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Original Research Article

Initial Validation of the Sleep Disturbances in Pediatric Cancer Model

Lauren C. Daniel,[1] PhD, Lisa A. Schwartz,[1,2] PhD, Jodi A. Mindell,[2,3,4] PhD, Carole A. Tucker,[5] PhD, and Lamia P. Barakat[1,2] PhD

[1]Division of Oncology, The Children's Hospital of Philadelphia, [2]Department of Pediatrics, Perelman School of Medicine of the University of Pennsylvania, [3]Division of Pulmonary Medicine, The Children's Hospital of Philadelphia, [4]Department of Psychology, Saint Joseph's University, and [5]College of Public Health, Departments of Physical Therapy and Electrical and Computer Engineering, Temple University

All correspondence concerning this article should be addressed to Lauren C. Daniel, PhD, Division of Oncology, The Children's Hospital of Philadelphia, 3501 Civic Center Boulevard, 10300 CTRB, Philadelphia, PA 19104, USA. E-mail: daniell@email.chop.edu

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Abstract

Objective The current study evaluates content validity of the Sleep Disturbance in Pediatric Cancer (SDPC) model using qualitative and quantitative stakeholder input. **Methods** Parents of children (aged: 3–12 years) with acute lymphoblastic leukemia ($n = 20$) and medical providers ($n = 6$) participated in semi-structured interviews about child sleep during cancer treatment. They also rated SDPC model component importance on a 0–4 scale and selected the most relevant sleep-related intervention targets. **Results** Qualitatively, parents and providers endorsed that changes in the child's psychosocial, environmental, and biological processes affect sleep. Stakeholders rated most model components (parent: 32 of 40; provider: 39 of 41) as important (>2) to child sleep. Parents were most interested in interventions targeting difficulty falling asleep and providers selected irregular sleep habits/scheduling, though groups did not differ significantly. **Conclusions** Stakeholders supported SDPC content validity. The model will inform subsequent measure and intervention development focusing on biological and behavioral factors most salient to sleep disturbances in pediatric cancer.

Key words: cancer; qualitative data; sleep.

Sleep disturbances are common during pediatric cancer treatment (Crabtree & Brimeyer, 2014; Rosen, Shor, & Geller, 2008); however, prevalence rates vary widely across studies, partly due to measurement variability and validity for use in cancer, with estimates ranging from 25% (Jacob, Hesselgrave, Sambuco, & Hockenberry, 2007) to 87% (Zupanec, Jones, & Stremler, 2010). Children with cancer and parent-proxy reporters indicate low sleep efficiency (Orsey, Wakefield, & Cloutier, 2013; Wright, 2011), less restful sleep (Wright, 2011), long sleep onset latencies (Rosen & Brand, 2010; Wright, 2011), and high levels of daytime sleepiness (Rosen & Brand, 2010) across cancer diagnoses. Although caregiver report of child sleep durations is similar to healthy control groups (Wright, 2011), actigraphy (a device worn on the wrist that provides an estimate of sleep time) indicates that children with cancer receive far less sleep than normative values (Orsey et al., 2013), suggesting that even though children may spend an adequate amount of time in bed, awakenings can reduce sleep duration. Children with cancer also face increased risk for sleep disturbances due to some common treatments that may affect sleep–wake regulation (steroids and radiation), disease location and central nervous system involvement, pain, medication side effects, and psychological response to cancer treatment (Rosen & Brand, 2014).

Children with acute lymphoblastic leukemia (ALL), the most common childhood cancer (Altekruse et al., 2010), are particularly vulnerable to sleep disturbances due to frequent corticosteroid administration [medications known to impact mood, behavior, and sleep propensity (Rosen et al., 2008)]. ALL treatment consists of three phases: induction, consolidation, and maintenance. Maintenance therapy lasts approximately 2–3 years, during which time the patient receives a combination of oral and intravenous agents typically on an outpatient basis, including monthly 5-day corticosteroid courses (Pui, 2008). Parents of children with ALL report frequent sleep disturbances relative to normative values across all phases of ALL treatment (van Litsenburg et al., 2011; Zupanec et al., 2010). Studies examining the impact of steroids on sleep in ALL maintenance therapy have found changes to sleep duration (Hinds, Hockenberry, Gattuso, et al., 2007; Rosen et al., 2015), increased daytime sleep (Hinds, Hockenberry, Gattuso, et al., 2007; Rosen et al., 2015), and fatigue (Daniel, Kloss, Szabo, Reilly, & Barakat, 2011), and indicators of a less robust circadian sleep–wake activity such as reduced physical activity and less regularity in sleep/wake patterns during steroid courses (Rogers, Zhu, Ancoli-Israel, & Hinds, 2014). Sleep disturbances in ALL maintenance therapy are negatively correlated with health-related quality of life (HRQL; van Litsenburg et al., 2011) and positively correlated with fatigue (Zupanec et al., 2010). Furthermore, these sleep disturbances persist into long-term survivorship for as many as half of ALL survivors (Meeske, Siegel, Globe, Mack, & Bernstein, 2005).

Although the occurrence of sleep disturbances in pediatric cancer has been described, examination of mechanisms of sleep disturbances is lacking and, to our knowledge, no models of sleep disturbances in pediatric chronic health conditions have been proposed. Therefore, we developed the Sleep Disturbance in Pediatric Cancer (SDPC; Figure 1) model to summarize the key components related to disrupted sleep identified in the pediatric oncology and general pediatric literature. The SDPC development was informed by socio-ecological theory (Bronfenbrenner, 1986), adult cancer models (Savard & Morin, 2001), infant sleep models (Sadeh & Anders, 1993), and expert review. This model is intended to inform sleep measurement and intervention development for pediatric cancer.

The SDPC model incorporates psychosocial, environmental, and biological mechanisms that may contribute to changes in sleep observed in child cancer. Psychosocial factors, such as family behaviors around sleep, often change during treatment. Parents report using co-sleeping shortly after diagnosis to comfort the child, but over time this behavior becomes difficult to discontinue, often leading to child and caregiver sleep disturbances (Williams & McCarthy, 2014). For some parents, difficulty setting limits during cancer treatment (Enskar, Carlsson, Golsater, Hamrin, & Kreuger, 1997; Williams, Lamb, & McCarthy, 2014) may also affect the child's sleep habits and patterns. Such changes in parental behaviors around the child's sleep during cancer treatment may affect the child's ability to fall asleep and return to sleep independently, potentially resulting in long-term sleep onset and sleep maintenance difficulties. Furthermore, psychosocial responses to cancer, such as anxiety and depression, can further disrupt sleep. Although the impact of psychological distress on sleep has not been examined during childhood cancer treatment, sleep disturbances are common in children with anxiety and depression (Chorney, Detweiler, Morris, & Kuhn, 2008).

Environmental changes related to cancer diagnosis and treatments, such as hospital stays, changes in sleeping location, and medical interventions at night, are another mechanism of sleep disturbances. Frequent hospitalizations can affect sleep hygiene and sleep patterns at the hospital and after returning home (Meltzer, Davis, & Mindell, 2012), especially due to the high frequency of awakenings for nursing care (Hinds, Hockenberry, Rai, et al., 2007). Hospital noise and light can also contribute to poor sleep quality. Changes in daytime routines and environments further alter sleep habits and can affect circadian sleep/wake cycles. For children not attending school regularly, inconsistent sleep schedules and the ability to nap freely during the day may affect nocturnal sleep by reducing the drive for sleep at bedtime (Berger et al., 2005).

Several biological processes may also contribute to disrupted sleep. Children with cancer often experience pain related to disease processes and treatment. The pediatric pain literature suggests that pain can result in reductions in sleep efficiency, slow wave sleep, and rapid eye movement sleep (Onen, Onen, Courpron, & Dubray, 2005). Disrupted or insufficient overnight sleep can, in turn, interfere with pain coping and perception of pain the following day (Lewin & Dahl, 1999). Studies in adults with cancer suggest that alterations to melatonin secretion, abnormalities in cortisol levels, and changes in the production of cytokines may contribute to sleep–wake disruptions (Berger et al., 2005). Cancer-related fatigue can also affect sleep–wake patterns, disrupting nighttime sleep habits and sleep propensity (Rosen, 2007). Corticosteroids (Hinds, Hockenberry, Gattuso, et al., 2007), antiemetic medications (Luginbuehl & Kohler, 2009), analgesics (Onen et al., 2005), and chemotherapy (Gedaly-Duff, Lee, Nail, Nicholson, & Johnson, 2006) may alter sleep architecture and propensity. Furthermore, misalignment between the circadian system and homeostatic sleep needs (which are based on timing and quality of recent sleep periods) due to to irregular sleep schedules, awakenings for medical care, daytime

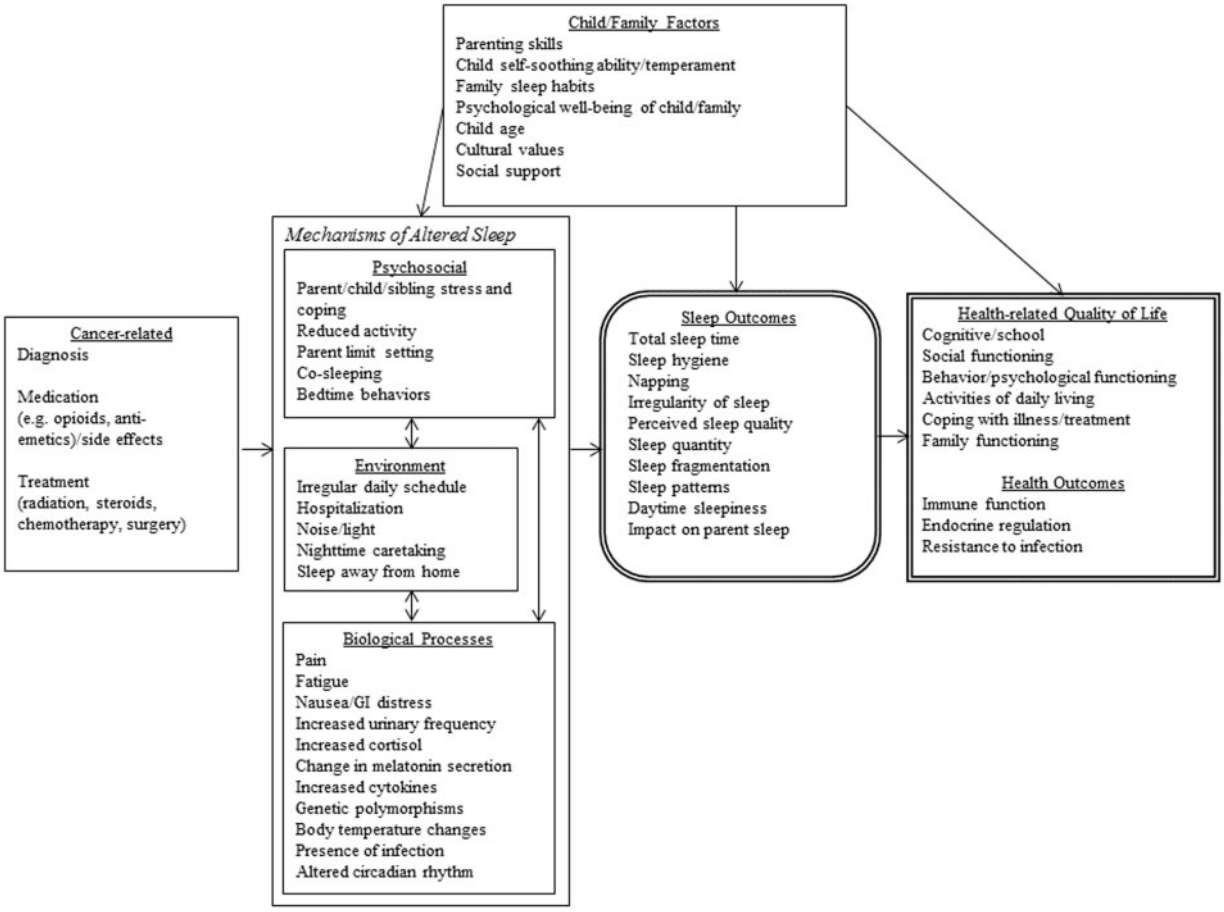

Figure 1. Sleep Disturbances in Pediatric Cancer model.

sleep, and treatments that affect sleep propensity may contribute to difficulty attaining a regular sleep–wake schedule during treatment (Rosen, 2007). Such dysregulated sleep cycles are biomarkers for poor cancer outcomes and high symptom burden in adult cancer (Innominato et al., 2014).

The current study seeks to validate the SDPC model, which will inform cancer-specific assessment and intervention development. Using quantitative and qualitative methods, the aims of the current study were to (1) validate the SDPC model via qualitative (semi-structured interviews) and quantitative (content validity ratings of SDPC Model components) data from relevant stakeholders and (2) assess interest in sleep interventions and intervention targets. Feedback will inform a cancer-specific sleep screening measure to guide systematic, universal sleep assessment in oncology and the development of behavioral interventions that are viewed as important and acceptable to key stakeholders.

Methods

Participants

Families were purposefully sampled to include those with and without sleep concerns from a larger study describing sleep in children during ALL maintenance therapy. Twenty-six families were approached for the current study (23 families enrolled, 3 families refused, 2 families declined further participation due to caregiver health concerns, and 1 family was lost to follow-up) to yield a final sample of 20 parents (85% mothers, 10% father, 5% both parents) of children aged 3–12 years ($M = 6.25$; $SD = 2.60$). Purposeful sampling targeted caregivers of children with and without sleep concerns as indicated by the parent's report of their child's sleep on the Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000), using the clinical cut-point for referral (total score of 41). Sample demographics are presented in Table I.

Six oncology providers were also invited to participate in semi-structured interviews. Purposeful sampling was used to include oncology providers with a range of experience treating children with cancer (three attending physicians, one nurse practitioner, one instructor, and one fellow) and a range of time practicing in pediatric oncology (range: 2–18 years).

Procedures

The SDPC model was developed before data collection for the current study; it was informed by literature

Table I. Sample Demographics

Demographic variable	*M*	*SD*	Range
Child age	6.25	2.6	3.85–12.74
Caregiver age	36.65	7.41	20.00–52.00
Month in maintenance	12.62	9.55	1.04–32.07
Barrett measure of social status	44.49	12.54	21.83–63.33
Children's Sleep Habits Questionnaire total score	44.82	7.01	36.09–64.97
	n		%
Child gender female	9		45
Caregiver participating in the interview			
Mother	17		85
Father	2		10
Mother and father together	1		5
Caregiver race/ethnicity			
White	13		65
Asian	3		15
Black	2		10
Hispanic	1		5
Other	1		5
Number of siblings in the home			
Only child	5		25
1	6		30
2	7		35
3	1		5
>4	1		5

review, existing sleep models, and expert reviews. Experts in pediatric sleep medicine (physician and psychologist board certified in sleep/behavioral sleep medicine, nurse scientists), pediatric oncology (nurse scientist, oncologist), and qualitative methods (nurse scientist and clinical child and adolescent psychologists) reviewed the model for structure, completeness, and clarity. The model was revised based on expert reviews before use in semi-structured interviews conducted for this study, and revisions were made after data collection based on stakeholder data and feedback.

After institutional review board approval, parents were approached during their child's monthly oncology follow-up appointment. Parents provided informed consent before completing baseline measures of demographics and child sleep. Families who agreed to the qualitative study participated in semi-structured interviews at a subsequent oncology follow-up appointment. To enhance feasibility, semi-structured interviews were selected rather than focus groups so that interviews could be conducted during an oncology appointment. Oncology providers also provided informed consent before interviews.

Semi-Structured Interviews

Parents and providers were interviewed by the principal investigator using a semi-structured interview guide that was developed to maintain consistency across interviews. Interview guides were developed with input from experts in pediatric sleep, oncology, and qualitative interviewing. Interviews were audio-recorded to allow for later qualitative analysis. Interviews lasted approximately 30 min.

Parent interviews focused on the child's sleep during cancer treatment, family management of sleep, and potential interventions. First, parents were asked about sleep when the child was diagnosed, how sleep has changed, and to discuss a time during treatment when the parent's efforts to facilitate their child's sleep were unsuccessful. Next, parents were asked about managing the child's sleep during treatment, their interest in interventions to manage sleep (behavioral and/or medical), and the potential timing of these interventions. Lastly, parents reviewed the SDPC model with the interviewer, gave feedback on model modification, and completed the SDPC component rating form, including intervention targets.

For the oncology provider interviews, providers were asked when sleep disturbances are most prevalent during treatment, how they manage these disturbances, and the anticipatory guidance they provide for families (e.g., steroid-related disturbances, changes to sleep and fatigue during/after radiation). Next, providers were asked about the need for and timing of sleep interventions during cancer treatment. Lastly, providers reviewed the SDPC model with the interviewer, gave feedback on modifications, and completed the SDPC component rating form.

Measures

Parent Measures

Demographic Information. After study enrollment, parents completed a family demographic information form.

Barratt Measure of Social Status. This measure is a simplified version of the Hollingshead Four Factor Form of Social Status (Hollingshead, 1975) that has been used in pediatric cancer research previously (Jurbergs, Long, Ticona, & Phipps, 2009). Parent and grandparent education and occupation were used to estimate social class (Barratt, 2006). Total scores for the measure range from 8 to 66, with higher scores indicating higher social status.

The Abbreviated CSHQ. This 33-item parent-report measure describes the child's last week of sleep, with higher scores indicating more difficulty with sleep (Owens et al., 2000). Internal consistency was adequate for the current sample (Cronbach's $\alpha = .65$). CSHQ total score was used to describe the sample and to guide purposeful sampling of caregivers of children with and without sleep problems.

Parent and Provider Measures

SDPC Component Rating Form. Parents and oncology providers completed parallel forms to rate the importance of the SDPC model components (parents rated 40 components and providers rated 41 components because radiation is in the model but was an exclusion criterion for families to participate in this study) on a 0–4 scale, with higher scores indicating greater importance to their child's sleep (parents) or sleep of children with cancer (provider). The form did not include rating of the health and quality of life outcomes component of the model. Parents and providers also selected the three most important model components and three most relevant sleep intervention targets.

Data Analysis

Qualitative Data

Saturation was demonstrated, that is, no new themes attained in the data (Strauss & Corbin, 1990), in both parent and provider interview qualitative analyses. Parent and provider interviews were systematically evaluated by two graduate-level trained reviewers coding transcripts in three rounds using the principles of directed content analysis, with codes inductively derived from the data (Hsieh & Shannon, 2005). In the first round, each reviewer coded all interviews separately according to the interview guide. After primary coding, discrepancies were discussed and resolved with the principal investigator (L.C.D.). Themes and subthemes were developed with the reviewers and principal investigator between the first and second coding based on the first round of data coding and SDPC model components. In the second round, the data were coded for subthemes. Each reviewer served as the primary coder for half of the interviews and a second coder for the remaining interviews. The reviewers and the principal investigator resolved any discrepancies in the subthemes together. In the third round, coding involved reviewers and the principal investigator working together to finalize codes and subcodes.

Quantitative Data

Descriptive statistics were computed for demographic data, CSHQ, and ratings of the SDPC model component importance. Content validity criterion for each SDPC model component was defined as an average parent and provider rating of >2 (on a scale of 0–4; Polit & Beck, 2006). Items that did not meet the threshold for both parent and provider stakeholders were removed from the model. Parent and provider ratings of the three most important model components and the three most relevant intervention targets were compared using Wilcox sign-ranked tests.

Results

Description of Child Sleep

CSHQ scores ranged from 36 to 64 ($M = 44.82$, $SD = 7.01$). In all, 60% ($n = 12$) of the sample reported clinically significant child sleep concerns (total score > 41) and 40% ($n = 8$) of the sample were below the clinical cut-point.

Aim 1: Validation of the SDPC Model—Qualitative Results

Interview Themes

Qualitative data supported the SDPC Model. Within the qualitative data, two approaches to child sleep and behavior management emerged as interview themes: accommodating child needs (parent $n = 12$, 60%; provider $n = 0$) and maintaining consistency/normalcy (parent $n = 8$, 40%; provider $n = 4$, 67%). Parents endorsing accommodations reported, "It kind of worked not really putting him on a schedule. Some things you kind of just let go, and when we were going through extensive treatment, I just wanted him to be happy, so we just kind of worked into his pattern." Parents endorsing consistency stated, "Just continue to do what you usually do you'll be okay, don't change too much just because they are sick," and providers noted the difficulty families face with consistency "They want that magic fix, it is hard to establish a routine and keep it intact. It is hard for them to be firm with their child." Parents reporting accommodation ($n = 12$, 60%) were significantly more likely to report sleep disturbances above the clinical cutoff of the CSHQ (10 of 12 were above the CSHQ cutoff; $\chi^2 = 6.81$, $p = .019$). There were no differences between CSHQ sleep disturbances by parents endorsing the consistency attitude ($n = 8$, 40%) versus those who did not ($n = 12$, 60%; $\chi^2 = 0.46$, $p = .648$). Five parents (25%) endorsed both approaches.

SDPC Model Components

Model components, exemplar quotes, and the number of parents/providers endorsing each model component are detailed in Table II. Parents identified two child and family factors contributing to child sleep during cancer: child temperament ($n = 2$, 10%) and cultural values (e.g., co-sleeping beliefs, $n = 1$, 5%). Cancer-related factors were supported as potential causes of sleep disturbance. Parents and providers endorsed medications in general (parent $n = 4$, 20%; provider $n = 3$, 50%) and specifically steroids (parent $n = 10$, 50%; provider $n = 3$, 50%) and chemotherapy (parent $n = 1$, 5%; provider $n = 1$, 17%) as cancer-related factors that disturb child sleep during treatment.

With regard to mechanisms of disrupted sleep, parents and providers identified psychosocial, environmental, and biological factors that affect child sleep during cancer treatment. Specifically, the psychosocial subcomponents identified were parenting strategies to encourage the child to go to sleep, such as bedtime routine (parent $n = 14$, 70%; provider $n = 1$, 17%), co-sleeping (parent $n = 10$, 50%; co-sleeping—provider $n = 2$, 33%), and child coping with cancer affecting sleep (parent $n = 3$, 15%; provider $n = 5$, 83%). Of the 10 parents who indicated co-sleeping, 4 reported that co-sleeping is a problem on the CSHQ. Parents indicated that co-sleeping could alleviate parent anxiety ($n = 2$, 10%) and child anxiety ($n = 2$, 10%). Other parents reported that co-sleeping disrupts family sleep ($n = 2$, 10%). Oncology providers described co-sleeping as an indicator of anxiety ($n = 1$, 17%) and as a common practice ($n = 1$, 17%).

Environmental subcomponents identified were transition between home/hospital (parent $n = 7$, 35%; provider $n = 0$), hospital environment (parent $n = 5$, 25%; provider $n = 2$, 33%), and electronics (parent $n = 4$, 20%; provider $n = 0$). Biological factors identified were pain (parent $n = 5$, 25%; provider $n = 0$), night sweats (parent $n = 3$, 15%; provider $n = 0$), hunger (parent $n = 2$, 10%; provider $n = 0$), altered circadian rhythm (parent $n = 2$, 10%; provider $n = 2$, 33%), and nausea (parent $n = 1$, 5%; provider $n = 0$).

Parents described the specific sleep outcomes experienced by their child and themselves during the course of cancer treatment. Changes endorsed were more night awakenings (parent $n = 9$, 45%), poor parent sleep (parent $n = 3$, 15%), more daytime sleepiness (parent $n = 3$, 15%), long sleep onset latencies (parent $n = 2$, 10%), and increased restlessness (parent $n = 1$, 5%). Oncology providers were not asked directly about changes to sleep.

Management of Sleep During Cancer

Parents were asked about strategies used to manage their child's sleep during cancer. In addition to themes already discussed that reflect parenting approaches to sleep and behavior management (accommodation or consistency) and parent sleep behavior management strategies, such as using a bedtime routine (under the psychosocial mechanism), parents also endorsed strategies ranging from using Benadryl ($n = 2$, 10%) and melatonin ($n = 2$, 10%), using a light ("good morning light") to indicate when the child can get out of bed ($n = 1$, 5%), giving medication while the child was asleep ($n = 1$, 5%), using television for white noise ($n = 1$, 5%), increasing physical activity ($n = 1$, 5%), and additional comfort from parent ($n = 2$, 10%).

Providers reported managing patient sleep with Benadryl and/or Ativan ($n = 3$, 50%). Five providers reported using melatonin infrequently, typically in response to a family's request ($n = 1$, 83%) or if the family is using Benadryl/Ativan daily ($n = 1$, 17%). Two providers noted that they do not prescribe sleep aids and that if a patient needs something more than Ativan, Benadryl, or melatonin, then they need to consider anxiety or "some other issue." Sleep hygiene and a focus on consistency were also common methods of managing sleep concerns ($n = 3$, 50%).

Aim 1: Validation of the Model—Quantitative Results

Quantitative data supported the SDPC model. Parents and providers rated the majority of SDPC model components as important to child sleep during cancer treatment (>2), thus supporting the content validity of the SDPC Model (Table III). When asked to select the three most important model components, parents and providers endorsed cancer treatment-related factors with the greatest frequency (parent $n = 12$, 60%; providers $n = 6$, 100%). The second most frequently selected model component for parents was biological factors (parents $n = 11$, 55%; providers $n = 2$, 33%), while providers selected behavioral factors with the second greatest frequency (parents $n = 2$, 10%; providers $n = 3$, 50%). A Wilcox sign-ranked test indicated that selection of the most important model components did not differ significantly between parents and providers ($z = 0.68$, $p = .498$).

When asked to select the three most relevant intervention targets, parents endorsed difficulty falling asleep with the greatest frequency, while providers endorsed irregular sleep habits/scheduling (Table IV). Frequency of rankings of intervention targets did not differ significantly between parents and providers ($z = 0.08$, $p = .937$).

Aim 1: Data Integration and Model Modification

Qualitative and quantitative data were consistent and these data supported minor revisions to the SDPC model. Co-sleeping, bedtime behaviors, and parent limit setting were redefined from *sleep outcomes* to *behavioral mechanisms*, due to qualitative data

Table II. Stakeholder Qualitative Support of the Sleep Disturbances in Pediatric Cancer Model

Model component	Subcomponent	Parent ($N = 20$) Exemplar quote	Provider ($N = 6$) Exemplar quote
Child and family factors	Child temperament	$n = 2$ (10%): *But I remember very well that after she turned one my little angel decided to not let her mom sleep ever again.*	–
	Cultural	$n = 1$ (5%): *I think there is a cultural thing where most Asian parents, I'm from India, where sleeping with a parent until 7 or 8 is normal and I have my own colleagues right from day 1 have their own room.*	–
Cancer related	Medications	$n = 4$ (20%): *The medications do play a role in how well she sleeps and stays asleep and the quality of her sleep.*	$n = 3$ (50%): *Challenging part is using Ativan and Benadryl for other things outside of sleep. If a parent says taking Ativan every night, I suggest melatonin.*
	Steroids	$n = 10$ (50%): *Falling asleep is harder with the steroids … I still put her in bed by 9:30, brush her teeth, everything, she just wouldn't fall asleep.*	$n = 3$ (50%): *With steroids, if parents want to give their child Benadryl before bed, I say that's OK but not to do it every night.*
	Chemotherapy	$n = 1$ (5%): *Certain meds made patient have to go to the bathroom (high dose methotrexate), that was a big disruption because he would have to get up several times during the night, would need fluids with those meds.*	$n = 1$ (17%): *Within the first couple of cycles of chemo, [sleep disturbance] is pretty obvious in kids with bone/solid tumors. With ALL more so in clinic because they are moved to out-patient more quickly, not as prevalent in beginning as it is in maintenance because they are adjusting to going back to life.*
Mechanisms of altered sleep	Psychosocial	$n = 18$ (90%): *[It] is important he's always prepared for what's going on, I think that's helped out with us a lot from the beginning of this experience, it makes him prepared so I don't think it causes any anxiety and I think anxiety has a lot to do with sleep.* Subcategory—Coping	$n = 5$ (83%): *I feel like a lot of parents sleep with their kid because they are more nervous than the kid, because I have parents say "I can't leave them alone so I just put them in my bed" so is the kid initiating it? It's usually the parent because they're worried about something happening to the child in the middle of the night. So that's the parental anxiety and it's disrupting the child.* Subcategory—Co-sleeping
	Environment	$n = 13$ (65%): *When she was first admitted … I would always be sleeping next to her because she would not let me fall asleep away from her, so I would have to wait until she fell asleep to move onto the couch, so that was a safety blanket for her because she always knew I would be there.* Subcategory—Hospitalization	$n = 2$ (33%): *The two things that I've identified in my patients is the constant disruptions in the hospitals it's hard to get a good night sleep and the anxiety after being discharged can also make it hard to get enough sleep.* Subcategory—Hospitalization
	Biological processes	$n = 10$ (50%): *She would have night sweats, she didn't seem like she was comfortable to me, especially during the first 3 months.* Subcategory—Night sweats	$n = 2$ (33%): *I think when you're in and out of the hospital, your circadian rhythm can be thrown off, and some kids are never on a normal schedule.* Subcategory—Altered circadian rhythm
Sleep outcomes	Sleep onset latency	$n = 2$ (10%): *At first she slept a lot, maybe her sleep is different now, [it is] taking her much longer to fall asleep.*	–
	Impact on parent sleep	$n = 3$ (15%): *I noticed how horrible my sleep is, my sleep is truly horrible. It is not just that I don't sleep, I really don't get good sleep. I haven't tried to quantify, but he is an active sleeper so it disrupts my sleep.*	–
	Night awakenings	$n = 9$ (45%): *Even though she wakes up refreshed, she seemed to have a lovely night using me as a punching bag. She definitely moves a lot, you'll never find her in the same position in the*	–

(continued)

Table II. Continued

Model component	Subcomponent	Parent ($N = 20$) Exemplar quote	Provider ($N = 6$) Exemplar quote
		same hour. She always moves to somebody's bed. She has times where she wakes up, but now thankfully she doesn't get up screaming or crying.	
	Altered sleep schedule	$n = 2$ (10%): *Prior to this month, he was up all night and he would get up in the morning and I have to get up at 4-4:30 and he would be up … he would go to sleep at 8:30-9 in the morning and be asleep until 2 in the afternoon. Then I would come home from work and he would say he wants breakfast.*	–
	Daytime sleepiness/naps	$n = 5$ (25%): *There are times we notice [patient] is more sleepy, he may fall asleep and then we carry him upstairs. I know that even though he is not telling us he is tired, that is a sign that he is.*	–
	Restless	$n = 2$ (10%): *When she was first was diagnosed she had restless sleep for quite some time, I think it was probably about 8 or 9 months until she seemed to rest.*	–

Note. Providers were not asked directly about the specific changes to sleep during cancer treatment.

suggesting that these behaviors may occur in some families due to the cancer diagnosis and could affect sleep outcomes both positively (reduce parent/child anxiety) and negatively (poor parent sleep). Marital satisfaction and adherence to medical treatment were removed from the final model due to not meeting a priori content validity inclusion criteria (average rating of >2 across parents and providers) and because these model components were not identified as factors disrupting sleep in semi-structured interviews.

Aim 2: Interest in Intervention During Cancer Treatment

The majority of parents ($n = 14$, 70%) indicated interest in intervention for sleep disturbances that occur during cancer treatment, five parents (25%) were not interested in intervention, and one (5%) was unsure. Eight parents (40%) explicitly stated they would not want their child to have medications for sleep and three other parents (15%) expressed interest in medications. All providers indicated interest in providing education and behavioral interventions for sleep disturbances during cancer treatment.

Discussion

The SDPC model describes the impact of cancer and cancer treatment on child sleep through psychosocial, environmental, and biological pathways, taking into account preexisting child and family factors that may also impact sleep. Qualitative and quantitative data from parents of children with ALL and oncology providers supported the validity of the SDPC model. A valid model of sleep disturbances in pediatric cancer is essential to advancing intervention and measure development, and informing theoretically based research to improve HRQL.

Parent approaches to managing child sleep and behavior during cancer treatment were a strong theme present in most interviews. A larger number of parents endorsed the need to "accommodate/tend to the child's needs" (35%) than parents who endorsed the need to "maintain normalcy/consistency" (15%), with some parents endorsing both approaches (25%), and 25% of parents not endorsing either attitude. Both parenting approaches are important in the context of the unpredictable circumstances posed by cancer, but an overemphasis on accommodations may result in future challenges with behavior management. Prior research in ALL has indicated that parent "laxness" and/or overprotection is associated with increased child emotional and behavioral difficulties relative to children without cancer (Williams et al., 2014). Understanding a family's belief system about sleep is critical to developing effective interventions that families will adopt. Results indicating more sleep concerns

Table III. Stakeholder Ratings of Importance (on a 0–4 Scale) of Sleep Disturbances in Pediatric Cancer (SDPC) Model Components

SDPC model component	Subcomponent	Parent importance *M* (*SD*)	Provider importance *M* (*SD*)
Preexisting	Parenting strategies	2.75 (1.29)	3.00 (0.89)
	Child ability to calm self	2.75 (1.21)	3.17 (0.41)
	Family sleep habits	2.70 (1.34)	3.33 (0.52)
	Psychological well-being of child/family	2.80 (1.28)	3.33 (0.82)
	Child age	2.45 (1.28)	2.83 (0.41)
	Cultural values	1.11 (1.29)	2.17 (0.98)
	Social support	2.26 (1.63)	2.17 (0.98)
	Marital satisfaction	1.89 (1.76)	1.33 (1.21)
Cancer diagnosis		2.11 (1.24)	2.83 (0.41)
Cancer treatment	Medication	3.15 (1.04)	4.00 (0.00)
	Radiation[a]	–	3.00 (0.89)
	Chemotherapy	2.88 (1.11)	3.33 (0.82)
	Steroids	2.78 (1.52)	4.00 (0.00)
	Surgery	2.00 (1.83)	2.67 (0.52)
Mechanism	Parent/child/sibling stress and coping	2.50 (1.36)	3.33 (0.82)
Psychosocial	Adherence to medical treatment	1.95 (1.47)	1.83 (1.17)
	Reduced activity level	1.60 (1.31)	2.00 (0.89)
	Regular sleep habits	3.10 (1.12)	3.50 (0.34)
	Parent/child sleeping in same bed	2.15 (1.60)	2.50 (1.05)
	Child sleeping location	2.60 (1.31)	2.67 (0.82)
	Behaviors around bedtime	2.95 (1.36)	3.17 (0.98)
	Parent limit setting at bedtime	3.00 (1.41)	3.17 (0.75)
Mechanism	Irregular daily schedule	2.35 (1.53)	3.17 (0.75)
Environmental	Hospital stays	2.80 (1.54)	3.67 (0.52)
	Noise and light in hospital	2.45 (1.47)	3.83 (0.41)
	Nighttime caretaking	2.16 (1.42)	3.50 (0.84)
	Sleeping away from home	2.26 (1.59)	3.33 (0.82)
Mechanism	Pain	2.90 (1.25)	3.50 (0.55)
Biological	Fatigue	2.40 (1.31)	3.67 (0.52)
	Nausea	2.40 (1.35)	3.50 (0.58)
	Increased need to urinate	2.20 (1.54)	2.50 (0.84)
	Changes in hormones	1.37 (1.53)	2.33 (0.52)
	Night sweats	1.60 (1.67)	2.50 (0.55)
	Infection	1.67 (1.78)	2.40 (0.89)
Sleep outcomes	Total amount of sleep child receives	3.25 (1.16)	3.00 (0.63)
	Parent sleep quality	2.05 (1.47)	2.50 (1.22)
	Child sleep quality	3.32 (1.06)	3.33 (0.52)
	Daytime sleepiness	2.30 (1.38)	2.83 (0.75)
	Parent sleep	1.90 (1.41)	2.50 (0.84)
	Napping	2.05 (1.54)	2.20 (0.44)
	Changes to sleep since diagnosis	2.11 (1.37)	2.83 (0.75)

[a]Children in the current study did not receive radiation, thus parents did not rate this item.

for parents who endorse the attitude "accommodating child needs" suggest that supporting parents in maintaining limit-setting around sleep behaviors may be important to manage sleep effectively during cancer treatment.

Parents and providers were in agreement regarding cancer treatments as a primary cause of sleep disturbances exhibited in children with ALL. However, nonsignificant differences in secondary components may have clinical implications for treating sleep disturbances. Parents rated biological causes with high importance, and providers rated behavioral causes with high importance. The perception of some parents that their child's sleep is dictated by uncontrollable factors, such as steroids or pain, may result in families assuming a "learned helplessness" (Abramson, Seligman, & Teasdale, 1978) approach to parenting their child and managing their child's sleep. Alternatively, parents may choose to not address sleep concerns, assuming that they will resolve when the patient is no longer taking a specific medication or experiencing a specific symptom. Although behavioral strategies may not overcome significant side effects produced by steroids and chemotherapy, parent training in behavioral sleep strategies has proven efficacious in increasing parental competence (Wolfson, Lacks, & Futterman, 1992). The incidence of ALL peaks between 2 and 4 years of age (Pui, 2008), when parental behavior is critical to good child sleep. Thus, these children are at an optimal developmental phase for family-based sleep interventions (Mindell et al., 2006). Behavioral interventions encouraging

Table IV. Stakeholder Endorsement of Most Relevant Model Components and Intervention Targets

Model component and Intervention target	Parents endorsing $N = 20$ *n* (%)	Providers endorsing $N = 6$ *n* (%)
Model components[a]		
Cancer treatment	12 (60)	6 (100)
Biological mechanisms	11 (55)	2 (33)
Environmental mechanisms	6 (30)	2 (33)
Preexisting family factors	4 (20)	1 (17)
Psychosocial mechanisms	2 (10)	3 (50)
Cancer diagnosis	2 (10)	0 (0)
Intervention targets[a]		
Difficulty falling asleep	9 (45)	3 (50)
Nighttime awakenings	7 (35)	4 (67)
Irregular sleep habits	6 (30)	5 (83)
Co-sleeping	6 (30)	1 (17)
Parent not sleeping enough	6 (30)	0 (0)
Daytime sleepiness	5 (25)	0 (0)
Bedtime refusal	4 (20)	1 (17)
Moving sleeping locations	3 (15)	0 (0)
Poor sleep quality	3 (15)	4 (67)
Child not sleeping enough	3 (15)	0 (0)
Child sleeping too much	1 (5)	0 (0)
Parent difficulty falling asleep	1 (5)	0 (0)

[a]Parents and providers were asked to rate the three most important model components and the three most important intervention targets.

consistent routines, setting limits, and decisions regarding sleeping locations consistent with a parent's beliefs have the potential to increase parental efficacy and encourage adaptation.

Interestingly, parents appeared to be more amenable to behavioral strategies to manage sleep than medications, despite the majority of providers reporting that they use Benadryl, Ativan, or melatonin for sleep. Because of the long-term nature of cancer treatment, the use of Benadryl and Ativan to manage sleep is less than ideal due to side-effects of these medications and the potential for developing tolerance (Owens & Moturi, 2009). Current recommendations for the use of medications to manage symptoms of insomnia advise short-term use in combination with behavior strategies (Owens & Moturi, 2009). Because research in long-term ALL survivors suggests persisting sleep problems at higher rates than normative values (Gordijn et al., 2013; Meeske et al., 2005), behavioral interventions may be warranted. For example, providing families with behavioral strategies before medications become the primary sleep treatment may reduce long-term sleep disturbances by reinforcing good sleep hygiene earlier in their cancer trajectory.

The current study presents a mixed-method validation of the SDPC model based on the experiences of key stakeholders. There are, however, several limitations. Parents participated in qualitative interviews during ALL maintenance therapy; as such, sleep concerns may not have been as pronounced and may not fully capture the experience of a child during active treatment, requiring frequent hospital admissions. This study is also limited in the use of parent and provider report without objective estimates of sleep. The results may not generalize to all childhood cancers. Because of the interrelated nature of biopsychosocial factors described in the SDPC model, some subcomponents may be represented in two categories (e.g., co-sleeping could represent a behavioral mechanism, an environmental mechanism, or can be conceptualized as a sleep outcome). We attempted to categorize subcomponents based on qualitative data, conceptualizing mechanisms as potential intervention targets that could be improved to affect sleep. As such, further testing of the model will be important to understanding how the model components interact. Although saturation of themes was reached for both parents and providers, the small numbers of participants from one childhood cancer center may not be fully representative of views and experiences across children with cancer and their families. Finally, because the current study represents key stakeholder feedback on the observable influences on child sleep during cancer, we are limited in the conclusions we can draw with regard to the physiological and circadian factors that likely contribute to child sleep during cancer. Research in adult oncology has started to elucidate the important bidirectional relationship between a regular sleep–wake cycle and cancer outcomes, and further study in this area to incorporate into the SDPC model is warranted to fully represent the experience of children with cancer.

The SDPC model offers a theoretical framework to guide intervention and measure development seeking to improve sleep in children with cancer. Next steps in this program of research are to validate the SDPC with a larger sample of pediatric oncology families, including a wide range of ages, diagnoses, and diverse racial/ethnic backgrounds. Additionally, research seeking to understand the complex relationship between sleep physiology and cancer recovery in pediatrics is an important part of this broader program of research. Sleep can be overlooked in complex medical conditions, but the current data speaks to the importance of addressing sleep concerns to maximize HRQL. Behavioral sleep interventions are focused, effective, and durable (Mindell et al., 2006), and thus, their application to a pediatric cancer population offers a novel approach to improve child and family HRQL. Regular screening for sleep disturbances during cancer treatment is important to identifying sleep disturbances early and facilitating the provision of resources and interventions to support families in managing their child's sleep.

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