Case Study: A Case-Series Evaluation of a Behavioral Sleep Intervention for Three Children with Autism and Primary Insomnia

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Objective To assess the effectiveness of a manualized multi-component behavioral sleep intervention for children with autism spectrum disorder (ASD) and primary insomnia. Methods Three children (2 males and 1 female, aged 8–9 years) participated. The intervention consisted of a treatment handbook for parents; a distance treatment approach was used in which parents had weekly telephone contact with a therapist. The main behavioral strategies employed were Faded Bedtime with Response Cost and positive reinforcement. Within a case-series design, both subjective (parent-report questionnaires and sleep diaries) and objective (actigraphy) measures were used to record changes in children’s sleep and daytime behavior. Results For all 3 children, mean sleep onset latency was reduced following the intervention. These improvements were generally maintained at follow-up 12 weeks later. Conclusions The current study provides preliminary evidence for the effectiveness of a manualized behavioral sleep intervention program for improving insomnia in children with ASD.

Key words autism spectrum; disorder; intervention outcome; sleep.

Introduction
A growing body of research indicates significant sleep problems for many children with autism spectrum disorder (ASD), which is characterized by impairments in social interaction and communication skills and restricted patterns of behavior, interests, and activities (American Psychiatric Association [APA], 2000). Sleep problems documented in children with ASD include abnormal sleep–wake patterns, problems with sleep onset, early morning waking, poor sleep quality, shortened night sleep, night waking, and developmentally inappropriate co-sleeping (Richdale & Schreck, 2009). According to several studies (e.g., Paavonen et al., 2008), dyssomnias (disorders of initiating or maintaining sleep) occur significantly more often in children with ASD versus typically developing (TD) children. Indeed, most children with ASD appear to have sleep problems (Mayes & Calhoun, 2009).

In TD children, sleep problems are linked to daytime behavior problems, as well as attentional and other neuropsychological impairments (Sadeh, 2007). Objective measures of children’s sleep problems are also related to overall family stress (e.g., Sadeh, Raviv, & Gruber, 2000). A few studies have investigated the relationship between sleep problems and daytime child and family functioning in children with ASD. Among parents of children with ASD, those whose children have sleep problems experience higher levels of parenting stress (Doo & Wing, 2006). Studies also indicate that reduced child sleep also predicts higher autism symptom scores (Schreck, Mullick, & Smith, 2004).

Behavioral treatments for child sleep problems are more efficacious than medication in both the short and long term (e.g., Ramchandani, Wiggs, Webb, & Stores, 2000). According to a systematic review, behavioral treatments improve bedtime problems and night waking in TD children (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006). Of the 52 intervention studies reviewed, 94% demonstrated that treatment was efficacious and more than 80% of
Behavioral interventions for sleep problems in children with developmental disorders such as ASD have received little empirical attention. Piazza, Fisher and Sherer (1997) compared faded bedtime to bedtime scheduling in two groups of children with developmental disabilities (3 of n = 14 had ASD); faded bedtime improved sleep more. In this and five other studies of behavioral treatments for sleep problems in children with autism (mainly single-case studies, all with n<6), none of the treatments could be classified as well-established or probably efficacious (Schreck, 2001, using the criteria of Chambless & Hollon, 1998).

Since Schreck’s (2001) review, a handful of other case or multiple baseline studies on behavioral treatment strategies for children’s dyssomnias have included some children with ASD. For example, Weiskop, Richdale and Matthews (2005) treated 13 children (six with ASD and seven with fragile X syndrome) using parent training, positive bedtime routines, and extinction, and found improved settling, night waking and co-sleeping. Montgomery, Stores and Wiggs (2004) found that a parent handbook was as effective as traditional face-to-face treatment for sleep problems in 66 children with severe learning disabilities (21 with ASD). All of these studies are limited by heterogeneous samples (range of ASD and other diagnoses) and lack of diagnostic information. In addition, these studies relied on observational and parent report measures, and lacked an objective measure of sleep (e.g., actigraphy). In one recent study, Reed et al. (2009) demonstrated positive impact of parent education groups on the sleep and daytime behavior of 20 children with ASD using actigraphy in addition to parent report. However, more research is needed regarding the effectiveness of promising behavioral strategies for sleep problems in children with ASD.

The current study examined the effectiveness of a manualized multi-component behavioral sleep intervention program. This program, shown to be effective for treating sleep onset difficulties in children with Attention Deficit/Hyperactivity Disorder (ADHD; Mullane & Corkum, 2006), was tailored for children with ASD. It was delivered using a distance treatment approach (i.e., by telephone) to minimize burden on parents. The program combined a parent handbook with weekly telephone contact with a trained therapist (PhD student in Clinical Psychology). The primary objective was to reduce sleep onset latency (the time it takes to fall asleep after ‘‘lights out’’) in children with ASD. Potential secondary benefits included increasing the children’s sleep efficiency and sleep duration, and improving their daytime behavior. In a case-series design, both subjective (parent questionnaires and sleep diaries) and objective (actigraphy) measures were used to track children’s sleep and daytime behavior.

Method
Participants

Children previously diagnosed with ASD between 5 and 9 years old were recruited for the study from the autism service at a pediatric hospital, as well as from community sources. Children were eligible if they met DSM-IV criteria for primary insomnia (APA, 2000), were attending school and were cognitively able to benefit from the intervention. This age group was selected in order to ensure that the children would be young enough to require direct parental involvement in their bedtime routines and sleep habits.

Children with neurological disorders (e.g., epilepsy), were excluded, as were children who were not toilet trained, had nocturnal enuresis, or co-slept with their parents. Children who had received a behaviorally based sleep intervention in the past 6 months, and children who were currently receiving psychopharmacological interventions for sleep problems, or whose type and dose of other psychoactive medication might change during the study, were also excluded.

Nine potential participants were screened; four were ineligible (one each: not toilet trained, nocturnal enuresis, co-slept with parents, and no sleep onset problems). One additional child was excluded because parents were concerned that he would resist actigraphy recording. Thus, four children (three males) were enrolled. Of these, one child’s parents did not complete end-of-treatment or follow-up measures. All three children who completed the study had been diagnosed with ASD by the same clinical psychologist, using DSM-IV criteria based on the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore & Risi, 1999), Autism Diagnostic Interview – Revised (ADI-R; Lord, Rutter & Le Couteur,
1994), and clinical judgment. Each child had significant difficulty initiating sleep and met DSM-IV criteria for primary insomnia (APA, 2000), and scored above the cut-off of 41 on the total sleep disturbances subscale of the Child Sleep History Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000).

The participants were one girl, “Laurie,” (9 years, 3 months; Wechsler Full Scale IQ = 93), and two boys, “Sam” (9 years, 4 months; IQ = 128), and “Richard,” (8 years, 3 months; IQ = 92). All three children met both ADOS and ADI-R criteria for autism, except Richard, whose ADI-R repetitive and restricted behavior score fell 1 point below cutoff. All were Caucasian and from two-parent, multi-child households. All parents were reportedly involved in implementing the intervention, but mothers took all telephone calls from the therapist. During the study period, Laurie took no medication; both boys took previously prescribed consistent doses of stimulant medication (Sam: 30 mg/day Ritalin; Richard: 36 mg/day Concerta). The primary sleep problem for all three children was prolonged sleep onset latency. By parent report, Laurie required approximately 1 hr, Sam took between 0.5 and 1 hr, and Richard took 2 hr to fall asleep.

Materials
All measures were collected for 1 week during baseline, end-of-treatment (7 weeks for Richard, 8 weeks for Sam, and 9 weeks for Laurie) and follow-up (12 weeks after end-of-treatment), unless otherwise indicated.

Actigraphs
Actigraphs provide reliable and stable sleep recordings in school-aged children (e.g., Sadeh et al., 2000) and have high agreement with polysomnography (85–90%). Actigraphy has the advantage of being less intrusive and more feasible than polysomnography, especially with special populations such as children with ASD. Mini-Motionlogger actigraphs were worn on the child’s non-dominant wrist from the time the child got into bed until he/she got out of bed in the morning. Actigraph recordings were scored using software based on a validated sleep algorithm (Sadeh, 1994). Children’s initial bedtimes were manually entered into the program based on information recorded each night by parents in the child’s sleep diary.

Sleep Diaries
Parents recorded, on a nightly basis, detailed information about their children’s sleep onset latency and sleep duration. The diaries were developed by Mullane and Corkum (2006) for a study of a comparable sleep intervention for children with ADHD.

Children’s Sleep Habits Questionnaire
The Children’s Sleep Habits Questionnaire (Owens et al., 2000) is a widely used parent-report questionnaire that measures sleep problems in preschool and school-aged children. Higher CSHQ scores indicate more disturbed sleep. The CSHQ possesses adequate psychometric properties and differentiates between clinical samples of children with sleep problems and community samples of children without sleep problems (Owens et al., 2000).

Child Behaviour Checklist
This measure assesses social competence and a wide range of behavior and emotional problems in children and adolescents aged 6–18 years. The Child Behaviour Checklist (CBCL) has adequate reliability and validity (Achenbach, 1991).

Parent Satisfaction Questionnaire
This author-developed measure was administered to parents by telephone following end-of-treatment. It was designed to assess parents’ satisfaction with the intervention program.

This multi-component program has been shown to decrease the severity of dyssomnias in a small group of children with ADHD (Corkum, Mullane, & Moon, 2006a; Mullane & Corkum, 2006). The first of the five handbook chapters provided information about basic sleep physiology and sleep problems. The second chapter outlined principles of sleep hygiene and the importance of consistent bedtime routines. The third chapter introduced the two main behavioral strategies employed in the program: Faded Bedtime with Response Cost (FBRC) and positive reinforcement. The fourth chapter helped parents tailor these two behavioral strategies to their child. The final chapter provided instructions on how to fade these two behavioral strategies. For a more detailed description, please contact the corresponding author.

The first step in the FBRC strategy was to establish a consistent wake time and a new bedtime 30 min later than the time that the child typically fell asleep. If the child did not fall asleep within 20 min of the new bedtime, he/she was removed from bed and made to engage in a low intensity activity (e.g., reading) for 20 min. Removal from bed was the response cost component of the intervention. After 20 min, the child was returned to bed and the procedure was repeated until the child fell asleep within 20 min.
Once the child was able to fall asleep within 20 min of the new bedtime for two consecutive nights, bedtime was moved 20 min earlier. This process was repeated until the child reached his/her target bedtime. The positive reinforcement strategy consisted of a sticker chart with rewards for positive sleep behaviors, such as following the bedtime routine without resistance.

**Procedure**

Approval for this study was obtained from the Research Ethics Board at a tertiary pediatric hospital. Each family was enrolled and began the study as they were recruited (i.e., not concurrently). Interested parents completed a telephone screening to determine their child’s eligibility. Parents then met the therapist, provided consent to participate, and completed the baseline CSHQ and CBCL. For the baseline week, parents completed sleep diaries and the children wore actigraphs. For each of the five subsequent weeks, parents read one chapter in the treatment handbook and completed the associated exercises. At the end of each week, the therapist contacted the parents by telephone to review the content of the chapter and monitor progress with the exercises. The therapist followed a standardized protocol designed to complement the parent handbook (Corkum, Mullane, & Moon, 2006b).

Although the intervention program was designed to run for only 5 weeks, child illnesses and challenges implementing the FBRC strategy lengthened the program to 7 weeks for Richard, 8 weeks for Sam, and 9 weeks for Laurie. In all cases, weekly phone calls with the therapist continued and focused on trouble-shooting around the FBRC strategy. In the week following the end of the program (end-of-treatment), parents completed CSHQ and CBCL, and the children wore actigraphs. Twelve weeks following the end of the program, parents again completed CSHQ, CBCL, and sleep diaries, and the children wore actigraphs for a week (follow-up).

**Results**

In keeping with the case-series design, data from the three children were examined individually. Trends and patterns are outlined below.

**Sleep Onset Latency**

Mean sleep onset latency decreased after treatment for all three children, as measured by actigraphy (see Table I). For Laurie, actigraphy indicated a decrease of 23 min from baseline to end-of-treatment. At follow-up, her mean sleep onset latency remained improved. Sam’s mean...
sleep onset latency decreased by 9 min and decreased by another 2 min at follow-up. For Richard, mean sleep onset latency decreased by 24 min, and was maintained at follow-up.

Inspection of night-by-night sleep onset latency as measured by actigraphy revealed an overall reduction in sleep onset latency at end-of-treatment compared to baseline for all three children (see Figure 1). For Laurie and Sam, this reduction in night-by-night sleep onset latency was maintained at follow-up. Richard’s night-by-night sleep onset latency varied widely at follow-up. Of note, variability in night-by-night sleep onset latency appeared to increase following treatment for all three children.

Sleep diaries also indicated a decrease in mean sleep onset latency for all three children from baseline to end-of-treatment, with decreases maintained at follow-up (see Table I). Sleep onset latency as measured by the mean CSHQ Sleep Onset Delay subscale decreased for both Sam and Richard but was unchanged for Laurie (see Table I).

**Figure 1.** Night-by-night sleep onset latency (as measured by actigraphy) for Laurie (A), Sam (B), and Richard (C).
or remained the same (Sam). Interestingly, sleep diary and CSHQ data suggested increased sleep duration for Richard and Sam from baseline to follow-up.

**Sleep Efficiency**

There was no clear pattern of change in mean sleep efficiency (i.e., percentage of time asleep compared to time in bed) as measured by actigraphy (see Table I). Laurie’s sleep efficiency was high (over 96%) and remained stable across recording weeks. Sam’s sleep efficiency increased after treatment, but this was not maintained at follow-up. Surprisingly, Richard’s mean sleep efficiency decreased slightly after treatment and further decreased at follow-up.

**Daytime Behavior**

All three children showed small decreases in total (internalizing and externalizing) daytime behavior problems, as measured by CBCL scores (see Table I). For Laurie and Richard, Total Problems T-scores decreased from the borderline clinical range at baseline to the average range at end-of-treatment, and remained in the average range at follow-up. Sam’s Total Problems T-score decreased slightly from baseline to end-of-treatment and again at follow-up, but remained in the clinical range.

**Parent Satisfaction**

Parents provided ratings on a brief author-created satisfaction survey. Ratings were given on a 5-point scale (ranging from 0 = strongly disagree to 4 = strongly agree). Parents were satisfied with the sleep intervention (mean rating = 3.0) and indicated that they would encourage other families to participate in this intervention (mean rating = 3.67). Parents also found the program manual helpful (mean rating = 3.67).

**Discussion**

This study examined the effectiveness of a multi-component manualized behavioral sleep intervention program tailored for children with ASD. This program consisted of a parent handbook (Better Nights, Better Days: Treatment of Sleep Difficulties Parent Handbook) and weekly phone contact with a therapist. The effectiveness of this program was studied for three children with ASD and primary insomnia using a case-series design. The results add to the limited literature on the effectiveness of behavioral strategies for treating sleep problems in children with ASD. The current findings provide some evidence for the effectiveness of this treatment program for reducing sleep onset latency and improving daytime behavior in these children. For all three participants, mean sleep onset latency decreased following the intervention according to both actigraphy and sleep diary. These improvements were relatively small but were maintained at 12-week follow-up. Night-by-night sleep onset latency actigraphy data generally echoed the summary data; it showed a decrease in sleep onset latency for all three children following treatment. This decrease was maintained at follow-up for two children. The third child’s sleep onset latency varied widely during the follow-up week, but the sleep diary contained no explanation that would account for this (e.g., sickness, change in routine). Overall, variability in night-by-night sleep onset latency appeared higher in all three children during the end-of-treatment and follow-up weeks than during the baseline week. This pattern may indicate that for these children, prolonged sleep onset latency is the natural state or set point. The variability at end-of-treatment and follow-up may reflect families’ ongoing efforts to implement the treatment strategies they learned in order to adjust this set point.

Contrary to predictions, actigraphy data showed that average sleep duration decreased slightly for two of the three children from baseline to follow-up, but was stable for the third child. One possible explanation is that as part of the faded bedtime strategy, the children were put to bed later (in order to decrease time awake in bed; see Table I). This had a positive impact on sleep onset latency but the trade-off may have been slightly decreased sleep duration for two of the three children.

In terms of daytime behavior, total behavior problems decreased from the borderline range of severity at baseline to the average range at end-of-treatment for two of the three children, and these improvements were maintained at follow-up. The third child’s total behavior problems decreased yet remained in the clinical range. Thus, small changes in the expected direction were observed in parent-reported behavior problems. It is noteworthy that daytime behavior appeared to improve following treatment despite no increase in the children’s sleep duration or improvement in sleep efficiency. Parents’ adoption of more structured bedtime strategies and anecdotal reports of decreased bedtime conflict might help to account for this effect.

Several limitations apply to the results of this study. First, as with all case study designs, there was no control group. A multiple baseline design was considered; however, the burden of completing questionnaires and daily sleep diaries, as well as ensuring that the children wore the actigraph each night, was judged too heavy for this group of parents. Therefore, outcome data were collected during single weeks at baseline, end-of-treatment and follow-up. A second limitation is that the results may not be
generalizable to a wider population of children with ASD and primary insomnia. Further research is needed to determine whether older and/or younger children with ASD and those with below-average cognitive ability might benefit from such an intervention program. A third limitation is that the program was designed for 5 weeks but all three families of children with ASD needed at least two extra weeks to implement the treatment components. Although the length of treatment did not appear to have any systematic effect on outcome, it is important to consider that both the family’s implementation time and the total phone contact time with the therapist varied across cases.

Implementing behavioral strategies for child sleep problems is challenging for all families; however, a number of obstacles are specific to children with ASD. Finding effective rewards for the positive reinforcement component of the program was a challenge. One modification was that stickers earned for positive sleep behaviors were “cashed in” more quickly than for children with ADHD (Mullane & Corkum, 2006), in order to maintain motivation. It was also difficult to identify appropriate quiet activities in which the children with ASD could take part when they were removed from bed during the FBRC procedure. Participants required a lot of parental supervision during such activities, and often their preferred quiet activities were related to their special interests or preoccupations. These children often resisted the interruption of these activities when they were returned to bed.

In conclusion, the current study provides preliminary evidence for the effectiveness of a multi-component manualized behavioral sleep intervention program for improving insomnia in high-functioning children with ASD. Importantly, however, the study also highlights the unique challenges of treating sleep problems in children with ASD. Further large-scale research employing an experimental design is needed in order to determine the program’s clinical utility.

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