3-2022

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Recommended Citation

Ingram DG, Cruz JM, Stahl ED, Carr NM, Lind LJ, Keirns CC. Sleep Challenges and Interventions in Children With Visual Impairment. J Pediatr Ophthalmol Strabismus. 2022;59(2):77-86. doi:10.3928/01913913-20210623-01

Sleep Challenges and Interventions in Children With Visual Impairment

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ABSTRACT

Purpose: To examine sleep patterns in a large and heterogeneous group of children with visual impairment.

Methods: A cross-sectional survey of parents of children with visual impairment was offered via the National Federation of the Blind and the National Organization for Albinism and Hypopigmentation.

Results: Complete survey results were available for 72 participants, aged 1 to 16 years. Parents of 52 (72%) children reported that their child had cycles of good sleep and bad sleep, and 50 (69%) reported that their child's sleep patterns caused significant stress for them or their family. Scores on the Childhood Sleep Habits Questionnaire (CSHQ) increased (> 41) in 64 (89%) children, indicating a likely clinically significant sleep problem. When compared to normative data from children aged 4 to 10 years, children in the current sample scored higher (more sleep problems) on all eight subscales on the CSHQ. The presence of comorbid developmental delay was most strongly associated with sleep problems. Supplemental melatonin and improving daytime and nighttime schedules or routines were reported as the most helpful for sleep. Many families reported a need for further information regarding melatonin use as a supplement.

Conclusions: A high proportion of children with visual impairment experience clinically meaningful sleep problems, regardless of degree of light perception or visual acuity. There is a strong need for increased awareness and screening for sleep problems in this population. Potential treatment modalities, including supplemental melatonin, should be discussed with families.

[*J Pediatr Ophthalmol & Strabismus*. 2022;59(2):77-86.]

INTRODUCTION

Sleep problems are common in childhood, affecting approximately 25% to 50% of young children and approximately 40% of adolescents.[1] Individuals with neurodevelopmental disorders experience an even higher risk of sleep disorders, with some estimates up to 80% depending on the syndrome.[2] Sleep disruption or loss may cause or exacerbate problems in underlying neurobehavioral functioning, emotional regulation, and overall quality of life.[3]

Children with visual impairment specifically may be at increased risk of sleep problems given the potential disruption of circadian entrainment from disor-

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Submitted: February 22, 2021; Accepted: April 15, 2021; Posted online: August 26, 2021

Disclosure: Dr. Ingram has previously received support from Jazz Pharmaceuticals. The remaining authors have no financial or proprietary interest in the materials presented herein.

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doi:10.3928/01913913-20210623-01

dered light information processing. Entrainment refers to the matching of internal circadian rhythm with the environment. Indeed, previous studies in children with visual impairment have found a high prevalence of early morning waking, extensive daytime sleeping, increased need for physical contact during sleep, difficulty falling asleep, and increased nighttime awakenings.[4,5] The presence of optic nerve disease specifically has been shown to predict increased daytime napping.[6] Other predictors of abnormal sleep patterns in children with visual impairment include severe visual impairment, abnormal pupillary responsiveness, developmental delay, and multiple hormone deficiencies.[7] Melatonin and implementing strict schedules and routines have been suggested as helpful interventions for sleep in children with visual impairment, with limited evidence for light therapy.[4,8,9] Most of these previous studies have been limited by fairly small and local samples.

In the current study, our overall goal was to further elucidate sleep and its problems in children with a wide spectrum of visual impairment. Specifically, we sought to increase sample generalizability by inviting participation from families from across the country. We also attempted to include robust participation from families of children with oculocutaneous albinism, a much less studied population in terms of sleep. Finally, we investigated possible predictors of sleep problems in children with visual impairment.

PATIENTS AND METHODS

Participants

Research participants were recruited via an online survey. An invitation to participate in the survey was distributed via the National Federation of the Blind (NFB) and through the National Organization for Albinism and Hypopigmentation (NOAH). Parents were eligible to participate if their child was aged 1 to 17 years, English speaking, and had internet access. There were no direct benefits offered for participation.

Measures

The survey was web-based and anonymous with no identifiers collected, and responses were recorded and stored on a secure server in Research Electronic Data Capture (REDCap; Vanderbilt University). The survey consisted of several sections that asked about characteristics of the child, as well as sleep habits and problems. Parents were asked about general information including sex, level of visual impairment, level of light perception, eye conditions, neurological conditions, endocrine disorders, and other medical disorders.

In terms of sleep, parents were asked several specific questions regarding whether their child had previously undergone various sleep assessments. The presence of cycles of good and bad sleep was asked, as well as whether the child's sleep patterns caused significant stress for the family. Parents were asked about any mediations used for sleep, and specifically what they had been previously told about melatonin.

A comprehensive childhood sleep questionnaire, the Childhood Sleep Habits Questionnaire (CSHQ), was used. This 33-item questionnaire measures sleep problems in eight domains: Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing, and Daytime Sleepiness. Individual items were scored, and a total CSHQ score was calculated. Higher CSHQ scores indicate more sleep problems, and a total score cutoff of 41 was considered indicative of clinically significant sleep problems.[10] The CSHQ has been widely used in the past to assess sleep characteristics of children with a variety of neurodevelopmental disorders.[11-14] Although the CSHQ has been validated in children aged 4 to 10 years, it has also been used in previous studies of children as young as 1 year and into adolescence.[15-18] Finally, open-ended questions asked about challenges and helpful interventions for their child's sleep, as well as what they had previously been told regarding melatonin.

Statistical Analysis

Survey results were summarized by calculating mean and standard deviation for continuous variables and frequencies for categorical variables. Individual domains and total scores were calculated for the CSHQ, and results for children aged 4 to 10 years were reported separately for the purposes of comparison with a previous large national sample of typically developing 4- to 10-year-old children in the United States[16]; published means, standard deviations, and sample sizes made summary independent samples *t* tests possible. The assumption of equal variances was tested via the Hartley test in each comparison, and the *P* values reported are based on whether the assumption was accepted or rejected. A total CSHQ score of 41 was chosen as a cutoff to indicate clinically significant sleep problems. In ad-

dition, CSHQ scores and sleep characteristics were explored by selected participant characteristics to assess for the possible impact of level of visual impairment and comorbidities. For these comparisons, categorical variables were analyzed via chi-square tests, continuous variables with two levels via *t* tests, and continuous variables with more than two levels via analysis of variance with post-hoc pairwise comparisons as needed.

Responses to open-ended questions were analyzed in a qualitative fashion. Free-text answers were individually coded and categorized into representative themes using a grounded theory approach. Grounded theory stipulates that the collected data are systematically examined line by line, and key phrases are identified and coded into categories to uncover overarching themes.[19]

Statistical tests were two-sided with results considered statistically significant at a *P* value of less than .05. This study was approved by the Institutional Review Board at Children's Mercy Hospital. All data analyses were performed in SPSS software (version 23.0; IBM Corporation).

RESULTS

Survey Participation

The survey was distributed, and responses were collected from July 2020 until September 2020. Overall, 103 parents opened the survey, and 72 finished the survey in its entirety and were included in analysis. Seventy-one mothers and 1 father completed the survey.

Participant characteristics are delineated in **Table 1**. Children's ages ranged from 1 to 16 years (average of 5.92 ± 3.8 years). The sample was split almost evenly between boys and girls. There were a wide variety of levels of visual impairment, light perception, and underlying ocular history, likely reflecting the different parent organizations through which the survey was distributed. Only one child lacked both eyes. A majority of participants denied any significant neurological or endocrine history, and the most common medical comorbidities were developmental delay and seasonal allergies.

Sleep History

Fifteen children (21%) had previously had a sleep study, and nine (13%) had been diagnosed as having sleep apnea. Eight (11%) had undergone tonsillectomy and none had previously been receiving continuous positive airway pressure therapy. Seventeen children (24%) had been prescribed a hypnotic agent for sleep, and the same number had seen a sleep physician. None had previously undergone actigraphy, 4 (6%) had undergone melatonin level measurement, and 3 (4%) had ferritin levels checked. Forty (56%) children had never had a sleep evaluation. Parents of 52 (72%) children reported that their child had cycles of good sleep and bad sleep, and 50 (69%) reported that their child's sleep patterns caused significant stress for them or their family.

Medications for Sleep Problems

Parents were surveyed regarding numerous possible medications that could be used for sleep, and responses are presented in **Table 2**. Forty-five parents (63%) reported that they had given melatonin to their child as a medication to help sleep; of those who had used melatonin, 32 of 45 (71%) found it helpful. In contrast, no participants had ever tried the melatonin agonists ramelteon or tasimelteon. Clonidine was the second most helpful medication surveyed, although only 6 (8%) had used and found it to be helpful. Few other medications were reported as ever been tried for sleep or helpful. Interestingly, most parents were not interested in the majority of medications for sleep, with the exceptions of melatonin, iron, and vitamin D.

Parents were asked what they had previously been told about melatonin, and free text responses were coded into themes. The most frequent theme was "nothing" (n = 18), followed by melatonin helps with sleep initiation but not maintenance (n = 13). There were several comments that raised questions about melatonin safety (n = 6) and appropriate dosing (n = 7). Some parents had been told melatonin helps to reset sleep cycles (n = 5), whereas others had been told that giving melatonin can result in tolerance or decreased natural melatonin production (n = 4). Concerns were raised about the possibility of melatonin causing endocrine problems (n = 2), mood problems (n = 2), nightmares (n = 1), or worsening seizures (n = 1).

CSHQ

Scores on the CSHQ are reported in **Table 3**. Overall, 64 of 72 (89%) children had CSHQ scores greater than 41, indicating a likely clinically significant sleep problem. When compared to normative data from children aged 4 to 10 years, children in the current sample scored higher (more sleep prob-

TABLE 1

Participant Characteristics

Characteristic	Value
Age (years), mean ± SD	5.92 ± 3.8
Sex	
Male	37 (51%)
Female	35 (49%)
Visual impairment severity	
Able to see the top letter on the vision chart	33 (46%)
Unable to see the chart but can see to count fingers	11 (15%)
Unable to count fingers but can see shadows and hand movement	4 (6%)
Unable to see shadows but can see light	6 (8%)
Unable to see light	18 (25%)
Light perception	
No light perception	18 (25%)
Light perception only	6 (8%)
Photophobia or extreme light sensitivity	42 (58%)
Normal reactions to light	6 (8%)
Eyes	
Both	67 (93%)
One	4 (6%)
None	1 (1%)
Ocular history	
Albinism	32 (44%)
Aniridia	3 (4%)
Anophthalmia	3 (4%)
Cataracts	2 (3%)
Coloboma	0 (0%)
Glaucoma	6 (8%)
Lebers congenital amaurosis	4 (6%)
Nystagmus	41 (57%)
Optic atrophy	1 (1%)
Optic nerve hypoplasia	13 (18%)
Strabismus	13 (18%)
Other	17 (24%)
None of the above	4 (6%)
Neurological history	
Hypoxic ischemic encephalopathy	0 (0%)
Periventricular leukomalacia	0 (0%)
Cerebral palsy	4 (6%)
Traumatic brain injury	3 (4%)
Brain infections (meningitis)	1 (1%)
Seizures	8 (11%)
Hydrocephalus/shunt	1 (1%)
Polymicrogyria	1 (1%)
Lissencephaly	0 (0%)
William's syndrome	0 (0%)
Dandy-Walker syndrome	0 (0%)
Cytomegalovirus infection during pregnancy	0 (0%)
Other	8 (11%)
None of the above	51 (70%)
Endocrine history	
Growth hormone deficiency	7 (10%)
Thyroid hormone deficiency	6 (8%)
Adrenal hormone deficiency	6 (8%)
Diabetes insipidus	5 (7%)
Other	1 (1%)
None of the above	61 (85%)
Medical history	
Seizure disorder requiring daily medication	5 (7%)
Hearing impairment	4 (6%)
Gastroesophageal reflux	7 (10%)
Congenital heart disease	2 (3%)
Autism spectrum disorder	9 (13%)
Developmental delay	21 (29%)
Atopic dermatitis/eczema	9 (13%)
Seasonal allergies	14 (19%)
None of the above	32 (44%)

SD = standard deviation

lems) on all eight subscales on the CSHQ. In addition, although bedtimes were similar, waketimes were significantly earlier and sleep duration was less in children with visual impairment.

Sleep and Comorbidities

Several participant characteristics were explored as potential associations with sleep problems, with

TABLE 2

Medications for Sleep Problems

Medication	Yes, and Helpful	Yes, but Not Helpful	No, but Interested	No, and Not Interested
Melatonin	32 (44%)	13 (18%)	11 (15%)	16 (22%)
Iron	4 (6%)	6 (8%)	34 (47%)	28 (39%)
Vitamin D	1 (1%)	11 (15%)	35 (49%)	25 (35%)
Clonidine	6 (8%)	1 (1%)	13 (18%)	52 (72%)
Trazadone	2 (3%)	0 (0%)	8 (11%)	62 (86%)
Gabapentin	3 (4%)	3 (4%)	9 (13%)	57 (79%)
Zolpidem	0 (0%)	1 (1%)	9 (13%)	62 (86%)
Ramelteon	0 (0%)	0 (0%)	9 (13%)	63 (88%)
Tasimelteon	0 (0%)	0 (0%)	12 (17%)	60 (83%)
Doxepin	0 (0%)	0 (0%)	10 (14%)	62 (86%)
Suvorexant	0 (0%)	0 (0%)	10 (14%)	62 (86%)
Hydroxyzine	1 (1%)	2 (3%)	8 (11%)	61 (85%)
Diphenhydramine	2 (3%)	5 (7%)	7 (10%)	58 (80%)
Clonazepam	0 (0%)	1 (1%)	12 (17%)	59 (82%)
Eszopiclone	0 (0%)	0 (0%)	10 (14%)	62 (86%)
Zaleplon	0 (0%)	0 (0%)	11 (15%)	61 (85%)
Amitriptyline	0 (0%)	0 (0%)	10 (14%)	62 (86%)
Mirtazapine	0 (0%)	0 (0%)	10 (14%)	62 (86%)
Risperidone	1 (1%)	0 (0%)	8 (11%)	63 (88%)

results presented in **Table 4**. Of the comorbidities examined, developmental delay had the most impact, with significant associations found with sleep duration, parasomnia (abnormal behaviors during sleep), sleep-disordered breathing (sleep apnea or other breathing problems during sleep), daytime sleepiness, and total CSHQ scores. Presence of neurological or medical comorbidities were only associated with increased sleep-disordered breathing scores. Neither optic nerve hypoplasia nor endocrine comorbidities were associated with any sleep outcome. Albinism was associated with less daytime sleepiness. Those children with no light perception had significantly worse sleep duration scores and increased chance of sleep problems causing significant family distress. Children who were unable to count fingers but able to see shadows and hand movements had worse sleep-disordered breathing scores.

Challenges and Helpful Strategies

Parents were asked about their child's greatest sleep challenge and what they have found to be most helpful for sleep. The most common themes for challenges were sleep maintenance (n = 35), sleep initiation (n = 24), daytime napping or daytime sleepiness (n = 6), and the child requiring parental presence for sleep (n = 5). Less common themes were nighttime crying (n = 3), nothing (n = 3), restlessness (n = 2), poor sleep quality (n = 2), nightmares (n = 1), behavioral consequences of poor sleep (n = 1), bedwetting (n = 1), seizures (n = 1), and sleep apnea (n = 1). The most frequently helpful interventions parents reported were consistent schedule/routine (n = 22), melatonin (n = 20), cosleeping (n = 12), and relaxation exercises/music (n = 12). For clarity, cosleeping refers to the parent and child sharing the same sleep surface. Other responses included a cool and dark room (n = 9), sleep medications in addition to melatonin (n = 5), decreasing light exposure (n = 3), essential oils (n = 3), transitional object (n = 3), decreasing naps (n = 2), good morning light (n = 2), mineral supplements such as iron/calcium/magnesium/zinc (n = 2), sensory interventions such as tight swaddling or vibrating mattress pad (n = 2), ketogenic diet (n = 1), exercise, treatment of sleep apnea (n = 1), nighttime tea (n = 1), cannabidiol oil

TABLE 3

CSHQ Scores (Mean ± SD)

Question	Current Sample, Total (N = 72)	Current Sample, 4 to 10 Years (n = 40)	Typically Developing Sample, 4 to 10 Years (n = 357 to 415)	*P*
Bedtime, h	8.1 ± 0.9	8.3 ± 1.0	8.4 ± 0.5	.535
Waketime, h	6.5 ± 1.0	6.5 ± 1.2	6.9 ± 0.4	.042
Sleep duration, h	9.6 ± 2.6	9.4 ± 2.1	10.1 ± 0.6	.042
Bedtime resistance	9.3 ± 3.3	9.9 ± 3.4	7.0 ± 1.8	< .001
Sleep onset delay	1.9 ± 0.8	1.9 ± 0.7	1.2 ± 0.5	< .001
Sleep duration	6.0 ± 1.8	6.2 ± 1.9	3.4 ± 0.9	< .001
Sleep anxiety	6.8 ± 2.3	7.3 ± 2.4	4.8 ± 1.4	< .001
Night waking	5.3 ± 1.6	5.5 ± 1.6	3.4 ± 0.8	< .001
Parasomnias	10.5 ± 2.3	10.4 ± 2.3	8.1 ± 1.3	< .001
Sleep-disordered breathing	3.6 ± 1.2	3.5 ± 0.9	3.2 ± 0.6	.045
Daytime sleepiness	12.4 ± 2.8	12.3 ± 2.6	9.7 ± 2.8	< .001
Total CSHQ	52.4 ± 9.3	53.3 ± 8.2	38.7 ± 5.5	< .001

CSHQ = Childhood Sleep Habits Questionnaire; SD = standard deviation

(n = 1), and safe sleeper bed (n = 1). Several parents (n = 9) reported nothing helped their child's sleep.

DISCUSSION

Sleep problems are commonly encountered in children, particularly those with neurodevelopmental challenges. In the current study, we sought to add to the existing literature in the specific area of sleep in children with visual impairment. Previous studies have documented that children with visual impairment have an increased risk of having sleep problems, and this increased risk was largely attributed to lack of light perception resulting in disrupted entrainment of the circadian sleep wake cycle and classically a non-24 circadian rhythm disorder.[20] In this circumstance, individuals with a non-24 pattern will tend to have a cycling mismatch between their internal circadian rhythm and the outside world, resulting in disrupted nighttime sleep and daytime sleepiness. Although that classic description is certainly valid, characterizing sleep in children with visual impairment as a whole is a challenge given that they are a heterogenous group both in terms of severity of visual impairment, etiology of visual impairment, and comorbid medical conditions. Results from our study may provide a broader view of sleep problems in these children and potentially helpful interventions.

In our heterogenous, nationwide sample, we found a high overall prevalence of sleep problems. Most families reported cycles of good/bad sleep in their children, and most families reported sleep patterns causing significant distress in the family. Furthermore, approximately 90% of children had CSHQ scores indicating a clinically significant sleep problem, and the nature of sleep problems spanned all types, with significantly elevated subscale scores compared to published norms. The presence of comorbid developmental delay was most closely associated with worse sleep, which is in keeping with a robust literature documenting the high prevalence of sleep disorders in children with neurodevelopmental disorders.[2] The presence of underlying neurological or medical comorbidities was associated with an increased risk of sleep-disordered breathing in our sample. We speculate that this represents the increased risk of sleep apnea conferred by conditions such as reflux, aspiration, cardiac dysfunction, seasonal allergies, and epilepsy in medically complex children. Surprisingly, we did not find any significant association between the endocrinopathies and sleep problems. This finding is in contrast to a previous study of children with optic nerve hypoplasia,[7] and may be due to a relatively lower prevalence of hormonal deficiencies in our sample.

We examined the degree of visual impairment in relation to sleep problems and found that children who completely lacked light perception had significantly worse sleep duration. We suspect that

TABLE 4

Sleep Habits According to Vision Level and Comorbidities[a]

Parameter	Bedtime Resistance	Sleep Onset Delay	Sleep Duration	Sleep Anxiety	Night Waking	Parasomnias	Sleep-Disordered Breathing	Daytime Sleepiness	Total CSHQ	Alternating Good /Bad Sleep Cycles	Sleep Causing Family Distress
Sex											
Male (n = 37)	9.3 (3.2)	1.9 (0.8)	5.8 (2.0)	6.6 (2.3)	5.1 (1.6)	10.9 (2.3)	3.8 (1.5)	12.5 (2.8)	52.6 (9.7)	62%	68%
Female (n = 35)	9.3 (3.5)	1.8 (0.8)	6.2 (1.7)	7.1 (2.4)	5.6 (1.6)	10.2 (2.3)	3.3 (0.6)	12.3 (2.9)	52.3 (9.0)	83%[b]	71%
Visual acuity											
Able to see the top letter on the vision chart (n = 33)	9.4 (3.6)	1.9 (0.8)	5.7 (1.7)	7.6 (2.4)	5.6 (1.6)	10.7 (2.2)	3.3 (0.7)	12.0 (2.8)	56.1 (9.2)	70%	76%
Unable to see the chart but can see to count fingers (n = 11)	8.4 (2.3)	1.6 (0.7)	5.2 (1.7)	6.3 (2.2)	4.6 (1.4)	9.7 (1.3)	3.5 (0.8)	14.0 (2.9)	50.6 (7.9)	73%	64%
Unable to count fingers but can see shadows and hand movement (n = 4)	9.3 (4.3)	2.3 (1.0)	7.0 (2.3)	6.5 (2.6)	5.5 (1.0)	13.3 (1.7)	6.3 (2.8)[c]	13.8 (1.9)	60.5 (6.4)	75%	50%
Unable to see shadows but can see light (n = 6)	9.0 (3.3)	2.2 (1.0)	5.3 (2.3)	5.7 (1.6)	5.2 (2.3)	10.5 (1.9)	3.2 (0.4)	10.8 (2.5)	48.5 (11.3)	33%	17%
Unable to see light (n = 18)	8.8 (3.3)	1.7 (0.8)	6.9 (1.7)[c]	6.3 (2.0)	5.3 (1.5)	10.1 (2.7)	3.6 (1.0)	12.3 (2.8)	51.9 (10.0)	89%	83%[b]
Light perception											
No light perception (n = 18)	8.8 (3.3)	1.7 (0.8)	6.9 (1.7)[c]	6.3 (2.0)	5.3 (1.5)	10.1 (2.7)	3.6 (1.0)	12.3 (2.8)	51.9 (10.0)	89%	83%
Light perception only (n = 6)	10.0 (4.1)	2.3 (1.0)	6.2 (2.6)	6.3 (2.4)	5.8 (2.1)	11.2 (2.6)	3.1 (0.4)	11.5 (2.7)	52.8 (13.9)	33%	33%
Photophobia or extreme light sensitivity (n = 42)	9.3 (3.3)	1.9 (0.7)	5.7 (1.7)	7.0 (2.4)	5.3 (1.6)	10.6 (2.1)	3.6 (1.3)	12.5 (3.0)	52.3 (8.6)	69%	69%
Normal reactions to light (n = 6)	10.3 (3.6)	1.8 (1.0)	5.0 (1.8)	8.2 (2.3)	5.5 (1.6)	10.8 (2.0)	4.0 (1.3)	12.8 (2.1)	54.2 (9.8)	83%	67%
Albinism											
Yes (n = 32)	8.9 (2.9)	2.0 (0.7)	5.6 (1.8)	6.7 (2.0)	5.2 (1.6)	10.3 (1.9)	3.4 (0.7)	11.7 (2.4)	50.5 (7.6)	69%	72%
No (n = 40)	9.6 (3.7)	1.8 (0.9)	6.3 (1.8)	7.0 (2.6)	5.5 (1.6)	10.8 (2.5)	3.8 (1.4)	13.0 (3.0)[b]	54.0 (10.4)	75%	68%
Optic nerve hypoplasia											
Yes (n = 13)	9.5 (3.9)	1.8 (0.9)	6.5 (1.9)	7.2 (2.6)	5.5 (2.1)	11.2 (2.5)	3.5 (1.0)	12.0 (2.6)	53.6 (12.3)	62%	77%
No (n = 59)	9.3 (3.2)	1.9 (0.8)	5.9 (1.8)	6.8 (2.3)	5.3 (1.5)	10.4 (2.2)	3.6 (1.2)	12.5 (2.9)	52.2 (8.7)	75%	68%
Developmental delay											
Yes (n = 21)	9.2 (3.5)	2.1 (0.8)	6.9 (1.6)	6.8 (2.4)	5.3 (1.2)	11.9 (2.7)	4.4 (1.8)	13.6 (2.7)	56.8 (8.5)	81%	76%
No (n = 51)	9.3 (3.3)	1.8 (0.8)	5.6 (1.8)[b]	6.9 (2.3)	5.4 (1.8)	10.0 (1.9)[b]	3.3 (0.5)[b]	11.9 (2.7)[b]	50.6 (9.1)[b]	67%	67%
Any neurological comorbidity											
Yes (n = 21)	9.0 (3.5)	2.0 (0.9)	6.5 (1.9)	6.4 (2.3)	5.6 (1.5)	10.6 (2.5)	4.4 (1.7)	12.1 (2.4)	53.4 (9.3)	86%	67%
No (n = 51)	9.4 (3.3)	1.8 (0.8)	5.8 (1.8)	7.0 (2.3)	5.2 (1.6)	10.5 (2.2)	3.2 (0.5)[b]	12.5 (3.0)	52.0 (9.4)	67%	71%
Any endocrine comorbidity											
Yes (n = 11)	9.5 (4.1)	1.7 (0.8)	6.9 (1.8)	6.6 (2.5)	5.3 (1.7)	10.3 (3.2)	3.7 (1.2)	11.6 (2.4)	52.3 (11.9)	73%	73%
No (n = 61)	0.2 (3.2)	1.9 (0.8)	5.8 (1.8)	6.9 (2.3)	5.4 (1.6)	10.6 (2.1)	3.6 (1.2)	12.5 (2.9)	52.5 (8.9)	72%	69%
Any medical comorbidity											
Yes (n = 40)	9.4 (3.5)	2.0 (0.8)	6.3 (1.9)	6.7 (2.2)	5.4 (1.5)	10.9 (2.4)	3.8 (1.4)	12.5 (2.7)	53.4 (9.3)	80%	70%
No (n = 32)	9.2 (3.1)	1.8 (0.7)	5.6 (1.7)	7.1 (2.4)	5.3 (1.7)	10.1 (2.0)	3.3 (0.5)[b]	12.3 (3.0)	51.1 (9.3)	63%	69%

[a]Values are presented as mean (standard deviation).
[b]Significant at the level of $P < .05$ for chi-square or t test comparisons.
[c]Significant at the level of $P < .05$ for post-hoc analysis of variance comparisons.

this finding is related to the fact that the lack of light perception leads to disruption of the normal circadian entrainment that happens in sighted individuals, leading to more frequent nighttime awakenings and disrupted sleep. These findings are congruent with prior studies in children with visual impairment,[4,5] as well as literature on non-24 sleep disorders in adults.[21] Free response data from the current study also highlighted sleep maintenance as the most frequently encountered sleep challenge. Importantly, children with intact light perception still had a high prevalence of sleep problems, so clinicians should not assume normal sleep with normal light response. One recent study demonstrated that light-induced suppression of melatonin secretion remains intact in a minority of totally blind individuals with eyes, and also those individuals with inner retinal damage are at higher risk of lack of photoreceptor responsiveness; the take-home point for clinicians being that circadian entrainment cannot be reliably predicted by the underlying type of blindness.[22]

One aspect of the current study that was particularly novel was the inclusion of a large number of children with visual impairment related to albinism. In theory, children with albinism may have hypersensitivity to light, which in turn could affect circadian entrainment. Indeed, we did find a high prevalence of sleep disorders of every nature in these children in our sample. Children with albinism tended to have lower (less problematic) scores for daytime sleepiness compared to other children with visual impairment, which we speculate may be due to increased alerting effects of light during the day. In contrast, we did not find significantly different scores on any other sleep characteristics compared to other children with visual impairment, including sleep onset delay. This is clearly an area and group of children who would benefit from additional study.

In addition to delineating sleep challenges, the current study also highlights several potential avenues for therapy. Parents reported several strategies as helpful for their child's sleep, the most frequent of which were improving consistent schedule and routine, as well as supplemental melatonin. These findings are congruent with previously published studies,[9] and may represent interventions that help to entrain a child's circadian rhythm. In addition to its hypnotic effects, the administration of supplemental melatonin can terminate the drifting of sleep/wake rhythms in many individuals with visual impairment.[23] Parental reporting that routine and schedule were important for better sleep is not surprising, because those timed environmental cues likely represent zeitgebers that may also help entrain the circadian rhythm of children. Zeitgebers are time cues, which may be either behavioral (meals, social interactions, etc) or endogenous (melatonin levels, body temperature), that sustain our circadian rhythm and orient sleep schedule to desired routine.[24] Cosleeping has previously been reported to be associated with visual impairment in children.[25] The lack of many parents mentioning light therapy as an effective intervention is consistent with the limited and thus far negative published data in children with visual impairment.[9,26] Additional frequently reported helpful measures included relaxation exercises, soothing music, and ensuring a cool/dark sleep environment. A litany of less well studied interventions deserve further study, such as essential oils, mineral supplements, dietary changes, and cannabidiol oil.

Melatonin supplementation deserves special discussion. Our results indicate that although many families found melatonin to be helpful for their child's sleep, there were a lot of areas of uncertainty and questions that were unanswered. Parents were unsure about the appropriate timing and dosage to use. Based on our experience and available published literature,[27] we suggest melatonin be dosed based on age and intended effect. When used for hypnotic effects (to promote drowsiness at bedtime), melatonin should be given approximately 30 minutes before bedtime in higher doses of 1 to 3 mg in children weighing less than 40 kg and more than 5 mg in children greater than 40 kg. In contrast, when the intended use of melatonin use is as a chronobiotic (to shift circadian rhythm earlier), a smaller starting dose of 0.2 to 0.5 mg can be given 3 to 4 hours prior to bedtime. We typically do not initiate melatonin supplementation earlier than 6 months of age. Although data are limited, time-release versions of melatonin are also available and may be helpful in children with more difficulty with sleep maintenance insomnia.[28]

Furthermore, parents had several concerns about the safety of melatonin supplementation. Importantly, melatonin is not currently regulated by the U.S. Food and Drug Administration and therefore available over-the-counter preparations may vary more than 400% in purity.[29] In terms of long-term safety and side effects, there is concern

that because melatonin is a hormone, exogenous supplementation could affect the production of other hormones in the body and therefore pubertal development; this risk remains theoretical with no evidence of such an effect in several human studies.[30,31] There does not seem to be consistently reported major side effects due to melatonin supplementation,[32] and withdrawal does not occur with discontinuation.[31] Specifically, supplemental melatonin also has not been found to increase seizure risk in individuals with epilepsy, and in fact some studies demonstrated a decrease in seizures, possibly due to improved sleep.[32] Surprisingly, there were no children in our study who had ever tried (and little interest from families) the melatonin agonists ramelteon or tasimelteon, which are more widely used in adults with insomnia or circadian rhythm disorders.

The current study has many strengths. First, although we did not query the specific geographic location of respondents to preserve anonymity, the fact that the survey was distributed via national advocacy organizations suggests that our findings may be widely generalizable to children with visual impairment. Second, the sample was heterogenous in terms of type and severity of visual impairment, allowing us to explore associations with sleep concerns. Third, the inclusion of a substantial number of children with albinism allowed us to delineate sleep issues within this condition, something that had not previously been studied in detail. Fourth, our survey included open-ended questions that allowed for a more in-depth appreciation of family experiences in addition to the standardized questionnaire.

The current study has several potential limitations. First, the nature of the survey was subjective, using parent-reported symptoms rather than objective sleep measures. Future studies using actigraphy (measuring sleep/wake patterns in ambulatory settings) and polysomnography (overnight sleep study to evaluate sleep architecture, breathing, and limb movements) would augment these subjective results with objective sleep metrics. That said, the CSHQ is a widely used and validated sleep questionnaire in children that has normative data available. In addition, subjective sleep concerns are ultimately of most practical clinical import. Second, the study sample size was relatively modest, limiting power to detect significant associations and generalizability. Third, there could be a selection bias in our survey results, with those families who experience sleep challenges potentially more likely to respond. That said, sampling is a challenge in this population in other ways because of the low incidence of blindness and severe vision loss in children in the United States. It does not mitigate the possible bias, but even if our estimates are uncertain, they highlight the need for further research into this area to do more robust studies. Fourth, the nature of our survey did not allow for formal diagnosis of circadian rhythm disorders, such as non-24. These disorders are of particular interest in children with visual impairment, and we speculate that the high rates of "good and bad sleep cycles" and reported sleep maintenance issues may represent underlying circadian rhythm entrainment problems. Fifth, although developmental delay was most closely associated with sleep problems in our study, this comorbidity was based on parent report without additional degree or type of impairment specified.

Our results substantiate the high prevalence of sleep problems in children with visual impairment. These sleep problems spanned every type, with an especially high rate of sleep maintenance issues (difficulty staying asleep throughout the night). The presence of developmental delay further increases chances of sleep problems, and medical and/or neurological comorbidity confers increased risk of sleep-disordered breathing. Clinicians should be alert for and inquire regarding sleep problems in these patients, regardless of the child's type and severity of visual impairment, because they represent a potentially treatable condition that impacts child and family quality of life. Future studies are needed to further evaluate potential diagnostic and treatment modalities specific to children with visual impairment.

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