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Sleep Patterns in Children with Cystic Fibrosis

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Abstract

This study examined sleep patterns and the association between sleep and perceived health for children with and without CF. Ninety families (45 CF) completed questionnaires about the child's sleep and health. Significant group differences were found for sleep patterns (bedtime, wake time, total sleep time), symptoms of sleep disordered breathing, and sleep disturbances. Poorer perceived health was associated with sleep disturbances among children with CF, but not for children without CF. This study highlights the importance of including sleep in the evaluation of children with CF, as both medical and behavioral interventions can improve the sleep of children with CF.

Sleep disruptions in children with chronic illnesses are common (Lewandowski, Ward, & Palermo, 2011). There are a number of underlying etiologies for these sleep problems, including disease symptoms or characteristics such as coughing, itching, or pain, and/or disease management issues such as nighttime blood glucose monitoring. Further, when a child has been diagnosed with a chronic illness, there are a number of changes to daily routines and parenting behavior. In particular, parents of children with chronic illnesses tend to be more lenient with routines, which often results in increased co-sleeping (or bed sharing) (e.g., Chamlin et al., 2005; Williams et al., 2000).

Cystic fibrosis (CF) is one of the most common hereditary fatal diseases in European American populations, with an incidence of approximately 1 in 3500 live births in the United States. The treatment regimen is highly complex with inhaled medications and airway clearance, and can require children to spend several hours every day to manage their disease. As with other pediatric chronic illnesses, sleep for children with CF can be disrupted by both disease characteristics and disease management.

Several studies have examined objective sleep outcomes, including sleep architecture, sleep efficiency, and nocturnal oxygen saturation in children with CF. One study found an association between sleep disruptions and the severity of lung disease in children with CF (Naqvi, Sotelo, Murry, & Simakajornboon, 2007). Other studies have found nocturnal desaturations and a high prevalence of sleep disordered breathing (de Castro-Silva et al., 2010; Ramos et al., 2011). One study that used actigraphy, an ambulatory wrist-watch sized device that estimates sleep-wake patterns, found that sleep efficiency was associated with the severity of a child's lung disease in a sample of children and adolescents with mild to moderate CF (Amin, Bean, Burklow, & Jeffries, 2005). Together these objective findings suggest that sleep quality in children with CF is likely to be disrupted by their disease.

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Despite these clinical findings, reports of sleep patterns for children with CF, including bedtime and wake time, have been limited. The complex treatment regimen for children with CF may have to be completed in the early morning or late evening due to competing time demands (e.g., child's school schedule, parent's work schedule, or extracurricular activities for other children in the home). This may require the child with CF to wake early or go to bed later in order to complete their treatments, resulting in a shorter opportunity for sleep and consequently a shorter total sleep time overnight. In addition, the existing literature has been limited by a focus primarily on school-aged children and adolescents, with little attention on the sleep of preschool children. This is important because younger children require significantly more assistance with disease management than older children. This could potentially result in a later bedtime or earlier wake time due to parental assistance (e.g., CF treatments completed after other children in the home are asleep or before other children wake in the morning).

The primary aims of this exploratory study of preschool and school-aged children were: (1) to compare sleep patterns and sleep disturbances in children with CF and otherwise healthy children without CF, and (2) to examine the relationship between sleep and perceived health status for children with and without CF. We hypothesized that children with CF would have more sleep disturbances and obtain less sleep than children without CF, and that the child's health status would be associated with sleep problems and sleep disruptions for children with CF.

METHODS

Participants and Procedure

Children with CF were identified by a multidisciplinary medical team at a large tertiary care children's hospital. Children without CF were identified through a peer nomination process where parents of children with CF were asked to nominate a child the same age and sex as the child with CF (e.g., friend, neighbor, classmate). Peer nominated children were screened for chronic illness, developmental disorders, and diagnosed sleep disorders. A total of 104 families of children with CF or otherwise healthy children without CF (CF; n=54, without CF n=48) were invited to participate in a larger study examining the impact of a child's chronic illness on maternal sleep and daytime functioning (Meltzer & Mindell, 2006). Fifteen families (12 CF and 3 without CF) did not participate because of reported time constraints or stated lack of interest. This resulted in a final sample of 45 children with CF and 45 children without CF.

This study was approved by the Institutional Review Board at the children's hospital, and all participants provided informed consent. Parents completed a series of questionnaires via telephone interview. This methodological approach has been shown to be reliable and valid when compared to mailed questionnaires or in-person interviews (Aneshensel, Frerichs, Clark, & Yokopenic, 1982; Fournier & Kovess, 1993; McCormick, Workman-Daniels, Brooks-Gunn, & Peckham, 1993). Medical records were reviewed after study completion for children with CF 6 years and older to obtain pulmonary function test (PFT) results completed closest to the study interview (within 12 months).

Measures

Children's Sleep Habits Questionnaire (CSHQ)—The CSHQ is a 45-item parent report questionnaire focusing on children's sleep behaviors over a one-week period (Owens, Spirito, & McGuinn, 2000). For this study we examined the Total Sleep Disturbances score and the sub-scale scores of Bedtime Resistance, Sleep Onset Delay, Night Wakings, Sleep Disordered Breathing, and Daytime Sleepiness. In addition, parents provided information

about the child's typical bedtime and wake time. Total sleep time was calculated from bedtime to wake time. For all scales, a higher score indicates poorer sleep or more sleep problems. The CSHQ has shown adequate reliability and validity (Owens et al., 2000; Owens, Spirito, McGuinn, & Nobile, 2000).

Child Health Questionnaire (CHQ)—The CHQ is a measure of health-related quality of life (Landgraf, Abetz, & Ware, 1999). The measure is completed by parents and provides parental perception of a child's physical, emotional and social well-being. For this study, the *General Health Perception* and *Change in Health* scales were examined. For *General Health Perception*, lower scores indicate the parent believes the child has poor health that will likely get worse, and higher scores suggest the parent believes the child has excellent health that will continue. A high score for *Change in Health* suggests that a parent believes the child's health is much better now than one year ago. These scales have been shown to have excellent reliability and discriminate in populations of children with and without chronic illnesses (Raat, Botterweck, Landgraf, Hoogeveen, & Essink-Bot, 2005).

Data Analyses

One-way analysis of variance (ANOVA) was used to examine differences between children with and without CF for the CSHQ Total Sleep Disturbances Scale and CSHQ sub-scales. Pearson's correlation was used to examine the association between the CSHQ Total Sleep Disturbances Scale and both General Health Perception and Change in Health.

RESULTS

Sample Demographics

Children in this study ranged in age from 3 to 12 years inclusive (mean = 7.2 years, SD = 2.5). Participants were 60% male (n=54), 100% Caucasian, with 74% having their own room (n=67). The lack of racial diversity was due to the fact that CF is a disease that occurs primarily in Caucasians (Cystic Fibrosis Foundation, 2011). There were no significant demographic differences between the groups of children with and without CF. PFTs were available for children with CF 6 years and older (n=25), with a mean FEV_1 percent predicted of 107.2 (SD = 11.9), which suggests normal lung functioning in this population. Two children had PFT data that were invalid due to short exhalation time, and two children did not have PFT data available as they transferred to another medical center prior to the medical record review. Controlling for child age, no significant differences were found between children with and without PFT data for any sleep or health variable.

Group Differences in Sleep

Statisitically significant differences were found between children with and without CF for wake time, total sleep time, and the Sleep Disordered Breathing Scale, suggesting that children with CF wake on average 18 minutes earlier, sleep 30 minutes less, and have more symptoms of SDB (Table 1).

Because sleep patterns can differ between preschool children (3–5 years, n=28) and school-aged children (6–12 years, n=62), exploratory analyses were conducted to examine group differences within these two age groups. For preschool children, a significant group difference was found for total sleep time, suggesting that younger children with CF sleep on average 42 minutes less than younger children without CF. Although a statistically significant difference was not found for the Sleep Onset Delay, a medium effect size was found, suggesting that preschool children without CF have more stalling behaviors at bedtime. In addition, a clinically meaningful difference was found for bedtime, with

preschool children with CF going to bed 30 minutes later than preschool children without CF (Table 1).

For school-aged children, significant group differences were found for wake time and the Sleep Disordered Breathing Scale, suggesting that school-aged children with CF wake on average 23 minutes earlier and have more symptoms of sleep disordered breathing than school-aged children without CF (Table 1). Although not statistically different, the effect size also suggests that school-aged children with CF sleep on average 23 minutes less per day and have more parent reported sleep disturbances than school-aged children without CF.

Association between Health and Sleep Disturbances

For children with CF, a significant association was found between *General Health Perception* and the CSHQ Total Sleep Disturbances Scale ($r = -.37$, $p = .01$), suggesting that children with CF who were perceived to have poorer health also had more sleep disruptions. Similarly, a significant association was found between *Change in Health* and the CSHQ Total Sleep Disturbances Scale ($r = -.32$, $p = .03$), suggesting that children with CF who were perceived to have poorer health now compared to one year ago also had more sleep disturbances. For otherwise healthy children without CF, no association was found between the CSHQ Total Sleep Disturbances Scale and either *General Health Perception* ($r = -.17$, n.s.) or *Change in Health* ($r = .06$, n.s.),

DISCUSSION

This exploratory study examined the differences in sleep patterns among children with and without CF. The results of this study suggest that children with CF sleep less than children without CF, due to a later bedtime for preschool children and an earlier wake time for school-aged children. In addition, children with CF have more symptoms of sleep disordered breathing compared to children without CF. Finally, sleep disruptions were associated with parent perception of the child's current/future health and change in health over the past year.

The primary limitation of this study is the lack of an objective measure of sleep (i.e., actigraphy) and the use of parental report for both the child's sleep and health. The study is also limited by the sample characteristics, with a relatively healthy sample of children with CF, as well as a more sleep disrupted sample of children without CF (almost two-thirds of the children without CF scored above the clinical cutoff on the CSHQ of 41). However, the high rate of sleep disturbances (according to the CSHQ) in the general population has also been found in other studies (e.g., Holley, Hill, & Stevenson, 2010; Owens, et al., 2000).

Despite these limitations this study provides an important contribution to the literature by examining the sleep patterns for both preschool and school-aged children with and without CF. The later bedtime of preschool children with CF may be due to their disease management. Anecdotally, a number of mothers in this study reported that they would wait until the other children in the family were asleep, and then would perform chest physiotherapy (typically lasting 20–30 minutes) on the child with CF as a way to help that child relax prior to bedtime. This structured bedtime routine also provides an explanation for the lower bedtime resistance and sleep onset delay scores reported for children with CF.

The earlier rise time for school-aged children with CF is likely due to disease management requirements prior to school starting. Although not statistically different, the 23 minutes in total sleep time per day between school-aged children with and without CF is clinically meaningful and similar to a 24 minute difference in total sleep time found by actigraphy in another sample of children with CF (Amin et al., 2005). Even a modest amount of sleep restriction in school-aged children has been found to have an impact on multiple aspects of

functioning, including behavior and emotion regulation (Beebe, 2011; Sadeh, Gruber, & Raviv, 2003).

As for disease characteristics, the significant differences in symptoms of sleep disordered breathing and total sleep disturbances found in this study support previous findings of increased sleep disordered breathing, decrease sleep efficiency, and increased nocturnal wakings (de Castro-Silva, de Bruin, Cavalcante, Bittencourt, & de Bruin, 2009; Naqvi et al., 2007; Ramos et al., 2011). This study also showed that parental perception of poor child health (and changes to health) was associated with increased sleep disturbances. Thus it is important to consider that changes to sleep or an increase in sleep disturbances may suggest changes to the child's underlying health status. Alternatively, sleep disturbances may be reported when the parent feels the child's health is declining and may be an important point of intervention. However, longitudinal studies are needed to explore specific aspects of CF disease characteristics and disease management that may impact sleep quality.

Implications for Practice

The results of this study highlight the importance of recognizing that even in a relatively healthy population of children with CF, sleep disturbances exist, and optimal management of these children should include a sleep assessment as part of the standard (routine) evaluations of children with CF. Optimal medical management of disease symptoms (e.g., night time coughing and bowel movements) should result in a reduction in sleep disturbances, in particular night wakings, in turn improving sleep quality. Further, the importance of sufficient sleep and consistent sleep patterns should be emphasized and reviewed with families on a regular basis, especially since sleep patterns change over development. Although some aspects of disease management (e.g., morning treatments before school) may not be easily modifiable, it is important for families to be cognizant of the impact that treatment routines have on bed times, wake times, total sleep time and sleep behaviors throughout the child's development. Minor adjustments in the timing of morning or evening treatment routines may improve overall sleep quality and quantity. As fatigue is one of the indicators of a pulmonary exacerbation (Dakin, Henry, Field, & Morton, 2001; Flume et al., 2009), and is also a common symptom of poor sleep, the role that sleep disturbance plays in the cause or effect of declining health in CF should always be considered.

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Table 1

Means, Standard Deviations, and ANOVA Results for Sleep Patterns and Sleep Disturbances.

	Children with CF (n=45)	Healthy Children (n=45)	*F*	ES
Bedtime	20:55 (0:59)	20:47 (0:46)	.46	.01
Wake Time*	06:46 (0:43)	07:04 (0:37)	4.56	.05
Total Sleep Time (hours)	9.83 (0.9)	10.27 (0.8)	5.84	.06
Bedtime Resistance	7.96 (2.4)	8.04 (2.6)	.03	.00
Sleep Onset Delay	1.42 (0.8)	1.56 (0.7)	.73	.01
Night Wakings	4.07 (1.5)	3.69 (1.1)	1.93	.02
Sleep Disordered Breathing*	3.44 (0.8)	3.16 (0.4)	4.24	.05
Daytime Sleepiness	14.04 (3.8)	12.76 (3.4)	2.84	.03
Total Sleep Disturbances*	48.87 (7.0)	45.69 (9.4)	3.29	.04
Preschool Children (3–5 years)	(n=16)	(n=12)		
Bedtime	20:57 (1:25)	20:27 (0:37)	1.27	.05
Wake Time	06:58 (0:56)	07:08 (0:32)	.30	.01
Total Sleep Time (hours)*	10.0 (1.0)	10.7 (0.7)	4.06	.14
Bedtime Resistance	7.31 (1.5)	8.25 (3.7)	.84	.03
Sleep Onset Delay	1.31 (0.6)	1.75 (0.8)	2.92	.10
Night Wakings	4.69 (1.6)	4.17 (1.3)	.82	.03
Sleep Disordered Breathing	3.25 (0.6)	3.17 (0.4)	.19	.01
Daytime Sleepiness	13.0 (3.7)	11.67 (3.5)	.91	.03
Total Sleep Disruptions	47.31 (4.6)	43.50 (14.8)	.95	.04
School-Age Children (6–12 years)	(n=29)	(n=33)		
Bedtime	20:54 (0:40)	20:55 (0:48)	.004	.00
Wake Time*	06:39 (0:32)	07:02 (0:39)	6.41	.10
Total Sleep Time (hours)	9.73 (0.8)	10.12 (0.8)	3.31	.05
Bedtime Resistance	8.31 (2.8)	7.97 (2.2)	.29	.01
Sleep Onset Delay	1.48 (0.8)	1.48 (0.7)	.00	.00
Night Wakings	3.72 (1.3)	3.52 (0.9)	.55	.01
Sleep Disordered Breathing*	3.55 (0.9)	3.15 (0.4)	4.72	.07
Daytime Sleepiness	14.62 (3.8)	13.15 (3.4)	2.61	.04
Total Sleep Disruptions	49.72 (8.0)	46.48 (6.7)	3.02	.05

* $p \leq .05$

Bedtime and wake time expressed by 24-Hour clock, SD expressed in minutes.

Effect size (ES) is partial eta squared (.01 = small ES, .06 medium ES, .14 large ES)(Cohen, 1988)