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EXAMINING SLEEP AND FAMILY FUNCTIONING IN PEDIATRIC
CRANIOPHARYNGIOMA USING ECOLOGICAL MOMENTARY ASSESSMENT

A dissertation submitted in partial fulfillment of the requirements for the degree of Doctor of
Philosophy at Virginia Commonwealth University

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December, 2019

i

Acknowledgements

I would like to thank my mentors, Drs. Marcia Winter and Joshua Langberg, for their unparalleled encouragement. I still am unsure how I got so lucky to have you both as my mentors. I came into this program as an over-enthusiastic, under-experienced graduate, and under your guidance, patience, and care, I have grown as a clinical psychologist, as a researcher, and as a mother. I will forever be grateful for your mentorship. I would also like to thank my parents. Dad, you have instilled in me a profound love for learning, and mom, you have been there to cheer me on throughout every step of the way. My husband, this degree is yours as much as it is mine. Thank you for your never-ending encouragement, support, and understanding when graduate school demands got in the way of our weekend plans. I look forward to growing on internship and beyond with you. Dr. Valerie Crabtree is another person who I owe much gratitude to; following one email proposing this project idea, she became supportive of my dissertation study. Thank you, Dr. Crabtree, for the doors that you have opened up for me and for your patience with our technical difficulties. You continue to provide support even after this study has been completed. I would also like to thank my dissertation committee members. Dr. Rohan, I am very fortunate to have shared my passion for pediatric hematology/oncology with you last year. Dr. Dzierzewski, your statistics expertise has been very helpful, and I thank you for your support throughout the F31 grant writing process. Dr. Corona, thank you for your support as Director of Clinical Training and as a really compassionate faculty member. To Adam, you have changed the meaning and purpose of life for me. Thank you for teaching me to slow down and be fully present with you. I hope to make you proud to be my son just as much as you make me incredibly proud and honored to be your mother. I love you.

Table of Contents

List of Tables

List of Figures

Abstract

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Major Co-Directors: Marcia Winter, PhD., Assistant Professor, Department of Psychology &
Joshua Langberg, PhD., Associate Professor, Department of Psychology

Craniopharyngioma is among the most common brain tumors in children and is

associated with greater rates of sleep problems compared to other pediatric cancers. However,

research examining sleep among youth with craniopharyngioma has been limited by a reliance

on retrospective reports or sleep studies. Families also play a crucial role in children's

adjustment following a pediatric cancer diagnosis, yet remarkably little is known about

transactional associations between family functioning and sleep in pediatric cancer. This study

examined cross-sectional and daily associations among family functioning, affect, and sleep

difficulties for youth with pediatric craniopharyngioma using retrospective reports and ecological

momentary assessment (EMA). Thirty-nine youth who underwent partial resection and proton

therapy and their primary caregivers completed retrospective reports, and youth completed daily

electronic surveys over a one-week period. At the end of the week, youth were asked for

feedback about their overall experiences using EMA surveys. Ordinal least squares regression

suggested significant associations between youth-reported but not parent-reported family

functioning, excessive daytime sleepiness, and insomnia. Multilevel modeling did not suggest

significant associations between daily family functioning, negative affect, and sleep efficiency.

Youth reported overall satisfaction and minimal burden from completing EMA surveys. Findings inform clinical recommendations for systematic screening efforts to acknowledge youth perceptions of family functioning and excessive daytime sleepiness at routine follow-up visits. Results highlight the importance of using a multifaceted approach to assess and treat sleep difficulties in pediatric oncology and of identifying potential pathways explaining associations between family functioning and sleep.

Examining Sleep and Family Functioning in Pediatric Craniopharyngioma using Ecological

Momentary Assessment

Brain tumors (BT) are among the most commonly diagnosed pediatric cancers, with more

than 23,000 children and adolescents affected in the United States between 2009 and 2013

(Ostrom et al., 2016). Although recent medical advances have drastically improved the five-year

survival rates of certain brain tumors (BT) to more than 80% in some cases, neurocognitive and

medical late effects still place significant strains on affected children and their families (Laffond

et al., 2012; Zada, Kintz, Pulido, & Amezcua, 2013). Disrupted sleep and excessive daytime

sleepiness are among the many frequently experienced symptoms in pediatric BT patients

(Brimeyer et al., 2016). Thus, sleep dysfunction can significantly contribute to patients' and

survivors' quality of life.

Patients diagnosed with craniopharyngioma, an intracranial pediatric brain tumor

affecting surrounding hypothalamic regions, are particularly susceptible to sleep dysregulation

even after maximal surgical excision, with 65-80% of patients continuing to experience sleep

dysfunction, fatigue, and behavior changes following treatment (Kaleyias, Manley, & Kothare,

2012; Jacola et al., 2016). In fact, sleep complaints are more frequent in pediatric

craniopharyngioma survivors than in other pediatric cancer survivors (Armstrong et al., 2017).

Due to the critical role sleep plays in neural regeneration, neurocognitive recovery, and

psychosocial functioning, examining sleep in pediatric craniopharyngioma patients may provide

insight into remediable targets for future interventions (Hakim et al., 2014; Short et al., 2013).

The family is crucial to several aspects of child functioning in pediatric cancer and BT. In

fact, research suggests bidirectional associations between family functioning and the

neurocognitive and psychological sequelae of childhood BT (Hocking et al., 2011; Peterson &

Drotar, 2006). Nevertheless, remarkably little is known about the interrelationship between family functioning and child sleep patterns in pediatric cancer populations, especially in craniopharyngioma patients, and how these factors combine to predict overall well-being. A deeper understanding of the role daily sleep patterns and family functioning play in youths' adjustment may facilitate the development of targeted interventions that can improve long-term quality of life for patients with BT and their families.

The overarching goal of the proposed study is to examine sleep patterns in a sample of pediatric craniopharyngioma patients and to evaluate daily associations among family functioning, sleep patterns, and mood using ecological momentary assessment (EMA). Past studies have identified associations between family characteristics and youth psychosocial outcomes in pediatric cancer (e.g., Robinson, Gerhardt, Vannatta, & Noll, 2007). Similarly, researchers have delineated associations between children's sleep patterns and family functioning in the general child literature (e.g., El-Sheikh et al., 2012). Nonetheless, research has yet to examine a model specifically focused on interrelationships among daily family functioning and sleep patterns and how youth daily sleep patterns influence daily affect. Further, past research on sleep patterns and correlates in pediatric cancer patients has tended to be limited by a reliance on retrospective self-report ratings. A deeper understanding of the role daily sleep and family functioning play in youth's adjustment and outcomes may facilitate the development of interventions that improve long-term quality of life for patients with BT and their families. Furthermore, given the known sleep issues in youth with pediatric craniopharyngioma, they are an ideal population to assess correlates of sleep difficulties in, as findings may inform research examining sleep in children with other cancer diagnoses.

<center>**Literature Review**</center>

Pediatric Craniopharyngioma and Sleep Dysregulation

  Childhood craniopharyngiomas are benign intracranial tumors that affect the hypothalamic-pituitary region of the brain (Poretti et al., 2004). These tumors most often occur during childhood or adolescence, with a peak incidence between the ages of five and fourteen, and their ten-year survival rates are around 85 to 92 percent (Laffond et al., 2012; Poretti et al., 2004). Craniopharyngiomas currently account for five to ten percent of all pediatric brain tumors (Manley et al., 2012). Treatment for pediatric craniopharyngioma typically involves complete or partial resection of the tumor followed by focal radiotherapy (Poretti et al., 2004). Often, radical resection poses a considerable risk of hypothalamic and optical chiasm damage (Jacola et al., 2016). Therefore, proton beam radiation therapy is arguably better suited to minimize damage to adjacent brain structures (Mizumoto et al., 2017). Nevertheless, because of the tumor's critical location, craniopharyngioma and its treatment are often associated with morbidity, including hormone dysregulation, deficits in memory and processing speed, behavioral changes, obesity, and sleep difficulties (Manley et al., 2012). Symptoms of hypothalamic dysfunction, including sleep and obesity, persist in more than 65% of patients even after treatment is completed (Poretti et al., 2004). Because these changes adversely impact children's quality of life (Walter et al., 2015), research efforts have aimed to identify correlates of and contributors to these difficulties that may be the target of psychosocial interventions.

  Pediatric craniopharyngioma patients report particular difficulties with sleep dysregulation and daytime sleepiness (Laffond et al., 2012; Zada, Kintz, Pulido, & Amezcua, 2013). For instance, approximately one-third of pediatric craniopharyngioma patients experience clinically significant levels of daytime sleepiness (Muller et al., 2002). Even long-term survivors

<center>3</center>

continue to struggle with sleep difficulties. For example, Manley et al. (2012) found that nine years past diagnosis, more than 60% of survivors reported daytime fatigue and difficulty falling asleep or staying asleep. Effects of treatment on hypothalamic and pituitary functioning have been shown to potentially influence sleep outcomes; for instance, a decrease in melatonin production in youth with craniopharyngioma has been associated with excessive daytime sleepiness (Muller et al., 2002). Similarly, disruption of normal circadian rhythms, or patients' biological clock, from hypothalamic damage could result in dysregulation of the sleep-wake cycle (Walter et al., 2015). Although the extent of tumor resection has not been shown to be significantly associated with sleep dysfunction, craniopharyngioma patients who receive more frequent radiation and have a higher BMI are significantly more likely to report greater daytime sleepiness (Manley et al., 2012).

Because of the sleep difficulties that youth with brain tumors – and especially those with craniopharyngioma experience, researchers have advocated for studies evaluating factors associated with sleep disturbances in this population (Brimeyer et al., 2016). For instance, the extent of excessive daytime sleepiness may influence patterns of neural activation during attention tasks in youth with craniopharyngioma (Jacola et al., 2016). Similarly, adult survivors of pediatric brain tumors who reported greater sleep disturbances and daytime sleepiness have lower quality of life and general health scores (Gapstur et al., 2009). Sleep is also critical for the recovery of neural processes for pediatric brain tumors (Jacola et al., 2016). In animal studies, disrupted sleep patterns have contributed to accelerated tumor growth and progression (Hakim et al, 2014). However, research to date has been limited methodologically to case studies and retrospective reports (Gapstur et al., 2009; Jacola et al., 2016). This study aims to further the

field by examining correlates of overall and daily sleep problems in adolescents with a craniopharyngioma diagnosis.

Sleep Difficulties in Pediatric Cancer Model

The Sleep Difficulties in Pediatric Cancer (SDPC) model was developed by Lauren Daniel and colleagues (2016) to describe potential mechanisms associated with sleep difficulties in youth with pediatric cancer. This model summarizes factors related to sleep in pediatric oncology using research from oncology but also from the general literature (Daniel et al., 2016). It also provides specific direction for assessing correlates of sleep disruption and developing interventions in pediatric cancer. The model highlights psychosocial factors that may influence youth sleep patterns, including changes in child or family functioning. Some processes by which family functioning may influence sleep difficulties in youth following a cancer diagnosis are co-sleeping or laidback parenting habits that may be difficult to discontinue (Williams, Lamb, & McCarthy, 2014; Williams & McCarthy, 2015). Similarly, child characteristics, such as temperament, anxiety and depression symptoms, and coping strategies may also influence the development or maintenance of sleep difficulties. Research has yet to examine associations between sleep and psychological factors in pediatric cancer, although findings from the general literature suggest that sleep disturbances are common in children with anxiety and depression (Chorney et al., 2008). The SDPC highlights the importance of addressing sleep concerns in pediatric oncology in order to improve health-related quality of life and health outcomes (Daniel et al., 2016). However, in the proposed model, the authors highlight gaps in current research, including a dearth of studies assessing factors associated with sleep difficulties in pediatric cancer. Studies are particularly needed to examine youth reports of family functioning, variations in daily sleep patterns and difficulties, and directionality of associations among sleep and

associated psychosocial characteristics. Therefore, this study aims to extend the current knowledge in the field and examine specific mechanisms that may contribute to sleep difficulties, as guided by the SDPC model.

Sleep Difficulties and Mood during Adolescence

Adolescence has been identified as a particularly important developmental period for both sleep problems and mood fluctuations. During this period, youth undergo hormonal changes, build upon their emotion regulation skills, and work to gain autonomy from their parents (Maciejewski et al., 2014). Overall, adolescence is an emotionally difficult process, thus youth are particularly vulnerable to developing symptoms of anxiety or depression (Maciejewski et al., 2014). One mechanism through which these symptoms may develop is via heightened mood fluctuations; adolescents report experiencing greater mood swings than younger children and adults (Maciejewski et al., 2014). Similarly, adolescents are more likely to experience sleep difficulties (Fricke-Oerkermann, 2007). In fact, adolescence has been described by some researchers as the "perfect storm" for the development of poor sleep patterns, mood fluctuations, and associated negative sequelae (Carskadon, 2011). Nevertheless, the associations among these constructs for youth with pediatric craniopharyngioma have yet to be examined.

Sleep during adolescence. Sleep problems are common in youth, affecting 17 to 41% of otherwise healthy children (Fricke-Oerkermann, 2007). As children become adolescents, although their sleep needs do not change, many teens do in fact sleep less due to factors like later bedtimes and early waketimes (Iglowstein, Jenni, Molinari, & Largo, 2003). Youth most often report difficulties initiating and maintaining sleep and significant difficulties with daytime sleepiness (Brand et al., 2009; Fricke-Oerkermann, 2007). Some factors associated with disrupted sleep habits include later bedtimes, consistently early school start times, pronounced

differences in weekday and weekend sleep habits, and circadian rhythm delays around the time of puberty (Carskandon, 2011). The effects of developmental changes and hypothalamic disruption that pediatric craniopharyngioma adolescents and young adults experience make it particularly important to further understand sleep patterns and their correlates in this population.

Sleep is a crucial part of children's healthy development, and sleep difficulties are associated with severe functional morbidity in otherwise healthy youth (Dewald et al., 2010; Owens et al., 2014). Insufficient sleep, excessive daytime sleepiness, and poor sleep quality have all been significantly related to academic, behavioral, and emotional functioning in children and adolescents (Fallone et al., 2002; Owens et al., 2014). For example, adolescents who have difficulty initiating or maintaining sleep have been found to have significantly higher rates of clinical depression in longitudinal studies (Lovato & Gradisar, 2014). Similarly, evidence suggests that individuals with compromised sleep (e.g., poor sleep quality and sleepiness) have impaired executive functioning and poor school performance (Dewald et al., 2010; Mitru, Millrood, & Mateika, 2002). Further, sleep deficits (e.g., shorter sleep duration or later bedtime) have been linked to increased body mass index in children and adolescents (Magee, Caputi, & Iverson, 2013). Insufficient sleep in adolescents may also contribute to increased reports of pain and poor overall health in the general population (Moore et al., 2008). Finally, insufficient sleep has been shown to adversely impact resilience following exposure to a stressful situation (Walter et al., 2015). Nevertheless, it remains unclear how disrupted sleep may contribute to pediatric craniopharyngioma outcomes. This study focuses on examining sleep correlates for youth with craniopharyngioma, as these youths are an especially vulnerable group for experiencing sleep difficulties (Manley et al., 2012).

Associations between sleep and mood. One factor that could be especially undermined by sleep disruptions and may also influence subsequent sleep processes is mood (Fuligni & Hardway, 2006; Kouros & El-Sheikh, 2014). Sleep deprivation and disrupted sleep patterns have been proposed to reduce individuals' ability to regulate negative emotions (Dahl & Lewis, 2002). For instance, healthy adolescents with shorter sleep durations report on average increased negative affect and reduced positive affect across both weekday and weekend nights; similarly, inconsistencies in adolescent sleep patterns are associated with negative mood (Fuligni & Hardway, 2006). Further, adolescents who are sleep deprived, with fewer than 6.5 hours of sleep on two consecutive nights, report less positive affect and heightened anxiety in comparison to their rested peers (Talbot et al., 2010). Furthermore, greater shifts in weekday to weekend sleep patterns are significantly associated with negative mood, difficulty with concentration, and increased daytime sleepiness; this is particularly important as adolescents sleep on average three hours later on weekends, a shift associated with changes similar to jet-lag symptoms (Brand et al., 2009). Importantly, the association between sleep and mood is bi-directional with adequate sleep leading to better emotion regulation skills via enhanced prefrontal cortex functioning (Talbot et al., 2010). Further, adolescents in sleep promotion programs have shown to improve both in sleep habits and emotional distress ratings (John et al., 2016).

The reciprocal effects of mood on sleep have been less extensively studied in youth. What is clear from the literature is that there are associations between anxiety and depressive symptoms and sleep related difficulties (Peterman et al., 2016). For instance, excessive rumination or catastrophizing may lead youth to experience difficulty falling asleep; thus, cognitive behavioral interventions targeted at improving anxiety symptoms have also resulted in changes in parent-reported youth sleep problems (Peterman et al., 2016). Moreover, some

theories suggest that affect may influence sleep, by either influencing individuals' appraisals of stressful situations throughout the day or by directly relating to better sleep habits (e.g., less difficulty falling asleep or staying asleep; Ong et al., 2017). A systematic review identifying associations between positive affect/mood (PA) and sleep provided the following summary: seven studies reported independent associations between daily PA and sleep, two yielded bi-directional associations, and seven failed to find a significant association (Ong et al., 2017). The majority of these studies assessed these associations with adult populations, and thus, more ambulatory studies are needed with youth, especially those who are faced with daily stressful situations or hassles associated with cancer's treatment and its side effects.

Family Functioning

Assessing sleep difficulties in children with brain tumors requires acknowledging the critical role that families play in children's adjustment (Van Schoors et al., 2016). Families are faced with several challenges following a child's diagnosis, including the need to redistribute family responsibilities, accommodate medical appointments, communicate openly about difficult emotions, and promptly address issues with treatment side effects (Van Schoors et al., 2015). In fact, more than fifty percent of parents or caregivers experience high amounts of distress around the time of their child's diagnosis, and levels of distress are observed to be higher than parents of otherwise healthy youth (Trask et al., 2003). Generally, a family's response to the pediatric cancer diagnosis and treatment requirements affects children's adjustment (Van Schoors et al., 2016). This is particularly important as related to the child's adaptation to and management of treatment, medical side effects, and frequent physician appointments (Kazak, 1989). The majority of families report coping well after the initial post-diagnosis period, with no significant differences in comparison to families of youth without cancer; however, a subset of families and

their youth struggle in response to treatment demands and challenges, and these adjustment difficulties usually persist one year following the cancer diagnosis (Trask et al., 2003; Van Schoors et al., 2016).

Family systems theories (FST) stress the interdependence of family members' adjustment to a child's illness and the importance of shifting responsibilities to accommodate the child's needs while maintaining the family unit (Kazak, 1997). There are several models, guided by FST, that have attempted to explain how families of youth with chronic illness adjust in response to medical stressors, but these theories have rarely been used as the guiding framework in pediatric cancer studies (Van Schoors et al., 2015 & 2016). For instance, the Family Adjustment and Adaptation Response Model (FAARS; McCubbin & Patterson, 1983) posits that the way in which a family copes is influenced by the interaction between the stressors associated with the illness and the resources that the family has access to. Stressors of the illness on the family unit may include financial difficulties, worries about medical outcomes, caring for other members in the household, and keeping up with daily chores (Carnes & Quinn, 2005). Family resources may encompass the social support a family has, financial stability, and spiritual support. Finally, family members' threat appraisals and acceptance of the diagnosis also contribute to their coping abilities. The way in which a family may adjust in the first part of treatment and then adapt during the latter half of the coping process is usually dependent on the complex interplay of all these contributing factors. Over time, families go through various phases within these cycles of stability, adjustment to the stressor, and adaptation (McCubbin & Patterson, 1983). The FAARS and other FST theories stress the fact that certain family characteristics (e.g., overall functioning, cohesion, communication, conflict) influence adaptation of youth in response to a significant stressor (Van Schoors et al., 2016).

Family functioning and pediatric cancer. Research in cancer populations has identified significant associations between family functioning and child psychosocial outcomes (Robinson, Gerhardt, Vannatta, & Noll, 2007). For instance, greater parental distress within a year from adolescents' cancer diagnosis is associated with poor youth adjustment (Trask et al., 2003). Similarly, family functioning is related to youth adjustment following a cancer diagnosis (Van Schoors et al., 2016). Aspects of family functioning that are particularly associated with youth adjustment (e.g., internalizing, externalizing, and posttraumatic stress symptoms) include cohesion, communication, family support, and low conflict (Van Schoors et al., 2016). In clinical work, family problem-solving, roles, affective responsiveness, affective involvement, and behavior control are all important dimensions. Measures guided by FST, such as the Family Assessment Device, assess the functioning of a family system across these six dimensions (Epstein, Baldwin, & Bishop, 1983). Nevertheless, the majority of studies examining associations between family functioning and youth outcomes in pediatric cancer are cross-sectional and focus on retrospective parent reports of family functioning (Trask et al., 2003; Maurice-Stam et al., 2007). Studies that rely on retrospective parent report can be limited by recall and social desirability biases and may be affected by parents' most striking or recent family experiences (Lippold et al., 2014). Alternatively, focusing on daily assessments of family functioning provides the opportunity to test how fluctuations in family life may affect other aspects of youths' functioning (e.g., sleep or mood; Timmons & Margolin, 2014).

Family functioning and youth sleep. Family characteristics and processes (e.g., family conflict and unstructured routines) are associated with children's sleep/wake problems in otherwise healthy children (e.g., El-Sheikh et al., 2012), though this has not been studied in youth with pediatric brain tumor diagnoses or survivors. For instance, adolescents who have

parent-set bedtimes often report having extended sleep durations and less daytime fatigue (Short et al., 2011). Similarly, adolescents who report having parents with positive parenting styles also report having better mood, increased concentration, and fewer difficulties with excessive daytime sleepiness; these adolescents also report lower rates of anxiety and depression symptoms (Brand et al., 2009). For younger children with acute lymphoblastic leukemia, lax parenting practices are associated with significant youth sleep difficulties (McCarthy et al., 2016). In fact, family stressors and youth-perceived home environment can be among the most critical contributors to adolescents' sleep quality (Tynjala et al., 1999). For example, significant associations between family environment, as defined by chaos and conflict, and children's sleep problems exist (Boles et al., 2016). Family processes are also often a target of behavioral interventions for pediatric sleep problems because pediatric sleep interventions require high parent and family commitment (Moturi & Avis, 2010). Examining global as well as daily associations between family factors and youth sleep in pediatric cancers may provide additional insight for the refinement of targeted sleep interventions.

Ecological Momentary Assessment

Ecological momentary assessment (EMA) or daily diaries address the above-mentioned limitations associated with retrospective reports by providing patients with the opportunity to report on their day-to-day fluctuations in sleep, family functioning, and mood in their "real" environment (Hacker & Ferrans, 2007). Thus, EMA reduces biases associated with retrospective self-reports. Further, EMA offers a non-experimental way to more closely infer causal associations by providing a temporal sequencing of events in the naturalistic environment (Bolger, 2003). EMA data are analyzed using rigorous longitudinal statistical methods, and thus, allow for accurate estimates of within-person and between-person effects and variability over

time (Bolger, 2003). Thus, EMA methods are particularly well suited to capture variations in daily youth and family occurrences as well as sleep habits; in this way, EMA captures day-to-day observations and enhances ecological validity (Lippold et al., 2014).

Technological advances have allowed advances in real-time data collection, as they offer distinct advantages over paper-and-pencil methods (Heron et al., 2017). For instance, mobile EMA methods are time-stamped to track compliance, provide the opportunity for customized alarm schedules, and allow participants to use their own devices (Heron et al., 2017). Because more than seventy percent of adolescents have access to a smartphone in the United States, using the participants' own phones has been suggested as a mechanism to decrease participant burden of carrying another device (Brenner, 2015; Heron et al., 2017). Mobile electronic momentary assessment has been identified as a promising tool for collecting youth real-time data in comparison to paper diaries (Palermo et al., 2004). For instance, mobile EMA have been found to have higher compliance rates, greater satisfaction, increased data fidelity, and lower participation burden on participants than pencil-and-paper methods (Berkman et al., 2014; Palermo et al., 2004). Although mobile EMA methods offer a variety of advantages over traditional diary approaches, they have yet to be utilized or assessed for feasibility with pediatric cancer patients.

In research with adolescents with chronic health conditions using EMA (e.g., arthritis or diabetes), 71% to 83% of those approached agreed to participate (Borus et al., 2013; Connelly et al., 2012; Mulyaney et al., 2012). Adolescent participants who agreed to participate in these studies completed 66% to 85% of the daily diaries (Bromberg et al., 2016; Brannon et al., 2016; Palermo, Valenzuela, & Stork, 2004). Furthermore, 61% of participants reported being satisfied by providing electronic self-report ratings and 72% did not find EMA burdensome (Palermo et

13

al., 2004). Participants also reported a positive experience and minimal burden associated with EMA as assessed through open-ended questions administered after data collection (Borus et al., 2013; Schuster et al., 2015). Seventy percent of adolescents rated EMA data collection as not being burdensome at all (Palermo et al., 2004). Unfortunately, these studies lack a quantifiable rating of satisfaction/burden. Thus, EMA allows a more complete and valid depiction of individuals' daily experiences that are also well-tolerated and minimally burdensome to participants (Heron, Everhart, McHale, & Smyth, 2017; Heron & Smyth, 2010).

Sleep assessment via EMA. Assessments of sleep in pediatric cancer populations have thus far been restricted by reliance on parent and youth ratings in settings that may lack ecological validity (e.g., laboratory or hospital environments; Short et al., 2013). Cross-sectional designs have prevented researchers from evaluating potential cascading associations between sleep, children's daily experiences, and the environment (Kaleyias et al., 2012). However, it is clear that daily variations in sleep patterns during adolescence exist and that these daily changes are associated with health outcomes, such as differences in BMI, blood glucose levels, and pain ratings (Valrie et al., 2007; Turner et al., 2016). For instance, adolescents report delayed sleep times and longer sleep durations on weekends in comparison to weekdays (Moore & Meltzer, 2008). Similarly, although children's global sleep quality is rated as high with a retrospective measure, great variability exists in day-to-day ratings (Bromberg et al., 2016). For example, participants report difficulty initiating sleep on 29% of study days and having less than 4 hours of sleep on 7% of these days (Bromberg et al., 2016). Therefore, when adolescents are asked to describe their sleep habits using retrospective recall methods, it is difficult to capture daily variability in their sleep patterns (Fuligni & Hardway, 2006). This research suggests that

14

ecological momentary assessment (EMA) or daily diaries may be better suited to capture variability in sleep patterns.

Other measures of sleep include actigraphy, multiple sleep latency test (MSLT), and polysomnography. Actigraphy is particularly valuable for accurately evaluating circadian rhythm patterns, body movement, and sleep vs. wakefulness patterns (Martin & Hakim, 2011). For instance, actigraphy has been compared to polysomnography in pediatric research and agreement rates have been satisfactory (Martin & Hakim, 2011). Nevertheless, actigraphy has not correlated strongly with subjective measures of sleep quality. MSLT is a sleep laboratory test where electroencephalography (EEG) patterns measure rate of sleep onset and rapid eye movement as patients sleep and wake at different intervals throughout the testing day (Martin & Hakim, 2011). Although MSLT is an objective indicator of sleep difficulties, it does not always distinguish between daytime sleepiness that results from insufficient sleep and that from narcolepsy or hypersomnia; additionally, MSLT are time-intensive and require participants to stay in the clinic for a full day (Johns, 2000). Finally, polysomnography is another time-intensive overnight sleep study that records body movement, brain activity, eye movement, and other physiological parameters to diagnose sleep related disorders. Daily diaries are an alternative method to identify variations and correlates of youth sleep patterns without placing significant burden on participants (Jungquist et al., 2015). Daily diaries and actigraphy can be used interchangeably to assess sleep start and end times (Talbot et al., 2010).

Discrepancies in reports exist when assessing youth sleep habits. Parents often underestimate the extent of their children's sleep problems, and often, children stay up later and wake up more frequently than their parents say they do (Owens, Spirito, McQuinn, & Nobile, 2000). Similarly, parents report more "idealized" sleep patterns with significantly longer

adolescents' sleep duration estimates than actigraphy, adolescents' self-reported sleep diaries, and self-reported retrospective ratings; these findings suggest that adolescents may be having more restricted sleep than their parents believe (Short et al., 2013). For pediatric brain tumor survivors, parent and child reports of excessive daytime sleepiness result in poor concordance (Brimeyer et al., 2016). Adolescent self-report diary measures correspond to actigraphy measures more closely than adolescent retrospective ratings and parental proxy measures (Short et al., 2013). Because of this, it is particularly important to collect daily self-report when assessing adolescents and young adults' sleep concerns.

Family assessment via EMA. Family processes have also been captured using EMA in general child and adolescent populations (Lippold et al., 2014). EMA is particularly well-suited to capture family functioning in its natural environment and to capture family processes that occur on a daily basis by minimizing the amount of time between participants' responses (Bolger et al., 2003). For instance, EMA has identified significant associations between negative parent-youth daily interactions and youth Hypothalamic–pituitary–adrenal (HPA) axis functioning. In contrast, cross-sectional or retrospective designs have yielded mixed results regarding the presence of these associations (Lippold et al, 2014). EMA processes have also highlighted the effects of spillover, during which youth's experiences in one context (e.g., the family) influence their behavior in another context (e.g., academics). For example, more family stress predicted greater difficulties with school learning the following day, and these effects persisted for two days (Flook & Fuligni, 2008). Thus, EMA methods may allow researchers to more accurately infer the directionality of family-youth effects by providing a depiction of the temporal sequences of events (Flook & Fuligni, 2008).

Effects of daily family processes for youth with chronic illnesses have also been examined via EMA methods (Leibach & Everhart, 2017; Tobin et al., 2015). For instance, when youth with asthma were asked to complete daily diaries of their family climate, negative caregiver-child interactions predicted youth asthma symptoms (Tobin et al., 2015). Similarly, for youth with cystic fibrosis, daily phone diaries identified significant associations between the amount of daily parental supervision youth received and objective adherence to their medical regimen (Modi et al, 2008). Finally, studies that have assessed associations between stress and sleep for youth with sickle cell disease included arguments with parents and family members in the measurement of stress. In these studies, higher stress was associated with shorter sleep duration (Valrie et al., 2007). However, studies have yet to examine the correlates of and daily variations in family functioning for youth with pediatric cancer; EMA methods are particularly well suited to answer these research questions.

Statement of the Problem

Pediatric brain tumor (BT) survivors are 2.7 times more likely to report sleep problems in comparison to the general population, even after controlling for age and gender (Lipton et al., 2009; Nolan et al., 2013). Craniopharyngiomas are among the most common brain tumors in children, and while benign in nature, they profoundly impact the pituitary-hypothalamic regions of affected patients (Cohen, Guger, & Hamilton, 2011). As advances in medicine have significantly enhanced the survival rates of cancer patients, attention is turning to assuring quality of life (QoL) for survivors; sleep-wake pattern disturbances and excessive daytime sleepiness are among the most prominent sequelae (Manley et al., 2012; Muller et al., 2002). Psychosocial processes (e.g., executive functioning and school performance) are impaired by

17

sleep difficulties, highlighting the importance of examining sleep patterns as potential targets for treatment in pediatric craniopharyngioma patients (Dewald et al., 2010).

Similarly, as survivorship rates of pediatric craniopharyngioma increase, understanding the daily experiences of patients is important to guide the development of interventions. Family interventions and cognitive behavioral therapies for insomnia (CBT-I) have been tested separately in pediatric cancer populations. Each intervention domain has been separately effective. Nevertheless, family interventions (e.g., problem-solving skills training) have largely focused on parents and their experiences, whereas CBT-I sessions are primarily designed for patients and survivors (Sahler et al., 2005; Poggi et al., 2009). By examining the daily interrelationships between family functioning and child sleep difficulties in pediatric craniopharyngioma, the proposed study could help inform future integration of sleep and family therapies.

The first purpose of the present study is to examine associations among global family functioning and sleep difficulties for youth with pediatric craniopharyngioma, using retrospective parent and youth ratings. This study will also assess daily associations among children's sleep efficiency, family functioning, and overall mood by providing temporal sequencing of daily events via adolescents' self-reported EMA (see *Figure 1*). Finally, this study will investigate the feasibility of using mobile EMA to assess daily sleep, mood, and family functioning for youth with pediatric craniopharyngioma.
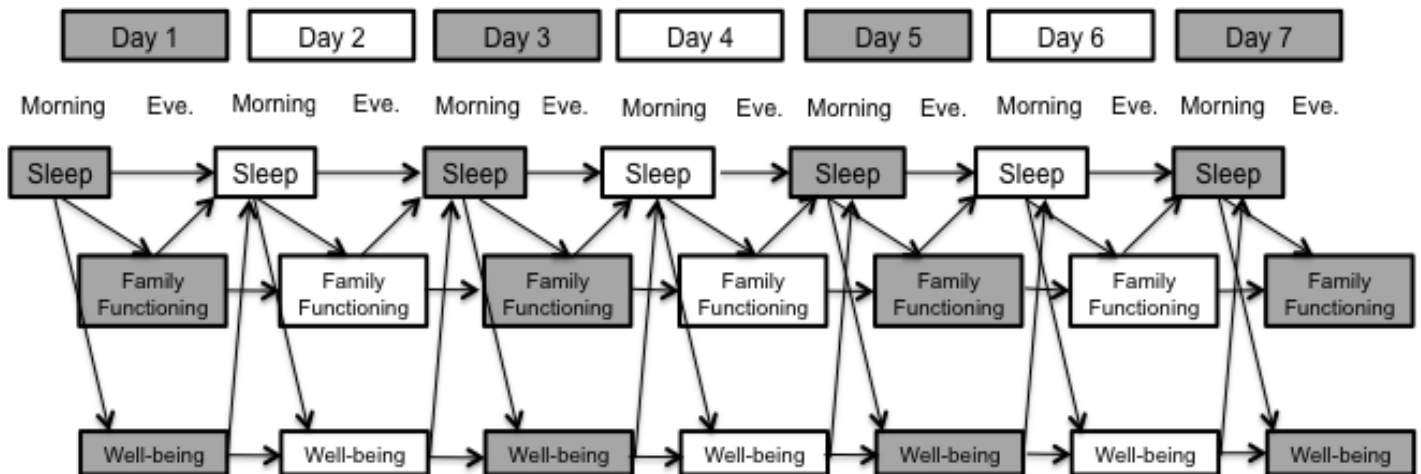
Figure 1. Conceptual model of associations among sleep, family functioning, and well-being (negative affect)

Aims and Hypotheses

    Aim 1. To examine whether children's global sleep habits and daytime sleepiness are associated with family functioning and routines using retrospective parent and youth reports.

    Hypothesis a: Based on previous research showing significant associations between children's sleep difficulties and family functioning (Boles et al., 2016; McCarthy et al., 2016), we hypothesize that children who have more sleep difficulties will have caregivers who also report poorer family functioning and less structured routines. We also hypothesize that there will be a significant positive association between children's self-reported sleep difficulties and their own ratings of overall family functioning.

    Aim 2. To examine the association between family functioning, sleep efficiency, and overall affect using EMA micro-longitudinal data to understand directionality of influence. In these analyses, negative affect is used as a measure of daily mood, as it has been shown to be an important contributor to overall well-being. The abbreviated Positive and Negative Affect Scale (PANAS) measure has also been previously tested and validated in an EMA context and with

youth (Ebesutani et al., 2012; Watson, Clark, & Tellegen, 1988). Thus, it will be used as a proxy of daily mood/well-being in this study.

Hypothesis a. Following days of poor family functioning and higher negative affect, children will report more sleep difficulties (i.e., poorer sleep efficiency). Similar to previous research on these associations, we hypothesize small but significant associations between family functioning and sleep and overall negative affect/well-being and sleep (Adam et al., 2007; Valrie et al., 2008).

Hypothesis b. Following nights with more sleep difficulties, as defined by poorer sleep efficiency, children will report poorer family functioning and higher overall negative affect. Without studies directly assessing associations between sleep efficiency and family conflict, this aim remains exploratory in nature. However, given findings from cross-sectional studies highlighting associations between family environment and sleep duration, we hypothesize that greater efficiency will be associated with better family functioning on subsequent days (Boles et al., 2016). Similarly, significant associations between sleep and daily mood have been documented in other pediatric populations (Valrie et al., 2008). Based on these findings, we hypothesize that nights with higher sleep efficiency will be followed by lower overall negative affect scores the next day.

Aim 3. To evaluate the feasibility and utility of using EMA surveys to measure fluctuations in family functioning and child sleep patterns over seven days, as stressors can vary widely from day-to-day. The feasibility of the study will be determined using the following criteria: compliance and completion rate of EMA diaries, overall satisfaction with EMA data collection methods, and overall self-reported burden by EMA data collection.

Hypothesis a. Consistent with past findings, we hypothesize that 70% of adolescents will complete at least 66% of EMA ratings across the one-week study duration (Bromberg et al., 2016).

Hypothesis b. In past studies of youth with a chronic illness, 61% of participants reported being satisfied by electronic daily diaries (Palermo et al., 2004). Based on this work, we hypothesize that at least 60% of participants will report being satisfied with the electronic surveys completed over the one-week EMA duration. We also hypothesize that 60% of participants will state that they are willing to participate in another study with electronic daily surveys.

Hypothesis c. Studies have only used open-ended questions to assess EMA burden on adolescent participants, and they have found an overwhelmingly positive experience and minimal burden with EMA data collection (Palermo et al., 2004). Unfortunately, these studies lack a quantifiable rating of satisfaction/burden. One study attempted to objectively measure burden of electronic diary ratings with an adult population and found that adults generally found three EMA surveys per day as causing somewhere between 'not at all' and 'slight' burden on a 4-point Likert scale ($M = .64$, $SD = .90$; Stone et al., 2003). With only limited literature in this area, we hypothesize that participants will, on average, rate burden in the lower 25th percentile of the rating scale (e.g., less than a 4.0 on a ten-point Likert scale), consistent with research on EMA being viewed favorably by adolescents.

Methods

Participants

Forty-one youth and their families, who were concurrently enrolled in a larger study, "A Phase II Trial of Limited Surgery and Proton Therapy for Craniopharyngioma and Observation

for Craniopharyngioma after Radical Resection" were recruited for this study (RT2CR; Merchant, PI; Crabtree, Co-I). Participants in the parent RT2CR study were youth with craniopharyngioma (ages 8-18 at diagnosis) and their families who were being treated at St. Jude Children's Hospital (Memphis, TN). Informed consent and assent were obtained for all participants and their parents for the parent study. Of the 41 participants who consented for the follow-up study, one withdrew from the study for being ill and one did not complete any baseline or EMA measures or respond to phone/email following the recruitment process. Thirty-nine participants and 35 caregivers completed baseline measures, and 39 participants participated in EMA portion of the study. Caregiver participation was lower than youth participation, given that youth attended the follow-up medical visit on their own and/or their caregivers did not respond to the REDCap surveys sent via email.

Research assistants contacted participants in the parent study who met inclusion criteria for participation in the current study. Specifically, participants were eligible for this follow-up study if they had a craniopharyngioma diagnosis, participated in the sleep study component of the RT2CR, had access to a smartphone or tablet, and agreed to download and use the Ilumivu mobile application. Exclusion criteria for this study included limited English language proficiency, as evidenced by an inability to read and respond to EMA survey test questions, visual or sensorimotor impairment, and cognitive or developmental diagnoses that would preclude understanding youth self-report questionnaires. Parental consent and participant assent (if age <18) or consent (if age ≥18) for the proposed study were obtained in-person. Since participants regularly visited St. Jude and had ongoing relationships with the research team of the parent project, minimal recruitment and participation burdens were expected.

Procedure

Institutional Review Board approval was obtained for this study at St. Jude Children's Research Hospital and at Virginia Commonwealth University. Prior to beginning data collection, and after consent and assent were obtained for the current study, staff helped the family download the Ilumivu mobile EMA (mEMA) application. Ilumivu is an established EMA tool that offers an in-mobile application for participants to respond to questionnaires in real time with limited burden. This application sent notifications to participants to remind them to complete surveys within chosen response times for each survey. Participants were trained in person in the appropriate methods to access and respond to mobile EMA surveys. Participants were also provided with the telephone number of research team members who were available at any time to answer questions or troubleshoot technical difficulties. In addition, a study team member contacted participants at least once via email and/or phone during the one-week period of data collection to check in regarding their understanding of the measures, to address potential barriers, and to ask about the general acceptability of the EMA modality. These efforts were proven to be effective in ensuring participants' adherence to the EMA protocol in past EMA studies (Bromberg et al., 2016).

Research participants and parents completed retrospective family functioning, mood, and sleep baseline measures via REDCap on the day of consent/assent (Day 0). Because most clinic visits occurred between 8 am and 5 pm, the mobile EMA assessment began on the day following enrollment (Day 1) and continued for the next week; Day 7 marked the last day of EMA data collection. On Day 8, the day following the final day of EMA assessment, participants completed a short survey assessing their overall satisfaction with electronic data collection. Consistent with

typical EMA procedures, participants were provided $50 for time spent completing baseline questionnaires and EMA surveys plus a bonus $15 for completing all EMA surveys.

Per the IRB-approved study protocol, we used Simon's (1989) two-stage design to evaluate feasibility of EMA data collection after the first ten participants completed all measures. Specifically, the percentage of total log completion was assessed to examine feasibility. If at least 70% of participants completed at least 66% of the administered EMA surveys, no changes were to be made to the protocol. If fewer than 47% of participants completed 66% or more surveys, it was decided that the study would undergo early termination. Even with one participant who was unenrolled due to significant health problems, 70% of participants completed more than 66% of the administered EMA surveys. Our average compliance rate for the first ten enrolled participants was 82%. All participants who completed the satisfaction survey prior to this checkpoint noted that they were satisfied with electronic data collection. Therefore, no changes were made to the protocol, and we continued recruiting participants.

To ensure that EMA data collection was not promoting elevated levels of anxiety and distress for participants, research assistants called families halfway through the weeklong data collection period (i.e., on Day 3). Participants were asked to rate, using a 10-point Likert scale, how distressing EMA completion was for them. If participants responded with a rating of 8 or above, indicating significant levels of distress caused by completing ratings, psychologists from the research team further assessed levels of anxiety and distress. We expected minimal levels of distress to be caused by this electronic data collection. No participants reported such elevated levels of distress or needed further intervention.

Measures

Retrospective Reports. Participants and their primary caregivers completed retrospective ratings of family functioning, mood, and sleep measures at baseline prior to beginning the seven-day EMA protocol. Baseline measures took approximately one hour for the youth and primary caregiver to complete.

Family functioning. The Family Assessment Device (FAD; Epstein, Baldwin, & Bishop, 1983) is a self-report scale based on the McMaster Model of Family Functioning (MMFF). It assesses the six global family domains of problem solving, communication, roles, affective responsiveness, affective involvement, and behavior control. The FAD provides 60 statements, for which caregivers and adolescents are asked to indicate how well each statement described their own family on a 4-point Likert scale. For this study, the General Functioning scale was used. It is an index of overall satisfaction with family functioning that consists of the mean response on twelve items of the FAD (Mansfield, Keitner, & Dealy, 2015); higher scores are indicative of poor family functioning. The FAD General Functioning scale has demonstrated high internal consistency (Cronbach's α = .83 to .86) and good construct validity (Hamilton & Carr, 2016; Kabacoff et al., 1990). The FAD has also been used to assess family functioning for youth with pediatric cancer with acceptable internal consistency (Cronbach's α = .78; Barakat et al., 2010). The FAD GF scale evidenced high internal consistency in this study for both parent and youth reports (Cronbach's α = .88 and .86 respectively).

Family routines. The Family Time and Routines Index (FTRI; McCubbin, McCubbin, & Thompson, 1996) is a 30-item scale that examines the activities and routines that families adopt and maintain together. Parents or caregivers rated how accurately each statement described their family activities on a 4-point Likert scale. The FTRI was initially based upon the Family Routines Inventory, but authors modified and expanded upon question content to include other

family life cycle changes (e.g., adolescence). The FTRI yields information regarding family routines on eight subscales: Child Routines, Couple's Togetherness, Meals Together, Parent-Child Togetherness, Family Togetherness, Relative's Connection, Family Chores, and Family Management. If certain routines do not apply for the family, the primary caregiver noted that this routine is "Not Applicable." The mean was calculated based on the endorsed items to yield a total Family Routines Score, with a higher score indicating the presence of greater family routines. The FTRI has evidenced high internal consistency (Cronbach's α = .88) in previous studies (McCubbin et al., 1996). Internal consistency for the FTRI was similarly high in this study (Cronbach's α = .88).

Daytime sleepiness. The Epworth Sleepiness Scale for Children and Adolescents (ESS-CHAD) is a measure used to assess daytime sleepiness in youth across a variety of situations (Janssen, Phillipson, O'Connor, & Johns, 2017; Wang et al., 2017). Both parents and youth rated the adolescent's tendency to fall asleep during eight scenarios on a scale of 0 to 3, with higher scores indicating greater chance of falling asleep. A total sum score was obtained, with higher scores demonstrating greater difficulty with EDS. The ESS-CHAD has demonstrated high internal reliability (Cronbach's α = .73) and good model fit as measuring a unidimensional construct (Janssen et al., 2017). Both self and parent proxy reports of the ESS-CHAD had high internal reliability in this study (Cronbach's α = .79 and .87 respectively).

Children's Report of Sleep Patterns (CRSP). The CRSP is a 60-item multidimensional self-report measure of sleep patterns, sleep hygiene, and sleep disturbances for youth and adolescents (Meltzer et al., 2013). Youth rated items describing their previous night's sleep and a typical night's sleep on a five-point scale, with 1 = never and 5 = always. The CRSP results in multiple scale scores, including those that assess insomnia, daytime sleepiness, electronics use,

caffeine usage, bedtime worries and sleep location. Higher scores indicate more difficulties. For this particular study, the insomnia scale was used to assess sleep difficulties. The insomnia scale has evidenced high internal reliability (Cronbach's α = .76 and .73) in previous studies and has also distinguished clearly between youth with clinically significant sleep difficulties and otherwise healthy youth (Meltzer et al., 2013; Meltzer et al., 2014). In this study, the CRSP had high internal reliability (Cronbach's α = .81).

Ecological Momentary Assessment surveys. Participants also completed EMA items twice daily over a one-week period. Questions were based on prior research with general child populations (Ebesutani et al., 2012; El-Sheikh et al., 2012). The morning survey took less than five minutes to complete, and the evening administration took less than ten minutes to complete.

Morning survey. The following items were assessed via the morning survey: bedtime, number of minutes it took to fall asleep (sleep onset latency), number and duration of nocturnal awakenings/fragmented sleep segments, overall youth-perceived quality of sleep (1 = very bad to 10 = very good), degree of "restedness" from the previous night's sleep (1 = exhausted to 10 = very refreshed), wake time after sleep onset, and time out of bed. For the purpose of this study, sleep efficiency was calculated as the percentage of the total time a participant was in bed that the participant was actually asleep. Sleep efficiency captures core problems of nighttime awakenings, short sleep duration, spending too much time in bed, and difficulty falling asleep (Reed & Sacco, 2016).

Evening survey. The evening survey consisted of items that assess mood, family functioning, medication use, and total screen time. The ten-item Abbreviated Positive and Negative Affect Schedule (PANAS) measured the degree of positive states and negative states the respondent felt throughout the day on a five-point Likert scale, with one indicating very

27

slightly or not at all and 5 indicating extremely (Ebesutani et al., 2012). For this study, the mean score of the five negative affect scale items was used to assess daily mood. The evening survey also consisted of eight items derived from the Child Home Data Questionnaire (CHDQ) that assessed youth perceptions of parent-child interactions throughout the day (Margolin, 1990; Robles et al., 2013). The mean score across these items was derived, with higher scores indicating poorer family functioning. Finally, participants were asked to report medications used, whether or not they napped, and total screen time each day.

Satisfaction survey. Participants completed a survey after their week-long EMA experience to assess their satisfaction with electronic daily diary completion (Borus et al., 2013). Specifically, participants were asked whether or not they were satisfied with data collection. They were also asked to rate the overall burden associated with completing online daily diaries on a ten-point Likert scale, with 1 being not burdensome at all to 10 being very burdensome.

Analyses

Power

Simulation studies using MLM to identify the sample size needed to detect effects revealed that a sample size of more than 30 families (resulting in more than 400 EMA data points) would provide sufficient power (Maas & Hox, 2005). Past EMA researchers have recruited anywhere between 6 and 303 youth in similar protocols (Dunton et al., 2015; Heron et al., 2017; Rofey et al., 2010). Similarly, EMA study durations have ranged from four days to one month, although there are no studies that we are aware of that have used mobile EMA with youth diagnosed with pediatric cancer. In the proposed study, assuming at least 60% of 36 participants would report being satisfied by the EMA data collection, the margin of error with 90% confidence interval was 13%. For the one sample t-test in burden, our initial proposed sample

28

size of 36 could detect minimal effect size 0.39 with two-side setting. Even using conservative retention and enrollment rates from past pediatric cancer studies, we were confident in our ability to recruit 36 participants and their families. To ensure sufficient power to examine patterns of sleep and family functioning and to assess interrelationships among constructs of interest, forty-one eligible participants from the parent study who were ages 8-18 at diagnosis were approached for recruitment.

Data cleaning and descriptive analyses

Initial data cleaning assessed for the presence of normality, multivariate outliers, and linearity. The Kolmogorov-Smirnov (K-S) test was used to determine whether the normality assumption was met. Multivariate outliers were detected using standardized residuals. Linearity and normality of residuals were assessed via a residual scatterplot. Robust maximum likelihood estimation methods provided accurate parameter estimates' in the presence of non-normality.

Descriptive statistics, using SPSS version 24.0, were used to examine characteristics of participants who completed all EMA forms in comparison to those who did not. Additionally, EMA days of daily log completion was assessed to examine feasibility. Similarly, descriptive statistics were used to examine the percentage of adolescents that noted overall satisfaction with the EMA data collection and willingness to participate in a future EMA study. Finally, a mean score of overall burden of participation was derived from participants' responses on the satisfaction survey. Descriptive statistics were also used to characterize the nature of sleep disturbances within this sample; this was done by comparing sleep problems of youth within this sample to statistics obtained from CRSP and ESS-CHAD validation studies. The prevalence of naps throughout the week was assessed as well.

We relied on a combination of empirical and theoretical approaches to systematically identify covariates for this study's models. Relevant socio-demographic (participant's race, participant's ethnicity, participant's age and sex, and number of caregivers in the household), disease-related (number of years since diagnosis and sleep-related medication use), and sleep hygiene (i.e., screen time, reported naps) variables that significantly and uniquely predicted outcome variables for each model were identified first. Given power considerations, we then relied on theory to determine which of these variables to include as potential covariates.

Multivariate statistics

Ordinary least squares (OLS) regression was used to examine the association between sleep difficulties (assessed via CRSP and ESS-CHAD) and family functioning/routines (assessed via FAD and FTRI). Sleep difficulties was used as a predictor in the first model with family routines being an outcome. Similarly, sleep difficulties' association with family functioning was assessed.

Multilevel modeling

Multilevel Modeling (MLM, also called linear mixed effects modeling or hierarchical linear modeling) was employed to examine individual variability in sleep, affect, and family functioning over the one-week period of data collection. MLM is flexible and well suited to analyze repeated measures as it accounts for dependency between observations and incorporates robust standard errors, thus providing a more accurate estimate of associations between variables (Bryk & Raudenbush, 1992). Repeated EMA measures were nested within participants. MLM analyses were conducted using SAS Version 9.4.

MLM allows for within and between person variability. Using the Intraclass Correlation Coefficient (ICC) provided a percentage of between-group variability in observed variance.

Multilevel lagged models were used to examine temporal patterns of effect between 1) family functioning and sleep efficiency and 2) affect (as defined by PANAS-C negative affect) and sleep efficiency. Time was included in these models as a fixed effect. First, the main effect of family functioning was entered in a model at time (t) to predict sleep difficulties at the subsequent timepoint (t+1). Next, a reverse path, testing sleep difficulties at time (t) predicting family functioning at the subsequent timepoint (t+1) was examined. Last, the same model structures were tested to examine interrelationships between sleep and affect/well-being, as measured by negative affect.

Results

A CONSORT flow diagram (see Figure 2) is included to summarize eligibility, recruitment, and consent processes. In summary, out of 63 potential participants, 43 children and families who were eligible to participate in this study were approached. Two families declined to participate, and 41 families consented to take part in this study. Following consent, one participant withdrew from the study for being critically ill, and one did not complete any baseline or EMA measures and did not respond to phone/email following the recruitment process (see Figure 2).

Thirty-nine youth and thirty-five of their primary caregivers who were enrolled in this study completed baseline measures of sleep, family functioning, and general demographics. Youth participating in this study were eight to 18.5 years old at the time of their craniopharyngioma diagnoses ($M = 12.15$, $SD = 2.88$) and 11 to 24 years old ($M = 15.86$, $SD = 3.05$) at the time of the current study. Time since diagnosis varied from two to twelve years, with youth having an average of 4.50 years since diagnosis at baseline ($SD = 1.94$). According to the youth-reported Epworth Sleepiness Scale (ESS), 32 percent of the sample reported clinically

elevated levels of excessive daytime sleepiness (EDS). Parents reported greater rates of EDS for their children, with a 48.5% prevalence of clinically significant EDS. Clinically significant levels of excessive daytime sleepiness, per parent reported ESS, for this sample at time of diagnosis and three months later were 40% and 28.5% respectively. Descriptive statistics for the study's baseline variables are presented in Table 1, and additional participant and family characteristics are presented in Table 2.
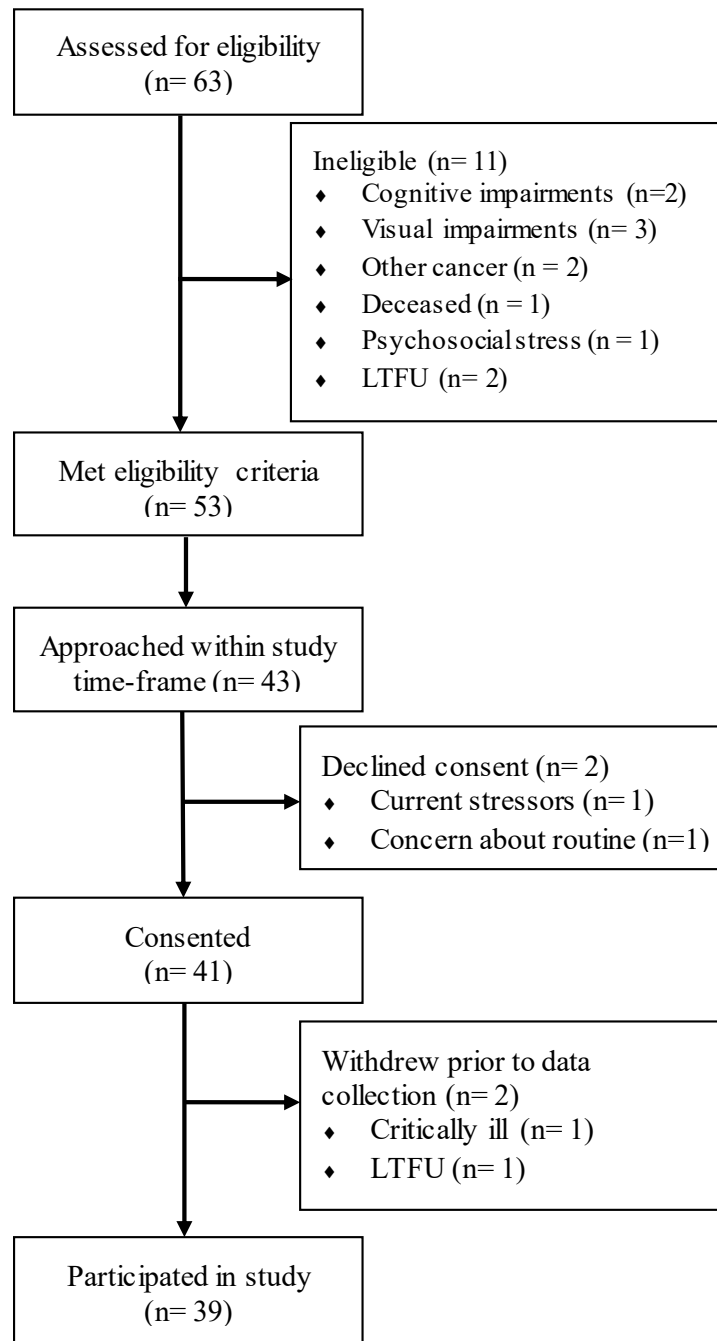
Figure 2. CONSORT Flow Diagram of Participant Eligibility and Recruitment

Table 1

Sample Descriptive Information (N = 39)

Variable	Mean (SD)
Child age (years)	15.86 (3.05)
Caregiver age (years)[a]	44.73 (6.54)
Time since diagnosis (years)[a]	4.50 (1.94)
Family general functioning (FAD PR)[a,b]	2.24 (.34)
Family general functioning (FAD YR)[b]	1.82 (.49)
Usual bedtime on weekdays[c]	21:16 (3:44)
Usual bedtime on weekends[c]	23:13 (1:42)
Usual waketime on weekdays[c]	06:53 (0:54)
Usual waketime on weekends[c]	08:49 (1:41)
	N (%)
Child sex (% male)[a]	21 (60.0)
Child race[a]	
African American/Black	9 (23.1)
Caucasian/White	28 (71.8)
Other	2 (5.1)
Child ethnicity[a]	
Hispanic or Latino	3 (8.6)
Not Hispanic or Latino	30 (85.7)
Other / Unknown	2 (5.7)
Child naps [c,d]	
Never naps	8 (21.6)
Never naps unless sick	5 (13.5)
Sometimes naps	17 (45.9)
Naps almost everyday	7 (18.9)

Note. [a] *N* = 35 due to missing caregiver-reported data. [b] Responses on the Family Assessment Device. PR indicates parent report and YR indicates youth report, and higher scores are indicative of poor family functioning. [c] Responses on the Children's Report of Sleep Patterns. [d] *N* = 37 due to missing youth-reported data.

Table 2

Additional Family and Participant Characteristics

Variable	N (%)
Participating caregiver[a]	
Biological mother	25 (71.4)
Biological father	7 (20.0)
Biological grandparent	1 (2.9)
Adoptive Parent	2 (5.8)
Number of caregivers in household[a]	
One caregiver	5 (14.3)
Two caregivers	29 (82.9)
Three caregivers	1 (2.9)
Number of people in household[a]	
2-3 people	9 (25.7)
4-6 people	22 (62.9)
7-8 people	4 (11.4)
Child age	
11-15 years old	19 (50.0)
16-19 years old	17 (44.7)
>19 years old	2 (5.26)
Child schooling[a]	
Middle School (5th-8th grade)	14 (40.0)
High School (9th to 12th grade)	14 (40.0)
College	5 (14.3)
Other	2 (5.8)
Child school status[b]	
Attending school	13 (36.1)
Summer/vacation	17 (47.22)
Homebound instruction	2 (5.6)
Not in school	3 (8.3)
Child ill (other than craniopharyngioma)[a]	
Yes	2 (5.71)
No	33 (94.3)
Child currently taking medication[a]	
Yes	1 (2.86)
No	34 (97.14)

Note. [a] $N = 35$ due to missing caregiver-reported data. [b] $N = 36$ due to missing youth-reported

data.

Table 3 presents bivariate correlations among participants' descriptive information, family functioning, and sleep characteristics. Correlations between baseline descriptive characteristics (e.g., youths' age, sex, and medication use) and sleep and family functioning variables were not statistically significant. Youth with longer time since diagnosis reported greater difficulties with excessive daytime sleepiness. As expected, there was a statistically significant concordance between parent-reported and youth-reported family functioning on the FAD. Poorer parent-reported family functioning was also associated with fewer parent-reported family routines. Youth retrospective reports of excessive daytime sleepiness were significantly associated with youth reports of family functioning. Similarly, parent-reports of family functioning were associated with youth-reported insomnia symptoms. Results did not suggest significant associations between youth reports of excessive daytime sleepiness and insomnia symptoms and parent-reported family routines.

Table 3

Correlations among Baseline Family, Sleep, and Demographic Variables

	1.	2.	3.	4.	5.	6.	7.	8.	9.	10.
1. Age	--									
2. Biological sex[a]	.12	--								
3. Time since diagnosis	.07	.07	--							
4. Medication use	.30	-.16	.12	--						
5. Poor Family functioning (YR)[b]	.09	.09	.10	.17	--					
6. Poor Family functioning (PR)[b]	.29	.06	.01	.01	.61**	--				
7. Daytime sleepiness (YR)[c]	.16	.26	.35*	.10	.35*	.15	--			
8. Daytime sleepiness (PR)[c]	.21	.23	.03	.15	.30	.07	.55	--		
9. Insomnia (YR)[d]	.05	-.10	-.15	-.18	.26	.50**	.04	.11	--	
10. Family Routines (PR)[e]	-.02	-.15	.07	-.07	-.22	-.36*	-.03	.07	-.26	--

Note. [a] Represents comparison of male to female biological sex. [b]Family Assessment Device scale scores: higher scores indicate worse family functioning. [c] Epworth Sleepiness Scale for Children and Adolescents (ESS-CHAD): higher scores indicate more difficulties with excessive daytime sleepiness. [d] Children's Report of Sleep Problems (CRSP): higher scores indicate more insomnia symptoms. . [e] Family Time and Routines Index (FTRI): higher scores indicate the presence of more structured routines, per caregiver report. YR indicates youth-report and PR indicates parent report.

Sleep characteristics for this sample were compared to those of other youth to identify similarities and differences in sleep experiences and identify potentially unique challenges faced by youth with craniopharyngioma. Analyses identified similar sleep characteristics in this sample to adolescents who participated in the Children's Report of Sleep Patterns (CRSP) validation study (see Table 4). Descriptive information from the CRSP validation study with adolescents captured youth-reported sleep characteristics from youth who presented to pediatric sleep clinics and from the community. Youth were asked to categorize themselves as great/good sleepers vs. OK/poor sleepers. ANOVA comparisons across the three groups (i.e., great/good sleepers, OK/poor sleepers, and youth with craniopharyngioma) yielded significant group differences in sleep location, bedtime electronic use, bedtime fears/worries, restless leg symptoms, insomnia symptoms, and parasomnias. Of important note, this study's sample had significantly lower bedtime electronic use than self-identified OK/poor sleepers from the validation study ($t(219) = 2.67$, $p = .0081$). Similarly, youth who participated in the current study had fewer self-reported insomnia symptoms than youth who identified themselves as OK/poor sleepers in the validation study ($t(219) = 5.59$, $p < .0001$). However, youth from this sample reported significantly greater bedtime fears/worries than both good/great sleepers ($t(382) = 6.45$, $p < .0001$) and OK/poor sleepers ($t(219) = 2.93$, $p = .0037$). Furthermore, youth reported significantly greater difficulties with restless leg symptoms than both groups from the validation study. It is important to note that the CRSP validation sample is diverse and consisted of youth who presented to pediatric sleep clinics, were pediatric oncology patients, attended two Australian schools, or participated in an Internet-based study for youth with and without asthma (Meltzer et al., 2014).

Table 4

Comparison of Sleep Characteristics in this Population to Those in CRSP Validation Study

Variable	Great/Good Sleepers (n = 345)	OK/Poor sleepers (n = 195)	Current sample (n = 39)
Caffeine	7.00 (2.34)	7.45 (2.79)	6.65 (2.62)
Activity Before Bed	17.70 (3.08)	17.74 (2.88)	17.40 (3.39)
Sleep Location[a]	2.80 (0.53)	3.15 (0.65)	2.87 (0.58)***
Bedtime Electronic Use	5.77 (2.68)	6.68 (2.82)	5.39 (2.28)***
Bedtime fears/Worries	3.18 (1.49)	3.95 (1.72)	4.86 (1.94)***
Restless Leg Symptoms	4.21 (1.35)	4.11 (1.27)	5.14 (2.21)***
Insomnia	8.28 (2.95)	11.88 (2.88)	8.87 (3.76)***
Parasomnias	3.90 (1.55)	3.83 (1.43)	3.08 (1.38)**

Note. All variables represent scale scores from the Children's Report of Sleep Patterns questionnaire.
ANOVA comparisons across three groups yielded the following significant differences.
[a]Square-root transformation used in accordance with Meltzer et al. (2015) article
* $p < 0.05$, ** $p < 0.01$, ***$p < .001$

Independent sample t-tests were also utilized to compare ratings of excessive daytime

sleepiness in this sample of youth with craniopharyngioma to youth who participated in the ESS-

CHAD validation study to identify differences in youth with craniopharyngioma in comparison

to same-age peers (Janssen et al., 2017). Results suggested significantly higher ratings of

daytime sleepiness in our sample in comparison to the sample of youth who participated in the

validation study (t(44) = -2.79, p < .01; 95 % CI = (-3.51, -0.57). The mean of self-reported ESS-

CHAD scores in the validation study (Janssen et al., 2017) was 5.36 (SD = 3.66). This was

compared to a mean overall score of 7.39 (SD = 4.30) reported by youth with craniopharyngioma

participating in this study. Of important note, parental proxy report provided higher ratings of

excessive daytime sleepiness (M = 8.43, SD =5.01) in comparison to their children's self-reports.

Results also suggest similar rates of parent and youth-reported EDS to those reported by families

of youth with ADHD diagnoses (Langberg et al., 2017). For instance, in a study examining

predictors of sleep difficulties for adolescents with ADHD, prevalence of clinically significant

EDS, per the Pediatric Daytime Sleepiness Scale, were 37.2 % and 42.1% for parent and

adolescent reports respectively (Langberg et al., 2017). In this study, rates of EDS were 48.5 %

percent and 32 % for parent and adolescent reports respectively. Results suggest that youth with

craniopharyngioma have more difficulties with EDS than same-age youth and that at least one-

third of patients report, by parent or proxy ratings, clinically significant levels of EDS.

Diary Completion

Youth completion diary entries ranged from two to 14 entries per participant during the

week-long EMA data collection period. Participants completed an average of 11.92 diaries (SD =

3.02). Nineteen participants completed at least one diary on each study day, and 14 participants

completed all 14 EMA diaries. On average, youth who completed daily EMA measures were

significantly older than youth who did not complete at least one diary per day (t(33) = 2.68, p = .012; Table 5). Independent samples t-tests and Chi-Square tests did not suggest any other significant differences on study variables between youth who completed all daily EMA diaries and those who did not. Similarly, bivariate correlations suggested an association between the number of EMA diaries completed and participants' age (r = .32), but results did not suggest the presence of significant associations between number of diaries completed and insomnia symptoms, family functioning, daytime sleepiness, or family routines.

Table 5

Comparison of Baseline Characteristics between EMA Completers and Non-Completers

Variable	EMA Complete Responders	EMA Incomplete Responders
Child Age	16.17 (2.68)	13.88 (2.34)*
Time since diagnosis	4.21 (1.25)	4.53 (2.67)
Family functioning (FAD-PR)	1.65 (.38)	1.66 (.38)
Family Routines-PR	2.22 (.29)	2.26 (.40)
Excessive Daytime Sleepiness- PR	8.39 (4.42)	8.47 (5.71)
Insomnia Symptoms (YR)	9.28 (3.61)	8.50 (3.94)
Family functioning (YR)	1.75 (.45)	1.89 (.53)
Excessive daytime sleepiness (YR)	7.06 (3.54)	7.70 (4.96)
	N (%)	N (%)
Ethnicity (Non-Hispanic/Latino)	16 (88)	14 (82.4)
Race (White/Caucasian)	15 (83)	13 (76)
Participant sex (Female)	10 (55.55)	6 (37.5)
Taking daily medication	18 (100)	16 (94.2)
Child currently ill/sick	1 (5.56)	1 (5.88)

Note. * $p < 0.05$, ** $p < 0.01$, ***$p < .001$; PR indicates parent report and YR indicates youth report.

Diary Descriptive Information

On average, youth indicated that it took them approximately 22.23 minutes ($SD = 29.55$) to fall asleep after going to bed. Across all EMA entries, youth reported a mean sleep quality rating of 7.23 out of 10 ($SD = 2.56$). Participants reported experiencing poor sleep (rating of four or less) on 16.37% of days during the study. Youth reported spending a mean of 47.77 minutes ($SD = 9.02$) on electronics prior to going to sleep. Youth reported taking at least one prescribed medication 81.7% of the time. Youth took medication to help them fall asleep on 20% of the total study days. Medications to help with sleep difficulties included: melatonin, trazodone, hydroxyzine, Benadryl, clonazepam, and Tylenol PM.

On average, youth napped on 31.7% of days during the study. They also confirmed the presence of a stressful situation on 18.8% of these days. Youth reported levels of excessive daytime sleepiness by noting they felt pretty sleepy or very sleepy on 19.3% of days during the study. Daytime naps ranged from several minutes to eight hours long, with youth napping an average of 1.52 hours ($SD = 1.34$) per day. Additionally, participants' electronic usage during the day ranged from zero to ten hours, with a mean of three hours ($SD = 9.02$ minutes). On average, youth rated the quality of their day as 7.68 ($SD = 2.33$) on a ten-point Likert scale, with higher scores indicating a better day. Similarly, youth rated their overall family conflict as 2.11 out of 10 ($SD = 2.11$; higher scores are indicative of greater conflict) and their overall family functioning as 7.72 ($SD = .17$; higher scores are indicative of better family functioning) on a ten-point Likert scale. See Table 6 for additional daily diary descriptive information.

Table 6

Descriptive Information from Daily Diaries

Variable	Mean	SD	Coefficient of Variation
Sleep efficiency	.94	.0052	.0056
Family functioning (CHDQ)[a]	1.29	.021	.016
Family functioning (self-rated)	7.72	.17	.021
Negative affect[b]	1.38	.043	.031
Positive affect[b]	3.33	.067	.020
Positive to negative affect ratio[c]	2.86	.087	.031
Total screen time (minutes)	180.70	9.02	.050

Note. [a] Mean score on the Child Home Data Questionnaire; higher scores indicative of poorer family functioning. [b] Mean scale score on the abbreviated Positive and Negative Affect Scale (PANAS). [c] Score derived by dividing the positive affect scale by the negative affect scale on the PANAS.

Baseline Associations

Consistent with our first aim, Ordinary Least Squares (OLS) regression tested associations between retrospective reports of daytime sleepiness and parent-reported family functioning at baseline. The results of the regression indicated that daytime sleepiness explained 2.4% of the variance in parent-reported family functioning ($R^2=.024$, $F(1,32)=.78$, $p = .38$). Results indicated that there did not appear to be a statistically significant association between youth-reported daytime sleepiness and parental reports of family functioning ($\beta = .15$, $p = .38$). For the model predicting parent-reported family routines, daytime sleepiness explained .1 % of the variance ($R^2=.001$, $F(1,32)=.018$, $p = .89$). Similarly, results suggested that youth-reported daytime sleepiness did not significantly predict changes in parent-reported family routines ($\beta = -.02$, $p = .89$).

Regression analyses also tested associations between youth-reported daytime sleepiness and youth-reported family functioning at baseline. The results of the regression indicated that daytime sleepiness explained 12.4 % of the variance in youth-reported family functioning ($R^2=.13$, $F(1,36)=5.09$, $p = .03$). Our results suggested the presence of a significant association between daytime sleepiness and youth-reported family functioning, whereby greater daytime sleepiness was associated with poorer family functioning ($\beta = .35$, $p = .03$). Similarly, higher insomnia symptoms explained 6.5 % of the variance ($R^2=.065$, $F(1,36)=2.52$, $p = .12$) in youth-reported family functioning. Trends suggested that greater difficulties with sleep, as indicated by the CRSP insomnia scale, were associated with poor family functioning ($\beta = .26$, $p = .12$). Nevertheless, directionality of effect cannot be inferred due to the cross-sectional nature of these associations.

Multilevel Modeling

A series of unconditional multilevel models were tested via SAS 9.4 to assess between and within-level variances for the following youth-reported variables: daily sleep efficiency, family functioning, and negative affect (Kwok et al., 2008). Table 7 summarizes the between and within level variances for each model as well as the intraclass correlation coefficient (ICC). The ICC characterizes how much of the total variance in each dependent variable is attributed to individual variability for each youth and how much is explained by nesting timepoints within individuals (Kwok et al., 2008). Similarly, when ICCs are greater than zero and lower than one, they suggest that multilevel modeling are more appropriate for analyzing the data accurately.

Table 7

Unconditional Models Predicting Youth Sleep Efficiency, Family Functioning, and Negative Affect

Model 1: Sleep Efficiency				
	Variance	SE	Z Value	*p*
Between Individuals	.0025	.00072	3.39	.0004
Within Individuals	.0030	.00033	9.10	<.0001
ICC	.45			

Model 2: Family Functioning (CHDQ)				
	Variance	SE	Z Value	*p*
Between Individuals	.046	.013	3.48	.0003
Within Individuals	.051	.005	9.52	<.0001
ICC	.47			

Model 3: Negative Affect (PANAS)				
	Variance	SE	Z Value	*p*
Between Individuals	.19	.054	3.50	.0002
Within Individuals	.22	.023	9.55	<.0001
ICC	.46			

To assess whether associations support Hypothesis A of our second aim, autoregressive (AR) multilevel models examined daily associations between 1) evening reports of family functioning and the following night's sleep efficiency and 2) evening reports of affect and the following night's sleep efficiency. Level 2 covariates in both models included time since diagnosis and age, and total daily screen time and sleep medication use were tested as Level 1 covariates. However, none of these covariates significantly predicted changes in sleep efficiency and were excluded from the final models predicting sleep. The autocorrelation parameter in both models indicated a significant association between participants' sleep efficiency on subsequent days ($r = .29$, $p = .008$). The significance of the autocorrelation parameter also indicates that MLM is suitable for predicting changes in daily sleep efficiency. MLM results suggested a marginally significant association between evening family functioning and the same night's sleep efficiency, whereby poorer family functioning related to lower sleep efficiency (Estimate = -.03, $p = .089$). However, models indicated that daily negative affect did not significantly relate to poor sleep efficiency on following nights (Estimate = -.012, $p = .19$). Table 8 summarizes random and fixed effect estimates for this model.

Table 8

Multilevel Random Effects Analyses Predicting Sleep Efficiency

Model 1: Family functioning predicting youth sleep efficiency

Random Effects	Variance (SE)	Z value
Between Individuals	.0017 (.00065)	2.57**
Autocorrelation Parameter	.29 (.11)	2.66**
Within Individuals	.0029 (<.00043)	6.59***
Fixed Effects	Estimate (SE)	t value
Intercept	.99 (.03)	28.45***
Family functioning	-.030 (.017)	-1.71†
Day of completion	-.0025 (.0023)	-1.07

Model 2: Negative affect predicting youth sleep efficiency

Fixed Effects	Estimate (SE)	t value
Intercept	.97 (.017)	55.40***
Negative affect	-.012 (.0090)	-1.33
Day of completion	-.0025 (.0023)	-1.08

Note. † p < .10 †* p < 0.05, ** p < 0.01, ***p < .001

Consistent with Hypothesis B of our second aim, a multilevel model also tested associations between nightly sleep efficiency and the following day's youth-reported negative affect. Level 1 (i.e., daily sleep medication use and screen time) and 2 (i.e., time since diagnosis and age) covariates were also tested in this model. Use of daily sleep medication uniquely predicted changes in daily negative affect, whereby use of sleep medication each night was associated with greater negative affect on subsequent days (Table 9; Estimate $= .37$, $p = .029$). Therefore, use of daily sleep medication was included in the final model predicting differences in negative affect. The autocorrelation parameter also was significant (r $= .42$, $p = .0002$), suggesting the presence of a positive association between negative affect on subsequent days. In the final model, results did not suggest that sleep efficiency significantly predicted differences in the following day's youth-perceived negative affect (Estimate $= -.92$, $p = .16$).

Table 9

Multilevel Random Effects Analyses with Sleep Efficiency Predicting Negative Affect

Random Effects	Variance (SE)	Z value
Between Individuals	.10 (.052)	1.97*
Autocorrelation Parameter	.42 (.12)	3.68**
Within Individuals	.23 (.043)	5.27***
Fixed Effects	Estimate (SE)	t value
Intercept	1.95 (.65)	3.02*
Sleep efficiency	-.92 (.65)	-1.43
Use of daily sleep medication	.37 (.16)	2.24*
Day of completion	-.05 (.021)	-2.21*

Note. † p < .10 †* p < 0.05, ** p < 0.01, ***p < .001; daily sleep medication use was included as a control variable in this model due to its significance in predicting daily negative affect.

The final multilevel model, summarized in Table 10, tested Hypothesis B of our second aim and assessed whether nightly sleep efficiency predicted differences in the subsequent day's youth reports of family functioning. As for covariates predicting differences in day-to-day family functioning, no Level 1 and Level 2 predictors resulted in significant associations with the outcome variable. Therefore, they were excluded from the final random effects model. This model yielded a significant yet negative autocorrelation parameter. To prevent inaccurate estimates of random and fixed effects with a negative covariance structure, the autocorrelation parameter was restricted to have a lower bound of zero. In the final model, there was no evidence to suggest that each night's sleep efficiency predicted significant changes in youth-perceived family functioning on following days (Estimate = -.46, p = .14).

Table 10

Multilevel Random Effects Analyses with Sleep Efficiency Predicting Family Functioning

Random Effects	Variance (SE)	Z value
Between Individuals	.047 (.014)	3.39**
Within Individuals	.041 (.0048)	8.47***
Fixed Effects	Estimate (SE)	t value
Intercept	1.77 (.31)	5.89***
Sleep efficiency	-.46 (.31)	-1.47
Day of completion	-.018 (.0079)	-2.21*

Note. † p < .10 †* p < 0.05, ** p < 0.01, ***p < .001; The MLM model for this analysis yielded a negative Autocorrelation (AR) Parameter, which implies a negative residual correlation structure. Therefore, we refit the model to place a lower bound of zero on the AR parameter.

Satisfaction and Acceptability of EMA

In support of our hypotheses for Aim 3, 79.49% ($n = 31$) of participants completed at least 66% (9.24 out of 14) of the EMA surveys. Confirming our second hypothesis of Aim 3, 100% of participants who completed the satisfaction survey ($n = 36$) indicated that they were satisfied with EMA data completion over the one-week EMA duration. Even when including all 39 participants and assuming that the three participants who did not complete the satisfaction survey purposely did not provide positive responses, 92.3% of participants shared that they were satisfied with EMA data completion. Additionally, 91.2 % of individuals who completed the survey indicated that they would be willing to participate in another EMA study. Participants rated their overall satisfaction an average of 8.88 ($SD = 1.67$), and consistent with our hypothesis, rated burden in the lower 25th percentile ($M = 2.69$; $SD = 2.18$) on a ten-point Likert scale.

Participants were also asked to comment on any aspects of EMA that may not have been captured by quantitative rating scales. For these responses, youth voiced both positive and negative experiences with EMA data completion. For instance, youth commented that the application was "actually very good and [helped] keep track of how [their] sleep was going" and that they "got to see how unhealthy it was for [them] the time that [they] went to sleep." Negative experiences with EMA primarily revolved around technical difficulties, and these technical difficulties were associated with either interrupted internet connectivity or the EMA application freezing. For instance, one participant reported that he "had one day that it completely froze" and another noted that it "would not" alert [her] with an evening notification. Results highlight the importance of practicing EMA survey completion and troubleshooting with adolescents in clinic prior to beginning the data collection period, since two adolescents

indicated that they "remembered [screen freezing] happened to [research staff] and just did what [research staff] told [them] to do."

Discussion

Even with five year survival rates greater than 90 percent, pediatric craniopharyngioma patients experience significant sleep difficulties and hypothalamic disruptions (Muller et al., 2006). Prevalent sleep problems in pediatric cancer populations include difficulty initiating or maintaining sleep, poor sleep quality, and short sleep duration (Walter et al., 2015). Sleep consequently affects other avenues of daytime functioning, including but not limited to quality of life, neural recovery, working memory, school performance, and executive functioning (Astill, Van der Heijden, Van Ijzendoorn, & Van Someren, 2012; Kopasz et al., 2010). Therefore, identifying factors associated with sleep difficulties in youth with craniopharyngioma is prudent to inform intervention development and family support initiatives. Similarly, with the significant improvement in pediatric cancer survival rates, recognizing and appropriately treating resulting sleep disruptions becomes crucial to optimize patients' quality of life following treatment. This study examined cross-sectional associations between sleep and family functioning for youth with pediatric craniopharyngioma diagnoses who have undergone partial resection and proton therapy, using both parent and child perspectives. To our knowledge, this is the first study to test daily associations between sleep, family functioning, and negative affect for youth with pediatric craniopharyngioma. Finally, feasibility and acceptability of EMA data collection were assessed via satisfaction surveys for youth following the seven days of daily diary administration.

Youth in this study reported sleeping an average of 8.96 hours (SD = 1.14) on weekdays and 9.54 hours (SD = 1.41) on weekends. Average sleep efficiency for this group was .94. The sleep duration falls within the recommended 8 to 10 hours for teenagers and 7 to 9 hours for

young adults (Hirshkowitz et al., 2015). Although sleep duration only represents one dimension of overall sleep, it is encouraging that youth with pediatric craniopharyngioma are on average able to sleep within what is considered a "normal range" for others their age (Hirshkowitz et al., 2015). In this study, about one-third of the sample self-reported clinically significant levels of excessive daytime sleepiness (EDS) compared to a prevalence of 48.5 % per parental proxy reports. Results mimic other pediatric populations (e.g., youth with anxiety, youth with cancer, and youth with IBD), in which parents either underestimate or overestimate the magnitude of sleep disruptions in comparison to self-report (Alfano, Patriquin, & De Los Reyes, 2015; Brimeyer et al., 2014; Pirinen et al., 2010). Nevertheless, parents and adolescents' ratings may be more concurrent when the chronic illness is more severe (Pirinen et al., 2010). Youth in this study may also be underestimating their sleep difficulties, since previous studies have identified that only one-third of adolescents who meet clinical criteria for a sleep problem actually endorse sleep problems themselves (Short et al., 2013).

Of interesting note, the prevalence of parent-reported clinically elevated symptoms of excessive daytime sleepiness increased from the three month post-diagnosis time period. This suggests that sleep difficulties, especially EDS, may persist or even worsen for brain tumor survivors for several years post diagnosis. Research has identified that altered patterns of daytime melatonin secretion with decreased night-time melatonin levels in pediatric craniopharyngioma may account for excessive daytime sleepiness and sleeping throughout the day (Cohen et al., 2011). Therefore, it is important to continue monitoring EDS symptoms even when youth have less frequent visits in survivorship clinics. Longitudinal studies are needed to further identify how various sleep disruptions develop over time following diagnosis so that

targeted screening and interventions could be developed and applied for brain tumor patients (Olson, 2014).

Although youth in this study reported less overall electronic screen time and nighttime screen time than youth from the CRSP validation study, they did report significantly greater bedtime fears and worries on the CRSP. This is consistent with research that indicates youth with craniopharyngioma and other brain tumors may have elevated levels of anxiety and worries in comparison to their same-age peers (Zyrianova, Alexander, & Faruqui, 2016). Similarly, youth with craniopharyngioma may have particular difficulties in modulating their emotions, and therefore, they may find it challenging to control their worries at bedtime (Cohen et al., 2011). Youth in this study also reported spending approximately three hours of screen time per day, which falls below the national average of screen time for this age range and significantly below youth from sleep, oncology, and community settings who participated in the CRSP validation study (Meltzer et al., 2014; Twenge & Campbell, 2018). Similarly, youth had significantly fewer ratings of insomnia symptoms compared to youth who classified themselves as "OK/bad" sleepers in the CRSP validation study. Because our study indicates that a significant proportion of youth with craniopharyngioma continue to experience clinically significant difficulties with excessive daytime sleepiness even five years after treatment completion, further research is warranted to identify what specific sleep factors influence EDS in this population in particular. Similar ratings of insomnia symptoms and sleep habits on the CRSP may also be attributable to the fact that a large proportion of the sample from the CRSP validation study was recruited from sleep and oncology clinics (Meltzer et al., 2014). Similarly, Greenfeld and colleagues (2011) highlight the likelihood that youth with CNS tumors may sleep more during the day, which may

explain how, because youth nap more, their experiences may not be fully captured by an overall insomnia symptoms score.

Correlation analyses did not yield statistically significant associations between youth demographic variables and EDS. However, there were significant associations between time since diagnosis and youth-reported excessive daytime sleepiness. In particular, a longer time since diagnosis was associated with greater EDS. Results suggest the importance of regularly assessing sleep difficulties in patients with youth craniopharyngiomas at routine medical visits even when transitioning off treatment and onto survivorship care, especially given the fact that EDS may result from disruption of melatonin production (Manley et al., 2012). Results also support previous studies that suggest that because of the suprasellar region location of craniopharyngiomas, youth are at a particularly elevated risk for long-term sleep difficulties (Mandrell et al., 2012). Finally, results imply that youth with craniopharyngioma may develop difficulties with EDS, even if they initially do not report significant sleep disruptions immediately following treatment completion. It is clear from this study that youth with craniopharyngioma experience greater difficulties with EDS than their same-age peers, even five years after diagnosis. The Epworth Sleepiness Scale is a brief survey that has strong test-retest reliability and may be administered at routine visits to monitor changes in EDS within this population to identify youth in need of further sleep assessment and intervention (Janssen et al., 2017).

Associations between Retrospective Ratings of Family Functioning and Sleep

Despite results suggesting the absence of significant associations between parent-reported family functioning or routines and youth difficulties with EDS, results suggested a significant association between youth-reported family functioning and EDS. In other words, youth who

reported poorer family functioning were also significantly more likely to endorse symptoms of excessive daytime sleepiness. Findings also suggested an association between poorer youth-reported family functioning and more insomnia symptoms. These cross-sectional associations support links between youth perspectives of family functioning and youth sleep patterns, as proposed by the Sleep Disturbances in Pediatric Cancer model (Daniel et al., 2016). Results also highlight the importance of assessing sleep difficulties in pediatric oncology from a family systems perspective (Daniel et al., 2016). Our findings reinforce the importance of assessing family functioning from children and adolescents' perspectives, because there are discrepancies in reporting even when using the same set of questionnaires. Finally, our results may inform the modification of current behavioral sleep interventions to acknowledge and incorporate family factors that may be associated with sleep difficulties to improve outcomes for patients and their families.

Associations between youths' own perspective of family functioning and sleep difficulties may be interpreted in one of two ways: youth who have more sleep difficulties may be more likely to report a more negative perspective of their family or youth who have poor family relationships may be more likely to develop sleep difficulties. Research has identified associations between family conflict and stress in children and adolescents (Flook & Fuligni, 2008). In particular, the family environment may influence sleep quality via anxiety, bedtime resistance, poor structure or routines, and/or stress (Boles et al., 2017). One plausible pathway for the reverse association may be explained.by greater sleep difficulties resulting in greater difficulties in adolescents' emotion regulation and thus poorer family interactions (Dahl & Lewin, 2002). Longitudinal studies assessing associations between adolescents' own perspectives of family functioning and youth sleep difficulties are needed to gain a clearer

picture of the directionality underlying these relationships. Similarly, studies should assess youths' perspectives of family routines and examine its associations with sleep outcomes.

Results also suggest differential associations between parent proxy reports and self-reports of family functioning and sleep difficulties. There is a line of research suggesting that discrepancies between parent and adolescent perspectives in family functioning may be indicative of adolescents' psychological adjustment and quality of life (De Los Reyes & Ohannessian, 2016; Human et al., 2016). Generally, studies to date have suggested that discrepancies between adolescent and parent perspectives of family functioning tend to widen with adolescents' age, as adolescents negotiate greater autonomy and family conflict increases (De Los Reyes & Ohannessian, 2016). Inconsistencies between parent and adolescent reports of the family environment have predicted cross-sectional and longitudinal ratings of poor adolescent adjustment (Human et al., 2016). Currently, family functioning is either not fully assessed in pediatric cancer contexts or assessed solely from caregivers' perspectives. Results highlight the importance of recognizing and soliciting adolescents' own perspectives of family functioning and routines, since they may be more closely related to health-related quality of life and sleep outcomes (e.g., EDS and insomnia symptoms) than caregiver reports. The fact that youths' own reports of family functioning, but not their parents', were related to sleep difficulties suggests that their own perspective of family functioning may affect bedtime worries or other sleep behaviors that in turn influence their sleep difficulties. Results also highlight the important information caregiver-youth discrepancies in ratings of family functioning and management patterns may provide for identifying difficulties in adjustment or sleep in pediatric oncology.

Daily Associations between Family Functioning, Sleep, and Affect

Daily diary data suggested a marginal association between family functioning and same night sleep efficiency, whereby poorer family functioning was associated with poorer sleep efficiency. The mechanisms by which family functioning affects sleep may include parent-set sleep bedtime, decreased parent-child conflict, and positive parenting styles (Brand et al., 2009; El-Sheikh et al., 2012; Short et al., 2011). It will be important for future studies to assess associations between such family factors and sleep outcomes for youth with pediatric craniopharyngioma in particular, given that findings have suggested significant associations between family functioning and youth adjustment in pediatric cancer (Van Schoors et al., 2016). Moreover, larger samples may be needed to detect statistically significant relationships given that family functioning is one of several factors expected to contribute to sleep health in youth with craniopharyngioma.

Our findings did not support our hypothesis that negative affect during the day would predict changes in the following night's sleep efficiency. Overall, youth reported lower rates of negative affect in this study compared to the general population, and therefore may not have elevated levels of negative emotion that disrupt sleep patterns. These null results may also be due to the large age range in our population, in that specific associations between affect and sleep may not be identified. Power limited us from dividing our sample based on developmental stages, but this may be an avenue for future research. Another explanation for our null findings may be attributed to our limited EMA timeframe. Increasing the EMA study duration may allow researchers to capture larger fluctuations in affect and sleep disruptions that may be due to stressors, such as traveling to and from medical appointments, receiving medical procedures, and shifting between the academic year and summer.

In models in which sleep efficiency predicted negative affect, findings suggested that sleep efficiency did not significantly predict ratings of negative affect on following days. Our findings may be due to the fact that youth primarily reported positive affect in this study and that there was limited variability in ratings of negative affect. What is unclear from this study is whether youth are reporting low levels of negative affect because they are coping well emotionally to being off treatment or whether they are underreporting negative emotions. Future studies would benefit from eliciting details from youth about their coping strategies as well as information from caregivers about youth behavioral and emotional functioning. Finally, our null findings may suggest that negative affect and sleep may not have immediate daily associations and that these associations occur over a longer period of time.

Finally, results suggest that sleep efficiency did not predict ratings of family functioning on following days. Results suggest that family functioning may be more predictive of later sleep efficiency than the reciprocal relationship. In other words, while poor family functioning during the day may undermine youths' sleep efficiency on following nights, poor sleep efficiency may not subsequently affect youths' perceptions of family functioning. Results are promising, given that interventions may target modifiable family factors to in turn improve sleep patterns. Future studies may lengthen the EMA period to capture greater variability in sleep efficiency and family functioning and to be able to capture these associations across transitions (e.g., school year vs. summer or medical visits vs. care at home).

EMA Feasibility and Acceptability

Study participants found electronic daily diary use acceptable and all youth who completed the evaluation survey were overall satisfied with EMA completion. Almost all participants indicated that they would be willing to participate in future studies with EMA data

collection. Some youth even indicated that reporting on sleep habits on a daily basis alerted them to some of their problematic sleep behaviors. Response rates in this study also concur with previous studies in supporting the feasibility and utility of using mobile technology for EMA to assess daily patterns and associations for children and adolescents (Heron et al., 2017). Because survivors of pediatric cancer often travel to attend medical visits and have less frequent contact with the medical team, mobile-based EMA studies may be particularly helpful in capturing real-life difficulties and strengths in order to identify youth who may be at risk for developing sleep or adjustment difficulties and to inform intervention delivery. Similarly, providing youth with an opportunity to complete EMA surveys remotely may increase participation rates from youth who may otherwise be unable or unwilling to participate in onsite longitudinal research studies.

Clinical Implications

Within the cancer context, sleep difficulties may be overlooked given the other medical needs that must be addressed. Nevertheless, results highlight the importance of assessing EDS at routine intervals in clinic visits, especially because of the elevated rates of EDS experienced among craniopharyngioma patients even as they enter the survivorship phase. It is also important to routinely monitor EDS difficulties because they can be indicative of other sleep problems. Similarly, past research has highlighted the importance of acknowledging the role of sleep concerns in maximizing patients' quality of life, providing helpful resources, and referring patients and their families to appropriate services (Daniel et al., 2016). Providers can also encourage patients and their families to monitor sleep habits on a daily basis in order to obtain more accurate data about experienced sleep difficulties. Self-monitoring of sleep habits and difficulties may also help in increasing patients' awareness about sleep habits. In fact, when youth were asked to provide additional comments about their experiences with EMA in this

study, some indicated that self-monitoring helped them identify problematic sleep habits (e.g., late bedtimes or excessive screen time). Behavioral sleep interventions are also brief, effective, and may be easily applied in pediatric cancer settings to improve adaptation following cancer diagnosis and treatment (Daniel et al., 2016; Mindell et al., 2006).

Our findings also support using a multifaceted approach to assess and treat sleep difficulties in pediatric oncology (Manley et al., 2012). Currently, patients may be prescribed stimulants (e.g., methylphenidate) to treat daytime sleepiness or fatigue, or melatonin to help with difficulties falling asleep (Rosen et al., 2008; Walter et al., 2015). As proposed by the model developed by experts in both oncology and sleep, the Sleep Disturbances in Pediatric Cancer (SDPC) model, there are several bio-ecological factors that influence sleep difficulties and in turn, health-related quality of life or other health outcomes. Therefore, a multidisciplinary approach is needed to assess and care for sleep problems experienced by youth with craniopharyngioma (Manley et al., 2012). By providing brief assessments and acknowledgement of bio-ecological factors (e.g., family conflict, stress, anxiety) in behavioral sleep interventions, one might increase the likelihood of sleep interventions being successful in pediatric oncology.

Finally, given the fact that adolescents reported high satisfaction with electronic data collection and often travel to attend routine visits in survivorship clinics, this study suggests that mHealth or eHealth interventions may be a promising avenue to pursue for adolescent and young adult patients with sleep difficulties. Because adolescents and young adults are increasingly relying on technology use, incorporating technology in care delivery for patients may help in improving engagement in medical care, increasing medication adherence, and improving health outcomes (Badawy et al., 2018). Similarly, research has yielded promising findings for a simple mHealth intervention working with parents of younger children with significant improvements

reported in sleep behaviors (Leichman et al., 2019). In this study, when parents used a free and publicly available phone application, they reported earlier bedtimes, fewer night awakenings, and increased total sleep duration in comparison to parents who did not receive this mHealth intervention (Leichman et al., 2019).This highlights the importance of researchers and clinicians working to modify electronic sleep interventions in order for them to be tailored to adolescents, young adults, and their families. eHealth interventions may also be tailored to target other behavioral factors that influence outcomes in pediatric cancer, including medication adherence, symptom monitoring, and coping strategies. However, clinicians must be careful to balance delivery of efficacious interventions electronically with maximizing feasibility and limiting burden on patients and their families.

Limitations

One limitation of this study is the reliance on youth-reported daily diaries for measuring all variables, including sleep difficulties, family functioning, and overall affect. Future micro-longitudinal studies should incorporate use of actigraphy measures in conjunction with EMA to assess sleep disruptions and experiences more accurately. Youth often underreport their difficulties with sleep, and therefore, capturing sleep difficulties with objective data would provide a better understanding of these associations (Short et al., 2013). Similarly, studies should incorporate behavioral observations of family functioning, which provide overall comparisons in observable family patterns that may translate to patterns and habits around sleep practices. Observations of family interactions, conflict, and functioning may increase ecological validity and decrease the susceptibility of findings to biases associated with retrospective reports (Waller et al., 2019).

Another limitation of this study is the limited EMA timeframe. While this is a pilot study, the EMA timeframe may not capture transitions to or from the school year, traveling to and from clinic visits, and potential medication changes that may disrupt sleep patterns. These events may be particularly impactful in children who are undergoing medical treatment that may influence sleep, family routines, and affect. Future studies should incorporate a larger timeframe to increase variability and capture associations across discrete timepoints to identify whether associations vary for specific events or transitions.

One of the characteristics associated with sleep that is proposed in the SDPC model that this study did not assess is cultural beliefs around sleep, illness, and family patterns. Although sleep problems are universal, there are cross-cultural differences that may account for variations in frequency and scope of children's sleep problems. Cultural beliefs around sleep (e.g., sleep habits, values, and parenting styles) often interact with children's developmental trajectories to define individuals' sleep practices, definitions of sleep problems, and effects of having a chronic illness on sleep difficulties (Owens, 2008). Experts have even argued that, by examining sleep correlates via a cross-cultural lens, one understands that sleep problems represent a complex interplay between physical, psychological, social, and cultural factors (Owens, 2008). To increase generalizability and increase the potential to identify and address important health disparities, a larger and more diverse population of youth with craniopharyngioma is needed. Similarly, examining the impact of cross-cultural variables like parental employment, competing time demands, socio-economic stressors, neighborhood environment, and parenting/discipline styles on sleep behaviors and difficulties will provide a more comprehensive view of a socio-ecological model of sleep determinants. Future research should address whether associations

between cultural beliefs around family sleep practices and sleep behaviors exist in pediatric cancer contexts and the magnitude of these associations.

Finally, technical difficulties limited our ability to collect consistent data from some participants despite strategies put in place to improve compliance and minimize data collection difficulties (e.g., ongoing compliance monitoring, calling participants mid-week, and practicing survey completion during recruitment). Nevertheless, inherent in all electronic studies are technical difficulties. In this study, participants voiced frustration when the application did not send notifications as it should or when the screen froze. According to a recent systematic review of studies utilizing mobile EMA for children and adolescents, most studies have not reported whether participants encountered any technical or logistical problems with EMA (Heron et al., 2017). Reporting these technical difficulties and their impact on mobile EMA studies is important as researchers and application software developers identify strategies to overcome these challenges in the future. Working to have multiple options for troubleshooting technical difficulties and continuing to partner with software companies to tailor EMA data collection for adolescents and young adults will ensure that future studies minimize the impact of technical difficulties on data collection. Similarly, providing greater opportunities to practice prior to beginning the EMA period (e.g., two or three test surveys at recruitment), may be helpful for youth to familiarize themselves with the EMA application and survey requirements.

Future Directions

Results highlight the importance of assessing family functioning and family management patterns within pediatric oncology, using both adolescent and caregiver perspectives. In the future, it is important to identify how family patterns, and in particular how the family organizes itself to manage the cancer diagnosis and treatment, influence youth sleep patterns. For instance,

Deatrick and colleagues (2018) identified that youth with family-focused management patterns, wherein families attempted to maintain homeostasis of family functioning in context of the cancer diagnosis, reported better overall quality of life in comparison to youth whose families focused more on managing the cancer condition itself. By identifying family management practices that influence difficulties for youth following diagnosis, and in particular following pediatric brain tumor diagnoses, one may identify potentially malleable mechanisms by which the family unit affects sleep and in particular nighttime worries or excessive daytime sleepiness (Deatrick et al., 2018). It will also be important for future studies to assess youth perspectives of family routines and to identify whether similar discrepancies in parent and youth reporting of routines and differential associations with sleep would exist.

Similarly, future EMA research studies should incorporate daily assessments of modifiable targets that may contribute to sleep disruption. For patients with craniopharyngioma in particular, it may be important to examine physical activity, sleep habits and routines, and eating habits. These factors may be influenced by the family environment and may in turn influence sleep characteristics. Patients and survivors with craniopharyngioma suffer from other morbidities secondary to hypothalamic dysfunction, including obesity (Manley et al., 2012). Given that elevated BMI can contribute to sleep difficulties, future studies should incorporate measures of daily eating and exercise habits to understand associations among BMI and EDS in youth with craniopharyngioma and identify behavioral targets for intervention to improve both health and sleep outcomes (Mandrell et al., 2012; Manley et al., 2012).

Future studies should also adopt mixed method approaches to further understand experiences associated with sleep disruptions and bedtime worries for survivors of pediatric brain tumors. Incorporating qualitative research to address these questions can provide additional

depth and richness to evaluating what factors influence sleep practices, why these factors affect sleep practices, and how these factors may disrupt sleep practices (Owens et al., 2006). Similarly, mixed method designs may provide additional information about barriers to treatment implementation and other potential targets for intervention (Owens et al., 2006). Similarly, developmental researchers have argued that using both quantitative and qualitative methods together can provide a better conceptualization of processes than either strategy can on its own (Yoshikawa et al., 2008). Because identifying factors that influence bedtime worries, sleep practices, and difficulties falling asleep are difficult to measure using either strategy alone, a mixed-method approach is indicated.

Finally, future directions for research include identifying factors associated with other sleep difficulties in pediatric craniopharyngioma. This study focused on sleep efficiency within a one-week timeframe, and it may be important to assess other specific variables of sleep that may be influenced by family or individual factors, including difficulties falling asleep, staying asleep, or waking up early. Similarly, it is important for research to examine associations between youth daily experiences and their subjective ratings of sleep quality. Whereas sleep efficiency may capture the ratio of time asleep to time spent in bed, subjective ratings of how rested youth feel and their sleep quality may be more prone to being affected by daily stressors and experiences. Because research has identified associations between worry and adolescents' perceptions of sleep quality and because youth in this study reported elevated rates of bedtime worries, it will be important to assess factors associated with their reported sleep quality (Lin et al., 2017). By examining factors associated with sleep quantity, sleep difficulties, and youth perceptions of sleep quality, research would provide a more comprehensive assessment of sleep in pediatric cancer.

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