Insomnia and Quality of Life in Children Referred for Limb Pain

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ABSTRACT. ***Objective.*** Children with limb pain have significantly diminished quality of life. Although this could result directly from the pain, we investigated the extent to which associated insomnia may contribute.
Methods. A consecutive series of pediatric rheumatology clinic patients (age 3–18 yrs) who presented for initial evaluation of limb pain were offered participation. Parents and children, as appropriate, completed the Pediatric Sleep Questionnaire and Pediatric Quality of Life Inventory (PedsQL 4.0). Validated measures of pain duration and current pain level were provided by the children. Subjects were judged to have substantial insomnia if they had at least 2 of the following symptoms: difficulty falling asleep at night, waking more than twice on average, trouble falling back to sleep, or waking in the morning feeling unrefreshed. Linear regression was used to model the total PedsQL 4.0 score on insomnia, pain duration, and pain level.
Results. Seventy-four subjects were recruited (47 girls, mean age 10 ± 3.9); 25 (33%) had juvenile idiopathic arthritis and 40 (54%) had insomnia. A low PedsQL 4.0 score was predicted by insomnia ($p < 0.001$), but not by pain duration or level (each $p > 0.10$). Neither pain level nor duration differed significantly between subjects with or without insomnia (each $p > 0.10$).
Conclusion. Significant insomnia may affect half of the children who present to a pediatric rheumatology clinic for limb pain. Quality of life in this setting may depend more on insomnia than on current level or duration of pain. (First Release Oct 15 2007; J Rheumatol 2007;34:2486–90)

Key Indexing Terms:
PAIN CHILD ARTHRITIS SLEEP INSOMNIA
SLEEP INITIATION AND MAINTENANCE DISORDERS QUALITY OF LIFE

Chronic musculoskeletal pain from inflammatory or noninflammatory causes affects up to 24% of children[1,2]. Juvenile idiopathic arthritis (JIA) is one of the most important inflammatory causes of limb pain in children. This disorder and others responsible for chronic pain have substantial adverse effects on quality of life[3,4]. The multiple aspects of chronic pain that impair quality of life are likely to include pain itself, disease activity, functional disability, school absences, social limitations, barriers to participation in sports, and depression[5-7]. The potential influence of sleep problems on quality of life in children with chronic pain or JIA has received relatively little attention. One recent study of adolescents with chronic pain, including JIA in addition to headache or sickle-cell disease, highlighted an association between quality of life and sleep disturbance[8]. Sleep problems of many types are common in children[9] and are particularly common in JIA[3,7,10,11]. Sleep problems such as insomnia and sleep apnea can have marked effects on quality of life[12-17]. Children with JIA are known to have frequent nighttime arousals, early morning awakening, and excessive daytime sleepiness[10,11]. Clearly, such children are at risk for secondary insomnia as a result of their pain[18]. However, the extent to which insomnia — usually defined as difficulty with sleep initiation, maintenance, or restorative quality — in turn influences quality of life in children with musculoskeletal pain has not been studied. We assessed the frequency of insomnia in a sample of such children and studied the association of insomnia with diminished quality of life.

MATERIALS AND METHODS

Procedures and participants. As approved by the Medical Institutional Review Board at the University of Michigan, the investigators recruited subjects from among patients aged 3–18 years who presented consecutively, between December 2002 and August 2004, for an initial evaluation of limb pain at the pediatric rheumatology clinic. Written informed consent was obtained from one parent, and assent from each child. Each family completed the Pediatric Sleep Questionnaire (PSQ)[19] and the Pediatric Quality of Life Inventory (PedsQL™ 4.0)[20,21]. Each participant received a routine clinical evaluation with history and physical examination by one of 3 pediatric rheumatologists who also determined the diagnosis and assisted if questions

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arose about the pain self-assessment items. Data were not collected on subjects who chose not to participate. However, during the recruitment period, feedback from the clinicians involved suggested that 70%–80% of patients and families approached about this study agreed to participate.

Measures. Insomnia was assessed using the PSQ, which includes simply-worded question-items that address each of the 4 defining features of insomnia[22], and these items have proven useful in previous studies of childhood insomnia across a wide range of ages[9,23]. Parents complete the PSQ, but are encouraged to obtain the assistance of their children. Although instructions impose no rigid timeframe that could be used to distinguish subacute from chronic insomnia, they specify that responses should "apply to how your child acts in general, not necessarily during the past few days since these may not have been typical if your child has not been well." For the purposes of this study, subjects were considered to have significant insomnia if answers were positive on at least 2 of the following question-items: difficulty falling asleep at night, waking more than twice on average, trouble falling back to sleep, or waking in the morning feeling unrefreshed.

Pain was assessed by duration (months, as reported by the parent) and level as reported by the child on the day of the evaluation (Wong-Baker FACES pain rating scale, 0–10, where 0 = No Hurt, 10 = Hurts Worst). This pictorial pain measure is designed for use in children aged 3 years and older; the instrument is valid and reliable in this age group; and this pain assessment method is preferred to others by the children themselves[24]. A validated, commonly used instrument, the PedsQL 4.0, was employed to assess quality of life[20,21]. Four age-appropriate versions were used. Parents completed the form for toddlers (aged 2–4 yrs); a clinician interviewed young children (5–7 yrs) to elicit appropriate responses; and older children (8–12 yrs) and teenagers (13–18 yrs) completed the appropriate questionnaires directly for their respective age groups. Each age-specific version of the PedsQL 4.0 includes only one item (among more than 20) that specifically asks about "trouble sleeping." Diagnosis was determined by one of 3 pediatric rheumatologists after a complete history and physical examination.

Seventy-eight families agreed to participate. The PSQ data and duration of pain were provided for 74 subjects. The PedsQL 4.0 was completed for 70 subjects, and the data from the Wong-Baker FACES pain rating scale were adequately completed for 59 subjects. Inadequate completion of the FACES pain rating scale did not appear to be age-related. The data from 4 subjects were excluded from all analyses because their families did not complete the PSQ.

Analysis. T-tests or chi-square tests, as appropriate, were used to compare age, sex, quality of life, pain duration, and pain level between subjects with insomnia and those without insomnia. The primary research question was whether insomnia predicts quality of life in children with pain: all other analyses were considered secondary, and correction for multiple comparisons was not attempted. Quality of life scores were computed and tested with and without inclusion of the one item that asks about "trouble sleeping." Linear regression was used to model the total PedsQL 4.0 score on insomnia, pain duration, and current pain level. All statistical analyses were carried out using Statistical Analytic Software version 8.02. The level of significance was set at $p < 0.05$.

RESULTS

Forty-seven (64%) of the 74 subjects with PSQ data were girls and the mean age of the subjects was 10.0 ± 3.9 years. Twenty-five (34%) of the subjects had received a diagnosis of JIA[25] at this initial evaluation. The remainder of the subjects were diagnosed with myositis (n = 1, 1%), arthralgia (n = 33, 45%), generalized pain syndrome and fibromyalgia syndrome (n = 6, 8%), hypermobility syndrome (n = 3, 4%), or growing pains (n = 6, 8%). Forty of the 74 subjects (54%) had insomnia based on the study criteria of at least 2 of 4 symptoms. Table 1 describes the number of positive responses to each insomnia question-item. Fifty-four subjects (73%) had at least one symptom of insomnia, 17 (23%) had at least 3 symptoms, and 9 (12%) had all 4 symptoms. The average duration of pain was 16.1 ± 22.4 months, and the median pain level was 4 on the scale of 1 to 10 (Figure 1). The mean total PedsQL 4.0 score was 63.5 ± 17.5 (Figure 2). The duration of limb pain ranged from 1 to 121 months and in only 2 subjects was the pain present for less than 6 weeks.

Table 1. Number of subjects with each insomnia symptom, and percentage positive among those who answered "yes" or "no" to the question-item.

Insomnia Symptom	No. (%)
Difficulty falling asleep at night	34 (47)
Waking more than twice on average	17 (23)
Trouble falling back to sleep	28 (39)
Walking in the morning feeling unrefreshed	41 (59)

Insomnia predicted significantly lower quality of life scores, whether the one question-item about "trouble sleeping" was included or not (Table 2). A regression model showed that insomnia (present vs absent) explained 18% of the variance in quality of life scores ($R^2 = 0.18$, ß = –14.7, SE = 3.9, $p = 0.0003$). This association was only mildly diminished in strength when the "trouble sleeping" item was omitted from the quality of life scores ($R^2 = 0.15$, ß = –13.3, SE = 3.9, $p = 0.0011$). Current pain level and pain duration were not significantly different in subjects with or without insomnia (Table 2). In this sample of patients, all of whom were referred to the pediatric rheumatology clinic, quality of life scores were not associated with current pain level or pain duration (each, $p > 0.10$ in simple linear regression models). When quality of life was regressed on insomnia along with pain level or pain duration, insomnia still independently predicted quality of life ($p = 0.002$ and $p = 0.001$, respectively).

DISCUSSION

This study of children referred for limb pain suggests for the first time that insomnia readily predicts generic quality of life in children with inflammatory and other musculoskeletal pain disorders. These disorders, and JIA in particular, are well known to be associated with impaired quality of life[26]. Insomnia also has a prominent effect on quality of life[12], although most of the studies that have demonstrated this have been performed in adults and none has examined the possibility that insomnia could significantly diminish quality of life in children with chronic musculoskeletal pain. Pain itself is a well recognized cause for insomnia, but adjustment for pain level or duration in our sample failed to eliminate the association of insomnia with quality of life. Although our cross-sectional study cannot prove causality, the results suggest that insomnia may make a prominent contribution, independent of associated pain, to diminished quality of life in children with chronic musculoskeletal pain.

Our findings are consistent with those of previous studies, some of which have used polysomnographic methods to study

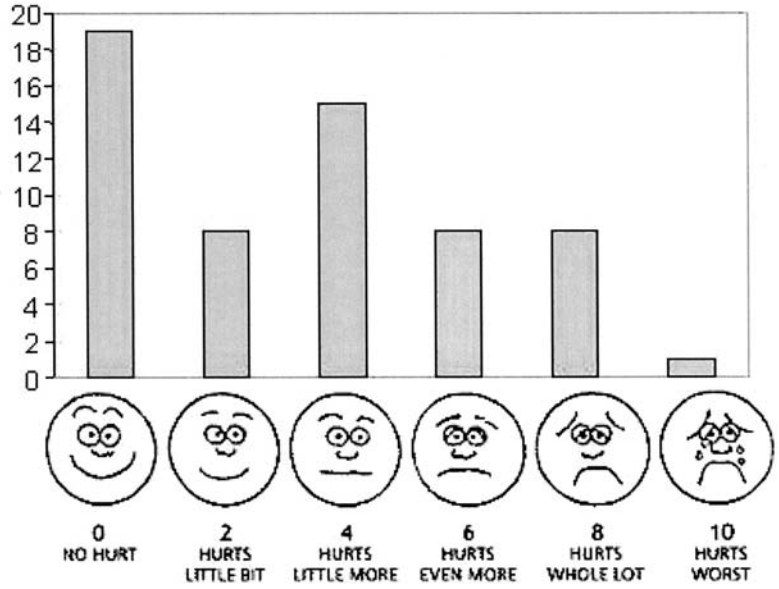

Figure 1. Numbers of subjects with indicated pain levels.

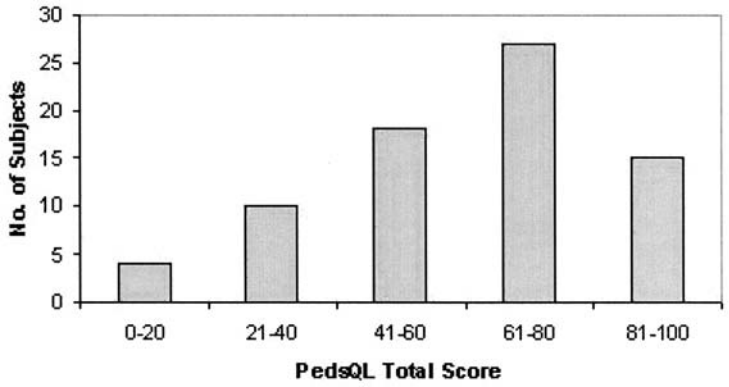

Figure 2. Distribution of scores for quality of life; the total score on the PedsQL 4.0 inventory.

Table 2. Demographic data, quality of life, and pain scores [mean (SD) or percentage] among subjects with and without insomnia.

	Insomnia, n = 40	No Insomnia, n = 34	p*
Age, yrs	10.6 (3.7)	9.3 (4.2)	0.17
Female (%)	25 (62)	22 (65)	0.84
Quality of life scores	57.0 (16.5)	71.7 (15.4)	0.0003
Quality of life scores (without "trouble sleeping" item)	58.1 (16.8)	71.4 (15.3)	0.0011
Pain duration, mo	18.1 (24.8)	13.9 (19.4)	0.42
Pain level	3.7 (2.7)	2.9 (3.2)	0.33

* T-test or chi-square, as appropriate.

smaller samples of children with JIA or other reasons for chronic pain. In comparison to 9 healthy controls, 16 children with JIA showed more arousals and awakenings, sleep-stage shifts, alpha-delta sleep that characterizes pain syndromes, and daytime sleepiness[11]. Among 21 patients with JIA, periodic leg movements, arousals, and alpha activity in non-rapid eye movement (REM) sleep were more prominent than among 20 controls[4]. Among the JIA patients, greater alpha

EEG activity was associated with greater joint involvement and self-rated pain, and periodic leg movements were associated with morning stiffness[4]. In a recent study of 26 adolescents with chronic musculoskeletal pain, participants in comparison to historical controls reported longer sleep onset latency, more night awakenings, a later morning wake time, and more symptoms of daytime sleepiness[27].

In addition to the likelihood that pain contributes directly to insomnia, recent data also suggest that insomnia could exacerbate chronic pain. Sleep deprivation, and perhaps REM sleep deprivation in particular, increases pain sensitivity and reduces pain tolerance[28-31]. Regulation of both REM sleep and nociceptive responses by modulation of acetylcholine release in the pontine reticular formation could potentially explain bidirectional interactions between REM sleep and pain[32].

The high frequency of insomnia symptoms we found in children with chronic pain, correlation of those symptoms with overall quality of life, and the possibility that insomnia could exacerbate pain suggest that identification and treatment of insomnia is warranted in clinical settings, in addition to treatment for the pain itself. Unfortunately, treatment of insomnia has been studied much less in children than in adults, and less in secondary insomnia than in primary insomnia. Several large, randomized clinical trials have shown that cognitive behavioral therapy is effective for adults with primary insomnia[33], and this approach is likely to have longterm advantages over hypnotics as a sole intervention. Virtually no data are available on use of hypnotics in children[34], but surveys of pediatricians show that medications are widely prescribed for childhood insomnia[35,36], even at ages during which nocturnal sleep cannot be expected to be consolidated from a developmental perspective. Behavioral approaches are believed to be optimal for insomnia problems encountered most frequently in children, such as limit-setting disorder and sleep-onset association disorder[37]. In the future, it may be that behavioral approaches or adequately tested hypnotics, or some combination of the 2, will eventually prove best for treatment of childhood insomnia secondary to chronic musculoskeletal pain.

Strengths of our study include its sample size relative to previous series, evaluation of all subjects by pediatric rheumatologists, use of well established survey measures of pain and sleep, and simultaneous use of a validated generic quality of life assessment for the first time in this population. However, the study has several limitations. Our measures of pain and sleep were subjective and could have been affected by recall bias. We did not obtain a Disease Activity Score in patients with arthritis, who made up one-third of the sample. Pain may not be the best measure of disease activity in patients with arthritis. However, as all subjects were recruited at their first visit to the pediatric rheumatology clinic, the majority of the children with arthritis most likely had active disease. Our investigation included no healthy controls, which may explain our inability to show an association between pain and insomnia, as found when another sleep habits questionnaire was used in a previous study of 25 children with JIA[3]. As in any cross-sectional study, we could not account for all possible confounds, including potentially important considerations such as depression or anxiety that could interact with insomnia or pain in our patients[8]. We also could not determine whether pain preceded development of insomnia, or perhaps insomnia predated the pain. Future studies of this type might employ more accurate and objective measures, such as an electronic diary for pain intensity, as the Wong-Baker FACES pain scale may measure pain affect more than intensity[38]. Finally, we may not be able to generalize our findings to all children with musculoskeletal pain, to the extent that our referral population could represent patients with different or more severe symptoms.

Insomnia symptoms may be quite common in children referred to a pediatric rheumatology clinic for limb pain. The quality of life experienced by patients seen in this setting may depend more on insomnia than on current level or duration of pain. Clinicians should inquire about insomnia and its potential causes, in addition to addressing pain and its causes, as bidirectional influences may exist between these conditions, and sleep disorders may well be amenable to treatment.

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