



Evaluation of a behavioral treatment package to reduce sleep problems in children with Angelman Syndrome

Keith D. Allen^{a,*}, Brett R. Kuhn^a, Kristi A. DeHaai^b, Dustin P. Wallace^c

^a Department of Psychology, Munroe-Meyer Institute for Genetics and Rehabilitation, 985450 Nebraska Medical Center, Omaha, NE 68198-5450, USA

^b Human Genetics Laboratory, Munroe-Meyer Institute for Genetics and Rehabilitation, 985450 Nebraska Medical Center, Omaha, NE 68198-5450, USA

^c Department of Pediatrics, University of Missouri – Kansas City, Children's Mercy Hospitals and Clinics, Integrative Pain Management, 2401 Gillham Road, Kansas City, MO 64108, USA

ARTICLE INFO

Article history:

Received 10 August 2012

Received in revised form 2 October 2012

Accepted 2 October 2012

Available online 1 November 2012

Keywords:

Angelman Syndrome

Sleep problems

Bed-time problems

Behavioral treatment

Disruptive behavior

ABSTRACT

The purpose of this investigation was to evaluate the effectiveness of a behavioral treatment package to reduce chronic sleep problems in children with Angelman Syndrome. Participants were five children, 2–11 years-of-age. Parents maintained sleep diaries to record sleep and disruptive nighttime behaviors. Actigraphy was added to provide independent evaluations of sleep–wake activity. The treatment package targeted the sleep environment, the sleep–wake schedule, and parent–child interactions during sleep times. Treatment was introduced sequentially, across families, and evaluated in an interrupted time series, multiple baseline design. Data show that prior to treatment, baseline rates of nighttime disruptive behavior were stable or increasing and none of the participants were falling to sleep independently. With the introduction of treatment, all participants quickly learned to initiate sleep independently. Gradual reductions were reported in disruptive behaviors and these improvements were sustained over time. Results were replicated with two participants when treatment was withdrawn and reinstated. Changes in disruptive bedtime behaviors and in sleep onset were found to be statistically significant. Parents indicated high satisfaction with the treatment. A behavioral treatment package was found to be effective with five children with long histories of significant sleep-related behavior problems. These results suggest that behavioral treatment may be a reasonable way to address sleep problems in some children with Angelman Syndrome.

© 2012 Elsevier Ltd. All rights reserved.

1. Introduction

Angelman Syndrome (AS) is characterized by developmental delay, happy demeanor, hypermotor behavior, stereotypies, and reduced adaptive skills. Behavior problems are frequently present in males and females of all ages, and include language deficits, excessive laughter, hyperactivity, short attention span, and problems sleeping (Summers, Allison, Lynch, & Sandler, 1995). Researchers have highlighted the importance of examining environmental influences on common behavior problems in genetic syndromes like Angelman and evidence suggests that some of these common problem behaviors in children with AS can be managed by systematically manipulating various aspects of the environment (Horsler & Oliver, 2006). Indeed, management of problematic behaviors has been based primarily on behavioral approaches that emphasize changing the environment, though psychoactive medication is sometimes required as well (Pelc, Cheron, Boyd, & Dan, 2008).

* Corresponding author at: Munroe-Meyer Institute, 985450 Nebraska Medical Center, Omaha, NE 68198-5440, USA. Tel.: +1 402 559 5756; fax: +1 402 559 6864.

E-mail addresses: kdallen@unmc.edu (K.D. Allen), brkuhn@unmc.edu (B.R. Kuhn), kdehaai@unmc.edu (K.A. DeHaai).

Sleep problems are one of the most common behavior problems associated with AS. In one study, severe sleep problems were reported in as many as 35% of individuals with AS (Didden, Korzilius, Smits, & Curfs, 2004). Another study found marked problems with independently initiating and maintaining sleep as well as frequent night awakenings and reduced total sleep (Bruni et al., 2004). In many cases, heightened sensitivity to the sleep environment has also been identified as a common problem (Conant, Thibert, & Thiele, 2009; Walz, Beebe, & Byars, 2005). Perhaps not surprisingly, sleep problems have been found to be more severe in younger children, peaking between 2 and 6 years-of-age (Clayton Smith, 1993). Indeed, some have found bedtime problems and night awakenings in as many as 90% of young children with AS (Summers et al., 1995), and children with AS have been identified by sleep experts to be among the most challenging populations to treat (Mindell, Emslie, et al., 2006; Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006b).

Not surprisingly, sleep problems also create detrimental effects for families. Most parents report that dealing with a child's ongoing sleep problems can result in fatigue, irritability, distress, and impaired social functioning (Didden & Sigafos, 2001). In addition, sleep problems in children with AS can create anger, anxiety and feelings of helplessness in family members who, because of their caregiving role for a child with AS, experience their own lack of sleep. Parents of children with AS report higher levels of psychological distress compared to parents of other children with rare genetic syndromes (Griffith et al., 2011).

Behavior therapy is currently the consensus "first-line" treatment for pediatric sleep disturbance, with numerous studies showing that this approach produces durable changes in more than 80% of young children (Mindell et al., 2006b; Morgenthaler et al., 2006). Parents serve as the active change-agent (Owens, 2006), and are coached in how to implement these principles to shape healthier sleep habits in their children. The basic elements of a quality sleep intervention include teaching parents how to (1) create a quality sleep-compatible environment, (2) adjust and regulate the sleep-wake schedule to consolidate sleep and (3) manage parent-child interactions to reinforce appropriate bedtime behaviors and to promote independent sleep initiation (Kuhn, 2007; Mindell & Owens, 2003).

These behavioral methods have empirical support and wide acceptance in the sleep literature and have been recommended in the management of sleep problems in AS (Pelc et al., 2008). Yet, few parents of children with AS report ever receiving this type of advice in treatment of sleep problems (Didden et al., 2004). This may be due, at least in part, to concerns about whether behavioral treatment of sleep problems is appropriate for children with AS. That is, some providers as well as parents may question whether behavioral interventions can be expected to solve sleep problems that are thought to reflect structural or chemical abnormalities (Walz et al., 2005), or the dysregulation of cortical interactions (Pelc et al., 2008).

Additionally, providers may be reluctant to recommend behavior treatments without clear empirical support. Indeed, practitioners and researchers alike have acknowledged the need for more research of behavioral methods for managing common sleep problems in children with AS (e.g., Didden et al., 2004; Pelc et al., 2008). To date, much of the empirical evidence to support the efficacy of behavioral sleep interventions has been derived primarily from research with more typically developing populations (e.g., Mindell, Emslie, et al., 2006; Mindell, Kuhn, et al., 2006). Numerous other studies have investigated behavioral treatments for children with other types of neurodevelopmental disabilities. For example, Piazza et al. have evaluated a procedure called faded bedtime with response cost (e.g., Piazza & Fisher, 1991; Piazza, Fisher, & Sherer, 1997). In the procedure, if a child does not fall asleep within 15 min of being placed in bed, the bedtime is pushed 30 min later until they do. This procedure has been found effective in children with intellectual disabilities and severe behavior disorders, but has not been evaluated for children with AS.

Only a single case study has been published evaluating a behavioral treatment in children with AS (Summers et al., 1992). Summers et al. treated a nine year old boy with AS who was sleeping less than two hours a night. The behavioral components included restriction of daytime sleep, installing a consistent sleep schedule, and reducing nighttime adult-child interactions. However, the treatment included administering the medication diphenhydramine HCl and it was conducted primarily in an inpatient behavioral treatment unit by staff. Treatment in the home by parents was only introduced after 55-days of inpatient treatment. Moreover, there was no actual experimental design. So, although the treatment worked well and the child increased nighttime sleep to over eight hours a night, the independent effects of the behavioral intervention or the parent's ability to implement it was never assessed. Thus, although behavioral sleep interventions are often recommended for children with AS (Pelc et al., 2008; Walz et al., 2005), there are no well controlled, empirical investigations of behavioral treatment of sleep problems in children with AS.

The primary purpose of this investigation was to evaluate a behavioral treatment package to address severe and chronic sleep disturbance in young children with AS.

2. Methods

2.1. Participants

Five children between 2 and 11 years-of-age with a diagnosis of AS (confirmed by genetic testing) served as participants. All were recruited from a general registry maintained by The Human Genetics Laboratory at the Munroe-Meyer Institute for Genetics and Rehabilitation at the University of Nebraska Medical Center and from the AS Foundation ListServ. Inclusion criteria were adapted from Mindell and Durand (1993) and included a minimum 4 week history of sleep disturbances as defined by bedtime resistance, delayed sleep onset and nighttime awakenings. More specifically, sleep disturbances were operationally defined as

Download English Version:

<https://daneshyari.com/en/article/10318089>

Download Persian Version:

<https://daneshyari.com/article/10318089>

[Daneshyari.com](https://daneshyari.com)