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Parent survey of sleep problems among children with CHARGE syndrome

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ABSTRACT

Sleep problems are common among children, especially those with developmental disabilities, visual impairments, and behavioral problems. Past research has indicated a particularly high prevalence of clinically-relevant sleep problems for children with CHARGE syndrome, who often possess all three of these qualities. To gather additional information regarding the nature of these sleep problems and how they are most commonly treated amongst parents, an explorative survey was conducted with 30 parents of children with CHARGE syndrome with comorbid sleep problems using the Sleep Disturbance Scale for Children, as well as demographic and sleep questionnaires developed for use in this study.

Our findings indicated that problems of sleep initiation and maintenance were most commonly reported, consistent with previous research. Parents most often reported the following factors suspected of contributing to sleep problems: self-regulation difficulties, teeth grinding, hormonal imbalance, problem behaviors, and anxiety. The most commonly administered treatments were reported to be the use of positive bedtime routines, melatonin treatment, the use of a weighted blanket, and prescription medications, respectively. While parents reported overall that they felt all three of these intervention strategies were slightly effective at improving their child's sleep problem, the use of positive bedtime routines and melatonin treatment were perceived as more effective by parents. These results aid professionals in the selection of future research and intervention strategies to recommend for parents of children with CHARGE syndrome.

1. Introduction

It is estimated that 25–30 % of American children are affected by poor sleep health (National Institutes of Health, 2011). Among children with developmental disabilities, sleep problems have been shown to be especially high (Polimeni, Richdale, & Francis, 2005; Souders et al., 2009). For example, one study investigated parent reports on sleep problems among children diagnosed with Autism Spectrum Disorders (ASDs) and found that 73 % of the children diagnosed with ASDs had a sleep problem as indicated by their parent (Polimeni et al., 2005). Another study, using actimetry sensors to measure gross motor activity during sleep, indicated that around 67 % of children diagnosed with an ASD, or a pervasive developmental disability, had a sleep difficulty (Souders et al., 2009). Parents of children with an intellectual disability, when compared to a typically developing control group, have been shown to report higher prevalence and chronicity of sleep problems for their children. For example, in one study 55.8 % of children with intellectual disabilities, according to parent report, had a notable sleep problem, and of those children 58.6 % had been experiencing their sleep problem for two or more years (Richdale, Francis, Gavidia-Payne, & Cotton, 2000).

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In addition, children with visual impairments are at high risk for sleep difficulties (Stores & Ramchandani, 1999), especially when those difficulties are combined with developmental or intellectual impairments, or hearing impairments (Khan et al., 2011), as is often the case for children with CHARGE syndrome, a genetic syndrome caused by a de novo mutation in the *CHD7* gene, a regulator gene suspected to have a role in gene expression, causing the mutation to affect multiple sensory systems, including vision, hearing, and balance (Blake et al., 2009). In a study of 20 adults with severe visual impairments who functioned within society, thus having access to many environmental cues, these adults still had abnormal circadian rhythms as indicated by body temperature, cortisol production, and melatonin production (Sack, Lewy, Blood, Keith, & Nakagawa, 1992). These findings highlight the importance of the dark-light cycle in regulating circadian rhythms, therefore sleep.

Sleep problems are important to address, as they are associated with many negative outcomes for children. According to one study with children diagnosed with Attention-Deficit/Hyperactivity Disorder (ADHD), children's sleep problems were associated with poorer quality of life, poorer caregiver mental health, poorer daily functioning of the child, and higher likelihood of missing school, or arriving late to school (Sung, Hiscock, Sciberras, & Efron, 2008). Caregiver mental health is important to address, as having a child with sleep problems is related to parental depression, which in-turn increases the likelihood of marital and family discord, child abuse, and child behavioral problems (Hiscock & Wake, 2002).

In their recent *Sleep in America* poll, the National Sleep Foundation indicated that those respondents who slept for a longer duration during the night also reported better general health, higher quality of life, and better physical and mental health (National Sleep Foundation, 2015). In addition, sleep problems occurring over a long period of time are likely to reduce a child's motivation and concentration (Durand, 1998). In fact, one study of children between 6 and 13 years old showed that insufficient sleep resulted in working memory impairments for these children during the day, indicating that sleep problems impair a child's ability to retain and use information in the daytime (Steenari et al., 2003). Sleep problems are also shown to impair a child's level of attention during the day, as well as their academic performance (Buckhalt, Wolfson, & El-Sheikh, 2009).

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There is very little research investigating sleep problems and their outcomes specifically with children with CHARGE syndrome. One study has indicated a high prevalence for sleep problems among this population, and suggested that sleep problems are related to problematic behaviors for these children, as well as to lower parent well-being over and above the contribution of problematic behaviors (Hartshorne et al., 2008). These findings are consistent with findings among children with other developmental disabilities, in that there has been a reported correlation between sleep problems and problematic behaviors (e.g., Richdale et al., 2000). Among children with CHARGE syndrome, difficulties initiating and maintaining sleep have been suggested to be most common (Hartshorne et al., 2008), consistent with the typically developing population.

Individuals with CHARGE syndrome may be predisposed to sleep problems for several reasons, including circadian rhythm abnormalities due to poor vision, difficulty with falling asleep and staying asleep due to behavioral traits difficulties with impulsivity and poor self-regulation, and problems due to many early hospital experiences and pain (Hartshorne, Stratton, Brown, Madhavan-Brown, & Schmittel, 2017; Heussler, 2011). Sleep problems have been correlated with problem behavior among this population (Hartshorne et al., 2008, 2016). Limited alternations between light and dark have been suggested as a contributing variable to sleep difficulties for individuals with a visual impairment (Palm, Blennow, & Wetterberg, 1997). Since melatonin is produced in darkness, limited alternations between light and dark may contribute to reduced or altered melatonin production and irregular circadian rhythms for these individuals as well. Problematic behaviors have also been associated with sleep difficulties (Hartshorne et al., 2008; Sadeh, Gruber, & Raviv, 2002), and may be a contributing factor in sleep difficulties for children with CHARGE syndrome. Side effects of other medications unrelated to the treatment of sleep could also contribute to nighttime awakenings (Durand, 1998).

Children with CHARGE syndrome may be especially predisposed for sleep-breathing problems due to enlarged tonsils or adenoids, or other craniofacial abnormalities (Blake et al., 2009). Hartshorne, Hefner, Davenport, and Thelin (2011) estimated that 50–60 % of individuals with CHARGE syndrome were born with choanal atresia or stenosis, 15–20 % with an orofacial cleft, and as many as 70–80 % were born with other distinctive facial features. These craniofacial abnormalities result in a higher risk for obstructive sleep apnea, which may contribute to poor concentration, hyperactivity, rapid mood changes, and impulsivity, thus poorer self-regulation (Heussler, 2011). Often infants with CHARGE syndrome undergo a large number of surgeries due to medical difficulties, including those to correct airway blockages due to choanal atresia or stenosis, cleft lip and palate, or problems with the larynx and trachea (Trider et al., 2012).

Parents and professionals working with children with CHARGE syndrome have begun to suspect that these children experience a great deal of pain, anxiety, and self-regulation difficulties, all of which may contribute to difficulties with sleep. Pain has been cited as a factor contributing to sleep problems (Breau & Camfield, 2011). Pain is, at the least, likely to make sleeping more difficult (Hartshorne et al., 2008) and has been associated with poorer overall health, lower sleep quality, more sleep problems, and greater sleep debt, with those individuals experiencing chronic pain reporting 30 min less sleep each night (National Sleep Foundation, 2015). The same survey found that respondents experiencing chronic pain reported that their sleep was more likely to be disturbed by environmental factors, such as noise, light, and temperature (National Sleep Foundation, 2015).

The association between childhood anxiety and sleep problems has been well documented. For example, in one study of children

Table 1
Phenotypical characteristics of sample.

Phenotypical characteristic	Current sample	Typically estimated (Hartshorne et al., 2011)
Ocular coloboma	76.7 %	80–90 %
Choanal atresia or stenosis	43.3 %	50–60 %
Cranial nerve dysfunction or anomaly	73.3 %	70–90 %
Ear anomalies	96.7 %	> 50 %
Genital hypoplasia	53.3 %	50 %
Heart malformations	70 %	75–85 %
Growth deficiency	63.3 %	70 %
Orofacial cleft	40 %	15–20 %
Distinctive facial features	66.7 %	70–80 %

with anxiety disorders, sleep problems were found among 88 % of the children, and severity of sleep problems was associated with severity of anxiety (Alfano, Ginsberg, & Kingery, 2007). While anxiety among children with CHARGE syndrome has not been well-researched, some evidence indicates that anxiety is relatively common among these children, and possibly the most common psychiatric diagnosis received by these children (Wachtel, Hartshorne, & Dailor, 2007). Additionally, self-regulation has been suggested as related to sleep problems (Kroese, De Ridder, Evers, & Adriaanse, 2014), and it has been suggested that children with CHARGE syndrome have difficulties with self-regulation (Hartshorne, Hefner, & Davenport, 2005). In a study comparing the sleep of 28 children with sleep disorders with 30 non-sleep disordered control children, it was demonstrated that “good sleepers” awakened as often during the night as sleep-disordered children, but were able to soothe themselves back to sleep much more quickly and often (Minde et al., 1993). Without this self-regulatory ability, greater difficulty with initiating and maintaining sleep can be expected.

The purpose of this study was to clarify the types of sleep problems that are most common among children with CHARGE syndrome, and the intervention strategies most often employed by parents in an attempt to resolve these problems.

2. Methodology

2.1. Participants

Parents of children between the ages of 3 and 18 with either a clinical or genetic diagnosis of CHARGE syndrome, as well as reported sleep difficulties, were surveyed. Participants were recruited from a CHARGE syndrome social media group operated by the CHARGE Syndrome Foundation in March and April of 2016. 30 respondents completed the survey.

Participant phenotypical characteristics are reported in Table 1 as a percentage of participants with those characteristics as reported by their parent on the CHARGE Syndrome Demographic Questionnaire, and percentages are displayed with estimated percentages reported by Hartshorne et al. (2011) as a comparison for typicality of the sample. Percentages are similar for most characteristics, suggesting a reasonably typical sample of children with CHARGE syndrome. In the current sample, there was a higher prevalence of ear anomalies and orofacial clefts than typically estimated.

2.2. Measures

2.2.1. CHARGE Syndrome Demographic Questionnaire

The CHARGE Syndrome Demographic Questionnaire (see Appendix A) is designed to be completed by parents who have a child with CHARGE syndrome. This form was designed for this study, and items include basic demographic information, as well as information about diagnostic characteristics, according to the Blake and Verloes diagnostic criteria (Blake et al., 1998; Verloes, 2005). In addition, age the child began walking was included, as it has been suggested to predict later developmental characteristics of children with CHARGE syndrome, including adaptive behaviors (Salem-Hartshorne & Jacob, 2005), and sleep difficulties (Hartshorne et al., 2008).

2.2.2. CHARGE Syndrome Sleep Questionnaire

The CHARGE Syndrome Sleep Questionnaire (see Appendix B) was designed for this study, and was meant to be completed by the parent of a child with CHARGE syndrome. This questionnaire gathers information on the frequency of the child’s sleep problems, description of the child’s sleep problems, current and past sleep interventions that have been attempted, and current bedtime routines. In addition, parents completed a checklist of factors they believe contribute to their child’s sleep difficulties.

2.2.3. Sleep Disturbance Scale for Children

The Sleep Disturbance Scale for Children (SDSC; Bruni et al., 1996; see Appendix C) is a 26-item questionnaire designed as a screening measure for identifying sleep problems for children. Items are rated on a 5-point Likert type scale, with the following descriptors: 1) Never or don’t know, 2) occasionally/1–2 times per month, 3) sometimes/1–2 times per week, 4) often/3–5 times per week, and 5) always/daily. A total sleep disturbance score is obtained by adding up ratings for each item. Bruni et al. (1996) suggest that a score of 55 is a good cut-off for clinical significance, and indicates that the internal consistency of this measure is acceptable to

be used for screening purposes. A cutoff score of 55 yields a sensitivity of .89, and a specificity of .74 (Bruni et al., 1996). This instrument was selected due to its past use in identifying sleep problems among children with CHARGE syndrome (Hartshorne et al., 2008). This study used a novel participant sample, though due to the limited published research on sleep among children with CHARGE Syndrome so far, the current sample of participants were compared to that used by Hartshorne et al., 2008 for a measure of typicality of the sample.

In addition, subsets of items may be added together to obtain scores for six factor-analytically derived subscales: disorders of initiating and maintaining sleep, sleep breathing disorders, disorders of arousal/nightmares, sleep wake transition disorders, disorders of excessive somnolence, and sleep hyperhidrosis. On the disorders of initiating and maintaining sleep subscale, items pertaining to sleep duration, sleep latency, going to bed reluctantly, difficulty falling asleep, falling asleep anxiety, night awakenings, and difficulty falling asleep after awakenings are included. For the sleep breathing disorders subscale, items pertaining to breathing problems, sleep apnea, and snoring are included. The disorders of arousal/nightmares subscale includes items pertaining to sleepwalking, sleep terrors, and nightmares. Items pertaining to jerking movements around sleep, rhythmic movement disorders, hallucinations while falling asleep, sleeptalking, and bruxism (i.e., confusion and screaming upon awakening) are included in the sleep wake transition disorders subscale. The disorders of excessive somnolence subscale includes items pertaining to difficulty waking, tired when waking, sleep paralysis, falling asleep in inappropriate situations, and bed wetting. Items related to nighttime sweating and sweating while falling asleep are included in the sleep hyperhidrosis subscale. Cutoff scores for the subscales are not specified.

2.3. Procedures

An invitation to participate in an online survey was sent to the CHARGE Syndrome Foundation's social media group on Facebook, and parents were sent a link to the online survey. Parents were required to consent to participating before being provided access to continue the survey. Personal information was included in the online survey as indicated in Appendix A, primarily to establish the age of the participant and determine whether the participant met inclusion criteria. Data were analyzed using descriptive statistics.

3. Results

Parent reported sleep problems, as reported on the CHARGE Syndrome Sleep Questionnaire, are displayed in Table 2 as a percentage of parents reporting the sleep problem for their child. The most frequently reported sleep problems were reported as waking at night, not enough sleep, falling asleep, and going to bed, respectively.

On the SDSC, 93.3 % ($n = 28$) of children met the cutoff score of 55 for a clinically significant sleep problem ($M = 74.9$, $SD = 13.89$). Scores ranged from 46 to 100. Table 3 indicates the percentage of participants who met each sub-category of sleep problem on the SDSC. The most common sub-category of sleep problem reported was sleep initiation and maintenance, which all but 2 participants met the cutoff score for. These ratings are consistent with parent reported sleep problems on the CHARGE Syndrome Sleep Questionnaire, which indicated going to bed, falling asleep, and waking at night are the most common sleep-related concerns.

Average sleep onset latencies, as reported by parents on the SDSC, are presented in Table 4. According to the SDSC, a non-disordered sleep onset latency is less than 15 min. For most participants ($n = 25$), average sleep onset latencies were reported to be longer than 15 min, with more than a quarter of participants ($n = 8$) requiring more than an hour on average to fall asleep after going to bed, providing further support that sleep initiation is problematic among many children with CHARGE syndrome.

Parents reported on factors they felt contribute to their child's sleep problems on the CHARGE Syndrome Sleep Questionnaire. These results are reported in Table 5. Parents most frequently reported that they suspect self-regulation difficulties, teeth grinding, hormonal imbalance, problem behaviors, and anxiety to contribute to their child's sleep problems. Breathing problems, pain, temperature regulation, muscular problems, and sleep schedule problems were also reported frequently as factors suspected of contributing to sleep problems. Parents did not report nightmares, bed wetting, sleep walking, night terrors, or depression as suspected factors contributing to their child's sleep problems.

Table 6 indicates sleep intervention strategies that parents have used either currently or in the past to address their child's sleep problems. Parents then rated on a 4-point Likert type scale within the how effective they felt each intervention strategy was/is in

Table 2
CHARGE Syndrome Sleep Questionnaire parent reported sleep problems.

Type of sleep problem	N	% of parents reporting
Waking at night	24	80.0 %
Not enough sleep	20	66.7 %
Falling asleep	20	66.7 %
Going to bed	12	40.0 %
Difficult behaviors	11	36.7 %
Tired after sleeping well	10	33.3 %
Sleep breathing	10	33.3 %
Waking in the morning	9	30.0 %
Sleeps too much	1	3.3 %
Excessive napping	1	3.3 %

Table 3
SDSC sub-categories of sleep problem based on cutoff score.

Type of sleep problem	N	% of participants
Sleep initiation and maintenance	28	93.3 %
Sleep-wake transition problems	20	66.7 %
Sleep breathing problems	19	63.3 %
Sleep hyperthyroidism	18	60.0 %
Excessive somnolence	14	46.7 %
Disorders of arousal	5	16.7 %

Table 4
SDSC reported average sleep onset latency (SOL).

Average SOL duration (min)	N	% of participants
Less than 15 min	5	16.7 %
15–30 min	8	26.7 %
30–45 min	3	10.0 %
45–60 min	6	20.0 %
More than 60 min	8	26.7 %

Table 5
Parent reported factors contributing to sleep problems.

Factor	N	% of participants
Self-regulation difficulties	13	43.3 %
Teeth grinding	13	43.3 %
Hormonal imbalance	12	40.0 %
Problem behaviors	11	36.7 %
Anxiety	11	36.7 %
Breathing problems	9	30.0 %
Pain	9	30.0 %
Temperature regulation	8	26.7 %
Muscular problems	6	20.0 %
Sleep schedule problems	6	20.0 %
Nightmares	0	0.0 %
Bedwetting	0	0.0 %
Sleep walking	0	0.0 %
Night terrors	0	0.0 %
Depression	0	0.0 %

Table 6
Parent reported intervention strategies.

Intervention strategy	N	% of participants
Positive bedtime routines	19	63.3 %
Melatonin treatment	15	50.0 %
Weighted blanket	15	50.0 %
Prescription medication	10	33.3 %
Continuous positive airway pressure (CPAP)	8	26.7 %
Over the counter medication	8	26.7 %
Visual schedule	6	20.0 %
Sleep diary	5	16.7 %
Relaxing noise	4	13.3 %
Surgery	3	10.0 %
Meditation/Yoga/Tai Chi	3	10.0 %
Bright light therapy	2	6.7 %
Diet	1	3.3 %
Essential oils	1	3.3 %

treating their child's sleep problems (1 = Not Effective, 2 = Slightly Effective, 3 = Moderately Effective, 4 = Very Effective). These results are presented in Table 7. Only intervention strategies reported by at least 5 respondents are presented. Parents reported using positive bedtime routines, melatonin treatment, weighted blankets, and prescription medication most often to treat their child's sleep problems, respectively. Overall, parents reported similar, but small efficacy for all these intervention strategies, with the exception of

Table 7
Parent reported efficacy of intervention strategies.

Intervention strategy	Average rating	Description
Prescription medication	2.9	Moderately effective
Positive bedtime routine	2.47	Slightly effective
Melatonin treatment	2.33	Slightly effective
Continuous positive airway pressure (CPAP)	2.25	Slightly effective
Over the counter medication	2.25	Slightly effective
Sleep diary	2.2	Slightly effective
Visual schedule	2.17	Slightly effective
Weighted blanket	1.93	Slightly effective

prescription medication which was reported as moderately effective. The use of positive bedtime routines and melatonin treatment were reported most often, and had slightly higher ratings of perceived efficacy than the other strategies reported.

4. Discussion

Previous research (Hartshorne et al., 2008) indicated that sleep problems are more likely to be present than not among children with CHARGE syndrome, and these sleep problems are likely to continue into adolescence and adulthood (Blake, Salem-Hartshorne, Abi Daoud, & Gradstein, 2005). This suggests that these sleep problems are not currently being effectively treated during childhood within this population. This explorative survey was conducted to determine the types of sleep problems most common among children with CHARGE syndrome, and what is typically done by parents and children to address these problems. Sleep initiation and maintenance were identified as significant issues for children with CHARGE syndrome. Types of sleep problems reported were consistent with Hartshorne et al. (2008), which reported sleep initiation and maintenance difficulties as the most common type of sleep problem among children with CHARGE syndrome. This information is important, as it highlights the need for intervention strategies to address specifically sleep initiation and maintenance problems among this population.

The use of positive bedtime routines and blankets has been recommended in the past by professionals (e.g., Heussler, 2011), and the use of melatonin treatment has been shown to improve sleep problems in a variety of other child populations, as noted above. Melatonin is also highly available over-the-counter at pharmacies and supermarkets, and due to its limited risk, this author speculates that physicians may recommend melatonin for treating sleep problems among this population. These strategies may also be commonly selected by parents for several reasons: professional recommendation, research support, availability, convenience, cost, or ease of implementation. While parents reported overall that they felt all three of these intervention strategies were slightly effective at improving their child's sleep problem, the use of positive bedtime routines (e.g., Adams & Rickert, 1989; Andersen, Kaczmarska, McGrew, & Malow, 2008; Malow et al., 2012) and melatonin treatment (e.g., Hoebert, Van Der Heijden, Van Geijlswijk, & Smits, 2009; Malow et al., 2012; Wirojanan et al., 2009) were rated more favorably. It is important for professionals to know what strategies parents are using, so that appropriate those strategies may be further studied, and research-based recommendations can be made to these parents.

Several limitations of this study should be noted. First, measures of sleep and of efficacy of intervention stentage of children with ear anomalies and orofacial clefts than expected. This may be significant, as congenital craniofacial anomalies are at a higher risk for sleep-disordered breathing problems. Additionally, while we attempted to compare our sample of participants with those reported in past research, recruitment online through social media may also have resulted in a sample that in not representative of the population of children with CHARGE syndrome. Several participants also reported the use of medications to treat sleep problems, and the relationship between these medications and their impact on reported sleep data were not explored.

Declaration of Competing Interests

This work was funded through the Department of Psychology at Central Michigan University. The authors have no competing interests. Data are available upon request.

Appendix A. Supplementary data

Supplementary material related to this article can be found, in the online version, at doi:<https://doi.org/10.1016/j.ridd.2020.103614>.

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