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Comorbid Sleep Disturbance in Adolescents with Functional Abdominal Pain

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Abstract

Objective/Background: Sleep disturbances have been commonly reported as comorbid in youth with pain conditions, but prior research specific to functional abdominal pain (FAP) is limited. This study describes individual factors associated with increased risk for sleep disturbance and characterizes the relationship between sleep disturbance and pain-related variables.

Participants: Participants included 278 adolescents (age 11 to 17 years, M age = 15 years; 89% Caucasian; 65% female) with FAP.

Methods: Participants reported on sleep disturbances, abdominal pain severity, functional disability, somatic symptoms, and healthcare utilization.

Results: Female adolescents reported greater sleep disturbance than male adolescents ($t(276) = 5.52, p < .001$, Cohen's $d = 0.70$) and increased age was associated with greater sleep disturbance ($r = .20, p = .001$). In hierarchical regressions controlling for age, sex, and abdominal pain, greater sleep disturbance was significantly associated with greater functional disability ($\beta = .32$), non-gastrointestinal somatic symptoms ($\beta = .35$), and emergency department visits ($\beta = .29$).

Conclusions: Results suggest that sleep disturbance is common and should be assessed in youth presenting with FAP and may be a potential target for intervention.

Introduction

Abdominal pain is one of the most common recurrent pain complaints of childhood, affecting approximately 20% of children worldwide (King et al., 2011). Functional

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abdominal pain (FAP) describes abdominal pain in the absence of an identified disease process (Drossman, 2016) and is associated with disruption of activity, reduced school attendance, and poor quality of life (Claar et al., 1999; Varni et al., 2015). FAP results in significant health care costs, accounting for 2–4% of all pediatrician visits and over 50% of pediatric gastroenterology visits (Rouster et al., 2015). Children and adolescents with similar chronic painful conditions – such as recurrent headache or musculoskeletal pain – commonly report disturbed sleep, and sleep disturbance increases risk for poorer physical, social, and emotional functioning, over and above the contribution of pain severity (Holley et al., 2017; Valrie et al., 2013). However, few studies have examined sleep disturbances in youth with functional abdominal pain (FAP).

Initial studies have suggested that rates of sleep problems may be higher in youth with FAP compared with healthy controls (Haim et al., 2004; Huntley et al., 2007). Less is known, however, about who is at risk for sleep disturbance within adolescent FAP populations. In healthy samples of youth, older adolescents and female adolescents report greater sleep disturbance compared to younger adolescents and males (Hysing et al., 2013). Previous studies of sleep in youth with FAP-related disorders have found inconsistent results (Haim et al., 2004; Huntley et al., 2007; Schurman et al., 2012). Limitations of these studies included small samples or very wide age ranges. In addition, to date, prior research has not examined the relationship between adolescent-reported sleep disturbance and abdominal pain-related variables such as functional disability, non-GI somatic symptoms (e.g., joint pain, fatigue, headache), and healthcare utilization (i.e., emergency department and outpatient medical visits). Consistent with a model of the pain-sleep relationship in children with persistent pain (Valrie et al., 2013), pain perception, sex, and sleep characteristics contribute to functional outcomes such as disability and health care use.

The purpose of this study was to (1) describe individual factors associated with sleep disturbance, and (2) examine the associations between comorbid sleep disturbance and pain-related disability, healthcare utilization, and non-GI somatic symptoms, over and above abdominal pain severity, within a large sample of adolescents with FAP. Findings may help clinicians to identify and target subgroups of adolescents with FAP who could benefit from sleep intervention. We hypothesized that (1) older age and female sex would be significant risk factors for higher levels of sleep disturbance, and (2) greater sleep disturbance would be associated with greater pain-related disability, more non-GI somatic symptoms, and higher rates of healthcare utilization, controlling for age, sex, and abdominal pain severity.

Methods

Participants and procedures

Data were drawn from the baseline evaluation of youth participating in a randomized controlled trial comparing an Internet-delivered cognitive-behavioral therapy program for managing chronic pain in youth with FAP with an Internet-delivered pain education control condition. Participants were adolescents (11–17 years) presenting to a tertiary pediatric gastroenterology clinic in the Southeastern United States for evaluation of abdominal pain. Inclusion criteria were: (1) able to read/write in English at the sixth-grade level, (2) have regular internet access, (3) experiencing episodic or chronic abdominal pain for at least the

past two months (the pain duration specified by Rome III Criteria for pediatric functional abdominal pain disorders) (Hyams et al., 2016), (4) not diagnosed with a serious medical condition (e.g., inflammatory bowel disease), and (5) no hospitalization within the month prior to enrollment.

All study procedures were approved by the Institutional Review Board; informed consent was obtained from parents and informed assent from adolescents. Potentially eligible patients were identified based on medical record review; research staff approached parents by telephone prior to or on the day of their clinic visit. Of 2028 potentially eligible patients, staff made contact with 62% of families (N = 1253) and 76% (N = 954) expressed interest. After assessing for eligibility, 43% (N = 411) met criteria and 344 were enrolled. Of this group, N = 42 did not complete baseline assessments, N = 22 were excluded due to subsequent diagnosis with organic pathology (e.g., inflammatory bowel disease), and N = 2 were excluded due to developmental delay not disclosed at screening. The final sample comprised 278 youth. Prior to randomization, adolescents and parents completed baseline surveys online.

Measures

Sleep disturbance—Youth reported about their sleep disturbance on the PROMIS Sleep Disturbance Short Form v1.0–8b (Cella et al., 2010; Yu et al., 2012) by rating the extent to which they had difficulty falling and staying asleep and their satisfaction with sleep over the past 7 days using a 5-point Likert scale (1 = *not at all* to 5 = *very much*); higher values indicated greater sleep disturbance. Eight items yielded a total score ranging from 8 to 40, which was converted to a T-score (M = 50, SD = 10) in order to characterize level of sleep disturbance, consistent with PROMIS recommendations (HealthMeasures, 2020). Scores were categorized as within normal limits (T-score <55), mild elevations in sleep disturbances (T-score 55–60), moderate elevations (T-score 60–70), and significant elevations (T-score 70 and above; HealthMeasures, 2020). The pediatric version of this PROMIS measure was not available at the time of data collection; therefore, the reference sample for generating T-scores was based on a nationally representative sample of adults. This measure has been validated in healthy samples of adolescents and has demonstrated content validity (Van Kooten et al., 2016) as well as initial convergent validity with both measures of daytime sleepiness as well as actigraphic sleep patterns (Hanish et al., 2017). The T-score conversion has been used in prior clinical samples of adolescents (Bian et al., 2017; Hanish & Han, 2018). Analyses were conducted using T-Scores. The measure demonstrated excellent internal consistency reliability in this sample ($\alpha = 0.94$).

Abdominal pain

Youth completed the Abdominal Pain Index (Laird et al., 2015), which assesses frequency, duration, and intensity of pain during the previous two weeks. Frequency is rated on a 6-point Likert scale (0 = *none* to 5 = *constant during the day*), duration on a 9-point Likert scale (0 = *none* to 8 = *all day*), and intensity on an 11-point Likert scale (0 = *no pain* to 10 = *the most pain possible*). The revised scoring method was used (Laird et al., 2015), which yields a mean ranging 0–4 that takes into account frequency, duration, and intensity with

higher scores indicating greater abdominal pain severity. The measure demonstrated acceptable internal consistency reliability in this sample ($\alpha = 0.78$).

Pain-related variables

Functional disability.—Adolescents reported on their functional disability using an abbreviated 10-item Functional Disability Inventory (FDI; Claar & Walker, 2006). The FDI was condensed to 10-items based on the most frequently endorsed items in a large database of FAP patients (items 2–5, 8–10, 12–14) where prior research demonstrated strong correlation with the 15-item total score ($r = .098$) (Stone et al., in review). Youth reported the degree of difficulty performing 10 activities on a 5-point Likert Scale (0 = *not at all* to 4 = *a lot*) and means were calculated (ranging 0–4). Internal consistency reliability was good in this sample ($\alpha = 0.86$).

Non-GI somatic symptoms.—Youth reported on non-GI symptoms (17 items; e.g., muscle soreness, headache, fatigue, blurred vision, pain in joints) using the Children's Somatic Symptom Inventory–24 (formerly the Children's Somatization Inventory–24; Stone et al., 2019; Walker et al., 2009; Walker & Garber, 2018). Youth rated the extent to which they were bothered by each symptom during the past two weeks on a 5-point Likert Scale (0 = *not at all* to 4 = *a lot*) and means were calculated (ranging 0–4). Internal consistency reliability was good in this sample ($\alpha = 0.88$).

Healthcare utilization.—Parents reported the number of emergency department (ED) visits and number of visits to outpatient providers (primary care, mental health, complementary/alternative medicine provider visits; each category could range 0–6 visits) over the past three months.

Statistical analyses

Statistical analyses were performed in SPSS v19.0 (SPSS, 2010). Descriptive statistics were used to characterize the sample. Skewness was <1 and kurtosis (κ) was <1 for all variables with the exception of ED visits ($\kappa = 9.90$) and outpatient visits ($\kappa = 1.84$). T-scores were used for analyses involving PROMIS Sleep Disturbance and are reported in the Results. Of note, analyses were also conducted with raw scores; patterns of significance and R^2 change did not differ. Age and sex differences in sleep disturbance were examined with Pearson's r (age) and independent samples t -tests (sex). Bivariate correlations (Pearson's r and Spearman's R) were conducted to examine associations among study variables. Hierarchical regressions were conducted to examine associations between sleep disturbance and pain-related variables. Linear regression was used for continuous dependent variables (functional disability, non-GI somatic symptoms) and negative binomial regression was used when the dependent variable was in the form of counts (outpatient visits, emergency room visits) to accommodate the detected overdispersion of these variables. Age, sex, and abdominal pain severity were controlled for in Step 1 and sleep disturbance scores were added in Step 2. Two-tailed p -values $<.05$ were considered significant. All participants had complete data for all variables with the exception of Health Service Use (ED Visits missing for $n = 2$ and outpatient visits missing for $n = 1$), for which negative binomial regressions accommodated the small proportion of missing values using maximum likelihood estimation.

Results

Preliminary analyses

Participants were 11- to 17-years-old ($M = 14.58$, $SD = 1.86$) and predominantly female (65.3%) and Caucasian (89.2% White, 6.5% Black, 3.2% Mixed/Other, 0.4% American Indian or Alaskan Native, 0.4% Asian, 0.4% not reported; 5.4% Hispanic). The majority of participants met Rome III criteria for irritable bowel syndrome (IBS; 58.3%, $N = 162$) and/or functional dyspepsia (FD; 15.8%, $N = 44$). Table 1 presents the descriptive statistics for the variables of interest. Fifty-five percent of the sample was within normal limits for sleep disturbance (T-score < 55); 19% had mild elevations (T-score 55–60), 18% moderate elevations (T-score 60–70), and 8% significant elevations (T-score 70 and above). Sleep disturbance scores did not differ significantly by race, ethnicity, or SES (p 's $> .10$). Table 2 presents bivariate correlations among study variables. Sleep was significantly correlated with abdominal pain severity ($r = .38$, $p < .001$).

Age and sex differences

Sleep disturbance scores were positively associated with age ($r = .20$, $p = .001$). Female adolescents reported higher levels of sleep disturbance than male adolescents (T-score $M_{\text{female}} = 55.13$, $SD = 10.89$; T-score $M_{\text{male}} = 47.52$, $SD = 10.23$; $t(276) = 5.52$, $p < .001$, Cohen's $d = 0.70$).

Associations between sleep disturbance and pain-related variables

Table 3 displays results from hierarchical regressions for each of the four dependent variables: functional disability, non-GI somatic symptoms, ED visits, and outpatient visits. After accounting for age, sex, and abdominal pain severity in Step 1, results from Step 2 indicate that sleep disturbance explained a statistically significant amount of additional variance for functional disability [$R = .08$, $F(1, 273) = 34.82$, $p < .0001$; small-medium Cohen's $f^2 = .09$], non-GI somatic symptoms [$R = .10$, $F(1, 273) = 40.37$, $p < .0001$; small-medium Cohen's $f^2 = .11$], and ED visits [Likelihood Ratio Test $\chi^2(1) = 6.22$, $p = .01$; small-medium Cramer's $V = 0.15$]. Additionally, abdominal pain severity was a significant covariate in Step 2 for models predicting functional disability, non-GI somatic symptoms, or outpatient visits (p 's $< .001$), with higher pain associated with poorer outcomes. Age was a significant covariate in Step 2 in the model predicting non-GI somatic symptoms, with older age being associated with higher levels of symptoms ($p < .001$). Sleep disturbance was a significant predictor in each of the final models for functional disability ($\beta = .32$), non-GI somatic symptoms ($\beta = .35$), and ED visits ($\beta = .29$) as dependent variables (p 's $< .05$).¹

¹In an exploratory manner, we also examined whether age or sex moderated the relation between sleep disturbance and pain-related variables. Of note, for analyses using the PROMIS Sleep Disturbance T-score, all interaction terms were not significant, and the models with interaction effects did not account for significant incremental variance in Step 2 of the regression for all dependent variables.

Discussion

The purpose of this study was to describe individual factors associated with sleep disturbance and to examine associations between comorbid sleep disturbance and pain-related variables in a large sample of adolescents with FAP. As hypothesized, older age and female sex were significant correlates of higher levels of sleep disturbance with effect sizes that were small to medium, respectively. Sleep disturbance was also significantly correlated with abdominal pain severity. Further, sleep disturbance was associated with greater functional disability, non-GI somatic symptoms, and ED visits, over and above the contributions of abdominal pain severity, age, and sex.

Eighteen percent of the sample reported moderately elevated sleep disturbance and 8% reported significant elevations; this finding is similar to a prior study that showed that 30% of youth with pain-related functional GI disorders reported significant elevations in disorders of initiating and maintaining sleep (Schurman et al., 2012). As hypothesized, girls and older adolescents reported greater sleep disturbance as compared to boys and younger adolescents. Previous investigations of youth with functional GI disorders have found mixed results, with one study showing that older age was associated with more insomnia symptoms, whereas another study reported that age was not significantly associated with sleep problems (Huntley et al., 2007; Schurman et al., 2012). Additionally, in previous studies of sleep in youth with FAP-related disorders, either sex was not examined (Haim et al., 2004) or was unrelated to sleep outcomes (Huntley et al., 2007; Schurman et al., 2012). Population-based studies, however, have found that older adolescent girls (e.g., ages 16- to 18-years-old) consistently report significantly poorer sleep duration and sleep efficiency than boys of the same age, and are at two-fold greater risk for insomnia (Hysing et al., 2013; Johnson et al., 2006). Results suggest that age and sex should both be considered when screening for sleep disturbances in youth presenting with FAP.

We also found that sleep disturbance was associated with more non-GI somatic symptoms (e.g., headaches, joint pain, dizziness) and greater functional disability, controlling for abdominal pain severity, age, and sex. Effect sizes were small to medium. Comorbid sleep disturbance has been tied to functional status in other pediatric pain populations, such as juvenile idiopathic arthritis, sickle cell disease, and mixed chronic pain conditions, such that disturbed sleep was associated with higher pain intensity, greater activity limitations, and poorer health-related quality of life (Valrie et al., 2013). Similarly, parents' report about their children's sleep disturbance was related to higher functional disability and GI symptoms in another study (Schurman et al., 2012). Our findings are consistent with Valrie et al. (2013) model of pain and sleep, which suggests that both pain perception and sleep characteristics jointly contribute to functional outcomes.

The evidence partially supported our hypothesis regarding sleep disturbance and health service use, highlighting the downstream effects of sleep disturbances over and above pain symptoms. Sleep disturbance was significantly associated with more ED visits but not outpatient visits (i.e., primary care, mental health, and complementary/alternative medicine providers) when controlling for abdominal pain severity, age, and sex. Other studies have shown that sleep problems are linked to greater healthcare use in pediatric primary care

(Meltzer et al., 2014) and to more ED visits in pediatric asthma (Daniel et al., 2012). The driving factors for increased health services are unclear but may be due to increased next-day pain severity (Holley et al., 2017; Lewandowski et al., 2010) or increased functional disability, or via mediators such as distress and impaired coping (Kouros & El-Sheikh, 2015; Wang & Yip, 2019). A recent systematic review in adults highlighted that poor sleep predicts decrements in global physical functioning, which may drive increased healthcare seeking (Afolalu et al., 2018). Future longitudinal studies are needed to better characterize these complex relationships. However, these findings suggest that addressing sleep problems may be an important component of comprehensive care to reduce costly healthcare utilization in pediatric FAP.

Strengths of this study include a large sample and assessment of pain-related variables across domains of functioning including sleep, disability, non-GI symptoms, and health service utilization. Limitations include the cross-sectional design and a predominantly Caucasian sample. A significant limitation is the measurement of sleep, as sleep was assessed subjectively via self-report with a measure normed on adults. Although we used a T-score conversion previously validated and used in clinical samples of adolescents (Bian et al., 2017; Hanish & Han, 2018; Hanish et al., 2017), future studies can use the pediatric version of the PROMIS Sleep Disturbance measure that is now available. Of note, six of eight items in the pediatric measure are also represented in the adult measure (Forrest et al., 2018). Although this study did not compare rates of sleep problems to healthy controls, prior studies have shown that sleep problems are higher in youth with FAP than healthy youth (Haim et al., 2004; Huntley et al., 2007). Finally, participants reported on non-GI somatic symptoms that may include pain in other anatomical sites (e.g., headache). Youth with functional abdominal pain are a heterogeneous group, and one source of this heterogeneity is the presence of comorbid non-GI pain and symptoms, which have been shown to be related to worse pain and functioning in other studies (Walker et al., 2012).

Results have important clinical and research implications. Given that sleep disturbance contributes to poorer outcomes over and above the influence of abdominal pain severity itself, clinicians may consider routinely screening for sleep disturbance when adolescents present with FAP, especially in older and female adolescents as these groups are at heightened risk. While there is evidence that psychological therapies for pain management leads to improvements in pain and disability for youth with chronic pain conditions (Fisher et al., 2018), previous research suggests that youth with comorbid sleep disturbances demonstrate poorer response to cognitive-behavioral pain interventions (Fales et al., 2015; Murray et al., in press). Taken together, there is a need to develop and test interventions that explicitly target comorbid sleep disturbance in youth with FAP such as cognitive-behavioral therapy for insomnia (Palermo et al., 2017).

In conclusion, our results highlight the importance of assessing sleep disturbance in youth with FAP, particularly older adolescents and girls. Findings from this study also suggest that a subgroup of youth with FAP may benefit from sleep interventions.

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Table 1.

Descriptives for primary variables (N = 278).

	M (SD)	Range
PROMIS Sleep Disturbance T-Score ^a	52.56 (11.43)	28.90–76.50
Abdominal Pain Index ^b	2.26 (.88)	0–4
Functional Disability Inventory ^c	1.07 (.74)	0–3.40
Child Somatic Symptom Inventory – Non-GI Symptoms ^d	.86 (.62)	0–2.88
Health Service Use – ED visits ^e	.53 (1.00)	0–6
Health Service Use – Outpatient visits ^f	3.68 (2.65)	0–15

GI = Gastrointestinal; ED = Emergency Department; N = 276 for ED visits, N = 277 for outpatient visits

^aPossible range 20–80;^bPossible range 0–4;^cAbbreviated FDI, Possible range 0–4;^dPossible range 0–4;^ePossible range 0–6, previous 3 months;^fPossible range 0–18, previous 3 months

Table 2.

Bivariate correlations among study variables.

	1	2	3	4	5	6
1. Sleep disturbance	-					
2. Abdominal pain	.38***	-				
3. Functional disability	.48***	.54***	-			
4. Non-GI somatic symptoms	.49***	.39***	.57***	-		
5. Outpatient visits	.14*	.26***	.19**	.18**	-	
6. ED visits	.16**	.10	.12	.12	.14*	-

Pearson's r reported for correlations among sleep, pain, disability, and symptoms; Spearman's R reported for correlations involving Outpatient and ED visits

GI = Gastrointestinal; ED = Emergency Department;

N = 276 for correlations with ED visits, N = 277 for correlations with outpatient visits

* $p < .05$,

** $p < .01$,

*** $p < .001$

Table 3.

Hierarchical linear regressions predicting pain-related variables.

	Functional Disability			Non-GI Somatic Symptoms			Outpatient Visits ^a			ED Visits ^a						
	B	β	t	R ²	B	β	t	R ²	B	β	z	2 × Log-Likelihood	B	β	z	2 × Log-Likelihood
<i>Step 1</i>				.30				.21				-1215.21				-538.76
Age	-.01	-.02	-29		.07	.22	3.93***		-.001	-.001	-.05		.12	.23	2.00*	
Sex	-.17	-.11	-2.02*		-.15	-.12	-2.05*		-.03	-.01	-.30		.13	.06	.50	
Abdominal Pain ^b	.42	.51	9.52***		.22	.31	5.62***		.22	.08	4.70***		.20	.18	1.56	
<i>Step 2</i>				.08				.10								
Age	-.02	-.04	-.88		.06	.19	3.57***		-.003	-.002	-.14		.10	.19	1.69	
Sex	-.07	-.04	-.87		-.06	-.04	-.82		-.01	-.001	-.08		.23	.11	.91	
Abdominal Pain ^b	.34	.41	7.81***		.14	.21	3.78***		.21	.07	4.13***		.11	.09	.78	
Sleep Disturb ^c	.02	.32	5.90***		.02	.35	6.35***		.004	.02	1.03		.03	.29	2.43*	

GI = Gastrointestinal; ED = Emergency Department; Disturb = Disturbance

N = 278 for models predicting functional disability and non-GI somatic symptoms; N = 276 for model predicting ED visits, N = 277 for model predicting outpatient visits

* $p < .05$,

** $p < .01$,

*** $p < .001$

^a Previous 3 months

^b Abdominal pain severity assessed with Abdominal Pain Index

^c Sleep disturbance assessed with PROMIS Sleep Disturbance Short Form v1.0 8b