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A Sleep Intervention for Children with Autism Spectrum Disorder: A Pilot Study

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A Sleep Intervention for Children with Autism Spectrum Disorder: A Pilot Study

Abstract

Background: Parents of children who have Autism Spectrum Disorders (ASD) commonly report sleep problems, which typically exacerbate daytime behavior problems. This pilot study sought to identify the short-term effects on sleep, behavior challenges, attention, and quality of life of children with ASD following use of the iLs Dreampad™ pillow, which delivers bone conducted music and environmental sounds. Aims were to demonstrate acceptability and feasibility, identify measures sensitive to change, and describe individual characteristics responsive to change.

Method: Parent report questionnaires assessed sleep behavior, attention, autism-related behaviors, and quality of life from 15 participants before and during intervention. A Sleep Diary documented average sleep duration and average time to fall asleep during the preintervention phase and the last 2 weeks of the treatment phase.

Results: Procedures were acceptable and feasible for families. All measures were sensitive to change. Children with ASD demonstrated significant change in sleep duration and time needed to fall asleep from pretest to intervention. Improvements were noted in autism-related behaviors, attention, and quality of life. Parent satisfaction was high.

Conclusions: The iLs Dreampad™ pillow may be one alternative intervention to pharmacological interventions for children with ASD who have sleep problems. Further study is warranted.

Comments

Conflict of Interest

We confirm that this manuscript has been read and approved by all named authors and that there are no other persons who satisfied the criteria for authorship but are not listed. Partial research support was provided by Integrated Listening Systems to the first author who was not involved in recruitment, administration of the intervention or data collection for this study. All authors attest no conflict of interest.

Keywords

Autism, Sleep behavior, Response to treatment

Cover Page Footnote

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Sleep problems have been reported in autism spectrum disorders (ASD) for many years (Sung, Hiscock, Sciberras, & Efron, 2008; Vriend, Corkum, Moon, & Smith, 2011), with prevalence estimates ranging from 44% to 83% (Konofal, Lecendreux, & Cortese, 2010; Reed et al., 2009). Common problems include difficulties initiating and maintaining sleep, short night sleep duration, early morning waking, tiredness upon waking, and daytime sleepiness. These sleep problems are chronic and stable over time, and they persist into adolescence if not treated (Humphreys et al., 2014; Sivertsen, Posserud, Gillberg, Lundervold, & Hysing, 2012).

Children with sleep problems show more challenging behaviors than those without sleep problems. In children with ASD, it is likely that sleep problems exacerbate their daytime behavior problems and ASD-related symptoms (Allik, Larsson, & Smedje, 2006), including more externalizing and internalizing behaviors, such as aggression, activity level, emotional reactivity, and anxiety, as well as poorer adaptive functioning and impaired health-related quality of life (Sikora, Johnson, Clemons, & Katz, 2012). Findings show more stereotypical behaviors, social difficulties, and emotional problems in children who have both ASD and sleep problems, as well as greater impairments in social and academic functioning (Allik et al., 2006; Bendz & Scates, 2010; Reynolds & Malow, 2011; Schreck, Mulick, & Smith, 2004).

In addition, the parents of children with sleep disorders are at a greater risk of sleep

deprivation. Consequences include increased maternal stress and impaired maternal mental health (Hodge, Hoffman, Sweeney, & Riggs, 2013). In these caregivers, there is also an increased rate of clinical depression and anxiety compared to the caregivers of children without sleep problems (Chu & Richdale, 2009; Sung et al., 2008).

Given the association between sleep problems, greater behavioral difficulties, and parental stress, there is a need for effective sleep intervention programs for this population. Recent systematic reviews of behavioral intervention for sleep problems in children with ASD concluded that the evidence for effective treatments is sparse (Turner & Johnson, 2013; Vriend et al., 2011). While there is an emerging evidence base and a wide variety of treatments, identifying appropriate interventions is imperative for this population. Behavioral methods have some reported success but these approaches can be time consuming (Reed et al., 2009; Weiss & Salpekar, 2010). Pharmacological interventions have the risk of side effects even though they are the most frequently offered treatment to families of children with ASD (Williams, Sears, & Allard, 2006).

One alternative approach to reducing sleep problems is the Integrated Listening Systems (iLs) Dreampad™ pillow. The iLs Dreampad™ pillow delivers bone conducted music and environmental sounds in a customized pillow. The sound selections are designed to emphasize frequencies associated with relaxation and sleep. Anecdotal evidence from clinical practice in occupational

therapy clinics suggests that the use of the iLs Dreampad™ pillow is gaining in popularity with parents of children with autism; however, there are no published studies that systematically examine this treatment in children with ASD.

Aims

The primary purpose of this pilot study was to explore parent perceptions of the impact of the potential association between the use of the iLs Dreampad™ pillow on the sleep behavior of children with ASD. As the first systematic investigation of this approach, our aims were to determine the feasibility and acceptability of the iLs Dreampad™ pillow, to identify measures sensitive to change, and to describe the characteristics of individuals that may impact responsiveness to the intervention. Our intent was to answer these questions in preparation for a more rigorous treatment study. Based on clinical practice/anecdotal evidence, we hypothesized that children with ASD would show improvement in sleep initiation and duration of sleep, reduction in night waking, and improved daytime behavior. In addition, we expected to see a reduction in ASD-related behavioral challenges and improvement in the child's quality of life.

Methods

Study Design

A quasi-experimental, single group, pretest/posttest design (Harris et al., 2006) was employed in this pilot study with two phases:

1. The preintervention phase, which had a 2-week observational period with no treatment.

2. The intervention phase during which the treatment was provided over a 4-week period. Pretest observations established how the participant slept without intervention. After the intervention was applied, the participants' sleep behaviors were then retested. Subjective feedback regarding feasibility and acceptability was also obtained from the families using a questionnaire.

Procedures

Procedures for this study were established and monitored by the principal investigator (PI). There were two participating sites: South Shore Therapies in Massachusetts and Knippenberg, Patterson, Langley, & Associates in Colorado. Each site recruited participants by obtaining confirmation of diagnosis and completing the screening form to ensure the child had a sleep disorder. For those who qualified, informed consent was obtained and the families were asked to complete the parent report measures. Each site received instruction regarding the use of the iLs Dreampad™ pillow so that they could monitor the participants during the 4-week intervention. They also were taught how to use the sleep diary so that they could guide the families.

The parents of the participants completed the demographics forms and parent report questionnaires. Each participating site instructed the families in filling out the sleep diary, which was used at the start and at the end of the study. Preintervention sleep data were collected during the first 2 weeks of the study. Following this 2-week phase, the families were given an iLs Dreampad™

pillow and instructed in its use. The second phase was the intervention phase, during which the participant used the iLs Dreampad™ pillow nightly for a 4-week period. A sleep diary was completed during the last 2 weeks of the treatment phase. Posttest measures were readministered at completion of the treatment phase. At the end of the study, the iLs Dreampad™ pillow was returned to those families who wished to keep it for future use.

The parents completed a subjective feedback form on which they answered questions related to their experience using the iLs Dreampad™ pillow, including what music or sound selections the child listened to, the child's enjoyment and comfort, any observed changes, and whether they would recommend it to others. All of the parents completed consent forms, and the report measures were sent to the PI for data analyses.

Participants

A convenience sample of self-selected subjects participated in this study. The parents of children with ASD received an "invitation to participate" flyer or an email invitation; these were sent to clients at South Shore Therapies and Knippenberg, Patterson, Langley, & Associates. If interested, the parents were asked to complete the research contact form or to contact the coordinator at the participating site. The families that signed the research contact form were contacted by a member of the research team staff to determine eligibility. Potential candidates were identified following

documentation provided by parent report of an ASD diagnosis.

Study inclusion criteria required validation of a sleep disturbance based on answers to two screening questions: one related to the severity of the sleep problem and the other related to the type of sleep problem (e.g., difficulty falling asleep, difficulty staying asleep, restlessness in bed, and tiredness upon waking). The parent report is considered a reasonably accurate method for identifying many types of sleep disturbances in children (Sadeh, Raviv, & Gruber, 2000; Tikotzky & Sadeh, 2001; Werner, Molinari, Guyer, & Jenni, 2008; Wolfson et al., 2003). Only those participants who had at least one type of sleep disturbance rated as moderate or severe were selected. The participants were excluded from the study if they had stressful life circumstances that could account for new onset sleep difficulties, comorbid medical or psychiatric illness that could be associated with insomnia or treatment with a medication known to cause insomnia or sedation, obstructive respiratory disorders and sleep apnea, were receiving behavioral treatments or medication for a sleep disorder, or could not comply with daily pillow use and/or keep a daily sleep log.

Twenty-two children with ASD met the inclusion and exclusion criteria and were enrolled in the study. Six children dropped out of the study before completing the intervention phase due to factors related to the time commitment to participate and/or a priority shift for the parents. One family dropped out because the child reported

not liking the music. The final sample contained 15 children: 10 boys and five girls ranging in age from 2 years 7 months to 15 years (mean = 8.1, SD = 3.56). All were Caucasian with at least one parent who had a high school degree or higher. Two participants had co-morbid attention deficit hyperactivity disorder (ADHD) and one participant had an anxiety disorder. Based on screening questions, characteristics of the final sample included the following type of sleep problems: difficulty falling asleep (87%), tiredness upon waking (67%), tossing or turning in bed (60%), and waking during the night (40%).

The Institutional Review Board of Rocky Mountain University of Health Professions approved all procedures. The parents provided informed consent and an assent was obtained for children 7 years of age or older. The parents were provided with both written and verbal information describing the iLs Dreampad™ pillow program and participation requirements.

Measures

The sleep diary was completed during each phase of the study. The sleep diary indicated the time the child went to bed, the time the child fell asleep, the time the child awoke in the morning, and if the child awoke during the night or took a nap during the day. A recording of a 2-week sleep diary has been described as adequate for identifying sleep patterns (Mindell & Owens, 2003). A total score was computed summarizing the number hr slept each day. Sleep diaries have been commonly used as an outcome measure in sleep research

(Honomichl, Goodlin-Jones, Burnham, Gaylor, & Anders, 2002; Weiss & Salpekar, 2010; Wright et al., 2011).

The Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000) is a parent report screening instrument that measures sleep habits and identifies common sleep complaints in children, including bedtime behavior, sleep behavior, and morning wake-up (Owens et al., 2000). The abbreviated 22-item version was used in this study. Although not designed for use with individuals with ASD, the CSHQ has frequently been used with this clinical group (Adkins et al., 2012; Malow et al., 2012; Malow et al., 2014; Rzepecka, McKenzie, McClure, & Murphy, 2011). Preliminary report of scale psychometrics suggests moderately strong internal consistency (total score, $\alpha = 0.78$) and the ability of items, subscales, and total scores to differentiate non-sleep disordered children from those with a suspected sleep problem. The CSHQ demonstrated adequate test-retest reliability ranging from 0.62 to 0.79 (Owens et al., 2000).

The Pediatric Quality of Life Inventory (PedQL; Varni, Seid, & Rode, 1999) is a measurement tool that reports health-related quality of life in children. The 23-item scale includes physical functioning, emotional functioning, social functioning, and school functioning. A recent study supports the responsiveness of the scale to measure outcomes in clinical treatment effectiveness research (Desai et al., 2014). In addition, it was shown to have adequate construct validity and

predictive validity (Desai et al., 2014). It has been used successfully to identify impaired health-related quality of life in various disease groups (Varni, Limbers, & Burwinkle, 2007; Varni, Seid, & Kurtin, 2001).

The Parental Concerns Questionnaire (PCQ; McGrew et al., 2007) assesses the presence and severity of developmental and behavioral concerns related to the core symptoms of ASD (e.g., language delay, self-injurious behaviors, hyperactivity, and compulsive behaviors). Parents are asked to designate to what extent a particular symptom has been a problem. High internal consistency was demonstrated ($\alpha = 0.93$ for the total sample) as well as stability over time ($\kappa > 0.6$). There was reported variability in item responses indicating that parents did not identify all items as concerns. Moderate to strong correlations with other parent-rated and clinician-rated instruments support the construct validity of the PCQ (e.g., Child Behavior Checklist and Autism Diagnostic Observation Schedule).

The Swanson, Nolan, and Pelham SNAP-IV (SNAP-IV; Swanson, 1995) is an 18-item checklist related to attention and hyperactivity used for diagnosing ADHD based on the symptom descriptions in the Diagnostic and Statistical Manuals (III, III-R and IV). One study reported good to excellent internal consistency (Stevens & Quittner, 1998), although agreement between parents and teachers was weak. A more recent study (Bussing et al., 2008) provides additional support of the internal consistency ($\alpha > 0.90$) and

factor structure of the SNAP-IV. Findings also suggest that the scale adequately differentiates varying levels of ADHD concerns.

Description of the Intervention

Intervention took place in the child's home. The version of the iLs Dreampad™ pillow used in this study required placement in the pillowcase of the child's preferred pillow. A selection of classical music or ambient sounds was played via a bone conduction unit embedded in the customized pillow. The sounds or music played continuously for 2 hr and then automatically shut off. The parents were instructed in use of the iLs Dreampad™ pillow by a study representative. Part of the instruction included how to adjust the volume and sound/music selection based on the child's preference.

Data Analyses

Significance in sleep duration and time to fall asleep, as well as change in standardized measures, were examined using Wilcoxon signed-rank tests for matched pairs. We chose not to correct for multiple comparisons due to the small sample size and the pilot nature of the study. Our interest was also in identifying scales and subtests that were responsive to change.

Descriptive analyses were conducted on the data to determine individual characteristics that might have affected responsiveness to the intervention. Average sleep duration and time to fall asleep were calculated for the participants in both phases of the study, such that a comparison could be made from pretest to intervention.

The subjective experiences of children and their families were summarized from completed parent feedback forms.

Results

The 15 participants completed sleep diary measures. One participant had partial data; only a sleep diary

was completed. See Table 1 for changes in measures described below from pretest to intervention using Wilcoxon signed-rank tests for matched pairs.

Table 1
Changes from Pretest to Intervention

Measures	Pretest		Intervention		Wilcoxon Signed Rank Test	p	Effect Size
	M	SD	M	SD			
Sleep Diary ^a (n = 15)							
Sleep Duration	8.95	0.96	9.65	1.07	-3.01	.003	.78
Time to fall asleep	0.94	0.60	0.54	0.40	-2.83	.005	.73
CSHQ Total (n = 13)	28.54	9.03	23.54	9.49	-2.63	.009	.73
Bedtime	13.46	4.45	11.00	4.93	-2.12	.034	.59
Sleep Behavior	6.64	3.20	5.77	2.62	-1.80	.072	.50
Night Waking	3.00	1.96	2.62	1.66	-1.51	.132	.42
Morning Wake	2.91	2.12	4.15	1.91	-1.70	.089	.47
PCQ (n = 14)	40.20	8.05	35.00	9.15	-2.28	.022	.61
PedQL Total (n = 13)	42.62	11.35	29.85	8.65	-2.94	.003	.82
Physical	12.31	6.55	12.31	6.55	-2.33	.020	.65
Social	10.54	3.97	7.85	3.89	-2.82	.005	.78
Emotional	10.31	2.66	6.85	3.16	-3.00	.003	.83
School	10.25	3.31	7.33	2.77	-2.59	.010	.72
SNAP-IV (n = 14)	31.71	6.12	27.00	9.66	-2.44	.015	.65

Note. ^atime reported in hours.

Sleep Diary

Average sleep duration and time to fall asleep for each phase of the study are reported in hr. At pretest, the participants slept an average of 9 hr each night and took approximately 1 hr to fall asleep. During the intervention phase (using the iLs Dreampad™ pillow), the participants slept 40 min longer and fell asleep 23 min sooner relative to the pretest phase. Wilcoxon signed-rank tests indicate that the change in sleep duration and time to fall asleep were significant from pretest to intervention.

Child Sleep Habits Questionnaire (CSHQ)

Sleep behaviors as measured by the total score on the CSHQ showed a significant change from pretest to intervention. There was also a significant reduction in behaviors on the bedtime subscale from pretest to intervention. Two of the three other sub-scores, sleep behavior and morning wake-up, improved from pretest to intervention but were not significant. Night wakings remained relatively unchanged. Several items showed significant change or a trend toward significance

from pretest to intervention, including ability to fall asleep in 20 min ($z = -2.27$; $p = .023$), sleeping alone in own bed min ($z = -1.89$; $p = .059$), not needing a special object to fall asleep min ($z = -1.71$; $p = .088$), less resistance to going to bed min ($z = -1.89$; $p = .059$), less restlessness during sleep min ($z = -1.90$; $p = .057$), and not moving to another person's bed during the night min ($z = -2.00$; $p = .046$).

Parental Concerns Questionnaire (PCQ)

A reduction in autism-related behavioral challenges was supported by change on the PCQ. A significant change was obtained from pretest to intervention.

Pediatric Quality of Life Inventory (PedQL)

The total score and all four subtests of the PedQL (physical, emotional, social, and school) showed significant improvements in children's quality of life from pretest to intervention.

SNAP-IV

Improvements in attention were supported by change in SNAP-IV scores. A significant difference was noted from pretest to intervention.

Postintervention Parent Feedback

Nine out of the 15 parents completed the parent feedback form. All reported improvement in their child's ability to fall asleep and improved quality of sleep. Only one parent reported no change in sleep behavior. Six parents reported a decrease in their child's stress level, four reported a decrease in their child's sensory sensitivity, and four reported improved overall behavior. All of the parents said they would recommend the iLs

Dreampad™ pillow to other children with ASD who have sleep problems.

The characteristics of three participants were examined in more detail because they showed the least amount of change on the standardized measures. These participants were reported to suffer from chronic nightmares, night terrors and sleepwalking, experienced extreme restlessness during sleep, and/or had significant gastrointestinal issues. Subjective parent report, however, confirmed improved sleep behaviors, such as increased sleep duration, reduced time to fall asleep, and improved quality of sleep.

Discussion

This pilot study suggests that the iLs Dreampad™ pillow may be an effective intervention for children with ASD who have sleep problems. Use of the iLs Dreampad™ pillow was feasible and acceptable for all children and families. All measures were sensitive to change. The characteristics of the children who showed less improvement in response to using the pillow were identified.

Most of the children in this study demonstrated improved sleep habits based on sleep diary data and the CSHQ. The children generally slept longer, fell asleep more quickly, and displayed less resistance to going to bed. There were fewer reported autism-related symptoms on the PCQ, including reduced anxiety, obsessive/compulsive features, aggression, mood swings, self-injurious behavior, hyperactivity, and attentional issues. The SNAP-IV further supported improvements in

attention. In addition, the parents reported positive changes in their child's quality of life as reflected on the PedQL, specifically that their child's physical, emotional, social, and school functioning were reported to improve. Parental satisfaction using the iLs Dreampad™ pillow was high, with parents recommending the pillow to other children and families. This subjective feedback supports the social validity of the iLs Dreampad™ pillow.

These findings are consistent with the literature suggesting that children with ASD and sleep problems tend to have exacerbated daytime behavior problems, such as increased stereotypic behaviors, social difficulties, and emotional symptoms (Allik et al., 2006; Bendz & Scates, 2010; Reynolds & Malow, 2011; Schreck et al., 2004; Sikora et al., 2012). Thus, with increased and better sleep, it is not surprising that the children in this study were reported to have fewer autism-related behavioral challenges and better attention, emotional, social, and school functioning.

This study contributes to the growing body of literature supporting varied sleep intervention programs for this population. Because parents play a key role in the implementation of these programs, recommended interventions should not only demonstrate efficacy but also acceptability and feasibility for implementation (Bramble, 1996; Keenan, Wild, McArthur, & Espie, 2007). This sleep intervention appears to be a good fit for the families who participated in this study, ensuring implementation and long-term maintenance (Turner & Johnson, 2013).

The individual data from three children with night terrors, sleep walking and chronic nightmares, GI issues, or extreme restlessness revealed that they did not respond as well to the use of the iLs Dreampad™ pillow in this study. Less improvement in this group may have been due to several factors, including (a) sleep problems of this nature may not benefit from the iLs Dreampad™ pillow as a stand-alone treatment, (b) more severe sleep problems may require use of the iLs Dreampad™ pillow for longer than 4 weeks in order to see an effect, or (c) more severe sleep problems may require use of the iLs Dreampad™ pillow for more than 2 hr each night. Scheduled awakenings have been shown to be effective in the treatment of night terrors (Durand, 2002) and therefore could be used in combination with the iLs Dreampad™ pillow.

Limitations

Results should be considered preliminary given the small sample size, the lack of a control group, and the fact the families were self-selected. The study participants were also homogenous with respect to ethnicity and parent education, and all of the measures used in this study relied on parent report. Although parental reporting is considered an accurate measure of sleep habits (Sadeh et al., 2000; Tikotzky & Sadeh, 2001; Werner et al., 2008; Wolfson et al., 2003), it is possible that the positive results obtained in this study were due to parental bias or placebo effects. Future studies need to control for expectancy and attention, such as using a placebo pillow without sound. Use of additional

objective measures should also be considered to supplement the parent report, including actigraphy or polysomnography. Replication of this study is needed to confirm the efficaciousness of this intervention as well as increasing the duration of use of the iLs Dreampad™ pillow beyond the time constraints of this study.

Conclusions

This study contributes preliminary information to evidence-based practices for children with ASD who have sleep problems. The iLs Dreampad™ pillow is a non-invasive intervention that is acceptable to children and families and can be incorporated into daily use. Outcome measures used in this study were sensitive to change and have potential for use in future clinical or research endeavors. Thus, the iLs Dreampad™ pillow may be one alternative intervention to pharmacological interventions for improving sleep problems that impact daytime behavior in children with ASD. Children with severe sleep disturbances, such as night terrors or extreme restlessness, may not be good candidates for this intervention. This study can be used to guide a more rigorous study of the iLs Dreampad™ pillow.

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