

# Sleep Characteristics of Youth with Functional Abdominal Pain and a Healthy Comparison Group

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**Objective** To describe sleep problems among youth with and without functional abdominal pain (FAP). **Methods** Participants were 8–15 years of age diagnosed with FAP ( $n = 67$ ) or healthy pain-free comparisons ( $n = 80$ ). Parents and participants completed instruments assessing sleep, psychiatric status, and FAP symptoms. Parent and child reports of sleep problems were compared across groups and the association of FAP to behavioral sleep problems was assessed controlling for psychopathology. **Results** Children with FAP were reported to have more symptoms of behavioral sleep disorders (BSD), as well as increased nightmares and daytime tiredness than the comparison group. There were no group differences in total sleep time. Logistic regression analysis indicated that FAP was associated with a significantly increased risk of BSD symptoms [Odds ratio (OR): 4.17] after controlling for psychopathology. **Conclusion** Sleep problems in youth with FAP warrant clinical attention and future research should continue to explore sleep problems that co-occur with and independent of psychopathology.

**Key words** abdominal pain; adolescents; children; depression; sleep problems.

## Introduction

Sleep plays an important role in optimal physical and mental functioning. During the past decade there has been increasing awareness and interest in the role of adequate sleep and the impact of sleep disorders on health and behavior (Colten, Altevogt, & Institute of Medicine (U.S.) Committee on Sleep Medicine and Research, 2006). The roles of sleep-specific changes in neurohormones and endocrine factors have been implicated in the control of pain (Andersen et al., 2006; Moldofsky, 2001, 2002), metabolism (Spiegel et al., 2004; Spiegel, Leproult, & Van Cauter, 1999), and immune function (Kapsimalis, Richardson, Opp, & Kryger, 2005; Opp, 2006). Changes in cognition, attention, and affect regulation arising from sleep disorders or inadequate sleep, are less well understood, but may also play a significant role in individuals' ability to cope with pain associated with specific medical disorders. This study is a preliminary

descriptive investigation of the sleep habits and prevalence of sleep disorder symptoms in children and adolescents diagnosed with functional abdominal pain (FAP).

FAP is a common disorder with symptoms that often persist into adulthood (Walker, Guite, Duke, Barnard, & Greene, 1998), and is associated with considerable functional impairment and increased utilization of healthcare services (Campo, Comer, Jansen-McWilliams, Gardner, & Kelleher, 2002; Zeltzer, Bush, Chen, & Rivala, 1997a, 1997b). It is estimated that the prevalence of FAP is between 9 and 25% in children and adolescents, with prevalence increasing with age into adolescence, particularly in females (Scharff, 1997). FAP is defined as at least three episodes of abdominal pain having no identified medical cause that occurs within a three-month period and is associated with use of healthcare services and/or disruptions in usual activities (e.g., school attendance) (Apley, 1967). Pain in

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patients with FAP is most commonly episodic, located in the periumbilical region, and described as sore, colicky, and/or sharp (Abu-Arafeh & Russell, 1995).

### ***Sleep Disturbance in FAP***

There are ~88 sleep disorders defined in the International Classification of Sleep Disorders, Diagnostic and Coding Manual (AASM, 2005). Common complaints arising from sleep disorders include objective and subjective reports of difficulty initiating and maintaining sleep, poor sleep quality, and various signs of daytime somnolence including difficulty waking in the morning and unplanned or increased need to sleep during the day. While research diagnostic criteria have been developed for most adult sleep disorders (Buysse, 2005; Buysse, Ancoli-Israel, Edinger, Lichstein, & Morin, 2006), comparable, reliable criteria have yet to be developed in children. This is the case for behavioral sleep disorders (BSDs) of childhood, which may be particularly common among children with medical and psychiatric disorders. BSDs generally involve the child having difficulty initiating and maintaining sleep and place a significant burden on the parents who are required to spend significant time with the child at the beginning or in the middle of the night.

To date there have been few studies of sleep disturbance in pediatric patients with FAP (Haim et al., 2004), although there have been reports of subjective complaints of insufficient sleep, poor sleep quality, and daytime fatigue in affected individuals (Garber, Zeman, & Walker, 1990; Heitkemper, Charman, Shaver, Lentz, & Jarrett, 1998; Hyman & Fleisher, 1997). Haim and colleagues (2004) reported abdominal pain prior to sleep onset was a unanimous complaint among study participants with FAP and that 29% of these participants reported that abdominal pain had aroused them from sleep. FAP is suspected to be a precursor of irritable bowel syndrome (IBS) in adults (Walker, 1999). IBS is characterized by FAP associated with bowel complaints such as changes in stool frequency or consistency, as well as relief with defecation (Hyams et al., 1995). The adult IBS literature suggests that associated complaints of subjective sleep problems are common. In the first of two studies of sleep in adults with IBS, participants experienced increased late night gastrointestinal disturbance and there was an objective decrease in the percentages of rapid-eye-movement sleep (Goldsmith & Levin, 1993). In the second study, participants with IBS reported a nighttime increase in pain symptoms

and poor sleep quality (Orr, Crowell, Lin, Harnish, & Chen, 1997).

### ***Pain***

Pain has been shown to have a deleterious impact on the initiation and maintenance of sleep (Drewes, Nielsen, Arendt-Nielsen, Birket-Smith, & Hansen, 1997; Lewin & Dahl, 1999), and the combined effects of chronic pain and illness are likely to have an even greater negative impact on sleep continuity and sleep quality. Insufficient sleep has been shown to increase the perception of pain (Moldofsky & Scarisbrick, 1976) and the cognitive and affective deficits resulting from chronic sleep disturbance may interfere with an individual's ability to manage pain and other stressors associated with chronic illness. These effects may be amplified in pre-adolescents whose coping skills may be eroded by nascent and developing cognitive competencies and abilities to regulate affect and attention. An understanding of the reciprocal effects of pain and insufficient sleep is likely to have important implications for intervention strategies.

Sleep disturbances have been documented in clinical samples of youth with chronic childhood illnesses associated with chronic pain. For example, parents of children with juvenile rheumatoid arthritis (JRA) reported higher rates of nighttime wakings, parasomnias, sleep-disordered breathing, and daytime sleepiness (Bloom et al., 2002). In addition, based on overnight polysomnographic recordings youth with JRA had more nighttime arousals and were significantly sleepier on multiple sleep latency tests (a validated measure of daytime sleep propensity) the following day (Zamir, Press, Tal, & Tarasiuk, 1998). Children with migraine headaches took longer to fall asleep (Bruni, Russo, Violani, & Guidetti, 2004) and headache duration predicted sleep anxiety, parasomnias, and bedtime resistance based on parental reports comparing migraines to healthy children (Miller, Palermo, Powers, Scher, & Hershey, 2003). Female adolescents with chronic musculoskeletal pain took longer to fall asleep, exhibited more night awakenings, had more subjective reports of poor sleep quality and higher rates of daytime sleepiness compared to a normative sample (Meltzer, Logan, & Mindell, 2005). Taken together, youth with chronic pain have higher rates of sleep problems and increased daytime fatigue. Sleep plays an important if not a critical role in the maintenance of optimal physical and mental health. For example, inadequate sleep and greater weekend bedtime delay has been associated with academic difficulties in adolescents (Wolfson & Carskadon, 1998). In addition,

specific deficits in cognitive performance and increased prevalence rates of psychiatric disorders have been associated with specific sleep disorders (Gozal, 1998; Picchietti et al., 1999).

### ***Psychopathology and FAP***

Psychiatric comorbidity with FAP is particularly relevant as sleep disturbance is often associated with anxiety and depressive disorders (Kirmil-Gray, Eagleston, Gibson, & Thoresen, 1984; Simonds & Parraga, 1984). Several studies have documented an association of FAP with anxiety and/or depressive symptoms and disorders in primary care (Campo et al., 2004) and specialty care samples (Garber, Zeman, & Walker, 1990; Liakopoulou-Kairis et al., 2002). One study found that patients with FAP have significantly higher rates of anxiety and internalizing symptoms (e.g., social withdrawal and sadness) than healthy controls, but that participants with FAP did not differ from patients with presumably “organic” gastrointestinal illness (e.g., inflammatory bowel disease) or psychiatric patients (Walker, Garber, & Greene, 1993). There is little data to support the notion that pain causes the associated emotional disturbance or that FAP is simply the consequence of emotional distress. One unconfirmed retrospective study suggests that anxiety may precede the development of FAP (Campo et al., 2004).

The goal of this study was to describe the sleep habits and prevalence of sleep problems in youth with FAP compared to a healthy pain-free comparison group, and to explore the extent to which FAP is associated with BSD symptoms while controlling for the presence of psychiatric disorders. We hypothesized that FAP would be associated with increased symptoms of sleep disorders, decreased sleep duration, and decrements in sleep quality.

## **Methods**

### ***Participants***

A consecutive sample of 67 children and adolescents with FAP and a comparison group of 80 pain-free participants aged 8–15 years were recruited from four primary care pediatric practices in western PA affiliated with a large Children’s Hospital. (Campo et al., 2004). An office-based screening procedure administered during a two-year period was used to identify potential participants. Standard criteria for FAP included three or more episodes of abdominal pain sufficient to interfere with activities or function during the previous three months in the absence of explanatory physical disease.

Comparison group participants were free of abdominal pain or other gastrointestinal problems, headache, chest pain, and limb pain during the previous three months. Exclusion criteria included physical disease considered sufficient to explain the child’s FAP, serious nongastrointestinal chronic physical disease (e.g., diabetes mellitus), pregnancy, mental retardation, and “atypical” symptoms or findings considered indicative of causal physical disease (e.g., persistent nighttime awakenings as a result of abdominal pain in patients who also have evidence of physical disease such as fever or blood in stool) (Boyle, 1997).

### ***Procedure***

The Human Rights Committee of the Children’s Hospital of Pittsburgh approved the study and all participants and their parents completed informed consent and assent documents. Children who presented for routine pediatric care and their parents were introduced to the study in the primary care practice waiting area via a brief letter. Parents interested in participating then completed a visit questionnaire assessing whether abdominal pain was a reason for the visit and whether the child had any history of recurrent pain in the past three months. A research assistant (RA) reviewed the questionnaire and identified children who presented with FAP as a reason for the visit. Physician referrals were not accepted to minimize the possibility of referral bias. To recruit a healthy comparison group with no history of recurrent pain, we utilized frequency matching based age and gender. Three potential comparison subjects were identified for each FAP participant. The comparison subjects were identified among children presenting for minor illnesses or well-child visits within the practice of origin. These children were identified from visit questionnaires collected sequentially after enrollment of the FAP patient and contacted sequentially until at least one agreed to participate in the comparison group. This sampling procedure resulted in a larger comparison group for several reasons; including instances in which a potential abdominal pain subject turned out to be ineligible after a potential control family had been recruited. In addition, there were times in which more than one potential control family sometimes expressed interest in participating in close proximity to another, therefore both families were recruited and subsequently enrolled in the study. Therefore, we agreed to include as many controls as we had contacted who agreed to participate.

The psychiatric interviewer was blind to subject status and interviewed the parent(s) first, then the child

alone, followed by a meeting with parent(s) and child if any areas of discrepancy were identified. While extensive self and parental questionnaires assessing pain, disability, and psychiatric function were completed (Campo, et al., 2004), the focus of this report is on parental and child-report sleep questionnaires. The medical record was reviewed, and the child's primary care physician was contacted when appropriate to clarify whether the presenting symptoms were truly medically unexplained or functional. A total of 220 children were identified on screening as potential FAP subjects. Of these, 67 (30.5%) were enrolled in the study, 67 (30.5%) could not be contacted (e.g. telephone calls were not picked up or returned after the initial screening), 44 (20%) declined participation, and 42 (19%) did not enroll because further review suggested they did not meet eligibility criteria.

## Measures

### Sample Characteristics

Demographic Information included age, ethnicity, gender, and socioeconomic status (SES) which was derived from the Four-Factor Hollingshead (Hollingshead, 1975). In addition, a thorough medical history was taken.

### Sleep Characteristics

Sleep variables, were derived from a 31-item child sleep questionnaire (CSQ) that is in development and is currently used as a clinical screen for common sleep problems in children ages 2–18 (Werthwein, Michaelidis, & Lewin, 2005). The CSQ items are derived from the International Classification of Sleep Disorders (ICSD) and assess sleep habits (usual bedtime, sleep onset latency (SOL), number of awakenings, and time awake after sleep onset), general questions regarding the frequency of symptoms of sleep disorders (bedtime resistance, snoring, sleep walking, etc.). The CSQ also contains a 7-item modified Epworth Sleepiness Scale (ESS) (scores range from 0 to 21) that assesses daytime sleep propensity (e.g., likelihood that a child will fall asleep in common environments, in a car, while in school, etc.). When answering questions parents were asked to report on actual bedtimes and wake times as well as the frequency of relevant events associated with sleep habits, sleep schedules, and the presence of common sleep disorder symptoms occurring in the past month using a 5-point Likert-type scale (1 = never, 2 = less than once a week, 3 = 1 or two times a week, 4 = 3 or 4 times a week, and 5 = 5 or more times a week). Cronbach's alpha for this scale was .76 which is within the .70 to .80 range and considered respectable (DeVellis, 2003) for reliability estimates. In addition to the parent report items, there

<b>Diagnostic criteria used to classify child sleep problems</b>	
<b>For each of the categories below one or more symptoms was required with a frequency <math>\geq 3</math> days/week for each variable:</b>	
<b>Behavioral sleep disorder (BSD)</b>	
Difficulty initiating sleep with a sleep onset latency (SOL) > 30 minutes	
Difficulty maintaining sleep with a wake time after sleep onset (WASO) > 20 minutes	
Bedtime resistance	
Calling out for parent or caregiver during the night	
<b>Excessive daytime somnolence (EDS)</b>	
Difficulty waking in the morning	
Daytime nap frequency	
Modified Epworth Sleepiness Score > 8	
<b>Insufficient sleep</b>	
Sleep Time (TST) < 7 hours	
<b>Sleep Disordered Breathing (SDB)</b>	
Snoring	
Child gasps for breath while sleeping	
Child stops breathing for short periods	

**Figure 1.** Diagnostic criteria used to classify child sleep problems

were several child report items using 5-point Likert-type scales which addressed sleep quality and stress associated with bed time, as well as forced choice (no/yes) questions about pre-sleep cognitions (bedtime worries).

While it was not our intention to formally diagnose study participants with sleep disorders, several categories of sleep problems were defined based on ICSD definitions. These sleep problems, which are common among children, include BSD, excessive daytime somnolence (EDS), sleep disordered breathing (SDB), and insufficient sleep (Fig. 1). Participants experiencing one or more symptoms of these sleep problems with a frequency  $\geq 3$ –4 days per week for the past month were categorized as probable for meeting criteria for the respective sleep problem. A BSD problem was coded as probable if a participant had one or more of the following symptoms for 3 or more days a week: difficulty initiating sleep with a SOL >30 min; wake time after sleep onset (WASO) >20 min which is the average amount of time that a child is awake before returning to sleep following an arousal from sleep; and bedtime resistance. Likewise, patients were categorized as having problems with EDS, if they napped, had difficulty waking in the morning three or more days a week for the past month and/or had a modified ESS > 8. SDB included snoring, gasping for breath, and pauses in breathing while sleeping with a frequency  $\geq 3$ –4 days a week for the past month. Insufficient sleep was defined as a total sleep time <7 h with a frequency  $\geq 3$ –4 days a week for the past month which is significantly lower than normative estimates of the average total sleep time for youth ages 8–15 which range from 10.4 (0.7) to 8.4 h (0.7), respectively with a mean of 9.4 h(0.70) (Iglowstein, Jenni, Molinari, & Largo, 2003).



### Psychiatric Disorders

The Schedule for Affective Disorders and Schizophrenia for School Age Children, Present and Lifetime Version (K-SADS-PL) (Kaufman et al., 1997) was administered to parents and children. The K-SADS-PL is a semi-structured psychiatric interview designed to determine present episode and lifetime history of psychiatric disorder according to *DSM-IV* criteria with established interrater and test-retest reliability as well as convergent and discriminant validity.

### Statistical Analyses

Data from the assessments were collected using scannable forms, then scanned, verified, and entered into a database under the supervision of the database administrator who checked for missing data and outlying values. Both parametric and nonparametric tests were used to compare the participants with FAP to the comparison group on each outcome variable. The scale of the CSQ is not continuous and the intervals are not equal so the Mann-Whitney U-test was used to compare groups on individual CSQ items. The association between group and BSD problems was evaluated by fitting logistic regression models. We have decided to focus on the association of FAP and BSD rather than insufficient sleep, EDS or SDB symptoms because pain associated with chronic pediatric medical conditions has typically been shown to have a significant impact on the initiation and maintenance of sleep which account for several of the BSD symptoms we have described in Fig. 1. Logistic regression models were chosen because the analyses have utilized a binary endpoint (e.g., the absence vs. the presence of one or more of the sleep problems outlined in Fig. 1). Significance was calculated by the likelihood ratio test. The odds ratios (OR) were computed with their corresponding 95% confidence intervals (95% CI). Statistical significance was achieved if  $p < .05$ . In addition, it should be noted that several multiple comparisons among the sleep questionnaire items were made to broadly characterize the range of potential sleep problems in youth with FAP. In cases in which multiple comparisons have been made critical  $p$ -values are often adjusted to minimize Type I error. However, given the nature of this study the critical  $p$ -values have not been adjusted to balance the potential loss of power and the possibility of inflating Type II errors (Curtin & Schulz, 1998; Garamszegi, 2006; Perneger, 1998). The Statistical Package for Social Science (SPSS Inc, version 12.0.2 for Windows, Chicago, IL) was used for statistical analyses. Effect sizes were calculated using Cohen's  $d$  for

**Table I.** Demographic Characteristics of the Study Sample ( $n = 147$ )

Variable	Comparison ( $n = 80$ ) (%)	FAP ( $n = 67$ ) (%)	Statistic
Gender			$\chi^2 (1) = .63$
Female	56.25	62.69	
Male	43.75	37.31	
Race			$\chi^2 (1) = 5.06$
African American	10.00	10.45	
Asian	1.25	1.49	
Caucasian	86.25	79.10	
Hispanic/Latino	0	5.97	
Mixed Ethnicity	2.50	2.99	
Age (years) <sup>a</sup>	12.13(2.17)	11.26(2.10)	$t (144) = -2.45^{**}$
SES <sup>†</sup>	2.03 (1.00)	2.50 (1.23)	$t (144) = -2.46^{**}$

Note. <sup>a</sup>Mean (SD); SES indicates socioeconomic status.

\*\* $p < .01$ .

continuous data and Phi,  $\Phi$ , for Chi-square test and Cliffs'  $d$  for Mann-Whitney U-test (Cliff, 1996).

## Results

### Sample Characteristics

The final study sample included 147 participants, 67 participants with FAP and 80 pain-free comparison participants with a mean age of 11.73 years ( $SD: 2.17$ ). Similar to previously reported clinical samples (Chitkara, Rawat, & Talley, 2005; Walker, Garber, & Greene, 1993; Walker, Garber, Smith, Van Slyke, & Claar, 2001) our study sample was 83% Caucasian and 59% were female (Table I). The two groups did not differ with regard to gender or ethnicity. However, the FAP group was significantly younger (FAP 11.26 (2.10) years vs. the comparison mean 12.13 (2.17) years) and had lower SES ratings based on the Hollingshead scale (Table I).

### Parent Reported Sleep Characteristics

Parental reports indicate that there was an average bedtime of 21:07 on weekdays and 22:57 on weekends and a wake time of 6:48 on weekdays and 8:58 on weekends. Based on parental reports on the CSQ, total sleep time on weeknights and weekends was comparable between the FAP and comparison groups (Table II). The FAP group had significantly longer SOL and WASO. In addition, following a nighttime awakening the FAP group took almost twice as long return to sleep as the comparison group. In addition, parents indicated that both groups maintained a comparable and regular bedtime and bedtime routine (Table III).

Based on the results of Mann–Whitney U-tests, participants with FAP had significantly more frequent symptoms of BSD, EDS, primary snoring, and nightmares (Table III). Participants with FAP had greater difficulty falling asleep ( $U=1735, z=-3.64, p<.001, d=.34$ ), and had a higher frequency of nocturnal awakenings ( $U=1796, z=-3.16, p=.001, d=.28$ ) than the comparison group. Participants with FAP also called out for parents more frequently throughout the night ( $U=2174, z=-1.96, p<.05, d=.16$ ), resisted going to bed more frequently ( $U=1824, z=-3.44, p<.00, d=.32$ ), had more difficulty waking up in the morning ( $U=2152, z=-1.97, p<.05, d=.19$ ), and took more frequent naps ( $U=1824, z=-2.20, p=.01, d=.18$ ) than the comparison group. In addition, parents of participants

with FAP reported a higher propensity to fall asleep during the day on the modified ESS than the comparison group ( $U=2159, z=-2.04, p<.05, d=.19$ ). Significantly higher rates of snoring, ( $U=1921, z=-3.04, p=.001, d=.27$ ) and nightmares ( $U=2122, z=-2.18, p<.05, d=.20$ ) were also reported for the participants with FAP, but there were no significant differences between groups for parasomnias (e.g., sleepwalking and night terrors).

Because CSQ items are rated on an ordinal scale, the association of age and SES with the CSQ items was assessed through nonparametric correlations. While SES was not associated with any of the CSQ variables, age was associated with total sleep time on school nights ( $r=-0.67, p<.001$ ), napping frequency ( $r=.35, p<.001$ ) and the frequency of call outs to parent or caregiver ( $r=-.31, p<.001$ ). Therefore, to clarify the influence of age on these variables, the study sample was divided into two age groups (children  $\leq 12$  years of age and children  $>12$  years of age) and between-group comparisons (FAP vs. comparison) were made within each age group. Age group had no influence on total sleep time for school nights, with no differences observed between groups for total sleep time. In addition, age group had no influence on napping frequency, with the FAP participants having a higher napping frequency in each age group (as reported above). However, the higher frequency of call outs in the night to a parent or caregiver

**Table II.** Parent Reported Sleep Characteristics

Variable	Comparison ( <i>n</i> = 80)	FAP ( <i>n</i> = 67)	<i>t</i> (147)	Effect Size <sup>a</sup>
	<i>M</i> ( <i>SD</i> )	<i>M</i> ( <i>SD</i> )		
Average hours slept (School Nights)	9.28 (1.75)	9.50 (1.76)	-.75	.13
Average hours slept (Nonschool Nights)	10.22 (1.97)	10.03 (1.75)	.85	.10
SOL (min)	17.88 (12.59)	23.85 (16.32)	-2.47**	.41
WASO (min)	8.04 (9.15)	15.34 (18.28)	-2.77**	.52

Note. SOL indicates sleep onset latency; WASO, wake after sleep onset.

\*\* $p < .01$ .

<sup>a</sup>Cohen's *d*.

**Table III.** Parent and Child Reported Sleep Characteristics

Sleep Problem	Comparison ( <i>n</i> = 80)		FAP ( <i>n</i> = 67)		Statistic	<i>p</i> value	Effect Size
	Mean ( <i>SD</i> )	Median	Mean ( <i>SD</i> )	Median			
<b>Parent Reported Sleep Characteristics</b>							
Difficulty falling asleep	1.43 (1.51)	1	2.38 (1.57)	3	MWU	<.001	.34
Nocturnal awakenings	0.61 (0.98)	0	1.31 (1.40)	1	MWU	<.001	.28
Calling for caregiver throughout the night	0.58 (1.45)	0	1.53 (3.71)	0	MWU	<.05	.16
Bedtime resistance	1.33 (1.51)	1	2.18 (1.53)	2	MWU	<.001	.32
Difficulty waking in the morning	1.69 (1.54)	2	2.21 (1.60)	3	MWU	<.05	.19
Nap frequency	0.54 (0.97)	0	1.05 (1.37)	0	MWU	<.05	.18
Modified Epworth Sleepiness Score	3.11 (2.69)	3	4.00 (2.88)	4	MWU	<.05	.19
Snoring frequency	0.88 (1.36)	0	1.76 (1.84)	1	MWU	<.05	.27
Nightmare frequency	0.73 (0.89)	0.5	1.06 (1.02)	1	MWU	<.05	.20
Night terrors	0.10 (0.34)	0	0.13 (0.39)	0	MWU	.53	.03
Sleepwalking	0.20 (0.46)	0	0.22 (0.49)	0	MWU	.76	.02
Frequency of regular bedtime	4.01(1.59)	5	4.24 (1.30)	5	MWU	.86	.02
Variability in bedtime routine	1.85 (0.80)	2	1.96 (0.71)	2	MWU	.30	.09
<b>Child Reported Sleep Characteristics</b>							
Sleep Quality	2.11 (0.95)	2	2.60 (1.03)	3	MWU	<.05	.27
Bedtime stress	1.98 (1.20)	2	2.60 (1.36)	3	MWU	<.05	.24
Total distressing thoughts	2.84 (2.29)	2.5	3.77 (2.98)	3	$t = -2.10$	<.05	.35

Note. The CSQ scale reads: 1 = never, 2 = less than once a week, 3 = 1 or two times a week, 4 = 3 or 4 times a week, and 5 = 5 or more times a week.

among the FAP participants (as reported above) was not maintained after dividing the sample into the two age groups, with the FAP and comparison groups reporting a similar frequency of call outs.

There was no difference in the use of over the counter sleep aids in the two groups; overall, 5.2% of study participants used these medications on at least an occasional basis. No significant associations were found between sleep disturbances and use of psychiatric and gastrointestinal medications. If group differences for use were associated with parent reported sleep disturbances. However, no significant associations were found between sleep disturbances and use of medications.

In summary, the results for individual items of the CSQ indicate higher levels of sleep problems in youth with FAP. However, when these items are considered individually their clinical significance is difficult to determine, therefore we have utilized well-defined clinical criteria described in Fig. 1 to categorize these problems in clinically relevant terms. Overall FAP participants had higher rates of BSD, EDS, and SDB compared to the comparison group (Table IV).

### Child Reported Sleep Problems

Between group comparisons of child-reports of sleep quality (on Likert scales) and sleep related worries

**Table IV.** The Percentage of Participants Meeting Sleep Problem Criteria

Variable	Comparison (n = 80)	FAP (n = 67)	$\chi^2$ (1)	Effect Size $\phi$
	%	%		
BSD	37.5	71.43	13.81***	0.34
EDS	18.75	32.84	3.84*	0.16
Insufficient Sleep	3.75	7.81	1.12	0.09
SDB	6.25	24.24	9.51**	0.26

Note. EDS-excessive daytime somnolence; BSD-behavioral sleep disorders; SDB-sleep disordered breathing.

\* $p < .05$ , \*\* $p < .01$ , \*\*\* $p < .001$ .

**Table V.** Distressing Thoughts at Sleep Onset

Distressing thought	$\leq 12$ years				$> 12$ years			
	Comparison (n = 38)	FAP (n = 42)	$\chi^2$ (1)	Effect Size <sup>†</sup>	Comparison (n = 38)	FAP (n = 22)	$\chi^2$ (1)	Effect size <sup>†</sup>
	%	%			%	%		
Scared of the dark	7.89	33.33	7.71**	.31	5.26	9.09	.33	.06
Noises bother me and keep me up	34.21	42.86	.63	.09	13.16	36.36	4.42*	.24
Someone in my family will get sick	2.63	19.05	5.34s	.26	2.63	13.60	2.71	.20
Someone in my family is going to die	13.16	30.95	3.62s	.21	2.63	9.09	1.22	.13

\* $p < .05$ . \*\* $p < .01$ .

<sup>†</sup> $\phi$ .

yielded several group differences. The FAP group's sleep quality ratings were significantly lower than the comparison group's ( $U = 848$ ,  $z = -2.42$ ,  $p < .01$ ,  $d = .25$ ) and the FAP group's ratings of stress associated with sleep periods were significantly higher ( $U = 872$ ,  $z = -2.38$ ,  $p < .01$ ,  $d = .24$ ) (Table III). The total number of sleep related worries reported prior to sleep onset was higher in participants with FAP ( $M = 3.8$ ,  $SD = 2.9$ ) than in comparison participants ( $M = 2.8$ ,  $SD = 2.3$ ) ( $t = -2.10$ ,  $p < .05$ ,  $d = .35$ ). When age was included as a covariate, group differences were not maintained ( $F(1,141) = 2.60$ ,  $p = .11$ ). Given the contribution of age, individual fears were examined between groups for two separate age groups (children  $\leq 12$  years of age and children  $> 12$  years of age). Older children tended to report being concerned about noises in their home, while younger children were more concerned about parental loss and illness and fear of the dark (Table V). Differences were also noted for ruminating about pain, with the FAP group reporting more pain related cognitions (33%) than the comparison participants (6.58%),  $\chi^2 1$ , ( $n = 142$ ) = 16.42,  $p < .001$ .

### Association of FAP, Sleep Disturbance, and Psychopathology

Logistic regression models were used to explore the association between FAP and BSD while accounting for potentially influential variables (e.g., psychopathology). To construct the most parsimonious logistic regression model, analyses were conducted in two steps. Step one of this analysis involved using univariate models to estimate the effects of potential risk factors without the complication of colinearity. The risk factors analyzed included group (FAP vs. comparison), demographic (age, gender, and SES) and psychiatric (diagnoses of depression and/or anxiety derived from the K-SAD-PL) variables. Of the potential risk factors, only group and psychiatric history (Table VI) were significantly associated with an increased

**Table VI.** Results of Logistic Regression Analysis Predicting Meeting Behavioral Sleep Problem Criteria

Variable	B	SE	Odds Ratio	95% CI	Wald Statistic
Age	-0.08	0.09	0.92	0.78-1.09	0.94
Sex	-0.21	0.38	0.81	0.39-1.70	0.31
SES	0.32	0.17	1.37	0.98-1.93	3.31
Group	1.43	0.39	4.17	1.93-9.00	13.21***
Depression	1.26	0.51	3.52	1.29-9.59	6.05*
Anxiety	1.29	0.4	3.62	1.66-7.90	10.46**
Group, adjusted for					
Depression	1.24	0.41	3.46	1.55-7.73	9.16**
Anxiety	1.04	0.48	2.84	1.11-7.24	4.76*
Depression and Anxiety	0.96	0.49	2.60	1.00-6.74	3.88*

\* $p = .05$ . \*\* $p = .001$ . \*\*\* $p < .001$ .

risk for BSD symptoms, therefore only group and the psychiatric variables were included in subsequent analyses. Step two involved using multivariate models to examine the association between the group main effect and BSD symptoms after adjusting for depression, anxiety and their interaction. The association persisted between the group main effect and BSD problems after controlling for the co-occurrence of psychopathology (Table VI).

## Discussion

The aims of this study were: (a) to evaluate sleep habits and the prevalence of sleep disorder symptoms in children diagnosed with FAP and; (b) to test whether a diagnosis of FAP was associated with an increased risk of having BSD symptoms after controlling for the presence of psychiatric symptoms and disorders. Symptoms of BSD, EDS, and SDB were most prevalent in the FAP group while bedtimes, bedtime routine, and total sleep times were not statistically different in the two groups. In addition, medication use was not associated with the increased prevalence of sleep problems. These findings are consistent with the results of previous studies suggesting that children with abdominal pain experience sleep disturbances (Huang, Palmer, & Forbes, 2000) and poor sleep quality (Haim et al., 2004).

When compared to the healthy, pain-free group, the participants with FAP, on average, had increased: (i) difficulty initiating sleep; (ii) difficulty maintaining sleep; (iii) distressing thoughts related to pain or nighttime fears; and (iv) self ratings of poor sleep quality. These clinically significant BSD symptoms include several of the primary symptoms of insomnia (AASM, 2005), a debilitating disorder involving increased arousal and sleep disruption that can be both cause and consequence of disturbances of mood and quality of life

(Walsh, 2004). Heightened arousal, rumination, and pain can also lead to increased sleep onset latency. Therefore, it is possible that increased rumination about pain and illness, nighttime fears, and decreased sleep may perpetuate insomnia symptoms. However, given the cross-sectional nature of this study it is not possible to establish the direction of causality between FAP and BSD symptoms.

It is not immediately clear why the participants with FAP had increased rates of SDB symptoms. While speculative, there are several plausible explanations that will require further investigation. It is possible that increased somatic complaints and somatic focus is common to both FAP and SDB (Lewin, Rosen, England, & Dahl, 2002). This may be due to the disinhibitory effects of sleep disruption caused by SDB and the increased frequency of physical symptoms in both FAP and SDB. Increased symptoms of EDS may be attributed to a greater frequency of sleep disturbances related to SDB and BSD symptoms in the participants with FAP. It is also possible that some underlying causes of SDB such as adenotonsillar hypertrophy, gastroesophageal reflux, and allergies may have some direct links with FAP. However, additional research will be necessary to expand our understanding of these associations.

As reported previously (Campo et al., 2004), participants with FAP exhibited higher levels of psychiatric disorders (anxiety and depression) than pain-free comparison subjects. Based on logistic regression analyses, the association between group and the occurrence of BSD symptoms remained significant after adjusting for symptoms of both depression and anxiety. These data suggest that children with FAP are at risk of developing BSD symptoms, although they do not identify the specific mechanisms or determinants of the association or direction of causation.



These data raise the possibility that there are potentially important and complex relationships between sleep, emotional disturbances, and FAP symptoms. Disruptions in sleep quality and continuity may lead to decrements in focused attention and adaptive coping, both critical for an individual's ability to deal with pain and other physical symptoms of FAP (Lewin & Dahl, 1999). The effects of inadequate sleep may further lead to maladaptive or inadequate coping, which, in turn, may lead to increased rumination and increased somatic complaints, which in turn could heighten arousal and erode sleep quality and continuity. Interventions targeting each contributing factor (e.g., sleep disorders, pain, rumination, and pain coping and management skills) may be the best approach to improving functional outcomes in children with FAP.

### ***Clinical Implications and Directions for Future Research***

The present results have several clinical implications. First, the evidence that children with FAP are at increased risk for symptoms of sleep disorders suggests that evaluating and treating sleep disorders in this population is warranted. Educating physicians and other healthcare professionals about the importance of evaluating sleep disturbances may help reduce the risk of future sleep problems, improve affective disturbances, and improve coping with physical pain. Second, these data suggest that children with comorbid FAP and emotional disorder are at higher risk for onset of BSD. This is further evidence to support the development and evaluation of multifactorial (e.g., pain, mood, and sleep) interventions.

There are several important limitations in the present study. The data collected in this study relied on parent and child report and, therefore, are subject to retrospective recall bias. It would be optimal to use both subjective and objective measures of sleep disturbance (e.g., actigraphy and polysomnography) because it is possible that times provided by parents may underestimate variables like WASO time or total sleep time. In addition, with self report the potential exists for there to be a discrepancy between child and parent reports of sleep problems, as children with chronic pain may over report sleep problems, while their parents may be unaware of the child's sleep difficulties and underreport sleep difficulties. The sleep questionnaire and algorithms used to define the probable sleep disorders have not been validated in a clinical sample, and, therefore, it is important to acknowledge that the study only points

toward increased rates of symptoms of these common disorders. Given the cross-sectional nature of this study it is not possible to establish the direction of causality between FAP and BSD symptoms and psychiatric disorders. It is therefore possible that the BSD symptoms observed within this sample are associated with physiological (e.g., abdominal pain) and psychological (e.g., depression and anxiety) variables. However, the BSD symptoms may also be a function of an additional unidentified factor. As noted in the methods, several multiple comparisons among the sleep questionnaire items were made to characterize the range of potential sleep problems in this sample. We believe that the balance we struck between control of type 1 error rates and control of type 2 error rates was appropriate for this stage of research in this area, as the desirability of casting a wide net is high to broadly describe the sleep habits and prevalence of sleep problems in youth with FAP as compared to a health comparison group. Subsequent studies can zero in on a smaller set of variables and thereby use more stringent type 1 error rate control without unduly sacrificing power.

### **Conclusions**

Despite these limitations, these data suggest that there are important, yet complex interactions between sleep and FAP. There is evidence of an increased prevalence of sleep disturbances among patients with FAP. Sleep disturbances in patients with FAP appear to be exacerbated by pain, however, the sleep-pain relationship is often considered to be bi-directional and it remains unclear as to whether pain is the antecedent to sleep disruption or if the inverse relationship is true. Increased arousal could result in disruptions in sleep quality and continuity or pain, with each of these then influencing or exacerbating the other. For example, symptoms of insomnia may cause chronic sleep disruption that leads to decrements in focused attention and adaptive coping, both critical for an individual's ability to deal with pain and other physical symptoms of FAP. Thus it is important to consider that sleep disturbances may be a precursor to chronic pain disorders such as FAP that have no identified organic cause.

Future research should focus on testing multivariate etiologic models that specify how the various risk factors might contribute to functional outcomes. For example, it would be useful to test whether increases in anxiety or depressive symptoms mediate the relationship between FAP symptoms and the onset of BSDs. Finally, it will be vital to conduct randomized trials that test the efficacy of

treatment strategies targeting adaptive coping for FAP, and therapy for mood and sleep disorders. An improved understanding of the etiologic processes that give rise to sleep disorder symptoms and the groups that are at greatest risk for BSD should ultimately facilitate the design of effective prevention programs for this common pediatric health problem.

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