (1 minute) and were maintained during follow-up (1 minute). Holly and Keith’s CSHQ-A sleep onset delay subscale scores of 1 were maintained throughout the study.

Waking after sleep onset

Mean values of night awakenings after sleep onset (WASO) in minutes, as measured by sleep diaries, revealed a progressive reduction in night-awakenings after sleep onset for Keith, whereas Holly’s and Bill’s mean WASO increased from baseline levels at the end of treatment (see Table 5). Keith’s WASO in minutes decreased from 16
minutes at baseline to 6.4 minutes during treatment and to zero during follow-up. Holly’s WASO increased from 0 minutes at baseline to 29 minutes at the end of treatment, followed by a decrease to 11.3 minutes during follow-up. Visual analysis of the data showed WASO maintained duration of 0 minutes on the first three out of four nights of follow-up and increased by 45 minutes on the final night. This increase corresponds with the uptick of FNA on the same night and may also be contributed to a change in Holly’s bedtime. Bill’s WASO increased by 6 minutes from baseline (35 minutes) to treatment (41 minutes) and decreased to 0 minutes at follow-up. Akin to his FNA sleep diary data, Bill’s night-by-night data showed a progressive decrease in mean WASO during treatment. Specifically, Bill experienced four consecutive nights of 0 minutes awake after sleep onset toward the end of treatment. The following night, WASO increased by 180 minutes (3 hours), which may have resulted from the unexpected change in his daytime routine and activity level.

WASO recorded by actigraphy decreased from baseline to the end of treatment for Holly and Bill (see Table 5). As previously reported, actigraphy treatment phase data for Keith were lost due to a software malfunction. Holly’s mean WASO decreased by 101 minutes from baseline (192 minutes; 3.2 hours) to treatment (91.3 minutes; 1.5 hours), with a return toward baseline levels (132.5 minutes; 2.2 hours) at the end of follow-up. Bill’s mean WASO decreased by 55 minutes from baseline (105 minutes; 1.8 hours) to treatment (50 minutes), and the decrease was maintained at follow-up. Keith’s remaining actigraphy data indicated his mean WASO increased slightly from baseline (51 minutes) to follow-up (57 minutes).

Time in bed
Time in bed (TIB) means measured in minutes by sleep diaries revealed an overall reduction in time spent in bed from baseline through follow-up for all three children. This calculation included the total amount of time the child was physically in bed during a 24-hour period. Holly and Keith’s TIB progressively reduced throughout the study, whereas Bill’s TIB slightly increased during follow-up (see Table 5). Bill’s mean TIB decreased by 1 hour and 14 minutes from baseline (595 minutes; 9.9 hours) to treatment (521 minutes; 8.7 hours). His mean TIB slightly increased – by 27 minutes – from treatment to follow-up (548 minutes; 9.1 hours). Holly’s mean TIB baseline (671 minutes; 11.2 hours) decreased by 45 minutes at the end of treatment (626 minutes; 10.4 hours), followed by an additional decrease of 11 minutes during follow-up (615 minutes; 10.3 hours). Keith’s mean TIB decreased from baseline (638 minutes; 10.6 hours) by 13 minutes at the end of treatment (625 minutes; 10.4 hours), and another 12 minutes at the end of follow-up (613 minutes; 10.2 hours). Overall, TIB means reduced from baseline to follow-up by 56 minutes for Holly, 47 minutes for Bill, and 25 minutes for Keith.

Correspondingly, TIB recorded by actigraphy decreased for all three children (see Table 5). Holly’s mean TIB decreased by approximately 1 hour from baseline (667 minutes; 11.1 hours) to treatment (605 minutes; 10.1 hours) and increased by 37 minutes from treatment to the end of follow-up (642 minutes; 10.7 hours). Similarly, Bill’s mean TIB decreased by 1 hour and 41 minutes from baseline (609 minutes; 10.2 hours) to treatment (508 minutes; 8.5 hours) and increased by 41 minutes throughout follow-up (549 minutes; 9.2 hours). Holly experienced an overall reduction in TIB of 25 minutes (31 minutes less than recorded in sleep diaries), whereas Bill’s TIB reduced by 1 hour (13 minutes more than recorded in sleep diaries). Much the same, Keith’s mean TIB
decreased from baseline (598 minutes; 10 hours) to follow-up (567 minutes; 9.5 hours) by 31 minutes (6 minutes more than recorded in sleep diaries).

Total sleep time

Total sleep time (TST) means measured by sleep diaries decreased from baseline to the end of treatment for all three children (see Table 5). TST was calculated by adding the durations of daytime and nighttime sleep within a 24-hour period. None of the parent dyads scheduled naptimes for their child; therefore a majority of sleep took place during the night. Holly’s baseline mean TST (653 minutes; 10.9 hours) decreased by the end of treatment (551 minutes; 9.2 hours) by 1 hour and 42 minutes and then increased by 27 minutes during follow-up (578 minutes; 9.6 hours). Her average decrease in mean TST was 1 hour and 15 minutes. Bill’s baseline mean TST (503 minutes; 8.4 hours) decreased by 57 minutes by the end of treatment (446 minutes; 7.4 hours) and reverted above baseline levels by 30 minutes (533 minutes; 8.9 hours) during follow-up.

Although none of the parents were interested in adding naps to their child’s sleep routine, Keith’s parents reported he had been allowed to nap during the day occasionally due to not sleeping either most or all of the night. This decreased the likelihood of him successfully sleeping through the night. Keith took two naps throughout the entire study, each on separate days during baseline. The first nap (15 minutes, 11:45 a.m. to 12:00 p.m.) occurred after sleeping for only 8.25 hours the night before with a terminal wake time of 2:00 a.m. Two days later, the second nap (3.75 hours, 8:15 a.m. to 12:30 p.m.) occurred after sleeping only 4.5 hours the night before with a terminal wake time of 11:00 p.m. Naps were eliminated for Keith at the onset of BFRC; this practice was
BEDTIME FADING WITH RESPONSE COST

maintained through follow-up. Both naps included, Keith’s mean baseline TST (599 minutes; 10 hours) decreased by 20 minutes by the end of treatment (579 minutes; 9.7 hours). His TST also reverted above baseline levels by 10 minutes (609 minutes; 10.2 hours) during follow-up.

Mean TST recorded by actigraphy increased for Holly and Bill and decreased for Keith from baseline to follow-up (see Table 5). Holly’s mean TST actigraphy data increased by 1 hour and 32 minutes from baseline (399 minutes) to treatment (491) and decreased by 16 minutes by the end of follow-up (475). Her overall TST increased by 1 hour and 16 minutes. Bill’s mean TST decreased by 13 minutes from baseline (415) to treatment (402) and increased by 1 hour and 2 minutes by the end of follow-up (464). His overall TST increased by 49 minutes. Keith’s mean TST decreased from baseline (525) to follow-up (502) by 23 minutes. Bill’s CSHQ-A sleep duration subscale scores did not change from baseline (5) to treatment (5) and decreased during follow-up (3), whereas Holly’s CSHQ-A sleep duration subscale scores did not change from baseline (3) through follow-up. Keith’s CSHQ-A sleep duration subscale scores decreased from baseline (6) to treatment (3) and did not change during follow-up (3).

Sleep efficiency

Sleep efficiency (SE) outcomes extrapolated from sleep diaries data indicate all three children overall maintained or improved the percentage of their sleep efficiency. Holly’s baseline SE of 97% decreased to 88% at the end of treatment and increased to 94% during follow-up. Bill’s baseline SE increased slightly from 85% to 86% at the end of treatment, and further increased to 97% during follow-up. Keith’s baseline SE of 94%
decreased slightly to 92% at the end of treatment and increased to 99% throughout follow-up. Compared with these data, SE outcomes extrapolated from actigraphy data were lower for each child. For example, Holly’s baseline SE of 60% increased to 82% at the end of treatment, and decreased to 75% during follow-up. Bill’s baseline SE increased from 68% to 80% at the end of treatment, and further increased to 85% during follow-up. Keith’s baseline SE of 89% was maintained during follow-up.

Children’s Sleep Habits Questionnaire, Abbreviated Version

BFRC led to a significant reduction in the Children’s Sleep Habits Questionnaire, Abbreviated Version (CSHQ-A) total sleep disturbance scores for all three children. Total sleep disturbance scores drastically decreased from the clinical range during baseline to the non-clinical range by follow-up for two children, whereas the third child’s follow-up score was only 1 point above the proposed clinical cutoff score (41). Specifically, Bill’s baseline CSHQ-A total score of 48 decreased to 43 at the end of treatment, and further decreased to 36 during follow-up, with an overall decrease of 12 points. Keith’s total baseline score of 60 significantly decreased to 46 at the end of treatment, with an additional decrease to 42 during follow-up, with a decrease of 18 points, which represents the greatest improvement among the study participants. Likewise, Holly’s CSHQ-A total score at baseline (56) decreased by the end of follow-up (40), leaving her with the second best reduction in score of 16 points.

As previously mentioned, Holly’s CSHQ-A subscale scores for sleep onset delay (1), sleep duration (3), and night wakings (5) remained unchanged from baseline throughout follow-up (EOT data not available), whereas bedtime resistance significantly decreased from 14 to the minimum score of 6 (see Table 5). Similarly, Keith’s CSHQ-A
subscale score for *sleep onset delay* remained unchanged throughout the study at a baseline score of 1. In contrast to Holly’s score, Keith’s CSHQ-A subscale score for *sleep duration* decreased from baseline (6) by half at the end of treatment (3) and remained unchanged during follow-up. His CSHQ-A subscale score for *night wakings* also decreased from baseline (7) to treatment (4) but then slight increased at follow-up (5). Keith’s CSHQ subscale score for *bedtime resistance* decreased from baseline (12) to the end of treatment (7), with an additional decrease at follow-up (6).

Bill’s CSHQ-A subscale score for *sleep onset delay* decreased from baseline (3) to the minimum score at the end of treatment (1) and did not change during follow-up. His CSHQ-A subscale score for *bedtime resistance* followed a similar pattern with a decrease from baseline (9) to the minimum score at the end of treatment (6), which did not change during follow-up. Bill’s CSHQ-A *sleep duration* subscale score maintained at baseline through end of treatment (5) and decreased to the minimum score (3) at the end of follow-up, whereas *night wakings* slightly decreased from baseline (7) to end of treatment (6), with a near minimal level decrease during follow-up (4). CSHQ-A *sleep disordered breathing* scores were reported within 1 point (range 3 to 4) of the minimal level (3) and determined non-clinically significant for all three children.

*Goal Achievement Scale*

Parents set sleep-related goals for their child and worked with a member of the investigative team to refine and determine the levels of success for each goal (see Table 4). Keith had four goals, whereas Holly and Bill had two goals each. The only similar parent-set goal across all three children was to reduce undesired co-sleeping in the parent’s bed by having the child sleep alone in their own bed. The percentage of nights
during the treatment phase when each child slept in their own bed dramatically increased for all three children as measured by the sleep-related behavior diaries. Bill’s goal of *sleep through the night in [his] own bed* dramatically increased from 0% success (0 out of 6 nights) during baseline, to 100% success (14 out of 14 nights) at the end of treatment phase, which remained at 100% success (4 out of 4 nights) through follow-up.

Similarly, Holly’s goal of *sleep in [her] own bed (every night)* increased from 0% success (0 out of 4 nights) during baseline to 100% success (11 out of 11 nights) at the end of the treatment phase and remained at 100% success during follow-up (4 out of 4 nights). Keith’s goal of *falling asleep on own (in [his] own bed)* increased from 0% success (0 out of 14 nights) during baseline to 100% success (14 out of 14 nights) at the end of the treatment phase and remained at 100% success (7 out of 7 nights) during follow-up. Keith’s goal of *sleeping through the night in [his] own bed (return to sleep during the night by himself)* went from 0% success (0 out of 14 nights) during baseline to 93% success (13 out of 14 nights) at the end of treatment. This goal’s success decreased to 75% (5 out of 7 nights) during follow-up due to briefly engaging in undesired co-sleeping in the parent’s bed (mean of 9 minutes) on two separate nights.

Bill’s and Keith’s parents had different goals related to the time their child went to sleep each night. Bill’s parents’ goal to *go to sleep sooner (reduce SOL each night)* was set at equal to or less than 15 minutes in order to achieve 100% success. He improved from 0% success (average of 57.5 minutes per night) during baseline to 75% success (average of 25.7 minutes per night) at the end of the treatment phase, which further increased to 100% success (average of 11.3 minutes per night) during follow-up. Keith’s parents’ goal of *bedtime between 7:00 and 7:30 p.m.* was set to 7 out of 7 nights
to achieve 100% success. This goal increased from 1% success (1 out of 14 nights) during baseline to 43% success (6 out of 14 nights) at the end of the treatment phase, and further increased to 86% success (6 out of 7 nights) during follow-up, thus indicating BFRC provided the structure necessary to achieve success.

Holly’s and Keith’s parents had similar goals for the amount of sleep their child received each night. Members of the investigative team used sleep norms (Iglowstein, et al., 2003) to help parents set the goals. Norms for total sleep duration per 24-hour period range between 9.6 and 11 hours for children ages 8 and 9 years and between 10.5 and 12.8 hours for children ages 4 to 5 years (Iglowstein, et al., 2003). Based on these norms, Holly’s goal to get required amount of sleep for [her] age (every night) decreased from 50% success (2 out of 4 nights) during baseline to 18% success (2 out of 11 nights) at the end of the treatment phase, followed by a return to baseline level 50% (2 out of 4 nights) during follow-up. Keith’s goal to get enough sleep (developmentally appropriate amount per night) also decreased from baseline 36% success (5 out of 14 nights) to 21% success (3 out of 14 nights) at the end of the treatment phase and increased to 43% success (3 out of 7 nights) during follow-up.

In contrast to the recommendations published by Iglowstein and colleagues (Iglowstein, et al., 2003), the National Sleep Foundation recommendations on sleep duration per 24 hours are between 10 to 13 hours of sleep for children ages 3 to 5 years and between 9 and 11 hours for children ages 6 to 13 years (Hirshkowitz, et al., 2015). Even though these recommendations were not available at the beginning of our study, these norms were compared with Holly’s and Keith’s total sleep time (TST) values. The results of this comparison revealed a steady improvement in sleep duration goals for both
children. Holly’s baseline level of 50% success (2 out of 4 nights) increased to 64% success (7 out of 11 nights) at the end of the treatment phase, and further increased to 100% success (4 out of 4 nights) during follow-up, whereas Keith’s baseline level of 57% success (8 out of 14 nights) remained stable through the end of the treatment phase and further increased to 71% success (5 out of 7 nights) during follow-up.

Although sleep duration norms for children with ASD have yet to be established, Hodge and colleagues (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014) found that children with ASD between ages 3 and 17 years experienced significantly less overall sleep (mean, 8 hours and 47 minutes; standard deviation, 93 minutes) compared with typically developing peers (mean, 9 hours and 22 minutes; standard deviation, 76 minutes). Compared with these findings, Holly’s baseline level of 25% success (1 out of 4 nights) increased to 82% success (9 out of 11 nights) at the end of the treatment phase, and further increased to 100% success (4 out of 4 nights) during follow-up, whereas Keith’s baseline level of 50% success (7 out of 14 nights) increased to 64% success (9 out of 14 nights) at the end of the treatment phase and decreased to 29% success (2 out of 7 nights) during follow-up (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014).

Kansas Marital Satisfaction Scale

Parent-completed Kansas Marital Satisfaction Scale (KMSS) indicated a possible ceiling effect for two of the three parent dyads (see Table 6). Bill’s and Keith’s parents each reported being extremely satisfied (7) on all three KMSS probes (i.e., 1) How satisfied are you with your marriage?; 2) How satisfied are you with your husband/wife as a spouse?; and 3) How satisfied are you with your relationship with your
husband/wife?) at baseline, end of treatment, and follow-up. Therefore, Bill’s and Keith’s parents’ total individual KMSS scores were maintained at the ceiling level of 21 for each parent throughout the study, thus staying above the cut-off score of 17 for distressed couples. In contrast, Holly’s parents each endorsed being somewhat satisfied (5) with their marriage and very satisfied (6) with their husband or wife as a spouse and with their relationship with their husband or wife at baseline and end of treatment. Holly’s parents’ KMSS scores with regard to their marriage increased by 1 point each from somewhat satisfied to very satisfied (6), while the other scores remained unchanged through to follow-up. Total individual KMSS scores for Holly’s parents remained stable from baseline (17) through end of treatment and increased at follow-up (18), also staying above the cut-off score indicating marital distress.

Parent’s Customer Satisfaction Questionnaire

Parent’s Customer Satisfaction Questionnaire (PCSQ) mean-ratings indicate high levels of satisfaction with treatment implementation and outcomes across all three families. Holly’, Bill’s, and Keith’s parents endorsed that the major problems that originally prompted treatment and the problems treated during this study had greatly improved (mean rating of 7). All three parent dyads endorsed feeling very positive (mean rating of 7) about the treatment program, feeling very satisfied (mean rating of 7) about their child’s progress, and would strongly recommend (mean rating of 7) the treatment program to a friend or relative. Parental expectations for a satisfactory outcome of the treatment at the end of follow-up were optimistic (Keith) or very optimistic (Holly and Bill; mean rating of 6.66).
Each parent endorsed being *very confident* (mean rating of 7) in their ability to manage *future* in-home sleep problems based on what they learned during the study, whereas only Holly’s and Bill’s parents also endorsed being *very confident* (7) in their ability to manage *current* in-home sleep problems independently, resulting in an overall mean ratings of 5. This discrepancy in endorsements by Keith’s parents may due to the reverse selection of the response options for this particular question, suggesting a rating of 7 may have been intended as opposed to 1 (i.e., 1 = *Very unconfident* verses 7 = *Very confident*). Problems related to the children *not treated* during the study have either remained *the same* (Holly and Keith) or *slightly improved* (Bill; mean rating 4.33), whereas the degree to which the treatment program helped parents with other personal or family problems have either *helped* (Holly and Keith) or *helped slightly* (Bill; mean rating 5.66).

In the open-ended questions, parents were invited to share their opinions on the best and worst components of the treatment program. The *most helpful* components included ‘sleeping in parents’ bed stopped completely’ for Bill; ‘getting her to sleep in her own bed’ for Holly; and ‘creating strict bedtime rules and sticking to it’ for Keith. Parents reported *liking most* the ‘results; treatment solved problems’ (Bill); ‘easy to follow/impliment’ (Holly); and ‘having us have rules to establish a better bedtime routine’ (Keith). Bill and Keith’s parents *liked least* the ‘lack of sleep a few nights’ (Bill) and ‘first few days were difficult, but it got better. those days were tough! and data entry.’ (Keith). Holly’s parents indicated that there was nothing they *liked least* about the treatment program, whereas each parent dyad endorsed none of the treatment program
components as least helpful. Likewise, none of the parents reported any part of the treatment program as not helpful, nor did they offer recommendations for improvement.

Discussion

The results of this study add to the literature supporting BFRC as an effective treatment option for co-occurring sleep problems experienced by children with ASD. This study evaluated the effectiveness of BFRC as an in-home treatment with parents as change agents in decreasing parent-defined sleep disturbances, such as undesired co-sleeping, night awakenings, difficulty falling asleep, and bedtime disturbances experienced by children with ASD. While this study centered on improving the sleep of children diagnosed with ASD by means of BFRC, innovative concepts of the study were to utilize parents to determine and implement treatment according to parent-derived sleep goals for their children.

Previous BFRC research on sleep disturbances in children with ASD (DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997) had not considered such novel aspects simultaneously. For example, two studies (DeLeon, Fisher, & Marhefka, 2004; Piazza, Fisher, & Sherer, 1997) were restricted to institutionalized settings with BFRC implemented by trained professionals, one study (Moon, Corkum, & Smith, 2010) involved training parents to implement BFRC in the home setting, and one study (Piazza, Fisher, & Sherer, 1997) used sleep norms in conjunction with parent goals to establish ideal sleep and wake times for their child. The current study extends both the science and practice of increasing parental agency while providing BFRC as a treatment option for children with ASD and co-occurring sleep disturbances.
The primary sleep problem across all three families – frequency and duration of undesired co-sleeping – improved for each child. Visual analysis showed improvement in baseline levels immediately following treatment onset. These improvements were maintained at one-month follow-up; thus, BFRC was effective in reducing (Keith) and eliminating (Holly and Bill) the frequency and duration of undesired co-sleeping. Neither frequency nor duration of undesired co-sleeping was negatively impacted by changes in routine for any of the children. Furthermore, parent goal for their child to sleep in their own bed were accomplished after the onset of treatment, and these outcomes were maintained across all children. Each of these outcomes offers compelling evidence for experimental control in a single-subject, non-concurrent multiple baseline design and meaningful implication for the effectiveness of BFRC as a parent-implemented home-based intervention.

This is the first study to demonstrate BFRC as an effective intervention for the elimination of undesired co-sleeping in children with ASD. Previously, Ashbaugh and Peck (1998) found that BFRC eliminated undesired co-sleeping with a typically developing child, whereas others (Piazza and Fisher, 1991b; Piazza, Fisher, and Moser, 1991) noted reductions in undesired co-sleeping among children diagnosed with non-ASD neurodevelopmental disorders. Previous BFRC studies in children with ASD primarily focused on changes in disruptive sleep-related behaviors, sleep latency, night awakenings, or sleep duration (DeLeon, Fisher, & Marhefka, 2004; Moon, Corkum, & Smith, 2010; Piazza, Fisher, & Sherer, 1997). The current study evaluated additional parent-determined sleep goals (i.e., reduce night awakenings and sleep onset, set bedtime, and obtain developmentally appropriate amount of sleep) as primary sleep problems and
sleep problems not identified as parent goals (i.e., BR, DBC, DSO, WASO, TIB, TST, SE) as secondary sleep problems. Nevertheless, these outcomes provide further evidence for a positive change following BFRC.

Above and beyond the complete elimination of undesired co-sleeping, Holly also demonstrated meaningful improvements in sleeping in her own bed, falling asleep independently, and bedtime resistance. At the onset of treatment, Holly’s parents no longer had to remain next to her to get her to fall asleep or to sleep in her own bed throughout the night. Moreover, Holly no longer engaged in disruptive behaviors during her bedtime routine or after she was placed in her bed for the night. Time spent engaging in bedtime resistance dramatically improved after the onset of BFRC. These changes were clinically important to Holly’s parents because a substantial portion of their evenings were no longer occupied by conflict over sleep. Occasionally, she continued to come out of her bedroom after being put in bed for the night; however, frequencies of these minor disturbances remained below baseline levels at follow-up.

Depending on the measurement method, Holly’s overall total sleep time decreased (sleep diary) or increased (actigraphy) from baseline to the end of follow-up. Unlike the other two children in this study, Holly did not experience difficulties with night awakenings prior to treatment. Nonetheless, both the number and the duration of night awakenings varied greatly during treatment and eventually returned to near baseline levels. This putative suppression of night awakenings during baseline may stem from her sleeping through the night in her parents’ bed with at least one parent in the room. Therefore, Holly’s night awakenings began after access to co-sleeping with a parent and dependent sleep onset were eliminated. It is worth noting that Holly experienced a change
in routine the last night of follow-up, which might have contributed to sleep problems experienced that night.

In addition to the elimination of undesired co-sleeping, Bill also experienced noteworthy improvements in sleep latencies, bedtime resistance, and the number of night awakenings and started sleeping in his own bed and falling asleep independently. After the onset of BFRC, Bill no longer sought parental presence to initiate or reinitiate sleep and he was able to sleep through the night in his own bed independently. He also engaged in fewer disruptive behaviors during his bedtime routine that continued to improve throughout treatment and follow-up. By the end of follow-up, Bill was able to consistently achieve sleep onset within 15 minutes of bedtime. This outcome was clinically significant because prior to intervention Bill often took one to two hours a night to fall asleep, which may have inadvertently disrupted his parents’ sleep.

According to both subjective and objective measures, Bill’s overall total sleep time increased from baseline to the end of follow-up. Night awakenings also progressively improved throughout treatment, however, Bill might have achieved greater improvement had it not been for a change in schedule and lack of daytime activity the second to last day of treatment. That same night, he experienced more frequent and longer lasting disruptive behaviors than he had the previous ten nights of treatment and spent the least amount of time sleeping out of any night during the entire study. Bill also took longer to fall asleep than he did the two nights before and had more night awakenings than he did the previous four nights. Nonetheless, his night awakenings were completely eliminated during follow-up.
Keith also demonstrated considerable improvements over and above a significant reduction in undesired co-sleeping, specifically in night awakenings, sticking to a set bedtime, sleeping in his own bed, falling asleep independently, and bedtime resistance. After the onset of BFRC, Keith experienced fewer night awakenings, which completely disappeared toward the end of treatment and remained absent throughout follow-up. As opposed to Holly’s data, Keith’s overall total sleep time from baseline to the end of follow-up increased, based on sleep diary data, or decreased, based on actigraphy data. Furthermore, Keith no longer required a parent in the room or in bed with him to fall asleep at bedtime or after night awakenings. Keith also engaged less often in bedtime refusal, following an expected extinction burst the first night of BFRC, when he was no longer permitted to sleep in his parents’ bed or with a parent in the room.

Overall, the occurrence of Keith’s sleep-related disruptive behaviors gradually declined throughout treatment and remained rare at follow-up, with the exception of an unexplained uptick. A steady upward trend in the length of time Keith engaged in bedtime resistance began the third night of treatment, reaching its peak on the sixth night. The following night marked the descent to near baseline levels. Unfortunately, no information was provided that could reasonably account for this change in trend. As for sleep latencies, Keith’s data showed a sharp increase on the first night of treatment, followed by a progressive decline that was maintained during follow-up. This pattern likely indicates that for this child, the onset of treatment temporarily altered his sleep latency, and he was able to eventually adjust to his original set point of sleep onset within 15 minutes of being placed in bed. Furthermore, this improvement was not clinically
meaningful as he was already achieving sleep within an acceptable range of latency (average within 10 minutes of being placed in bed) before treatment.

In all BFRC studies with children on the autism spectrum, researchers determined treatment onset and primary sleep outcomes, with the exception of Piazza and colleagues (1997), who used sleep norms in conjunction with parent preferences to establish children’s sleep and wake times. In the current study, two children (Holly and Keith) had parent-determined goals for sleep duration based on sleep norms (Iglowstein, et al., 2003). At the time of our study, the most current and complete sleep duration norms available were based on the combined results of two longitudinal studies conducted with Swiss children (Iglowstein, et al., 2003). Based on these norms and lofty parent goals, it is not surprising that neither Holly nor Keith attained total sleep time goals after the onset of treatment. This may be partly because children with ASD tend to sleep less compared with typically developing peers (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014). Compared with other studies with children on the autism spectrum (Durand & Mindell, 1999; Moon, Corkum, & Smith, 2010; Schreck & Mulick, 2000), Holly’s and Keith’s sleep durations appeared to fall within the normal range for their age. Therefore, both parent dyads might have overestimated their child’s need for sleep.

As previously described, two children (Bill and Keith), whose parents had a goal of reducing the number of night awakenings, experienced an overall improvement during treatment and total elimination during follow-up. These results are consistent with the findings of two studies (Piazza, Fisher, and Sherer, 1997; DeLeon, Fisher, and Marhefka, 2004) in that night awakenings were nearly eliminated following BFRC in children with ASD. Consistent with findings by Moon, Corkum, and Smith (2010), mean sleep onset
latency as measured by both actigraphy and sleep diary revealed an overall decrease from baseline following BFRC for two children (Bill and Holly) with ASD. BFRC was also effective in reducing the duration and frequency of disruptive sleep-related behaviors for all three children. Moreover, BFRC produced a clinically significant improvement in CSHQ-A total scores for three children with severe ASD.

Prior to treatment, parent-reported total sleep disturbance scores indicated that their children experienced dyssomnia-type sleep problems in the clinical range. Two children (Holly and Bill) greatly improved, their problems moving from the clinical range to the non-clinical range by the end of follow-up. However, one child (Keith), who experienced the greatest improvement with the largest reduction in scores out of all three children, was only one point above the clinical cut-off score by the end of the follow-up. These parent-reported scores on each subscale are consistent with night-by-night data from sleep and behavior diaries, indicating an overall positive change with less bedtime resistance for all three children, fewer night awakenings and longer sleep duration for Bill and Keith, and less sleep onset delays for Bill. Actigraphy data further support these positive changes in sleep onset delays and sleep duration for Bill. These outcomes offer strong support for BFRC successfully treating a wide variety of multiple co-occurring disruptive sleep and related problem behaviors in children on the autism spectrum.

The total amount of time spent sleeping in a 24-hour period was greater when recorded in sleep diaries vs. by actigraphy across all three children. Furthermore, the average amount of time spent in bed as recorded by both sleep diaries and actigraphy significantly reduced from baseline to follow-up for all three children. This reduction in time spent in bed likely produced the observed improvements in sleep efficiency.
According to sleep diary data, each child was already achieving sleep efficiency within the low-end of normal (85%) to high (97%) range prior to treatment. However, actigraphy data produced lower sleep efficiencies, compared with sleep diary data, for each child.

In contrast to findings by Moon, Corkum, and Smith (2010), there was a pattern of change in mean sleep efficiency as measured by actigraphy for two children. Holly’s and Bill’s sleep efficiency increased after treatment, and mild improvements were maintained at follow-up, whereas Keith’s mean sleep efficiency remained at 89% throughout the study. For all three children, the amount of time spent awake after sleep onset recorded by actigraphy was greater than time recorded in sleep diaries, which was nearly zero. Similarly to the improvements of night awakenings recorded in sleep diaries, two of the three children spent no time awake after sleep onset by follow-up, whereas the third child might have continued the pattern of zero time awake after initially falling asleep had she not experienced a change in routine on the final night of follow-up.

In the present study, all parent dyads reported high levels of satisfaction with BFRC and its results and were willing to strongly recommend this treatment to others. This outcome is similar to high parent satisfaction with treatment reported in the one other BFRC study that used parents as change agents in the home setting with children with ASD (Moon, Corkum, & Smith, 2010). In our study, parents reported the use of structured guidelines and the elimination of undesired co-sleeping as being the most helpful components. In sum, these parent opinions suggest that benefits outweighed the challenges and support the social importance of BFRC as an effective parent-implemented treatment. Only one parent satisfaction item was rated negatively. This
discrepancy might be explained by a change in response item value and topographical similarity between Very unconfident and Very confident rather than a true endorsement of low confidence in their ability to manage their child’s current problem behaviors. For example, these parents wrote, “The treatment was awesome. It gave us the tools we needed.” Therefore, this score should be interpreted with caution. As such, this minor inconsistency does not diminish the overall high level of parent satisfaction across all three families and the social validity of BFRC as a feasible parent-implemented in-home treatment for children with ASD and multiple co-occurring sleep problems.

We explored the impact of changes in sleep problems on marital satisfaction. Holly’s parents each experienced some minor improvement in marital satisfaction at the end of treatment. This slight improvement does not provide sufficient evidence to support the impact of BFRC on marital satisfaction; however, previous findings (Fisman, Wolf, & Noh, 1989; Mihaila & Hartley, 2016) suggest various explanations for this change. For instance, an increase in marital satisfaction might indicate that Holly’s parents’ sleep also improved, thus increasing each parent’s positive affect (Mihaila & Hartley, 2016). Likewise, removing Holly from her parents’ bed might have had a positive impact on the quality of their marriage by allowing for more time alone with each other (Fisman, Wolf, & Noh, 1989). Bill’s and Keith’s parents consistently endorsed being extremely satisfied with their marriage, his or her spouse, and his or her relationship with their spouse. Although a ceiling effect left little room for improvements in marital relations, these results do indicate that BFRC did not produce a decline in couple’s marital satisfaction.

promising intervention for sleep disturbances experienced by children with ASD. Our study expands upon this research in a number of ways. First, the current study addresses the lack of agency parents have been given when children participate in treatment-related research. Parents, not researchers, determined the treatment goals and the primary outcome variables. The current method enhances the social validity of BFRC and the social significance of the positive findings. Second, the current study obtained actigraphy data as an objective measure of the sleep-related outcome variables.

Third, a non-concurrent multiple baseline across subjects design was employed in place of a more rigorous design due to its feasibility in applied settings and in an attempt to avoid potential temporal delays in participant enrollment (Watson & Workman, 1981). Nonetheless, we met experimental design requirements (Harvey, May, & Kennedy, 2004) by staggering baseline durations across the three participants and planning for the implementation of BFRC. For example, BFRC was introduced after baseline data on the frequency of the primary target behavior (i.e., undesired co-sleeping) were determined stable (4 consecutive days for Holly and 6 consecutive days for Bill) or after 14 consecutive days of data collection, as done for Keith. The maximum of 14 consecutive days represents a potential methodological weakness; however, this two-week limit was chosen based on clinical considerations for the participants and their families. Experimental control was demonstrated by a functional relationship between the implementation of BFRC and observable changes in the frequency of undesired co-sleeping for each child.

In addition, results from a small sample might not generalize to a wider population. We did not evaluate the extent to which each parent dyad worked together.
We did encourage both parents to implement treatment and designated one parent responsible for data recording; therefore, it is unclear if a single parent could effectively implement treatment and achieve similar results. In addition, this study focused on preschool and school-aged children, who largely depend on parents to structure their sleep routine and sleep environment. It would be beneficial to determine if parents of adolescents with ASD can effectively implement BFRC with comparable outcomes. Nevertheless, BFRC was effective for three young children with severe autism and multiple co-occurring sleep problems, who were living in a two-parent household. A randomized controlled trial would strengthen the external validity of these findings and help to determine effective levels of parental participation.

Lastly, we did not include an objective measure such as video recording to confirm procedural integrity or collect IOA to document the reliability of data collection procedures. Based on clinical considerations (Burke, Kuhn, & Peterson, 2004; Durand & Mindell, 1999; Vollmer & Sloman, 2008), we selected telephone checks, a less intrusive method, to ensure ongoing and timely data collection and to obtain data on treatment integrity.

Behavior analysts have a great appreciation for precise terminology that conveys information effectively. *Response cost* is best described as a punishment procedure in the form of a penalty, which may not be the most accurate label for the response extinction components of the BFRC sleep intervention. Although terminological refinement may be warranted, perhaps the more salient endeavor lies in conceptualizing the effectiveness of BFRC using behavioral processes and concepts. Respondent conditioning, operant conditioning, and the value altering effects of motivating operations all contribute to
understanding the socially significant phenomenon of sleep. As such, the process of respondent and operant conditioning and motivating operations may all contribute to the effectiveness of the BFRC procedure.

Respondent conditioning may contribute through a stimulus-stimulus pairing process, in which an unconditioned stimulus (UCS) becomes a conditioned stimulus (CS) that elicits the conditioned response (CR). From a respondent conditioning standpoint, the UCS is somnolence (i.e., physiological state of sleepiness), which is often associated with delaying bedtime and limiting access to the unconditioned response (UCR) of sleep. Delaying the initial bedtime component of the BFRC procedure increases the likelihood of the child achieving sleep onset within 15 minutes of being placed in bed. The behavior of lying down in bed may have become a CS that functions as a signal for initiating sleep. If the child falls asleep soon after being placed in bed, the CS effectively elicited the CR of sleep.

Operant conditioning, that is, the basic process of learning that occurs when a stimulus changes immediately following a response, resulting in changes in the behavior when under similar conditions in the future, may also contribute to the effectiveness of the BFRC procedure. Prior to treatment, the child had access to preferred activities, parental attention, and the opportunity to fall asleep whenever he or she became sleepy, whereas during treatment the parent controls such accessibility and opportunities. In the BFRC treatment, the child has 15 minutes to fall asleep once being placed in bed, otherwise he or she is removed from bed and does not have another opportunity to sleep for 30 minutes. The stimulus change of being removed from bed occurs immediately following the child’s response of being awake in bed. Although the child has access to
preferred activities and parental attention, access to sleep is restricted until the next scheduled bedtime. As the child’s sleepiness increases, being removed from bed becomes an unpleasant event. The child learns to avoid being removed from bed by engaging in sleep-compatible behaviors or achieving rapid sleep onset.

Motivating Operation (MO) is a behavioral concept used to explain variations in the strength of value-altering and behavior-altering effects (Michael, 1982; Michael, 2000). Sleep deprivation is one of the primary MOs identified in humans. It is the current strength of sleepiness that can alter the reinforcing value of consequences and change the probability of behaviors. That is, extreme sleepiness can increase the effectiveness of sleep as reinforcement and increase the current frequency of sleep-compatible behavior (e.g., lying down in bed) that has been reinforced by sleep. As such, MOs alter the effectiveness of both respondent and operant conditioning. Delaying the child’s bedtime and restricting access to sleep alters the reinforcing value of sleep and changes the probability of achieving rapid sleep onset and engaging in sleep-compatible behaviors. When the duration of time children spend awake in bed is reduced, the experience of sleepiness and sleep onset comes under stimulus control of the bedtime routine, the bed, and other aspects of the bedroom environment. Therefore, the BFRC procedure makes the most out of naturally occurring sleep processes (i.e., motivating operations) while teaching more appropriate sleep associations (i.e., stimulus control).

In summary, this investigation expands research on BFRC as a home-based, parent-delivered intervention for undesired co-sleeping with co-occurring sleep problems experienced by children with severe ASD. This investigation sets the stage for empowering parents to function as the agents of change by utilizing their own treatment
goals and making them responsible for implementing BFRC. Moreover, this study provides evidence that elevates BFRC toward to a new classification tier as a well established empirically supported treatment (Chambless & Hollon, 1998). In conclusion, the results of this study provide evidence for parent implementation of BFRC as a behavioral treatment option for children with ASD and multiple sleep problems. Future research should center on generalizing these favorable outcomes.
References


doi:10.1111/cns.12651


Wirojanan, J., Jacquemont, S., Diaz, R., Bacalman, S., Anders, T. F., Hagerman, R. J., Goodlin-Jones, B. L. (2009). The efficacy of melatonin for sleep problems in
children with autism, fragile x syndrome, or autism and fragile x syndrome.

*American Academy of Sleep Medicine, 5*, 145-150.


Table 1

*Participant Information*

<table>
<thead>
<tr>
<th></th>
<th>Holly</th>
<th>Bill</th>
<th>Keith</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age in years</td>
<td>8.5</td>
<td>7</td>
<td>4.5</td>
</tr>
<tr>
<td>Gender</td>
<td>Female</td>
<td>Male</td>
<td>Male</td>
</tr>
<tr>
<td>ADOS-2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Overall total</td>
<td>13</td>
<td>24</td>
<td>24</td>
</tr>
<tr>
<td>Comparison score</td>
<td>6</td>
<td>10</td>
<td>10</td>
</tr>
<tr>
<td>Classification</td>
<td>Autism</td>
<td>Autism</td>
<td>Autism</td>
</tr>
<tr>
<td>CARS-2</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Raw score</td>
<td>31*</td>
<td>44</td>
<td>39</td>
</tr>
<tr>
<td>T-score; percentile</td>
<td>51; 54&lt;sup&gt;th&lt;/sup&gt;%</td>
<td>57; 76&lt;sup&gt;th&lt;/sup&gt;%</td>
<td>51; 54&lt;sup&gt;th&lt;/sup&gt;%</td>
</tr>
<tr>
<td>Severity group</td>
<td>Severe</td>
<td>Severe</td>
<td>Severe</td>
</tr>
<tr>
<td>Sleep problems meeting selection criteria</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Requires parent in room to fall asleep</td>
<td>☑</td>
<td>☑</td>
<td>☑</td>
</tr>
<tr>
<td>Refusal to sleep in own bed</td>
<td>☑</td>
<td>☑</td>
<td></td>
</tr>
<tr>
<td>Undesired co-sleeping</td>
<td>☑</td>
<td>☑</td>
<td>☑</td>
</tr>
<tr>
<td>Bedtime resistance</td>
<td>☑</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep onset latency</td>
<td>☑</td>
<td>☑</td>
<td></td>
</tr>
<tr>
<td>Night awakenings</td>
<td>☑</td>
<td>☑</td>
<td>☑</td>
</tr>
</tbody>
</table>

*Note:* An ADOS-2 Comparison score of 6 indicates a moderate level of autism spectrum, while a score of 10 indicates a high level of autism spectrum. * Scores for this participant were taken from the CARS-2 High Functioning (HF) version, as opposed to the Standard (ST) version of the measure.
Table 2
*Dependent Variables on Sleep Diary & Sleep Related Behavior Diary*

<table>
<thead>
<tr>
<th>Dependent Sleep Variables</th>
<th>Definitions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Frequency of Night-Awakenings (FNA)</td>
<td>Total number of times child awoke during the night and got out of bed; Marked by time with an up arrow, not including final rising</td>
</tr>
<tr>
<td>Waking-up After Sleep Onset (WASO)</td>
<td>Total minutes the child is awake after initially falling asleep; Non-shaded in boxes of time (small box = 15 min.; large box = 1 hour) after initial sleep onset, until time of final rising</td>
</tr>
<tr>
<td>Sleep Onset Latency (SOL)</td>
<td>The time between initially being placed in bed awake, marked by time with a down arrow, and the time of sleep onset each night, marked by shaded in boxes</td>
</tr>
<tr>
<td>Total Sleep Time (TST)</td>
<td>Total duration of nighttime and daytime sleep acquired within 24-hours; Shaded in boxes for when the child is asleep (small box = 15 min.; large box = 1 hour)</td>
</tr>
<tr>
<td>Time in Bed (TIB)</td>
<td>Amount of time the child was physically in bed during within 24-hours; Time child gets into bed marked by a down arrow until final rising</td>
</tr>
<tr>
<td>Dependent Sleep Onset (DSO)</td>
<td>The achievement of sleep with the required assistance of a parent; Marked by the letter D next to a down arrow</td>
</tr>
<tr>
<td>Independent Sleep Onset (ISO)</td>
<td>The achievement of sleep without the assistance of a parent; Marked by the letter I next to a down arrow</td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Dependent Sleep Related Behavior Variables</th>
<th>Definitions</th>
</tr>
</thead>
<tbody>
<tr>
<td>Undesired Co-Sleeping (UC-S)</td>
<td>Unwanted co-sleeping with a parent or sibling in any room during a 24 hour period; Frequency and duration of undesired co-sleeping with parent(s) or siblings by child</td>
</tr>
<tr>
<td>Sleeping in a Non-Designated Area</td>
<td>Frequency of child sleeping in a location other than the child’s bed (e.g., couch, car, floor, etc.) during a 24 hour period</td>
</tr>
<tr>
<td>Location of Non-Designated Sleep</td>
<td>Location of undesired co-sleeping with child</td>
</tr>
</tbody>
</table>
**Session Timeline**

<table>
<thead>
<tr>
<th>Weekly Sessions</th>
<th>Activities</th>
</tr>
</thead>
<tbody>
<tr>
<td>Session 1:</td>
<td>Intake interview, CARS-2, ADOS-2, KMSS, and consent</td>
</tr>
<tr>
<td>Session 2:</td>
<td>Identify parent sleep goals, teach data collection, give actigraphy, and</td>
</tr>
<tr>
<td></td>
<td>start baseline data collection</td>
</tr>
<tr>
<td>Session 3:</td>
<td>Review week one baseline data and determine treatment or cont. baseline</td>
</tr>
<tr>
<td>Session 4:</td>
<td>Review week two baseline data or week one treatment data. Cont. or</td>
</tr>
<tr>
<td></td>
<td>discontinue treatment</td>
</tr>
<tr>
<td>Session 5:</td>
<td>Review week one or week two of treatment data. Cont. or discontinue</td>
</tr>
<tr>
<td></td>
<td>treatment</td>
</tr>
<tr>
<td>Session 6:</td>
<td>Review week two of treatment data or begin follow-up data collection</td>
</tr>
<tr>
<td></td>
<td>(4-weeks post treatment)</td>
</tr>
<tr>
<td>Session 7:</td>
<td>Begin (4-weeks post treatment) or end (7 days) follow-up data collection.</td>
</tr>
<tr>
<td>Session 8:</td>
<td>End follow-up data collection</td>
</tr>
<tr>
<td>Goals</td>
<td>Phases</td>
</tr>
<tr>
<td>----------------------------------------------------------------------</td>
<td>--------</td>
</tr>
<tr>
<td>Bill’s parent-stated goal and (parent-therapist collaborated goal); (GAS %)</td>
<td></td>
</tr>
<tr>
<td>1. Sleep through the night in own bed; (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>2. Go to sleep sooner (reduce SOL each night); (% of minutes per phase)</td>
<td></td>
</tr>
<tr>
<td>Holly’s parent-stated goals and (parent-therapist collaborated edit to goals); (GAS %)</td>
<td></td>
</tr>
<tr>
<td>1. Sleep in own bed (every night); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>2. Get required amount of sleep for age (every night); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>3. Get enough sleep (get developmentally appropriate amount of sleep each night); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>4. Fall asleep on own (in own bed); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>Keith’s parent-stated goal and (parent-therapist collaborated goal); (GAS %)</td>
<td></td>
</tr>
<tr>
<td>1. Get enough sleep (get developmentally appropriate amount of sleep each night); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>2. Fall asleep on own (in own bed); (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>3. Bedtime between 7:00-7:30 p.m. (% of nights per phase)</td>
<td></td>
</tr>
<tr>
<td>4. Sleep through the night in own bed (return to sleep during the night by himself); (% of nights per phase)</td>
<td></td>
</tr>
</tbody>
</table>

Note: BSL = Baseline; EOT = End of treatment; FU = Follow-up; SOL = Sleep onset latency; Developmental sleep norms according to<sup>a</sup> Hirshkowitz, et al., 2015; <sup>b</sup>Iglowstein, Jenni, Molinari, & Largo, 2003; <sup>c</sup>Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2013.
<table>
<thead>
<tr>
<th>Dependent Variables</th>
<th>Holly</th>
<th>Bill</th>
<th>Keith</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Actigraphy</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>SOL (mean min.; SD)</td>
<td>43.5; (39.72)</td>
<td>13.81; (17.89)</td>
<td>31.5; (16.98)</td>
</tr>
<tr>
<td>TST (mean min.; SD)</td>
<td>399.25; (68.06)</td>
<td>491.27; (75.69)</td>
<td>475.25; (48.32)</td>
</tr>
<tr>
<td>SE (%; SD)</td>
<td>60.03%; (10.10)</td>
<td>81.08%; (10.73)</td>
<td>74.75%; (11.09)</td>
</tr>
<tr>
<td>WASO (mean min.; SD)</td>
<td>192; (69.2)</td>
<td>91.27; (74.10)</td>
<td>132.5; (102.86)</td>
</tr>
<tr>
<td>TIB (mean min.; SD)</td>
<td>667.25; (53.74)</td>
<td>605.18; (40.07)</td>
<td>642; (70.24)</td>
</tr>
<tr>
<td><strong>CSHQ-A Subscale raw scores</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep onset delay (range: 1-3)</td>
<td>1</td>
<td>N/A</td>
<td>1</td>
</tr>
<tr>
<td>Sleep duration (range: 3-9)</td>
<td>3</td>
<td>N/A</td>
<td>3</td>
</tr>
<tr>
<td>Night wakings (range: 3-9)</td>
<td>5</td>
<td>N/A</td>
<td>5</td>
</tr>
<tr>
<td>Bedtime resistance (range: 6-18)</td>
<td>14</td>
<td>N/A</td>
<td>6</td>
</tr>
<tr>
<td>Sleep disordered breathing (range: 3-21)</td>
<td>4</td>
<td>N/A</td>
<td>3</td>
</tr>
<tr>
<td><strong>CSHQ-A Total scores</strong></td>
<td>56</td>
<td>N/A</td>
<td>40</td>
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<tr>
<td>Sleep diary</td>
<td></td>
<td></td>
<td></td>
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<tr>
<td>SOL (mean min.; SD)</td>
<td>18.75; (7.5)</td>
<td>17.72; (22.06)</td>
<td>26.25; (22.5)</td>
</tr>
<tr>
<td>TST (mean min.; SD)</td>
<td>652.5; (46.63)</td>
<td>550.90; (48.41)</td>
<td>577.5; (25.98)</td>
</tr>
<tr>
<td>SE (%)</td>
<td>97.18%</td>
<td>87.85%</td>
<td>94%</td>
</tr>
<tr>
<td>WASO (mean min.; SD)</td>
<td>0; (0)</td>
<td>28.63; (32.48)</td>
<td>11.25; (22.5)</td>
</tr>
<tr>
<td>TIB (mean min.; SD)</td>
<td>671.25; (49.36)</td>
<td>625.90; (11.79)</td>
<td>615; (36.74)</td>
</tr>
<tr>
<td>FNA (mean freq.; SD)</td>
<td>0; (0)</td>
<td>0.72; (0.94)</td>
<td>0.5; (1)</td>
</tr>
<tr>
<td>DSO (mean freq.; SD)</td>
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<td>0; (0)</td>
<td>0; (0)</td>
</tr>
<tr>
<td><strong>Behavior diary</strong></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>UC-S (mean min.; SD)</td>
<td>652.5; (46.63)</td>
<td>0; (0)</td>
<td>0; (0)</td>
</tr>
<tr>
<td>UC-S (location)</td>
<td>Parents’ bed; None</td>
<td>None</td>
<td>Parents’ bed; None</td>
</tr>
<tr>
<td>N-D S (mean freq.; SD; location)</td>
<td>0; None</td>
<td>0; None</td>
<td>0; None</td>
</tr>
<tr>
<td>BR (mean scores; SD)</td>
<td>15; (0)</td>
<td>0.36; (0.23)</td>
<td>0.25; (0.23)</td>
</tr>
<tr>
<td>DBC (mean scores; SD)</td>
<td>1.25; (0.5)</td>
<td>1.18; (0.98)</td>
<td>0.5; (0.57)</td>
</tr>
</tbody>
</table>

*Note: BSL = Baseline; EOT = End of treatment; FU = Follow-up; SOL = Sleep onset latency; TST = Total sleep time; SE = Sleep efficiency; WASO = Waking-up after sleep onset; TIB = Time in bed; CSHQ-A = Children Sleep Habits Questionnaire Abbreviated; FNA = Frequency of night awakenings; DSO = Dependent sleep onset; UC-S = Undesired co-sleeping; N-D S = Non-designated sleep; BR = bedtime resistance; DBC = Disruptive behavior composite; N/A = Not available; INC = Incomplete data due to software malfunction.*
### Marital Impact Variable, Kansas Marital Satisfaction Scale (KMSS)

<table>
<thead>
<tr>
<th>Scores</th>
<th>Holly's Parents</th>
<th>Bill's Parents</th>
<th>Keith's Parents</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>BSL</td>
<td>EOT</td>
<td>FU</td>
</tr>
<tr>
<td>KMSS total score</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Maternal score</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(range: 3-21)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17</td>
<td>17</td>
<td>18</td>
<td></td>
</tr>
<tr>
<td>Paternal score</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>(range: 3-21)</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>17</td>
<td>17</td>
<td>18</td>
<td></td>
</tr>
</tbody>
</table>

*Note: BSL = Baseline; EOT = End of treatment; FU = Follow-up; KMSS = Kansas Marital Satisfaction Scale.*
Table 7

Parent’s Consumer Satisfaction Questionnaire (PCSQ)

<table>
<thead>
<tr>
<th>Questions</th>
<th>Parent Scores</th>
</tr>
</thead>
<tbody>
<tr>
<td>1. The major problem(s) that originally prompted me to begin treatment for my child is (are) at this point</td>
<td>7 7 7</td>
</tr>
<tr>
<td>2. My child’s problems that have been treated during the study are at this point</td>
<td>7 7 7</td>
</tr>
<tr>
<td>3. My child’s problems that have <em>not</em> been treated during the study are</td>
<td>4 5 4</td>
</tr>
<tr>
<td>4. My feelings at this point about my child’s progress are that I am</td>
<td>7 7 7</td>
</tr>
<tr>
<td>5. To what degree has the treatment program helped with other general personal or family problems not directly related to your child?</td>
<td>6 5 6</td>
</tr>
<tr>
<td>6. At this point, my expectation for a satisfactory outcome of the treatment is</td>
<td>7 7 6</td>
</tr>
<tr>
<td>7. I feel the approach to treating my child’s sleep problems in the home by using this type of parent implemented treatment program is</td>
<td>7 7 7</td>
</tr>
<tr>
<td>8. Would you recommend the treatment program to a friend or relative?</td>
<td>7 7 7</td>
</tr>
<tr>
<td>9. How confident are you in managing current sleep problems in the home on your own?</td>
<td>7 7 1*</td>
</tr>
<tr>
<td>10. How confident are you in your ability to manage future sleep problems in the home using what you learned from this study?</td>
<td>7 7 7</td>
</tr>
<tr>
<td>11. My overall feeling about the treatment program for my child and family is</td>
<td>7 7 7</td>
</tr>
</tbody>
</table>

Note: *Adapted from Helping the Noncompliant Child (2nd ed.) by Robert J. McMahon and Rex L. Forehand. Copyright 2003 by The Guilford Press. Permission to photocopy this form is granted to purchasers of this book for personal use only. Research questionnaire was rated using a 7-point Likert scale (1 = Very dissatisfied; 7 = Very Satisfied) and response options varied by question (example: questions 1, 2, and 3 offer 1 = Considerably worse; 7 = Greatly improved); *Questions 8 and 9 are worded and scored in reverse order and may be responsible for this score inconsistency.
Frequency of Undesired Co-sleeping

Figure 1. Frequency of undesired co-sleeping per night with parents across children.
Duration of Undesired Co-sleeping

Figure 2. Duration of undesired co-sleeping with parents across children.
Figure 3. Duration of bedtime resistance across children.
Disruptive Behavior Composite

Figure 4. Disruptive behavior composite scores across children.
Figure 5. Frequency of night awakenings across children.
Figure 6. Frequency of dependent sleep onset (DSO) per night across children.
Appendix A
Sample sleep diary data sheet.

Instructions: Please complete the sleep diary during the evening and morning each day within the same 24-hour period. The same person should complete all diaries. Record the date and the following information:

1. Mark the time your child gets into bed with a down arrow “▼”;
2. Mark the time your child gets out of bed with an up arrow “▲”;
3. Shade in boxes when your child is asleep (small box = 15 minutes; large box = 1 hour);
4. Mark a “W” next to the arrow if your child was awaken by a parent or alarm; mark an “S” if your child awakened by his/herself; and
5. Mark a “D” if your child falls asleep dependently (with parental presence) or an “I” if your child falls asleep independently (without parental presence).
Appendix B
Sample sleep related behavior diary data sheet.

**Instructions:** Please complete the sleep related behavior diary during the evening and morning each day within the same 24-hour period. The same person should complete all diaries. Mark a tally in the corresponding box and record the duration of each instance the child engages in a disruptive behavior during the bedtime routine and once placed in bed, undesired co-sleeping, and sleep in a non-designated area. Record the date, the time the child gets ready for bed, the time and reason for removing the actigraphy, and the locations of undesired co-sleeping and non-designated sleep. Please use the following definitions:

1. Disruptive behaviors are defined as one or more verbal protests and/or physical protests a night when instructed by a parent to go to bed or go to sleep;
2. Sleeping in a non-designated area is defined as one or more occurrences of sleeping in a location other than the child’s bed during a 24 hour period; and
3. Co-sleeping is defined as unwanted sleeping with a parent or sibling in any room during a 24-hour period.

<table>
<thead>
<tr>
<th>Date</th>
<th>Time child gets ready for bed</th>
<th>Total duration of disruptive behaviors during bedtime routine</th>
<th>Total duration of disruptive behaviors once child is in bed</th>
<th>Time parent(s) went into bedroom after “lights out” (including chosen every 15 min.)</th>
<th>Total number of times child came out of bedroom once child is put to bed</th>
<th>Total number of times child co-slept with parent(s) or sibling (unattended)</th>
<th>Duration child co-slept with parent(s) or sibling (attended)</th>
<th>Location of undesired co-sleeping with child</th>
<th>Location of non-designated area (e.g. couch, crib, floor, etc.)</th>
<th>Time actigraphy is taken off and reason</th>
</tr>
</thead>
<tbody>
<tr>
<td>May 6, 2023</td>
<td>8:30 pm</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
</tr>
<tr>
<td>May 7, 2023</td>
<td>8:30 pm</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
</tr>
<tr>
<td>May 8, 2023</td>
<td>8:30 pm</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
</tr>
<tr>
<td>May 9, 2023</td>
<td>8:30 pm</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
<td>[ ]</td>
</tr>
</tbody>
</table>
Appendix C
Sample integrity-monitoring data sheet.

**Instructions:** Record parent responses as reported in the corresponding boxes. Record “Y” for “yes”, “N” for “no”, or “N/A” for not applicable to the context.

<table>
<thead>
<tr>
<th>Tri-weekly phone-check</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Date and time of call (e.g., 8/14/13; 8:00am)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Indicate the caller’s initials</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Parent answer the phone (Y, N, or N/A)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Return call by 5:30 p.m. (Y, N, or N/A)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Bedtime Fading (BF)</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Was BF implemented? (Y, N, or N/A)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time placed child in bed last night?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time checked to see if child was sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>How did you determine if your child was sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>What did you do after your child was or was not sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

<table>
<thead>
<tr>
<th>Response cost (RC)</th>
<th>1</th>
<th>2</th>
<th>3</th>
<th>4</th>
<th>5</th>
<th>6</th>
<th>7</th>
</tr>
</thead>
<tbody>
<tr>
<td>Was RC implemented? (Y, N, or N/A)</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time child was taken out of bed and bedroom?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>What did your child do during that time?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time returned child to bed last night?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Time checked to see if child was sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>How did you determine if your child was sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>What did you do after</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>your child was or was not sleeping?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>-------------------------------------</td>
<td>---</td>
<td>---</td>
<td>---</td>
<td>---</td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>How many times was RC repeated?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>What time did your child finally fall asleep?</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

Call #

Notes:

Barriers:

Solutions:

Call #

Notes:

Barriers:

Solutions:

Call #

Notes:

Barriers:

Solutions:

Call #

Notes:

Barriers:

Solutions:

Call #

Notes:

Barriers:

Solutions:
Appendix D

Sample vignettes of sleep patterns and related behaviors.

Please complete the sleep diary and sleep related behavior diary using the information presented below. If you have any questions, please do not hesitate to ask the investigator before leaving your session.

**Vignette 1.**

On Saturday 6/22/13, James went to bed by himself at 9:00 p.m. and fell asleep at 9:45 p.m., alone. He awoke on his own at 12:30 a.m. and was put back to bed at 1:45 a.m. by his mother. James fell asleep at 2:00 a.m. by himself and slept till his father woke him at 7:30 a.m..

**Vignette 2:**

On Sunday 6/30/13, Chelsea was put to bed by her father at 8:30 p.m. and fell asleep at 10 p.m. with her father in the room. Chelsea awoke at 2:30 a.m. and went to sleep by herself around 3:30 a.m.. She woke up at 5:00 a.m. on her own, but did not leave her bed until 6:00 a.m.. Chelsea fell asleep in the car on the way to daycare from 7:30 a.m. till 8:00 a.m. and woke-up independently. On the way home from daycare she slept in the car from 5:30 p.m. till her father woke her up at around 6:35 p.m..
(a) Bedtime procedures:

(i) Follow your regular bedtime routine and place your child in his or her bed at the initial adjusted bedtime.

(ii) After 15 minutes, check to see if your child is awake by standing within one foot of the bed and whispering his or her name. If there are no observable motor or verbal responses (i.e. opening eyes, talking, etc.) it is safe to assume your child is asleep.

(iii) If you determine your child is awake (i.e. he/she responded to your whisper), implement response cost.

(b) Response cost:

(i) Remove your child from the bed and bedroom and allow him or her to partake in activities not related to sleep for 30 minutes, such as playing with toys or watching T.V.

(ii) At the end of 30 minutes, return your child to his or her bed.

(c) Repeat the procedure if your child does not fall asleep within 15 minutes of being returned to bed.

(d) Address night time awakenings by returning your child to his or her bed with minimal parental attention and interaction.

(e) Bedtime fading:

(i) Adjust the bedtime for the following night according to your child’s previous time of sleep onset by 15 minutes. For example, if your child’s initial bedtime was 10:30pm and he/she fell asleep within 15 minutes of being placed in bed, fade the bedtime to 10:15pm. If your child did not fall asleep within 15 minutes of the initial bedtime, push back the bedtime to 10:45pm.