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BRIEF REPORT

Brief, Family-Centered, Videoconference-Based Behavioral Insomnia Program in Elementary School-Aged Pediatric Cancer Survivors: A Proof-of-Concept Study

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ABSTRACT

Pediatric cancer survivors are at heightened risk for insomnia. Though behavioral interventions are the recommended approach, there are not enough trained clinicians. No known published trials have been conducted among school-aged survivors, despite them having unique age-related sleep issues. Here, we tested a videoconference-based behavioral insomnia intervention program enrolling 15 caregivers of cancer survivors (5–12 years) with insomnia. Staff without prior sleep education/training delivered intervention sessions providing evidence-based strategies tailored to family needs. Self-report measures were collected before intervention and approximately 2 weeks post intervention. Caregivers reported significant sleep and quality-of-life improvements. Thus, we show preliminary support for a brief intervention that does not require significant resources or clinical expertise.

1 | Introduction

More than 85% of American children diagnosed with cancer will survive [1, 2]. However, in this growing survivor population, poor sleep is common. Approximately 30% report clinically significant insomnia and are significantly more likely to use sleep medication compared to their siblings [3–5]. Sleep disturbances are associated with neurocognitive impairment, emotional distress, fatigue, and worse quality-of-life in survivors [5–7]. The longer a child struggles with sleep, the more likely they are to develop these problems [8–10]. Thus, it is imperative that pediatric cancer survivors receive evidence-based sleep treatment.

As there are no U.S. Food and Drug Administration-approved medications for pediatric insomnia and those used off-label have daytime side effects [11, 12], the American Academy of Sleep Medicine (AASM) has stated that behavioral interventions (e.g., extinction methods, standardized bedtime routines) are the gold standard for insomnia in healthy children [13]. The duration of these interventions varies considerably, with most ranging

Abbreviations: AASM, American Academy of Sleep Medicine; CSHQ, Children's Sleep Habits Questionnaire; PedsQL, Pediatric Quality of Life Inventory; PROMIS, Patient-Reported Outcomes Measurement Information System; Survivor-SHIP, Survivor Sleep Health Information Program.

between 2 weeks to 2 months. The AASM concluded that "even relatively short interventions (one to three sessions) can be very effective in improving sleep in early childhood" [14]. Unfortunately, there are not enough trained behavioral sleep medicine providers [15, 16], and likely far fewer with expertise in pediatric oncology. Further, the child's age impacts behavioral intervention strategies, as there are different sleep challenges and treatment strategies depending upon developmental stage [17]. We are not aware of a published trial evaluating behavioral interventions for insomnia in elementary school-aged (approximately 5–12 years of age) survivors [18].

Our trial endeavors to address these limitations in the literature by assessing whether a virtual intervention has the promise to be an effective treatment to reduce a known sequela (insomnia) for a vulnerable pediatric cancer survivor population. As clinicians working in survivorship specialty clinic programs often lack sleep training [19, 20], and because there is a dearth of pediatric behavioral sleep medicine providers [15], we sought to determine if non-behavioral sleep medicine cancer center staff could effectively deliver a manualized insomnia intervention to elementary school-aged survivors.

2 | Methods

2.1 | Participants

Participants were recruited at medical appointments, via provider referrals, or mailed/emailed letters. We enrolled a caregiver of a child who: (i) was between 5–12 years of age; (ii) was diagnosed with any cancer, except non-melanoma skin cancer; (iii) did not receive cancer treatment (e.g., radiation, surgery) other than chemoprevention in the past 6 months, and had no further treatments planned; (iv) met DSM-5 diagnostic criteria for insomnia disorder assessed through a structured diagnostic interview conducted by a research assistant [21]; (v) had no prior diagnosis of a seizure or developmental disorder; (vi) did not have untreated sleep apnea; (vii) had no plans to adjust sleep medications. The study was IRB-approved and registered at clinicaltrials.gov (NCT04863157).

2.2 | Intervention

Our Survivor Sleep Health Information Program (Survivor-SHIP) was adapted from a family-centered program designed to address sleep problems in typically developing children [22, 23]. Content was tailored for pediatric cancer survivors following discussions between the study authors, pediatric oncologists, and the study interventionists. This included information regarding treatment-related late effects that could impair sleep [24]. Study interventionists met individually with caregivers approximately every other week for three videoconference sessions, providing psychoeducation and home practice recommendations. Session 1 (approximately 45 minutes) provided an overview of common sleep issues for pediatric cancer survivors and a discussion of the caregiver's primary concerns about their child's sleep. The interventionist worked with the caregiver to set one or two manageable sleep goals (e.g., developing an appropriate bedtime routine, encouraging the child to fall asleep on their own), and provided them with targeted handouts. In Sessions 2 and 3 (approximately 30 minutes each), the interventionist provided tailored feedback and education based on how the child was responding to intervention efforts, addressed challenges, and discussed ways to support maintenance of changes.

2.3 | Study Interventionists

Interventionists were a program manager without a clinical background and a clinical social worker, both of whom worked with pediatric cancer patients but did not have sleep training. Before the trial, interventionists: (i) reviewed a pediatric behavioral sleep medicine textbook [25]; (ii) received a full-day Survivor-SHIP training; and (iii) met monthly for 3 months to receive training on pediatric sleep disorders and protocol implementation with the principal investigator.

2.4 | Measures

Study measures were completed by the caregiver at baseline (approximately 1 week before Session 1) and follow-up (approximately 1 week following Session 3).

Demographics and medical history: were collected by caregiver report and medical record review (baseline).

Sleep disturbance and impairment: The Patient-Reported Outcomes Measurement Information System (PROMIS) Sleep Disturbance and Sleep-Related Impairment [26] scales are each eight-item measures used to assess the extent and consequences of a child's poor sleep.

Sleep habits: The Children's Sleep Habits Questionnaire (CSHQ) [27] is a 33-item questionnaire that assessed the child's overall sleep habits.

Quality of life: The Pediatric Quality of Life Inventory (PedsQL) [28] is a 23-item scale measuring core physical, mental, and social health dimensions.

2.5 | Statistical Analysis

Descriptive statistics were calculated for study demographic, disease-specific, and outcomes data. Wilcoxon matched pair tests assessed baseline/follow-up changes in the child's sleep and quality of life, with Hedge's g calculated to determine effect sizes.

3 | Results

Of 15 enrolled participants, 10 completed all intervention sessions and assessments, providing data for these analyses. Two enrolled caregivers were lost to follow-up before the first session, two were lost due to increasing family demands, and one completed the intervention but not their follow-up assessment. Children were on average 9.7 ± 1.9 years old, and primarily non-Hispanic White (Table 1). At baseline, most were taking a sleep medication. At follow-up (Table 2), participants reported statistically significant pre/post-intervention improvements in sleep disturbance and

TABLE 1 | Demographic and cancer-related descriptives ($N = 10$).

	$\bar{x}$ (SD)	N (%)
Demographic		
Age (years)	9.7 (1.9)	
Gender		
Male		6 (60.0%)
Female		4 (40.0%)
Ethnicity		
Non-Hispanic White		8 (80.0%)
Multiple ethnicities		2 (20.0%)
Sleep medications		
None		2 (20.0%)
Over the counter		7 (70.0%)
Over the counter and prescription		1 (10.0%)
Cancer-related		
Primary diagnosis		
Brain tumor		1 (10.0%)
Leukemia		6 (60.0%)
Solid tumor		3 (30.0%)

TABLE 2 | Sleep and health changes following the intervention.

	Baseline ($N = 10$)	Follow-Up ($N = 10$)	
	Mean (SD)	Mean (SD)	Effect size (g)
Children's Sleep Habits Questionnaire			
Total summary score	54.3 (9.0)	36.2 (5.2)[a]	2.9
Patient-Reported Outcomes Measurement Information System Sleep Disturbance			
Total summary score	27.0 (4.4)	18.5 (5.5)[a]	3.1
Patient-Reported Outcomes Measurement Information System Sleep-Related Impairment			
Total summary score	18.8 (4.7)	14.6 (6.0)[a]	1.2
Pediatric Quality of Life Inventory			
Physical function	57.5 (27.6)	71.3 (25.5)[a]	0.9
Emotional function	55.5 (20.2)	78.5 (21.4)[a]	1.6
Social function	69.0 (23.2)	76.5 (20.0)	0.6
School function	65.5 (25.3)	74.5 (21.5)[a]	0.8

Note: Baseline data were collected approximately 1 week before Session 1. Follow-up data were collected approximately 1 week after Session 3. Hedge's *g* effect size range: small = 0.2, medium = 0.5, large = 0.8.

[a]Follow-up values that are significantly different ($p < 0.05$) from baseline.

sleep-related impairment, better sleep habits, and improved physical, emotional, and school quality of life.

4 | Discussion

Though behavioral interventions are recommended, many cancer programs do not have an expert to provide evidence-based treatment [19]. Our findings showing significant improvements in sleep and quality of life following the intervention support the novel approach of using a brief, family-centered videoconference intervention led by staff without sleep expertise. Although preliminary, the robust improvements seen in this study, with medium to large effect sizes across outcomes, are comparable to those seen for families receiving standard behavioral insomnia intervention treatment [29].

Long-term follow-up care guidelines for pediatric cancer survivors recommend annual screening for sleep disturbances and a referral for behavioral interventions if insomnia is identified [30]. While e-health interventions may play a role [31, 32], clinician-directed treatment can result in greater symptom improvements and lower attrition [33]. Thus, finding treatment pathways that do not require extensive prior clinician training is important to increase accessibility [34]. As our interventionists received only approximately 25 hours of training, pediatric cancer centers could consider how existing staff could provide insomnia treatment for families.

4.1 | Limitations

The results must be considered preliminary and viewed considering several limitations. Our small sample was comprised primarily of non-Hispanic White families. In addition, though our retention rate was consistent with studies of other psychosocial interventions in this population [35], the fact that several participants dropped out of the trial further limits generalizability and raised a concern of selection bias. Measurement in the trial was limited and did not include objective sleep measures or assessment of covariate that could have impacted outcomes (e.g., duration of sleep medication used prior to the trial). Finally, a limited follow-up period and lack of comparison group limit the ability to understand duration of observed effects or infer causation. Despite these limitations, however, the trial provides proof of principle results that support ongoing investigation of this novel approach to addressing the sleep problems commonly seen in pediatric cancer survivors. Specifically, we now plan to conduct a randomized trial with a larger and more diverse sample, including more comprehensive sleep assessment, and comparison with a control condition over an extended follow-up period.

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Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

The data that support the findings of this study are available from the corresponding author upon reasonable request.

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