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A Systematic Review of Behavioral Approaches to Treat Pediatric Sleep Disturbances

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Abstract

This paper begins with an overview of the history of behavioral treatments used to treat pediatric sleep disturbances. Next, the paper analyzes the results of a systematic literature conducted to identify trends in pediatric sleep disturbance research. Only peer-reviewed, experimental studies with participants ages 2-12, designed to treat sleep refusal using extinction, graduated extinction, or bedtime routine procedures were included. A total of 23 studies were included in the analysis; 12 of the 23 studies were extinction, 7 were graduated extinction, and 4 were bedtime routine. Data related to authors, year published, number of participants, participant ages, participant diagnoses, independent variable, dependent variable, experimental design, follow up measures, treatment measures, and treatment outcome were extracted. Trends related to the inclusion of functional analysis methodology, treatment acceptability measures, and long-term follow up measures are discussed. Trends related to demographic information and prevalence are also presented. The results highlight the need for experimentally sound studies that use single case methodology, include replications, collect objective measures, and include follow up measures at least one year post-treatment.

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Chapter I: Introduction and Review of Literature

Pediatric sleep disturbances are common. The prevalence literature shows that approximately 25% of infants and toddlers experience some form of sleep disturbance (Bixler, Kales, Scharf, Kales, & Leo, 1976; Jenkins, Bax, & Hart, 1980; Lozoff, Wolf, & Davis, 1985; Richman, 1981; Richman, Stevenson, & Graham, 1975). Sleep disturbances describe a variety of sleep problems and can be classified into two categories: parasomnias and dysomnias. Parasomnias include sleep disorders that interrupt sleep, such as sleepwalking and night terrors (Mindell, 1993). Dysomnias describe sleep disorders related to initiating and maintaining sleep, as well as daytime fatigue (Mindell, 1993). Dysomnias include narcolepsy, obstructive sleep apnea, and adjustment sleep disorders. Adjustment sleep disorders are environmentally-influenced sleep problems related to delayed sleep onset and bedtime resistance, affecting approximately 20-30% of infants and toddlers (Blader, Koplewicz, Foley, & Abikoff, 1997; Mindell, Kuhn, Lewin, Meltzer, Sadeh, & Owens, 2006; Owens, Spirito, McGuinn, & Nobile, 2000). This paper will focus on infant toddler sleep disturbances (ITSD) associated with difficulties in settling at bedtime and delayed sleep onset, the most common pediatric sleep problems (Johnson, 1991; Lozoff et al., 1985).

Research shows that pediatric sleep disturbances have serious consequences. Parents of children with sleep difficulties are less likely to sleep throughout the night themselves. Lack of sleep may be related to Rickert and Johnson's (1988) findings that show ITSD is correlated with low family satisfaction, fatigue, and stress. Further, children with sleep disturbances are more likely to be abused (Bax, 1980). Although parents are often told their child will "outgrow" the problem, this is not always the case, as sleep problems often persist. Kataria, Swanson, and

Trevathan (1987) surveyed a sample of children and found that sleep problems persisted after three years for approximately 84% of the children with sleep problems. Johnson, Chilcoat, and Breslau (2000) assessed 717 children at age 6 and 11 and found that mothers' reports showed that children at age 6 with sleep problems were more likely to have anxiety-depression at age 11. The research also showed that sleep deprivation is related to reduced alertness (Sadeh, Gruber, & Raviv, 2003). Reduced alertness may result in poor responding and low achievement in the educational setting. Left untreated, pediatric sleep disturbances might have long-lasting adverse effects for the child and the family.

Empirical support for ITSD treatments is lacking. Medical treatment typically involves prescribing drugs such as antihistamines, chloral hydrate, benzodiazepines, and melatonin (Kuhn & Weidinger, 2000). Pharmacological treatments are temporary fixes, however, and are often ineffective long term. Further, over 70% of prescription drugs in the United States are not labeled for children, as they have not been tested on children (Blumer, 1999). This causes physicians to rely upon experience and anecdotal evidence to guide their treatment of ITSD (Stojanovski, Rafia, Balkrishnan, & Nahata, 2007). Relying upon treatment that is not based upon controlled experimental research may have serious repercussions for children. For example, because pediatric doses are estimated, side effects are not uncommon. It is also important to note that currently, there is no medication to treat pediatric insomnia in the United States.

Behavioral Sleep Medicine (BSM) treats sleep disorders using empirically supported interventions. Behavioral approaches include unmodified extinction, graduated extinction, positive bedtime routines, and scheduled awakenings. The purpose of this paper is to review well-established behavioral procedures designed to eliminate ITSD. The paper will first present an overview of the history of behavioral interventions targeting delayed sleep onset and frequent night awakenings. The paper will then review the “Excuse Me” Drill (EMD), a multicomponent behavioral procedure designed to reduce the frequency of incompatible bedtime behaviors and to promote independent sleep initiation. Next, the paper will describe a systematic literature review on behavioral interventions designed to treat pediatric sleep disturbances. Finally, the paper will conclude with a synthesis of the systematic review results and suggestions for future research.

Extinction Procedures

Unmodified extinction. Unmodified extinction requires caregivers to put their child to bed, leave the room, and withhold attention when their child engages in behaviors that are incompatible with sleep. The procedure is implemented following the development of a consistent bedtime routine (explained in more detail below). Williams (1959) conducted the first published study using unmodified extinction for sleep problems in young children. The participant was approximately 18-months-old and engaged in bedtime tantrums that were suspected to be maintained by attention. The tantrums consisted of whining, crying, and screaming. The child was screened for physical abnormalities before participating in the study. The study used a B only design.

The family was instructed to first complete the child’s usual bedtime routine before placing the child in bed and leaving the room. The parent was instructed to record the frequency and duration of tantrums. The data showed that the child tantrumed for 45 minutes on the first night of intervention. Tantrum duration gradually decreased and ultimately stopped by the tenth day. Approximately one week after treatment, the child’s aunt was present at night and the child again tantrumed; the aunt provided attention to the child during this episode, which led to an increase in bedtime tantrums. A second extinction series ensued and tantrums were eliminated by the ninth night.

Graduated extinction. Graduated extinction involves withholding reinforcement for increasingly longer periods independent of responding. Rolider and Van Houten (1984) trained parents to implement a graduated extinction procedure to reduce the frequency of bedtime crying through a multiple baseline across participants' design. The participants consisted of three male toddlers aged 20-30 months. During baseline, parents were instructed to use a stopwatch to record the beginning and end time of each crying episode, the latency between when their child started crying, and when they picked their child up. The primary dependent variable was the number of minutes the child engaged in crying behavior after being placed in bed.

During baseline, parents were instructed to put the child to bed in his own bedroom and to leave the room. If the child began to cry, the parents were asked to tend to their child as they normally would and to leave the room once the crying stopped. The experimenters reviewed the baseline data with the parents to show them how long they ignored their child and asked the parents to ignore their child for the same length of time. Two of the three sets of parents agreed to comply while the parents of the third participant chose to ignore their child for 5 minutes longer than suggested.

During the graduated extinction phase, parents were again instructed to put their child to bed and to leave the room. This time, however, parents were instructed to increase time spent ignoring by 5 minutes more every two nights. During the first two nights of the intervention, two of the three families ignored their child for a total of 10 minutes and the third family ignored their child for total of 15 minutes. By the end of the intervention, two of the three families ignored their child for 25 minutes and the third family ignored their child for 30 minutes. The experimenters provided positive feedback contingent upon compliance with the procedure via telephone calls during the first four days of the intervention. After the first four days, the experimenters praised the parents intermittently.

The results of the study indicated that graduated extinction procedure was effective in reducing the duration of bedtime crying episodes. During the baseline condition, the average duration of crying for each participant lasted less than 22 minutes (Rolider & Van Houten, 1984). By Days 4 to 9 of the intervention phase, the average duration of crying episodes decreased to 0 or near 0 levels for each child (Rolider & Van Houten, 1984). None of the children engaged in bedtime crying at 55-, 65-, and 70-day follow ups. The results showed that the duration of bedtime crying was reduced by gradually increasing the amount of time parents ignored their child. The authors suggested that the procedure was effective because parent attention is systematically faded until time spent ignoring exceeds the duration of tantrum behavior.

Similarly, Adams and Rickert (1988) sought to compare the effectiveness of positive routines and graduated extinction in reducing bedtime tantrums. Thirty-eight children were recruited for participation through local advertisements and public notices. To be considered for inclusion, the child needed to have tantrum-related sleep problems at least once per week with no problems falling asleep. The researchers met with parents to complete the Dyadic Adjustment Scale (DAS), an assessment measuring relationship adjustment of married couples. The parents were instructed to collect data for seven weeks on when their child was put to bed, the time the tantrum began, the topography of behaviors observed, the time the tantrum ended, the time the child was noticed to be asleep, and the number of times the child left the bed. The experimenters monitored the data weekly via phone calls with the parents.

Following baseline, the families were randomly assigned to the positive routines group, the graduated extinction group, or the control group. Parents assigned to the positive routines group were instructed to select four to seven preferred activities for their child to complete preceding bedtime. The parents were instructed to provide their child with vocal praise after each activity. The parents then guided the child to the bathroom to brush his or her teeth and to use the toilet. Next, the parent helped the child into pajamas before reading a story. After story time, the parent provided tactile stimulation, such as scratching their child’s scalp or massaging his or her child’s back in their bed. The routine ended with a kiss goodnight. Initially, the bedtime was arbitrarily set later in the evening to increase the likelihood of quick sleep onset. The routine was implemented 5 to 10 minutes earlier each week until an appropriate bedtime was achieved.

Parents assigned to the graduated extinction group were instructed to place their child in bed and to ignore their child’s cries for a specified period. If the child began to cry, the parent was instructed to ignore the behavior for 10 minutes. Once the 10-minute period ended, the parent was permitted to check on the child for a short interval (15 seconds or less). Each week, the ignore interval required before checking on the child increased by 10 minutes. By the end of the sixth week of treatment, the parent ignored the child for 60 minutes before checking in.

Parents assigned to the control condition were informed that their child would “outgrow” the bedtime tantrums. Parents were also informed that if the tantrums did not decrease following the intervention, their child would have the opportunity to participate in another treatment.

Parents in each of the three groups were instructed to collect data as they had during the baseline phase.

The results showed that children in the two experimental groups engaged in fewer bedtime tantrums than the children in the control group. The positive routines and graduated extinction interventions were equally effective in reducing tantrum behavior; however, the changes in behavior were observed much faster in the positive routines group. Additionally, parents in the positive routines group reported higher marital satisfaction, according to the DAS, which was administered pre- and post-treatment.

Scheduled awakenings. McGarr and Hovell (1980) investigated whether parental attention reinforced sleep behavior in a 3-month-old infant who awakened frequently throughout the night. The participant was screened to rule out medical issues related to the concern. A 15-day baseline with normal bedtime routines required the parent to record total sleep duration each night. Based on data from baseline, the parent was instructed to wake the child 15-30 minutes prior to the previous night's wake up times. If the child was sleeping or quiet during scheduled awakenings, the parent was instructed to play pleasant music on the music box, change the child's diaper (if needed), feed, and cuddle the child for a few minutes before placing the child back in bed. The wake time criterion was increased by 15 minutes later after the completion of two consecutive night awakenings. A return to baseline saw the child sleeping less and crying more. Thus, the parent chose to reintroduce the treatment. The child began sleeping for longer, uninterrupted durations (six or more hours) compared to all previous phases combined. The parent stopped adhering to procedure protocols when the child began sleeping through the night. Toward the end of the study, however, the child awoke earlier than the scheduled wake up time.

When this occurred, the parent played music and provided attention before the child cried. The results indicated that the intervention was effective in increasing the length of uninterrupted sleep and decreasing the frequency of crying episodes. The authors suggested that this procedure may be effective because it teaches the child to sleep for longer periods and parent attention is gradually faded. This explanation is flawed, however, because it relies on the assumption that parent attention functions as a reinforcer for sleep behavior.

Johnson and Lerner (1985) sought to determine if scheduled awakenings would reduce the frequency of spontaneous awakenings and crying episodes for 12 children ages 6 months to 2 years and 6 months. The participants were recruited through a local newspaper advertisement. The experimenters interviewed the parents to assess the severity of spontaneous night awakenings and crying episodes. Medical screening ruled out possible physical causes for the awakenings and crying. During baseline, parents were instructed to record for 1 week when their child was placed in bed and when they awoke. During intervention, parents were instructed to wake their child 15 minutes before their child's average awakening time, which was based on baseline data. If the parents were asleep, they were instructed to set an alarm. The authors spoke with the parents over the phone semi-weekly to review the data and evaluate compliance with the procedure. The scheduled awakenings gradually reduced from an average of two awakenings per night to none. Parents were instructed to continue collecting data on spontaneous night awakenings. A return to baseline was employed for 3 of the 12 participants after only 3 weeks of treatment due to illness, a disruption in data collection, and unstable data. The treatment effects maintained for six of the participants at follow up collected 3 months later.

In another study, Rickert and Johnson (1988) sought to compare the effectiveness of scheduled awakenings and systematic ignoring on reducing nighttime awakenings. Systematic ignoring involves withholding attention for specified intervals. Thirty-three children aged 6 to 54 months were recruited through a local newspaper ad. Each participant awoke at least once per night. The participants were divided into three groups: scheduled awakenings, systematic ignoring, and control. Experimental group parents were instructed to record for 1 week when their child awoke each night. Control group parents were informed that their child would "outgrow" night awakenings. The control group parents were instructed to continue tending to their child as they normally did. Control group parents were also instructed to record for the 8 weeks of the study when their child awoke each night.

The experimenters used the baseline data to identify timeframes for the parents to wake their child. During scheduled awakenings, the parents were instructed to respond to their child as they previously did by rocking, feeding, or changing diapers. Throughout the study, parents were requested to record when the child went to bed, when the child awoke during the night, and at what time the child awoke to start his or her day. The fathers of three of the participants collected data for 1 day during baseline, treatment, and follow up to measure reliability. Parents in the systematic ignoring group were instructed to withhold attention during their child's spontaneous awakenings. If their child began to cry, parents were asked to ensure the child's safety and to avoid eye contact while changing the child's diaper. Parents in each group reported their data to the experimenters via phone call twice per week.

The results showed that children assigned to the experimental groups awoke less frequently than children assigned to the control group. Although decreases in night awakenings were observed in both experimental groups, the systematic ignoring group observed faster decreases than children in the scheduled awakening group. Overall, the number of spontaneous night awakenings decreased for children in all three groups. The treatment effect maintained during follow-up measures recorded 3 and 6 weeks after the intervention.

Faded or scheduled bedtime. Piazza, Fisher, and Scherer (1997) sought to replicate and extend a prior study (Piazza & Fisher, 1991) by comparing the effectiveness of faded bedtime with response cost (FBRC) and bedtime scheduling. Fourteen children with developmental disabilities were referred by an inpatient facility specializing in the assessment and treatment of individuals with severe destructive behavior. Each of the participants slept less than 10% of the expected time suggested for their chronological age. The observers were instructed to record whether the child was awake or asleep during 30-minute interval checks. The observers also collected data on when participants fell asleep at night, the exact time the participants awoke in the morning, the time the participants spontaneously awoke throughout the night, and when the participants fell back asleep after waking up at night. During the baseline phase, the observers prompted the children to go to bed once they completed their bedtime routines. If a child got out of bed, the observers prompted the child to return to bed at 30-minute intervals.

Seven participants were randomly assigned to the FBRC group and seven participants were assigned to the scheduled bedtime group. The experimenters used the baseline data to calculate the average sleep onset times for participants in the FBRC group. The experimenters added 30 minutes to the average sleep onset time to set the initial bedtime to increase the likelihood of drowsiness. If the child did not fall asleep within 15 minutes of being placed in bed, the staff implemented the “response cost component.” The response cost component consisted of removing the child from bed for 1 hour. During this time, the child was given free access to toys and television but was not permitted to sleep. At the end of the hour, the child was placed back in bed. The procedure was repeated until the child fell asleep within 15 minutes of being placed in bed. Bedtime schedules were faded by adjusting the child's bedtime by 30 minutes. For example, if the child fell asleep within 15 minutes of being placed in bed, bedtime was adjusted to 30 minutes earlier the subsequent night. If the child did not fall asleep within 15 minutes of being placed in bed, the child was placed in bed 30 minutes later the subsequent night.

Children in the scheduled bedtime group were put to bed after they completed their bedtime routines. The children were also woken up at specific times in the morning based on their baseline data. The participants were not given an opportunity to sleep during the day unless it was age-appropriate to nap. If the child did not fall asleep during naptime, sleep was prevented until the next designated sleep time.

The results showed that the FBRC procedure was more effective than the scheduled bedtime procedure in reducing the occurrence of disturbed sleep. The mean average of disturbed sleep for participants in the scheduled bedtime group was 1.37 hours during the baseline phase and 1.10 hours' post-treatment (Piazza, Fisher, & Scherer, 1997). The mean average of disturbed sleep for participants in the FBRC group was 1.44 hours during the baseline phase and 0.53 hours during the post-treatment phase (Piazza et al., 1997). The study added support to Piazza and Fisher (1991), demonstrating that the FBRC is an effective procedure in reducing the frequency of night wakings and inappropriate bedtime behaviors. It is important to note, however, that the response cost component of this study is a misnomer. True response cost procedures involve contingent removal of a previously earned reinforcer. In this study, the experimenters prevented sleep, though sleep was not previously earned, nor was sleep identified as a functional reinforcer.

The "Excuse Me" Drill

Thus far, the paper has reviewed extinction, faded or scheduled bedtimes, positive bedtime routines, and scheduled awakenings. Although extinction is well established in the literature, it is often difficult for both parents and children to endure. Graduated extinction is a supported, systematic procedure, easier for parents, but it is not based on child success. FBRC relies upon the assumption that sleep functions as a reinforcer and it does not teach the child pro-sleep behavior. Kuhn (2011) designed the "Excuse-Me" Drill to address the needs of families struggling with ITSD. The EMD is a response-dependent procedure that promotes independent sleep initiation skills and reduces maladaptive bedtime behaviors. This procedure is designed to treat children with delayed sleep-onset and difficulties settling at night using reinforcement and extinction.

The procedure begins by teaching the child to fall asleep in his or her bedroom. Parents are encouraged to place their child in bed drowsy and to continue implementing routine sleep-related activities such as lying next to the child in bed, rocking, and singing. The goal of this component is to help the child become comfortable in his or her sleep environment. Kuhn noted that between 5% and 10% of children do not require further treatment after completing this step (Kuhn, 2011). The next step requires the caregiver to select a start night for the EMD to begin. Kuhn recommends choosing a time when both parents are present and have the time to dedicate their efforts to implementing the protocol with fidelity.

The next component of the EMD involves temporarily delaying bedtime and managing the sleep schedule. By delaying bedtime, the child is more likely to be tired, thus increasing the likelihood of quick sleep onset and decreasing the likelihood of bedtime resistance. To select an appropriate bedtime, Kuhn (2011) recommended parents put their child to bed 20 to 30 minutes later than their average sleep onset time, based on the last 3 or 4 nights. Once the child begins falling asleep independently, parents may put their child to bed 15 minutes earlier every 2 days, or until the desired bedtime goal is reached. Kuhn emphasized the importance of waking the child at their usual morning wake time and maintaining their nap schedules. This strategy eliminates the opportunity for the child to make up for lost sleep, increasing the likelihood that the child will fall asleep without resistance at bedtime.

At this point, the child is consistently falling asleep in his or her bedroom. The point of the EMD, however, is to promote independent sleep initiation. Children that warrant treatment typically cry, call out, and get out of bed when the parent leaves the room. To begin, the parent puts the child to bed following the usual bedtime routine. Next, the parent explains to the child, "I have to go check on something" (or similar language) and leaves the room. After 2 to 3 seconds, before the child can engage in incompatible sleep behaviors, the parent returns to find the child lying in bed. The parent then offers specific praise, "Good job staying in your bed!" If the child remains successful at remaining in bed while the parent is out of the room, the length of time between parental visits increases. Kuhn advises parents not to enter their child's room unless the child is quiet, calm, and lying down in bed. Kuhn also emphasized the importance of the child being alone when he or she falls asleep. If the child engages in incompatible sleep behaviors such as making demands, getting out of bed, or calling out to parents, parent presence should be withheld. If the child leaves the room, the parents should physically guide their child back to bed, limit language to "go back to bed," and avoid making eye contact. Once the child is in his or her bedroom, the parent warns the child that if they leave the room again, the door will need to be closed. Kuhn advised parents to remove items that may cause harm to the child if he or she engages in maladaptive behavior with door secured shut. Once the child is quiet for 3 to 5 seconds, the parent should open the door immediately and put the child back in bed, if needed. This stage of the EMD is repeated until the child initiates sleep.

The underlying mechanism of the EMD can be explained using an analysis of stimulus function and motivating operations. The parent leaving the room serves as a stimulus signaling a worsening set of conditions for the child; the parent leaving is a reflexive motivating operation (CMO-R) (Michael, 1993). When the parent leaves the room, the value of conditioned negative reinforcement increases. The child is likely to engage in behaviors that have been correlated with termination of the aversive condition (parent out of room) and refrain from any behavior that would prolong the parent's absence (e.g., remaining calm in bed). When the parent leaves the room and quickly returns, the CMO-R is eliminated, thus abating behaviors such as crying and calling out, which historically resulted in parent presence. The schedule of parent presence is gradually thinned; the child no longer associates the parent leaving the room with a worsening set of conditions. Additionally, the child no longer engages in incompatible sleep behaviors, which increases the likelihood of independent sleep-initiation, the primary goal of the procedure.

Allen, Kuhn, Dehaai, and Wallace (2013) incorporated the EMD as a part of a behavioral treatment package designed to treat sleep disruptions in five children diagnosed with Angelman Syndrome. The EMD was included in this study to accommodate parents who declined to ignore their child's disruptive sleep behaviors. Disruptive sleep behaviors ranged from resistance to sleep, spontaneous night awakenings, and falling asleep in locations other than their specified sleep environment. The authors implemented a multiple baseline design across participants' experimental design. A reversal to the baseline condition was implemented for two of the five participants due to failure or inability to comply with the procedure. During the baseline condition, parents were asked to record for 2 to 6 weeks the time their child fell asleep and when their child awoke throughout the night.

The treatment package included three components: sleep environment, sleep schedule, and parent child interactions. The "sleep environment" component consisted of creating a sleep environment conducive to sleep. For example, parents were instructed to make their child's room dark, quiet, and to adjust the temperature, if needed. The "sleep schedule" component consisted of maintaining a consistent sleep-wake schedule for one week prior to intervention. Parents were instructed to delay their child's bedtime if their child had a long sleep onset time. If the child fell asleep within 15 to 20 minutes of being placed in bed, the child was put to bed 30 minutes earlier the subsequent night. The sleep schedule continued until an appropriate bedtime was achieved. During the "parent-child interaction" component, parents were asked to ignore inappropriate bedtime behavior such as crying and leaving the room. Parents that refused to ignore their child were instructed to implement the EMD.

Parents were asked to record sleep latency, total sleep time, the frequency and duration of spontaneous nighttime awakenings, and the total sleep time divided by the time spent in bed (sleep efficiency), daily. Parents were also asked to record the number of disruptive behaviors observed during the bedtime routine, the number of disruptive behaviors after "lights out," and the number of disruptive behaviors throughout the night. The dependent measures consisted of the disruptive behavior composite (DBC) and the independent sleep onset (ISO). The ISO was calculated by adding the total number of nights the child independently initiated sleep each night by seven and multiplied by one hundred. Each participant wore a MicroMini Motionlogger actigraph on their ankle to measure motor movement via accelerometer (Allen et al., 2013).

The results of the study indicated that the treatment package was effective in reducing disruptive bedtime behaviors for each participant. Before treatment, rates of disruptive behaviors ranged from 100 to 200 per night and each participant's DBC was increasing. Further, none of the participants initiated sleep independently (Allen et al., 2013). Rates of DBC decreased to zero or near zero levels by week 4 of treatment. Each participant initiated sleep independently after 1 week of treatment. The experimenters measured customer satisfaction using the Abbreviated Acceptability Rating Profile (AARP), an eight-item questionnaire rated on a Likert-type scale (Allen et al., 2013). The results showed that parents found the treatment to be effective and they would recommend it to others.

Synthesis of Results

This paper reviews some important findings in BSM. First, extinction is effective in treating ITSD (Williams, 1959). Extinction is a useful procedure as it produced rapid results. Graduated extinction may be a more appropriate treatment option for parents who report difficulties ignoring their child, however. Scheduled awakenings are an effective intervention to reduce frequent night awakenings (Johnson & Lerner, 1985; McGarr & Hovell, 1980; Rickert & Johnson, 1988). However, graduated extinction was found to decrease disruptive sleep behaviors quicker than scheduled awakenings (Rolider & Van Houten, 1984). The research shows that positive routines and graduated extinction are equally effective in reducing tantrum behavior; however, positive routines may produce quicker results (Adams & Rickert, 1989). In another study, Piazza et al. (1997) found that the faded bedtime with "response cost" (FBRC) procedure was more effective than scheduled bedtimes in reducing the occurrence of disturbed sleep time. This shows that putting a child to bed drowsy increases the likelihood of uninterrupted sleep. The reliance upon self-reporting in most the studies reviewed in this paper (Adams & Rickert, 1989; Johnson & Lerner, 1985; McGarr & Hovell, 1980; Rickert & Johnson, 1988; Rolider & Van Houten, 1984; Williams, 1959) jeopardizes the validity of the data. Future researchers should use independent observers or video technology in addition to self-reporting to strengthen experimental control.

Behavioral interventions designed to reduce ITSD are limited. Extinction-based procedures are most commonly used. Although the literature shows that extinction procedures are effective, extinction-only procedures are associated with low treatment acceptability. Further, extinction can be dangerous due to the increase in maladaptive behavior that is sometimes observed when reinforcement is removed. Extinction is also difficult to implement with fidelity; families must be cautious not to reinforce previously extinguished behaviors as this can result in the behavior becoming resistant to change. Interventions including both extinction and reinforcement components have proven to be effective and more acceptable in reducing pediatric sleep disorders.

Behavioral procedures are effective in treating pediatric sleep disorders but very few target independent sleep initiation. Unlike graduated extinction, the EMD procedure is response-dependent; parent presence is available contingent upon appropriate bedtime behavior. Over time, parent presence is faded until the child initiates sleep independently. With graduated extinction, the child is ignored for increasingly longer periods regardless of the child's success. Further, the EMD gradually increases sleep compatible behaviors instead of gradually reducing the frequency of crying. The EMD is understudied, however. Currently, only one controlled study using the EMD has been published (Allen et al., 2013). Before effort is devoted to future investigations of the EMD, it would be prudent to systematically measure the variables of the procedure, as this analysis does not currently exist. Parent presence consists of two variables, parent in room and parent out of room. It is unclear; however, which variable facilitates pro-sleep responding quicker. Additionally, it is unknown whether families value the EMD procedure or find it effective.

To better understand the literature, I propose to conduct a systematic review of the literature relating to pediatric sleep disturbances. The results of the search will guide a discussion regarding the strengths and weaknesses of the research and suggestions for future research.

Chapter II: Method

Procedure

A systematic review was conducted to identify trends in the literature on behavioral treatment of pediatric sleep disturbances. Only peer-reviewed, experimental studies with participants ages 2-12, designed to treat sleep refusal using established behavioral interventions were included in this analysis. Excluded materials included book chapters, due to the limited or non-existence peer-review, review articles, or descriptive studies that did not manipulate variables. Only extinction, graduated extinction, and bedtime routine interventions were included in the review. Interventions using scheduled awakenings were excluded from the review because these studies treat difficulties maintaining sleep and I am interested in bedtime resistance treatment. The terms “sleep problems,” “children,” “not infants,” and “extinction” were searched using the advanced Boolean search through EBSCO; this search produced 77 articles. Sixty-four of the 77 articles were excluded because they did not meet the inclusion criteria. The terms “sleep problems,” “children,” “not infants,” and “graduated extinction” were searched using the advanced Boolean search through EBSCO; this search yielded 30 articles. Twenty-three of the 30 articles were excluded because they did not meet the inclusion criteria. The terms “sleep problems,” “children,” “not infants,” and “bedtime routine” were searched using the advanced Boolean search through EBSCO; this search yielded 30 articles. Of these 30 articles, four met the inclusion criteria. One additional article was found by searching the reference sections of articles found in the searches previously described. A total of 24 articles met the inclusion criteria.

Data Analysis

I extracted the following components from the literature: authors, year published, number of participants, participant ages, participant diagnoses, independent variable, dependent variable, experimental design, follow up measures, treatment measures, and treatment outcome. I looked at the years the studies were published, the independent variables, and the authors to determine if certain types of interventions were being published more or less often during certain years by certain researchers. I looked at the dependent variables and treatment measures to determine whether objective measures were being collected. Information regarding participants, including age and diagnosis were analyzed to determine if particular age groups or diagnoses were being included or not included in the literature. By looking at the number of participants, experimental design, whether follow up measures were collected and when they were collected, I wanted to identify trends related to the quality and integrity of the studies. Finally, by looking at treatment outcomes, I wanted to determine if the treatments were effective in reducing disruptive sleep behaviors.

Chapter III: Results

Analysis of the literature yielded several important findings regarding experimental design. First, 16 of the 23 articles included single-case design methodology. Of these 16 studies, nine were extinction, five were graduated extinction, and two were bedtime routine (see Appendix A, Table 1 for a summary of design use across interventions). The seven studies that did not use single-case designs used pre-post designs. In these seven studies, statistical analyses for the group were presented as opposed to the individual. The results also show that 8 of the 16 single case design articles incorporated a withdrawal to baseline. Five of these studies were extinction, one was graduated extinction, and two were bedtime routine. The extinction withdrawal designs included ABAB, ABC, ACABAB and BAB; the graduated extinction designs included ABAB; the bedtime routine designs included ABC and ABCAB. The remaining 8 of the 16 single case designs used AB designs. Of these eight studies, four were extinction and four were graduated extinction (see Appendix A, Tables 2, 3, and 4 for article summaries).

The results show that follow up measures were prevalent, but rarely conducted a year or more post-treatment. Twenty studies included follow up measures. Of these 20 studies, three included follow up measures collected a year or more post-treatment. Of these three studies, all were extinction. Further, most the studies spent approximately 2 weeks in follow up (see Appendix B, Figure 1).

The results highlight some significant trends concerning treatment procedures and measures. First, functional analysis (FA) methodology was reported in six of the 23 studies. Of these six studies, five were extinction and one was graduated extinction. This is significant because FA methodology yields objective measures used to identify the function of behavior, and this information can inform treatment decisions. Second, all 23 studies included parent report measures. Parent report measures typically consisted of diary entries describing bedtime, the frequency and severity of problem behaviors, as well as the latency of sleep onset. Of the 23 studies, nine collected treatment acceptability measures. Of these nine studies, five were extinction, three were graduated extinction, and one was bedtime routine. Treatment acceptability measures generally asked the parent to rate the effectiveness of the procedure and whether they would recommend the procedure to a friend. Objective measures were not as prevalent in the research, however. Five of the 23 studies reported objective measures; 1 used audio recordings and 4 used actigraphy. Actigraphy measures typically consisted of total sleep duration, latency to sleep onset, and wake time (see Appendix A, Tables 5, 6, and 7 for summaries of treatment measures).

Overall, recruiting methods were similar across intervention types. Six of the studies report using advertisements and public notices; six of the studies reported referrals from clinics and health workers, including nurses, pediatricians, and community intervention programs; one study reported referrals from a school and another reported referral from head start teachers; one study used an independent market research firm to recruit; and 10 studies did not report who referred families to the researchers. Reporting recruiting methodology is important because it provides future researchers with effective, and in effective ways, to recruit participants (see Appendix A, Tables 8, 9, and 10 for summaries of prevalence).

Additional trends pertaining to prevalence were identified. First, 13 of the 23 studies were from the United States, five were from the Netherlands, three were from Australia, one was from Sweden and one was from England. Table 11 (Appendix A) shows that four of the 12 extinction studies are from the United States, five of the seven graduated extinction studies are from the United States, and all four of the bedtime routine studies are from the United States. Each of the five studies from the Netherlands were extinction based; four of these were extinction and one was graduated extinction. Second, patterns related to the publication years were found. Figure 2 (Appendix B) shows a cumulative record of articles published per year, per intervention type. The graph shows that the number of extinction studies flat lined after 2007 but graduated extinction and bedtime routine studies gradually increased from 2002 to 2015.

Chapter IV: Discussion

Several trends were identified in experimental design. For example, the results show that single-case design methodology is not uncommon in pediatric sleep disturbance literature; 16 of the 23 studies used single case designs. In single-case design methodology, the individual serves as their own control. This practice allows the researcher to demonstrate experimental control and to identify functional relations with more precision (Kennedy, 2005). Conversely, large-scale studies mask information about the individual with the use of statistical analyses. Normand (2016) argued that when the group's performance is averaged together, it makes it difficult to predict and control the behavior of the individual. Instead, statistical analyses measure the probability that a behavior will occur, though this probability is based on group behavior, not individual. Because functional relations are more informative to individual interventions than statistical relations are, future researchers of pediatric sleep disturbances should consider adopting single-case methodology.

Additional trends concerning methodological practices were identified. First, eight of the 23 studies incorporated a return to baseline. Of these eight studies, five were extinction, one was graduated extinction, and two were bedtime routine. A return to baseline helps to demonstrate a functional relation. The dependent variable is more likely to be controlled by the independent variable, as opposed to extraneous variables, when changes in the dependent variable are observed following the withdrawal and subsequent reapplication of the independent variable. AB designs were used in eight of the 23 studies. Of these eight studies, four were extinction and four were graduated extinction studies. AB designs show a snapshot of behavior change but a return to baseline is needed to demonstrate control of the independent variable. There are a few reasons why researchers might have chosen not to use withdrawal methodology. First, it is possible that families protested the inclusion of a withdrawal design due to fears of losing pro-sleep behaviors or delaying progress (Curfs, Didden, Sikkema & De Die-Smulders, 1999; McGarr & Hovell, 1980). It is also possible that the researchers found the inclusion of withdrawal methodology unethical due to the potential negative effects.

Future researchers may benefit from including multiple baseline designs when treatment withdrawal is not possible. In multiple baseline design methodology, two or more baseline conditions are implemented simultaneously. The independent variable is not removed once it is introduced. Instead, the independent variable is applied sequentially across the baselines to help the researcher identify patterns in responding (Kennedy, 2005). For example, during baseline, the frequency of calling out might be stable at about 10 episodes per night. When the intervention is applied, the frequency of calling out might decrease to an average of two episodes per night. If the remaining tiers of the baseline design remain unchanged, the changes in responding are likely attributable to the intervention. Multiple baseline design methodology was used in nine studies. Of these studies, four were extinction, three were graduated extinction, and one was bedtime routine. Eight of the studies used a multiple baseline design across participants; one study used a multiple baseline design across behaviors. Given the likelihood of carryover effects, BSM researchers should consider multiple baseline designs across persons as being an easier way of demonstrating control, though using functionally- and topographically-distinct behaviors in a multiple baseline across behaviors could demonstrate control (Barlow, Nock, & Hersen, 2009).

Another methodological trend in the research concerns the lack of long-term follow up measures. Of the 23 studies included in the review, only three of the studies included follow-up measures conducted at least 1 year post-treatment (see Appendix B, Figure 1). This finding is concerning because the literature shows that sleep problems may be long lasting (Kataria et al., 1987). Figures 1, 3, and 4 (Appendix B) show the percentage of studies that remain in each condition by month (see Appendix A Tables 12, 13, and 14 for the number of months spent in each condition). The data shows that most of the studies spend less than 2 weeks in follow up. Whether researchers can obtain an accurate measure of responding with less than two weeks of data is questionable because it often takes longer to establish stable patterns of responding. Long term follow ups conducted within 1-5 years' post-intervention may increase the likelihood of obtaining accurate accounts of treatment maintenance, particularly if those follow-ups measure other systemic issues like child health and well-being and family stress. Additionally, long-term follow up measures should include objective measures such as actigraphy to supplement parent reports. Parents might be more likely to participate in long-term follow ups if response effort is reduced. This might be accomplished by using mobile device technology to complete short nightly surveys, which are submitted to the researcher nightly. This practice is likely to result in more descriptive accounts of the behaviors over time. Treatment effects that maintain over long periods are likely to be associated with higher quality of care and increased treatment acceptability from families and health professionals.

Trends regarding consumer satisfaction were identified. For instance, of the 23 studies, 10 studies collected treatment acceptability measures. Of these 10, six were extinction, three were graduated extinction, and one was bedtime routine. Standardized instruments were used in four of 10 studies reporting treatment acceptability measures. In each study, participants expressed satisfaction with the treatment. It is surprising that more of the studies did not include treatment acceptability measures because consumers guide our practice. If a consumer does not value a procedure, it is not likely to be implemented with fidelity or recommended to others. Future researchers should measure treatment acceptability to help determine whether the study is worth replicating. If consumers do not rate the treatment as acceptable, the treatment should be modified. Treatment acceptability measures provide important information concerning the needs of the population served. If consumer input is ignored, quality of treatment risks being compromised (see Appendix A, Tables 5, 6, and 7 for summaries of treatment acceptability measures).

Of the 23 studies included in the analysis, six incorporated a functional analysis component. Five of these studies were extinction and one was graduated extinction. Interestingly, the data shows that extinction studies spend less time in intervention (see Appendix B, Figure 4). It is possible that extinction participants spend less time in intervention because five of the 12 extinction studies incorporated a functional analysis component. Functional analyses identify the function of problem behavior and assist practitioners in developing informed treatment. Considering the high prevalence of pediatric sleep disturbances, the lack of functional analysis methodology in the literature is noteworthy. It is concerning to find that most of the studies in this review did not identify the function of disruptive sleep behaviors objectively. Although parent interviews and standardized survey instruments are useful, they are subjective measures. FA methodology identifies the controlling variables of problem behavior, yielding objective measures, which equip the researcher to predict and control behavior with more precision. It may be possible that the researchers and children determined the likely function of disruptive sleep behaviors through structured interviews and parent reports. It is also important to note that FA's often evoke problem behaviors; the researchers may have elected not to include an FA component for this reason. Regardless, experimental research focusing on FA-informed interventions might help pave the way for better assessment tools that can tailor intervention strategies to the individual.

Analysis of the results produced interesting findings concerning the prevalence of pediatric sleep disturbance literature. For instance, of the 23 studies included, 13 of the studies came from the United States. All four of the BTR studies were from the United States. It is possible that Americans are less tolerant of extinction, thus influencing the study of treatment alternatives such as BTR's. Five of the 23 studies were from the Netherlands. Of these five studies, 5 used unmodified extinction and 1 used graduated extinction (see Appendix A, Table 10 for a summary of articles organized by country and intervention type). Cultural differences may be at play; perhaps families in the Netherlands are more accepting of interventions that require ignoring. Because culture may affect parenting styles, it is a variable that should be considered when treating pediatric sleep disturbances. Deater-Deckard, Dodge, Bates, and Pettit (1996) found a negative correlation with child aggression in African American children whose parents used physical discipline; the opposite effect was observed with European children whose parents used physical discipline. The research also shows that culture is likely to play a role in which types of interventions parents prefer. Borrego, Ibanez, Spendlove, and Pemberton (2007) measured treatment acceptability of child management behavioral procedures with 97 Mexican Americans. The results showed that a punishment-based procedure was preferred over reinforcement-based procedures. Studies that identify cultural trends empirically may serve as useful tools to researchers intervening on sensitive behaviors such as disruptive bedtime behaviors (see Appendix A, Tables 7, 8, and 9 for summaries of demographic information).

The search yielded some interesting findings related to the type and number of studies published per year. For instance, the number of studies across each intervention type plateaued during 1982-1988. In the mid 90s, the number of extinction studies increased, only to plateau in 2007. The number of graduated extinction studies increased during 2002-2004 and 2012-2014. The number of bedtime routine studies increased in 2002 and continued to gradually increase to 2015. Figure 1 (Appendix B) shows that the number of extinction studies has flat lined, unlike graduated extinction and bedtime routine studies. As mentioned previously in this paper, extinction is not for everyone; some parents refuse to ignore their child. This might be part of the reason why the number of graduated extinction and bedtime routine procedures have accumulated while extinction procedures have not.

Trends in recruitment practices were found. For example, three of the studies noted that participants were referred by clinics or medical practitioners. Six of the studies reported that participants responded to newspaper advertisements and/or notices posted locally. The remaining 14 studies reported that parents, colleagues, schools, and community-based centers referred the child. One study reported that an independent market research firm was used to recruit 264 participants. Future researchers should be aware of the difficulties associated with recruiting participants. Future researchers should strive to establish relationships with local pediatricians and early intervention programs, as they are often in direct contact with individuals reporting bedtime resistance.

In conclusion, future researchers should strive to adopt single-case design methodology, with replications and long-term follow-up measures. Unobtrusive technologies should be explored to increase the use of objective data collection, and data collection practices should include systemic variables. Future researchers should also consider measuring social validity measures including parent stress, child's academic success, and marital satisfaction which will provide more information about how sleep problems affect the child and family. Future researchers should also incorporate functional assessments to help guide treatment selection. In addition, future researchers should take note of the difficulties associated with participant recruitment in pediatric sleep disturbance research. Researchers might benefit from developing professional relationships with pediatricians and other medical professionals who can refer potential participants to them. The results of the systematic literature review indicate the need for more controlled, experimentally sound studies to treat pediatric sleep disturbances.

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Appendix A: Tables

Table 1

Summary of Design Use Across Intervention Types

Design Type	Intervention		
	E	GE	BTR
Single Case Designs	9	5	2
Group Designs	3	2	2

Note: E=extinction; GE=graduated extinction; BTR=bedtime routine

Table 2

Summary of Extinction Interventions

Author(s)/Year	N	Age(s)	Diagnosis	Independent Variable(s)	Dependent Variable(s)	Design	FA	Follow Up	Conclusion
Bramble (1996/1997)	15	• *M*= 7.2 yrs; *SD*=7.2±2.6 yrs	• Down Syndrome • Smith-Magenis syndrome • Carcinuria perinatal • Cerebral anoxia • Cerebral leucodystrophy • Cerebral palsy • Epilepsy	• E	• Sleep problem severity score	• Pre-post	No	• 4 mo+ • 18 mo+	• E+
Curfs, Didden, Sikkema & de Moor (1999)	1	• 6 yrs	• Wolf-Hirschhorn syndrome • Seizure disorder	• E	• Duration of nighttime disruptions (mins)	• AB	Yes	• 3 mo+	• E+
Didden, Curfs, Sikkema & de Moor (1998)	6	• 2-7 yrs	• Prader-Willi syndrome • Fragile X syndrome • ADHD • Spastic tetraphegia asthma • Seizure disorder • Spinal muscle atrophy • Spastic diplegia • Strabismus.	• E • D • M	• Duration of nighttime disruptions (mins)	• ABC • AB (3) • B (2)	Yes	• 3 mo+ • 6 mo+	• E+ • D+ • M+
Didden, Curfs, van Driel & de Moor (2002)	4	• 1:11-25 yrs	• Severely mentally handicapped • Seizure disorder • Visual/physical impairments • Severe to moderate mental handicap • Autism • mild seizure disorder • Downs Syndrome • Mild delays in expressive/receptive language	• M • M+E • E	• Duration of disruptive sleep behaviors (mins)	• Non-concurrent MBD across subjects; AB (2) • Non-concurrent MBD across subjects; ABC • Non-concurrent MBD across subjects; ABAB	Yes	• 6 mo+	• M- • M+E+ • E+

Table 2 Continued

Author(s)/Year	N	Age(s)	Diagnosis	Independent Variable(s)	Dependent Variable(s)	Design	FA	Follow Up	Conclusion
Didden, de Moor & Kruit (1999)	1	• 2:5 yrs	• Left side paresis	• E	• Duration of nighttime crying (mins)	• AB	Yes	• 3 mo+	• E+
Freeman (2006)	4	• 3 yrs		• Pass • Pass+E	• Frequency of calling out • Frequency of leaving bedroom	• Nonconcurrent MBD; ABAB (3) • Nonconcurrent MBD; ACABAB	No		• Pass • Pass+E+
Friman, Hoff, Schnoes, Freeman, Woods & Blum (1999)	2	• 3 yrs • 10 yrs		• Pass	• Frequency of crying • Frequency of leaving room per night	• ABAB (2)	No	• 3 wks+	• Pass+
Moore, Friman, Fruzzetti & MacAleese (2007)	19	• 3-6 yrs		• Pass+E	• Frequency of calling out • Duration of calling out (mins) • Latency to quiet (mins)	• Pre-post	No	• 3 mo+	• Pass+
Reid, Walter & O'Leary (1999)	16	• 1:4-4 yrs		• E • GE	• Expectancies of compliance • Expectancies of outcome • Expectancies of bedtime/nighttime compliance • Parent bedtime/nighttime stress • Frequency of "good" bedtime/nighttime	• Pre-post	No	• 2 mo+	• GE+ • E+
Thackeray & Richdale (2002)	3	• 4-12 yrs	• Severe range of intellectual disability • Slow-onset asthma • Trisomy-21 • Moderate range of intellectual disability • Mild range of intellectual disability • ADHD • Anxiety	• E	• Goal Achievement Scale • Frequency of bedtime disruptions	• MBD across subjects; ABC (3)	No	• 2wks+ • 3mo+	• E+

Table 2 Continued

Author(s)/Year	N	Age(s)	Diagnosis	Independent Variable(s)	Dependent Variable(s)	Design	FA	Follow Up	Conclusion
Weiskop, Matthews & Richdale (2001)	1	• 5:4 yrs	• Autism	• E	• Latency to sleep (mins) • Number of nights the child sleeps in own bed	• AB	No	• 3 mo+ • 12 mo-	• E+
Weiskop, Matthews & Richdale (2005)	12	• *M*= 5:1 yrs	• ASD • Asperger's syndrome • FXS	• E	• Latency to sleep (mins) • Frequency of pre-sleep disturbances • Frequency of sleeping in own bed alone • Frequency of night waking	• MBD across participants; AB (12)	Yes	• 3 mo+ • 12 mo+	• E+

Note. E=extinction; M=mean and medication; D=desensitization; PR=parent report; MBD=multiple baseline design. + denotes: positive treatment outcome. – denotes negative treatment outcome. Bramble (1997) measures treatment acceptability for Bramble (1996) and are thus combined. Numbers in parenthesis in the design column indicate the number of participants the design was used with.

Table 3

Summary of Graduate Extinction Interviews

Author(s)/Year	N	Age(s)	Diagnosis	Independent Variable(s)	Dependent Variable(s)	Design	FA	Follow Up	Conclusion
Adams & Rickert (1989)	36	• *M*=2:5 yrs		• GE • BTR	• Frequency • Duration of bedtime tantrums (mins)	• Pre-post	No	• 3 wks+ • 6 wks+	• GE+ • BTR+
Allen, Kuhn, Dehaai & Wallace (2013)	5	• 2-11 yrs	• Angelman Syndrome	• Behavioral treatment package: Sleep schedule, BTR, GE/EMD	• Disruptive bedtime composite mean frequency per wk • Weekly percentage of independent sleep onset	• MBD across participants; AB (3) • MBD across participants; ABAB (2)	No	• 1 mo+ • 3 mo+	• Behavioral treatment package+
Durand & Mindell (1990)	1	• 14 mo		• GE	• Frequency of night wakings • Duration of night wakings • Total duration of sleep • Latency to sleep • Time put in bed	• MBD across behaviors; AB	No	• 1 mo+ • 2 mo+ • 9 mo+	• GE+
Eckerberg (2004)	95	• *M*=14.2 mo		• GE	• Frequency of night wakings • Duration of night wakings • Total sleep duration • Total time awake during night	• Pre-post	No	• 1 mo+ • 3 mo+	• GE+
Knight & Johnson (2014)	3	• 4-5 yrs	• Autism Spectrum Disorders	• Behavioral treatment package: circadian rhythm management, BTR, white noise, GE	• Frequency of night wakings • Sleep onset latency (mins)	• MBD across participants; AB (3)	No	• 1 mo+	• Behavioral treatment package+
Vervloed, Hoevenaars & Maas (2003)	1	• 4:6 yrs	• Leber's congenitcal amaurosis	• GE	• Sleep onset latency (mins) • Duration of sleep	• AB	Yes	• 3 mo+	• GE+
Wade, Ortiz & Gorman (2007)	5	• 4-5 yrs		• GE	• Sleep onset latency (mins) • Number of minutes to quiet • Total sleep duration • Frequency of night wakings • Daytime Sleepiness	• AB (5)	No	• 2 mo+	• GE+

Note. GE=graduated extinction; BTR=bedtime routines; PR=parent report; MBD=multiple baseline design. + denotes: positive follow up results and positive treatment outcome. – denotes negative follow up results and negative treatment outcome. Numbers in parenthesis in the design column indicate the number of participants the design was used with.

Table 4

Summary of Bedtime Routine Interventions

Author(s)/Year	N	Age(s)	Diagnosis	Independent Variable(s)	Dependent Variable(s)	Design	FA	Follow Up	Conclusion
Christodulu & Durand (2004)	4	• 2.6-5:11 yrs	• CHARGE • PDD-NOS • Sensory integration disorder • Hypotonia • Immune deficiency • Autism	• BTR • Sleep restriction • BTR +sleep restriction	• Frequency/duration of bedtime disturbances • Frequency of night wakings • Duration of night wakings	• MBD across participants; ABC • MBD across participants; ABCAC • MBD across participants; AB • MBD across participants; AC	No	• 3 mo+ • 6 mo+	• BTR+ • Sleep Restriction+
Milan, Mitchell, Berger & Pierson (1982)	3	• 2.1-15.5 yrs	• Spastic quadriplegic • Mentally retarded • Hyperactive • Seizure disorder • Mild orthopedic problems • Profoundly retarded	• BTR	• Preretirement cooperation • Number of minutes past appropriate bedtime • Duration of in-bed resistance	• MBD across participants; ABC (3)	No		• BTR+
Mindell, Du Mond, Sedeh, Telofski, Kulkarni & Gunn (2011)	264	• 6-36 mo		• Algorithmic internet based intervention+BTR • Algorithmic internet based intervention	• Sleep latency (mins) • Frequency of night wakings • Duration of night wakings (mins) • Total nighttime sleep (hrs) • Longest continuous sleep period (hrs) • Frequency of naps	• Pre-post	No		• Algorithmic internet based intervention+BTR+ • Algorithmic internet based intervention+
Staples, Bates & Petersen (2015)	87	• 30-42 mo		• BTR	• Adherence to BTR • Nightime sleep minutes	• Longitudinal	No		• BTR+

Note. BTR=bedtime routines; PR=parent report; MBD=multiple baseline design. + denotes: positive follow up results and positive treatment outcome. Numbers in parenthesis in the design column indicate the number of participants the design was used with.

Tab le 5

Extinction Interventions: Treatment Measures

Author/Year	Measures	Treatment Acceptability	Total Participants Satisfied	Total Participants Unsatisfied
Bramble (1996/1997)	• PR	• Acceptability of Approach	• 12	• 3
Curfs, Didden, Sikkema & de Moor (1999)	• PR	• No		
Didden, Curfs, Sikkema & de Moor (1998)	• PR	• No		
Didden, Curfs, van Driel & de Moor (2002)	• PR	• No		
Didden, de Moor & Kruit (1999)	• PR	• No		
Freeman (2006)	• PR	• No		
Friman, Hoff, Schnoes, Freeman, Woods & Blum (1999)	• PR	• No		
Moore, Friman, Fruzzetti & MacAleese (2007)	• PR • Audio recordings	• Treatment Evaluation Inventory	• *M*=34.1/40; *SD*=2.5	
Reid, Walter & O'Leary (1999)	• PR • Child Behavior Checklist/2-3 • Beck Depression Inventory • Dyadic Adjustment Scale • Parenting Scale • Parenting Stress Index-Short Form	• Treatment Satisfaction	• E: *M*=5.9/7; *SD*=1.7 • GE: *M*=6/7; *SD* =.9	
Thackeray & Richdale (2002)	• PR • Frequency of naps • Duration of naps • Morning wake time • Restlessness of sleep • Duration of sleep time • Medication • Daytime sleepiness prior to bedtime • Bedtime • Latency to sleep (mins) • Frequency of night wakings • Behavioral Evaluation of Disorders of Sleep questionnaire • Sleep start/end time • Assumed sleep • Actual sleep time • Actual sleep percentage • Sleep efficiency • Actual wake time • Fragmentation Index • Goal Achievement Scale • MTS on-task daytime behavior/activity type • Developmental Behavior Checklist—Teacher Version	• Programme Evaluation Questionnaire	• 4	• 0
Weiskop, Matthews & Richdale (2001)	• PR • Goal Achievement Scale	• Griffin and Hudson (1978) Programme Evaluation Questionnaire*	• 1	• 0
Weiskop, Matthews & Richdale (2005)	• PR • Goal Achievement Scale	• Program Evaluation Questionnaire*	• 15	• 0

Note: PR=parent report   * Denotes standardized

Table 6

Graduated Extinction Interventions: Treatment Measures

Author(s)/Year	Treatment Measures	Treatment Acceptability	Total Participants Satisfied	Total Participants Unsatisfied
Adams & Rickert (1989)	• PR	• No		
Allen, Kuhn, Dehaai & Wallace (2013)	• PR • Actigraphy • Developmental Behavior Checklist-Parent Report Form • Abbreviated Children's Sleep Habits Questionnaire • Abbreviated Acceptability Rating Profile	• Abbreviated Acceptability Rating Profile*	• 5	• 0
Durand & Mindell (1990)	• PR • Bedtime routine video recordings • Beck Depression Inventory • Dyadic Adjustment Scale	• No		
Eckerberg (2004)	• PR • Visual Analogue Scales on daytime behavior • Flint Infant Security Scale • Swedish Parenthood Stress Questionnaire Scales	• No		
Knight & Johnson (2014)	• PR • Gilliam Autism Rating Scale-Second Edition	• Parent Satisfaction with Bedtime	• 3	• 0
Vervloed, Hoevenaars & Maas (2003)	• PR	• No		
Wade, Ortiz & Gorman (2007)	• PR • Early Childhood Inventory: Post-Traumatic Stress Disorder Scale • Generalized Anxiety Scale • Separation Anxiety Scale • Children's Sleep Habits Questionnaire • Eyberg Child Behavior Inventory • Beck Depression Inventory-II	• Therapy Attitude Inventory*	• 5	• 0

Note. PR=parent report. * denotes standardized instruments.

Table 7

Bedtime Routine Interventions: Treatment Measures

Author(s)/Year	Treatment Measures	Treatment Acceptability	Total Participants Satisfied	Total Participants Unsatisfied
Christodulu & Durand (2004)	• PR • Actigraphy • Albany Sleep Problems Scale	• Parental Sleep Satisfaction Questionnaire	• 4	• 0
Milan, Mitchell, Berger & Pierson (1982)	• PR	• No		
Mindell, Du Mond, Sedeh, Telofski, Kulkarni, & Gunn (2011)	• PR • Brief Infant Sleep Questionnaire • Pittsburgh Sleep Quality Index • Profile of Mood States • Customized Sleep Profile	• No		
Staples, Bates & Petersen (2015)	• PR • Actigraphy • Parenting Scale	• No		

Note. PR=parent report

Table 8

Extinction Interventions: Demographic Information

Author/Year	Country	Recruitment
Bramble (1996/1997)	England	Referred to research clinic by specialist community nurses, pediatricians, child psychologist colleague
Curfs, Didden, Sikkema & De Die-Smulders (1999)	Netherlands	Unknown
Didden, Curfs, Sikkema, & de Moor (1998)	Netherlands	Unknown
Didden, Curfs, Sikkema, & de Moor (2002)	Netherlands	Referred
Didden, de Moor, & Kruit (1999)	Netherlands	Referred by parents
Freeman (2006)	USA	Parents responded to fliers
Friman, Hoff, Schnoes, Freeman, Woods, & Blum (1999)	USA	Referred
Moore, Friman, Fruzzetti, & MacAleese (2007)	USA	Previous studies (Freeman, 2006; Friman et al., 1999)
Reid, Walter, & O'Leary (1999)	USA	Recruited through notices in local papers, pediatricians' offices, mothers' groups
Thackeray & Richdale (2002)	Australia	Recruited from Special Developmental School, Special School
Weiskop, Matthews, & Richdale (2001)	Australia	Referred by parents
Weiskop, Matthews, & Richdale (2005)	Australia	Recruited through advertisement in disability newsletter, referral from medical practitioner

Table 9

Graduated Extinction Interventions: Demographic Information

Author/Year	Country	Recruitment
Adams & Rickert (1989)	USA	Recruited through local newspaper advertisements, posted notices
Allen, Kuhn, Dehaai, & Wallace (2913)	USA	The Human Genetics Laboratory at the Munro-Meyer Institute for Genetics and Rehabilitation at University of Nebraska Medical Center and AS Foundation ListServ
Durand & Mindell (1990)	USA	Advertisement in local newspaper
Eckerberg (2004)	Sweden	Referred by Child Clinic in Falun and Child Health Centers
Knight & Johnson (2014)	USA	Parent seminar series on Autism and ASD early intervention program
Vervloed, Hoevenaars, & Maas	Netherlands	Unknown
Wade, Ortiz, & Gorman (2007)	USA	Parent meetings, distribution of flyers, Head Start teachers and supervisors

Table 10

Bedtime Routine Interventions: Demographic Information

Author/Year	Country	Recruitment
Christodulu & Durand (2004)	USA	Referred to Albany (NY Center)
Milan, Mitchell, Berger, & Pierson (1982)	USA	Referred to community-based intervention projects
Mindell, Du Mond, Sedeh, Telofski, Kulkarni, & Gunn (2011)	USA	Independent market research firm
Staples, Bates, & Petersen (2015	USA	Community sample of young children

Table 11

Number and Type of Intervention per Country.

Country	Intervention		
	E	GE	BTR
Australia	3	0	0
England	1	0	0
Netherlands	4	1	0
Sweden	0	1	0
USA	4	5	4

Note. E=extinction; GE=graduated extinction; BTR=bedtime routine

Table 12

Extinction Interventions: Months in Baseline, Intervention, and Follow-up Conditions

Author/Year	Time in Baseline	Time in Intervention	Time in Follow Up
Bramble (1996, 1997)	0.5	0.5	0.5
Curfs, Didden, Sikkema, & De Die-Smulders (1999)	25*	31.2*	Unknown
Didden, Curfs, Sikkema, & de Moor (1998)	0.3*	1.8*	1.6*
	0.8	0.1*	0.3*
	0.5*	1.6*	0.2*
	0	1.1*	0.3*
	0	1.5*	0.1*
	0.1*	1.5*, 3.4*	0.3*
Didden, Curfs, Sikkema, & de Moor (2002)	4.3*	1.9*	0.2*
	0.2*	0.9*	0.1*
	0.4*	4.1*	0.1*
	0.4*, 0.3*	0.4*, 0.3*	0.2*
Didden, de Moor, & Kruit (1999)	0.2*	0.7*	0.2*
Freeman (2006)	0.2*, 0.2*	0.1*, 0.6*	0
	0.2*, 0.1*	0.2*, 0.3*	0
	0.7*, 0.2*	0.3*, 0.4*, 0.5*	0
	0.2*, 0.2*, 0.4*	0.2*, 0.4*, 0.5*	0
Friman, Hoff, Schnoes, Freeman, Woods, & Blum (1999)	0.5*, 0.3*	0.3*, 0.2*	0.1*
	4.3*, 0.3*	0.3*, 0.2*	0.1*
Moore, Friman, Fruzzetti, and MacAleese (2007)	0.2	0.3	0.1
	0.5	0	0.1
	0.2	0.7	0.2
Reid, Walter, & O'Leary (1999)	0.2	0.7	0.2
Thackeray * Richdale (2002)	0.8*	0.1, 0.1*	0.2
	0.7*	0.2; 1.1*	0.2
	0.5*	0.2; 0.3*	0.2
Weiskop, Matthews, and Richdale (2011)	0.1	0.2	1
Weiskop, Matthews, & Richdale (2005)	0.1*	0.2*	0.1*
	0.1*	0.2*	0.1*
	0.1*	0.2*	0.1*
	0.1*	0.2*	0.1*
	0.1*	0.2*	0.7*
	0.1*	0.2*	0.7*
	0.1*	0.2*	0.7*
	0.1*	0.2*	0.7*
	0.1*	0.3*	0.7*
	0.2*	0.2*	0.7*

Note. ***** denotes estimated guess from visual inspection of graphs. Numbers separated by semicolons indicate multiple measures per participant.

Table 13

Graduated Extinction Interventions: Months in Baseline, Intervention, and Follow Up Conditions

Author/Year	Time in Baseline	Time in Intervention	Time in Follow Up
Adams & Rickert (1989)	1.6	0	0.5
	0.2	1.2	0.5
	0.2	1.2	0.5
Allen, Kuhn, Dehaai, & Wallace (2013)	0.5*	1.4*	0
	0.7*; 0.7*	0.7*; 1.2*	0.5
	1*	2.1*	0.5
	1.2*; 1*	2*; 1*	0.5
	1.4*	1*; 0.4*	0.5
Durand & Mindell (1990)	0.7*; 1.2*	10.5*; 8.6*	0.7*; 0.7*
Eckerberg (2004)	0.2*	14.3*	0.5*
Knight & Johnson (2014)	0.2*	0.9*	0.2
	0.5*	1*	0.2
	0.7*	1.1*	0.2
Vervloed, Hoevenaars, & Maas (2003)	0.9	0.1	0.4
Wade, Ortiz, & Gorman (2007)	0.5	0.9	0.2

Note. ***** denotes estimated guess from visual inspection of graphs. Numbers separated by semicolons indicate multiple measures per participant.

Table 14

Bedtime Routine Interventions: Months in Baseline, Intervention, and Follow Up

Author/Year	Time in Baseline	Time in Intervention	Time in Follow Up
Christodulu & Durand (2004)	2.1* 3.3*; 0.5* 3.3* 4.7*	1.6*; 4.4* 0.2*; 1*; 3.3* 0.7* 3.5*	0.5 0.5 0.5 0.5
Milan, Mitchell, Berger & Pierson (1982)	0*; 0.2*; 0.2* 0*; 0.2*; 0.2* 0*; 0.2*; 0.2*	0.2*; 0.2*; 0.2*; 0.2*; 0.2*; 0.2* 0.2*; 0.2*; 0.2*; 0.2*; 0.2*; 0.2* 0.2*; 0.2*; 0.1*; 0.3*; 0.3*; 0.3*	0 0 0
Mindell, Du Mond, Sedeh, Telofski, Kulkarni & Gunn (2011)	0.2	0.1	0
Staples, Bates & Petersen (2015)	0	0.7	0
Christodulu & Durand (2004)	2.1* 3.3*; 0.5* 3.3* 4.7*	1.6*; 4.4* 0.2*; 1*; 3.3* 0.7* 3.5*	0.5 0.5 0.5 0.5

Appendix B: Figures

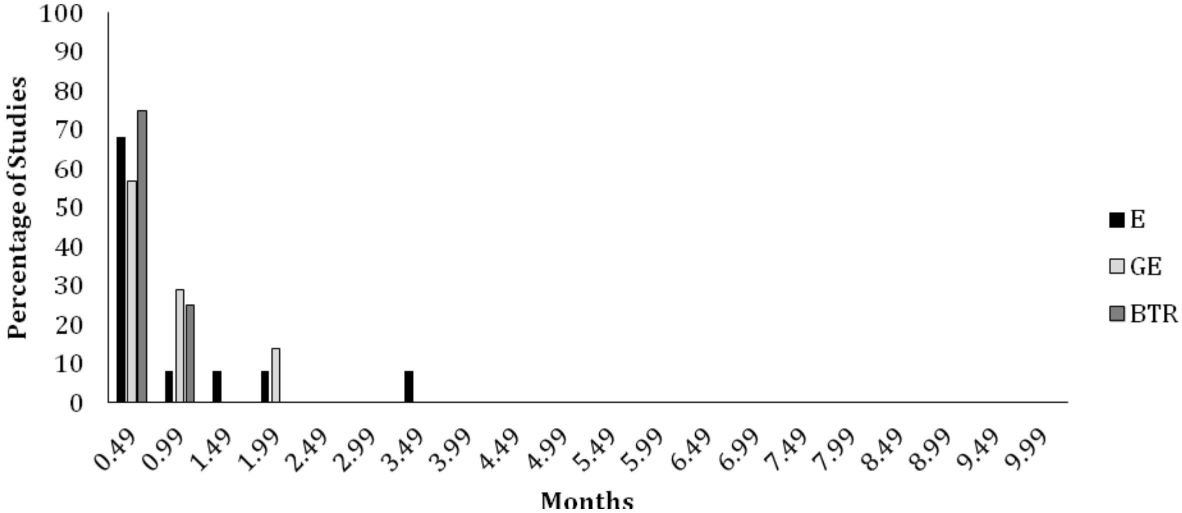

Figure 1. Bar graph displaying number (percentage) of interventions and time (months) spent in follow up.

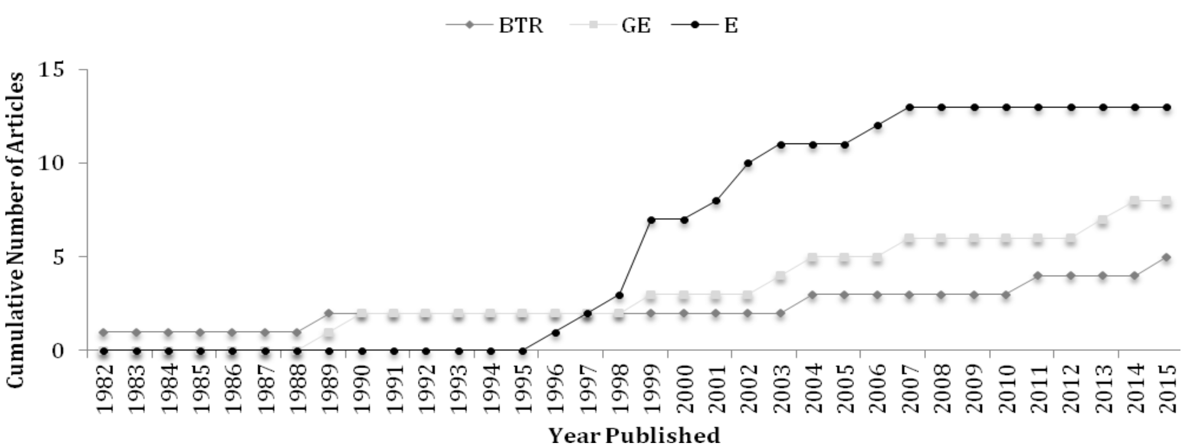

Figure 2. Cumulative record displaying number of studies, by intervention type, published from 1982 to 2015.

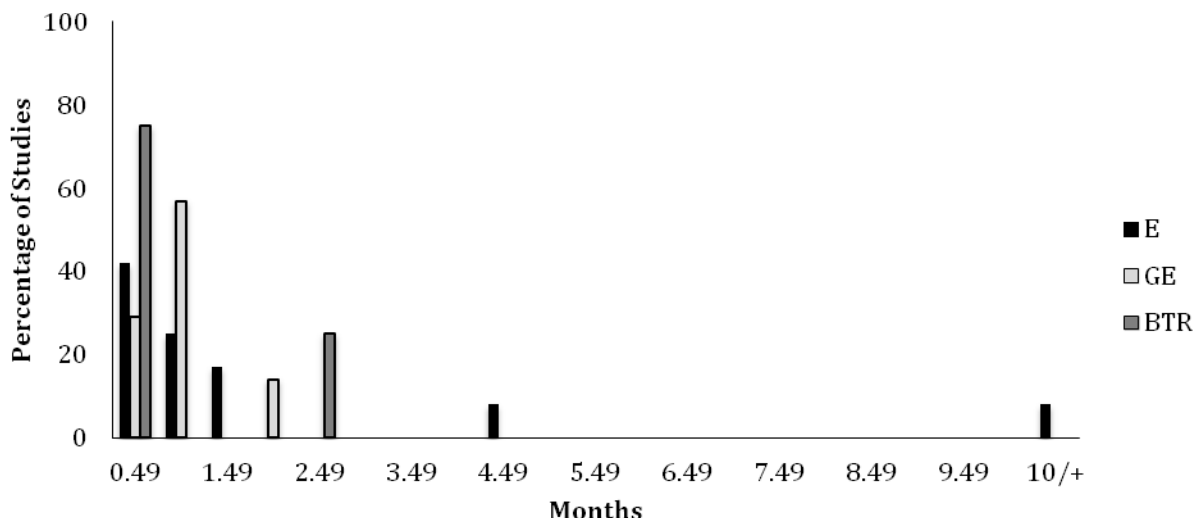

Figure 3. Bar graph displaying number (percentage) of interventions and time (months) spent in initial baseline.

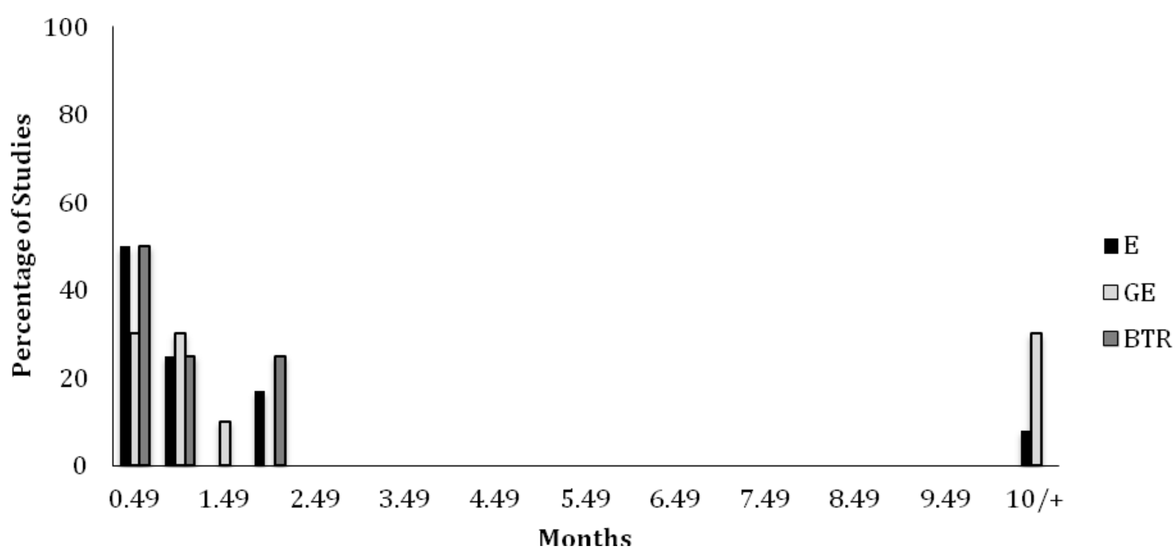

Figure 4. Bar graph displaying number (percentage) of interventions and time (months) spent in initial intervention.