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Short Communication

Acceptability and feasibility of a brief behavioral sleep intervention for youth with CF

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ABSTRACT

Youth with CF are at increased risk for physiological and behavioral sleep difficulties due to disease-specific symptoms and more general pediatric sleep challenges. Despite evidence suggesting that behavioral sleep interventions are effective for improving common sleep difficulties, no interventions exist for youth with CF. SLEEP-CF was designed to fill this gap by providing tailored, flexible behavioral sleep support to youth with CF. Results suggest that SLEEP-CF is an acceptable and feasible behavioral sleep intervention, even in a population with normative sleep habits. There may be benefit in terms of improving sleep knowledge and sleep hygiene. Technology use during and after bedtime is prevalent. CF care team members are encouraged to assess sleep as part of routine CF care, and to provide support as indicated.

1. Introduction

Sufficient, high-quality sleep is important for mental and physical well-being in children; however, many children and adolescents do not receive sufficient sleep [1,2]. Youth with Cystic Fibrosis (YwCF) are at increased risk for impaired sleep due to both disease-specific symptoms and more pervasive pediatric behavioral sleep challenges [3–6]. In YwCF, impaired sleep health may lead to negative effects on both physical and mental health [7–10]. Therefore, comprehensive assessment and treatment of sleep disorders, including strategies to improve behavioral sleep factors, is an important treatment target in YwCF.

Behavioral sleep interventions in adults are efficacious [11], and there is growing evidence for their impact in children and adolescents [12]. To date, there are no behavioral sleep interventions for YwCF. In a recent study regarding sleep needs and concerns in YwCF, participants expressed interest in learning and practicing evidence-based behavioral strategies to improve sleep, including cognitive behavioral techniques and sleep education [13]. Individualized plans and flexibility in implementation (including the use of telehealth) were noted to be important factors for YwCF. The primary aim of this study is to evaluate the acceptability and feasibility of an evidence-informed, behavioral intervention for YwCF, called SLEEP-CF.

2. Methods

SLEEP-CF consists of two "core" modules which provide: (1) basic sleep education (e.g., healthy sleep hygiene habits, recommended nightly sleep) and (2) tailored support based on specific participant needs (e.g., introduction of relaxation or cognitive-behavioral skills to assist with sleep onset). There are also four booster sessions, designed to be delivered remotely and focused on participant challenges or evolving needs. All intervention sessions were delivered by a Licensed Psychologist, Pediatric Pulmonologist/Sleep Medicine Physician, or Licensed Clinical Social Worker. The intervention is designed to be delivered over six months, with one module or booster session per month, although flexibility to accommodate participant preference is prioritized.

In the current study, measures of sleep health and hygiene were administered at three intervention timepoints to YwCF and their parents: baseline, midpoint (approximately 3 months after enrollment; varied based on intervention progress for individual participants), and post-intervention (approximately six months after enrollment; varied based on intervention progress for individual participants) . Measures of anxiety and depression, although outside

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the scope of this report, were also administered as part of the study battery. Data from baseline (time 1) and post-intervention (time 3) are reported.

2.1. Participants

Twelve YwCF completed the intervention between September 2019 and January 2021. Eleven parents provided proxy data to evaluate the intervention.

2.2. Measures

SLEEP-CF acceptability and feasibility. The primary intervention outcomes were acceptability and feasibility. These outcomes were assessed using the SLEEP-CF Evaluation Questionnaire, a 16-item questionnaire adapted from published tools to assess intervention acceptability, feasibility, and usability [14–17]. Four open-ended questions allowed for additional participant feedback.

Sleep health and hygiene. The Children's Report of Sleep Habits (CRSP) is a validated 62-item questionnaire that assesses sleep across three domains: Sleep Patterns, Sleep Hygiene, and Sleep Disturbance [18]. There is a 67-item parent-proxy report. The CRSP was administered at three study timepoints to YwCF and their parents. Each domain has a number of subscales; subscales addressed by SLEEP-CF are reported in this manuscript. As the intervention did not address parasomnias, restless leg syndrome, nighttime enuresis, or sleep disordered breathing, subscales measuring these areas were not computed.

The PROMIS Sleep Impairment and Sleep Disturbance scales were administered to parents at three study timepoints [19]. These are brief validated scales that are scored on a t-distribution. Technology use was also assessed via a series of investigator-derived questions about number of devices and patterns of use. The investigator-derived questions are available as Supplemental Online Materials.

Sleep knowledge. An 8-item questionnaire was developed to assess knowledge about sleep health at baseline and post-intervention. Participants received one point for each correct answer (maximum total score = 8). The investigator-derived questionnaire is available as Supplemental Online Materials.

2.3. Data analysis

Paired sample t-tests were computed for all validated instruments. Cohen's *d* was computed as a measure of clinical effectiveness, with commonly accepted interpretation guidelines (0.2 = small effect, 0.5 = medium effect, 0.8 = large effect) [20].

3. Results

The average intervention completion time for participants was 240 days, with an average of 104 days between Module 1 and Module 2. The majority of participants (n = 10; 83%) completed the intervention after the onset of the COVID-19 pandemic in March 2020.

3.1. Participants

YwCF were White (n = 12; 100%) and primarily female (67%). Average age was 14 years old (range = 10–17). Mean BMI percentile was 46.87% (range = 11.23% - 94.65%), mean FEV_1% predicted was 86.05 (range = 39–110), and 92% were taking CFTR modulators. All parent participants were White mothers. Intervention participation occurred during the COVID-19 pandemic for many participants; notably, many participants experienced transitions from in-person to virtual learning over the course of their participation. Retention was high (75%), with two participants dropping out after completing baseline measures but before completing any intervention sessions, and two additional participants dropping out over the course of the intervention.

3.2. Acceptability and feasibility

The majority of YwCF and their parents rated the SLEEP-CF intervention very favorably. See Figs. 1 and 2 for item-level responses. On open-ended questions, YwCF expressed that it was helpful to set goals related to increasing sleep, and also noted that the tailored help related to scheduling sleep and other activities was beneficial. One parent did not provide responses on the acceptability and feasibility questionnaire.

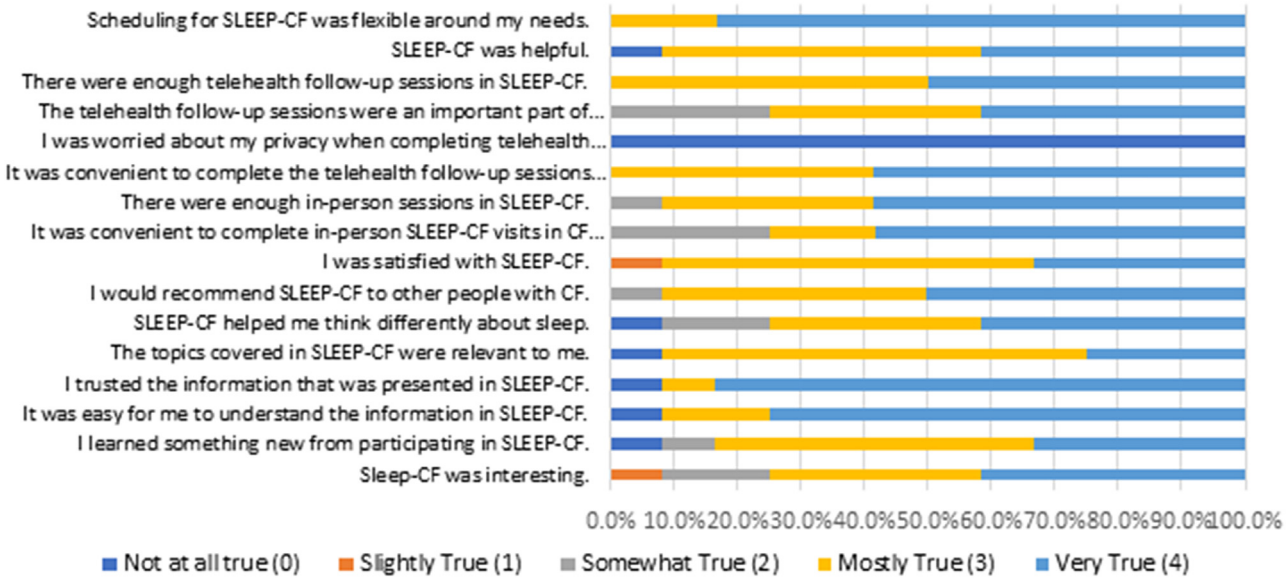

Fig. 1. SLEEP-CF Acceptability and Feasibility: Youth (n = 12).

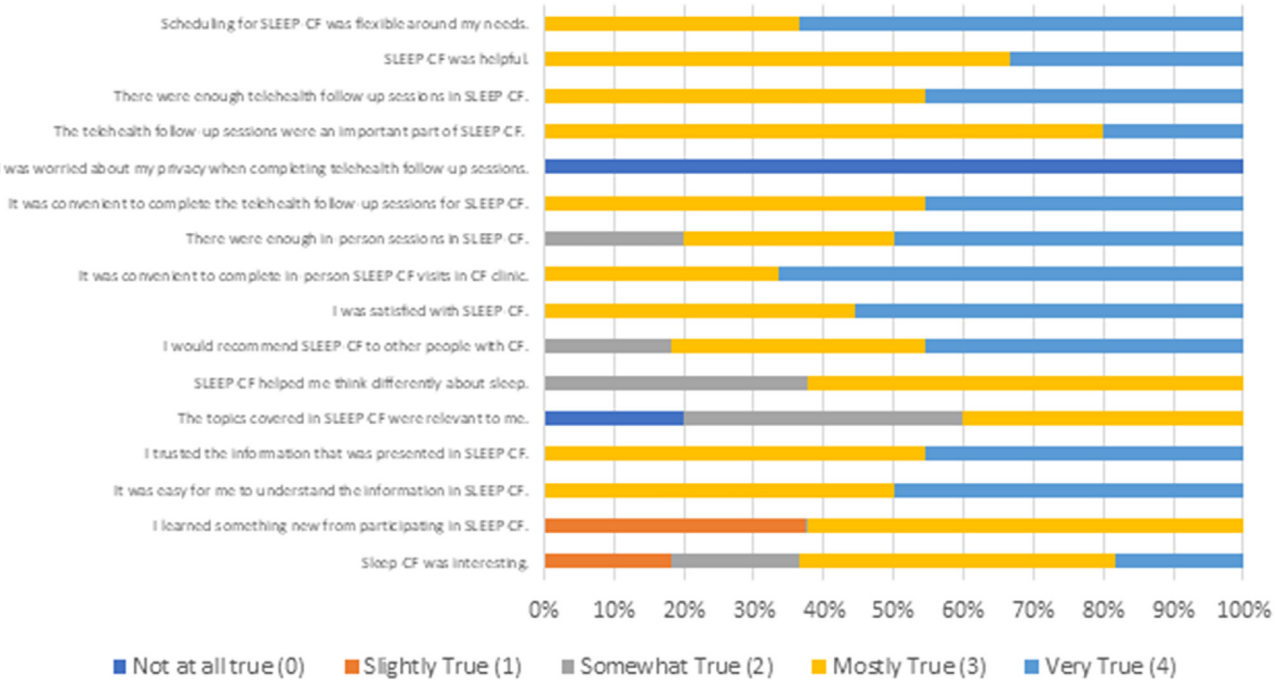

Fig. 2. SLEEP-CF Acceptability and Feasibility: Parents (n = 11).

Table 1
Mean score changes and effect sizes for validated exploratory sleep outcome measures.

Outcome	Baseline (T1) Mean (SD)	Post-Intervention (T3) Mean (SD)	Effect Size	p-value
Youth Report				
CRSP Caffeine Index	6.41 (2.35)	5.92 (1.98)	0.23	0.48
CRSP Activities Before Bed Index	17.92 (3.53)	19.67 (2.74)	0.55^{+}	0.06*
CRSP Sleep Location Index	8.92 (4.32)	7.58 (2.19)	0.39	0.23
CRSP Electronic CRSP Use at Sleep Onset Index	6.33 (2.64)	7.25 (3.52)	0.30^{+}	0.18
CRSP Bedtime Fears/Worries Index	5.75 (1.82)	4.67 (1.61)*	0.63	0.04*
CRSP Insomnia Symptoms Index	6.00 (1.81)	6.75 (3.19)	0.29^{+}	0.52
CRSP Sleepiness Index	9.75 (2.56)	8.83 (3.13)	0.32	0.14
Parent Report				
CRSP Caffeine Index	5.33 (1.67)	4.92 (1.83)	0.23	0.21
CRSP Activities Before Bed Index	17.5 (3.23)	17.42 (3.37)	0.02	0.95
CRSP Sleep CRSP Location Index	7.50 (3.45)	7.25 (1.71)	0.09	0.82
CRSP Electronic Use at Sleep Onset Index	5.75 (2.56)	6.50 (2.47)	0.30^{+}	0.17
CRSP Bedtime Fears/Worries Index	2.75 (0.97)	2.58 (1.00)	0.17	0.50
CRSP Insomnia Symptoms Index	9.58 (3.32)	9.83 (3.35)	0.07^{+}	0.86
CRSP Sleepiness Index	6.12 (1.34)	5.58 (1.24)	0.42	0.24
PROMIS Sleep Disturbance	54.93 (6.73)	51.40 (8.31)	0.47	0.12
PROMIS Sleep Impairment	54.85 (9.15)	47.55 (7.01)	0.90	0.01*

Note. CRSP stands for the Children's Report of Sleep Habits measure.
* = statistically significant value of p<.05. All p values reflect paired sample t-tests. Effect sizes reported as Cohen's d. $^{+}$= effect size suggests poorer function.

3.3. Sleep habits and hygiene

Refer to Table 1 for subscale data from the CRSP and PROMIS scales. The majority of indexes on the CRSP did not reflect a statistically significant change from baseline to post-intervention, with the exception of the YwCF report for the Activities Before Bed and Bedtime Fears/Worries indices. For YwCF, small-medium improvements were reported for caffeine, sleep location, and sleepiness. A larger effect was reported for bedtime fears/worries. For parents, a similar pattern was reported. Several subscales worsened or marginally improved from baseline to post-intervention: activities before bed, electronic use at sleep onset, and insomnia symptoms. The PROMIS Sleep Impairment scale showed statistically significant improvement.

3.4. Technology use

Technology use was highly prevalent. On an investigator-derived survey, over half of the study participants reported using an electronic device nightly within one hour of bedtime (85% at baseline, 67% at post-intervention). Similarly, many participants reported using their devices after turning their lights off at night (69% at baseline, 50% at post-intervention). These results are consistent with the electronic use at sleep onset subscale on the CRSP.

3.5. Sleep knowledge

Sleep knowledge improved marginally over the course of the intervention (M_{T1} = 5.3; M_{T3} = 5.6). The change in score for YwCF

from baseline to post-intervention ranged from −2, reflecting a decrease of two points, to 3 (reflecting an increase of 3 points).

4. Discussion

In this study, an evidence-informed, behavioral intervention for YwCF is acceptable and useable. Study retention and completion of all sessions was high, and the majority of participants provided favorable responses, especially in regard to the flexibility, convenience, and use of tele-medicine for implementation. These findings suggest that the number of modules is not overly burdensome for youth with chronic disease, and the use of telemedicine is well-accepted for this type of intervention.

Although not reaching statistical significance, there were small to medium changes in caffeine usage, bedtime location, and sleepiness with a larger change in bedtime fears/worries. In a larger sample size including children with higher levels of baseline sleep impairment, this effect may be greater. Moderate – large effects were observed on parent-proxy measures of sleep disturbance and impairment. Future studies are needed to understand this effect, particularly given the added complexity of the COVID-19 pandemic and its likely impact on sleep health and hygiene.

The effect size for certain indices (technology usage, activities before bed, and insomnia) worsened during the study, which was unexpected. This finding may similarly represent changes in sleep behaviors and mental health secondary to the Covid-19 pandemic with a shift to virtual schooling and decreased extracurricular and physical activities secondary to the lockdown.

Although the majority of participants did not report disordered sleep, overall study results suggest that the intervention was rated favorably by participants. This suggests the potential benefit of incorporating sleep education and sleep hygiene support into routine clinical care for YwCF. Future work should also explore the potential benefits of providing targeted behavioral sleep interventions to individuals with CF who report more clinically significant sleep impairment, including adults.

Declaration of Competing Interest

All correspondence regarding this article can be directed to Kimberly Canter, Ph.D., at Kimberly.canter@nemours.org. There are no conflicts of interest, financial or otherwise, to report.

CRediT authorship contribution statement

Kimberly S. Canter: Conceptualization, Methodology, Formal analysis, Investigation, Resources, Data curation, Writing – original draft, Writing – review & editing, Visualization, Supervision, Project administration, Funding acquisition. **Abigail Strang:** Methodology, Writing – original draft, Writing – review & editing, Supervision, Project administration. **Sophie Wilks:** Formal analysis, Investigation, Project administration. **Katherine Okonak:** Formal analysis, Investigation, Project administration, Writing – review & editing. **Aaron Chidekel:** Conceptualization, Methodology, Writing – review & editing, Supervision, Project administration, Funding acquisition.

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Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:10.1016/j.jcf.2022.07.012.

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