SLEEP AND PAIN IN ADOLESCENTS WITH FIBROMYALGIA

Relationship between Sleep and Pain in Adolescents with Juvenile Primary Fibromyalgia Syndrome

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INTRODUCTION

Chronic pain is associated with significant morbidity in children and adolescents, including functional disability1–2 and symptoms of depression and anxiety.3,4 One of the most disabling co-morbidities is poor sleep quality, endorsed by more than 50% of children and adolescents with chronic pain5 and nearly all children with juvenile primary fibromyalgia syndrome (JPFS).6 Subjective sleep problems include difficulty falling asleep, frequent awakenings, waking unrefreshed, and daytime somnolence.7,8 These subjective complaints are supported by objective sleep measures, including polysomnography (PSG) and actigraphy, which demonstrate shorter sleep duration, poorer sleep efficiency, more arousals/awakenings and limb movements, and longer sleep onset latency among children and adolescents with chronic pain compared with healthy controls.9–13

Patients with fibromyalgia also exhibit electroencephalographic (EEG) abnormalities, particularly alpha intrusions into slow wave sleep (SWS).14–16 Alpha-delta sleep (ADS) has been associated with pain, decreased energy, mood disturbances, and unrefreshing sleep in adults with fibromyalgia,14,17,18 although it is not specific to the disorder.19 The direction of causality between ADS and pain remains unclear. Some suggest that chronic pain leads to poor sleep quality,20,21 whereas others propose that improving sleep quality will decrease pain.16,17 The relationship between chronic pain and disrupted sleep is widely considered to be bidirectional.22 However, the strength of association between ADS and unrefreshing sleep is uncertain.23

The relationship of ADS to pain and subjective sleep quality has been less well studied in children. Children with juvenile rheumatoid arthritis24 and JPFS11 have demonstrated significantly more ADS and subjective sleep difficulties than healthy controls. Mothers with fibromyalgia have more ADS, nonrestorative sleep, and morning fatigue than children with JPFS,11 suggesting these symptoms may worsen over time. Elucidation of relationships among pain, ADS, and subjective sleep quality in JPFS might lead to earlier and more effective interventions.

Exercise therapy is an established treatment for fibromyalgia, with efficacy studies in adults documenting significant improvements in physical fitness, pain threshold and intensity, and sleep.25–28 However, whether and how exercise therapy affects sleep architecture has not been well studied. Exercise and physical fitness have been associated with greater SWS in both children and adults29–32 and it has been suggested that exercise therapy may improve sleep in fibromyalgia specifically.
by reducing ADS. \(^{16,33}\) To test this hypothesis, we investigated whether successful treatment of pain via exercise therapy was associated with improved subjective and objective sleep quality, including ADS, in adolescents with JPFS. We had three main hypotheses: (1) at baseline, adolescents with JPFS will have poor objective sleep quality compared with healthy controls, including more ADS; (2) ADS will be associated with greater pain, disability, and subjective sleep difficulties; and (3) after treatment, pain intensity, subjective sleep quality, and ADS will improve.

**METHODS**

**Participants**

Patients between the ages of 13 and 17 years were recruited from a multidisciplinary pain treatment program that includes intensive exercise therapy. Patients were eligible for participation if they met diagnostic criteria for JPFS\(^{34}\) and reported pain intensity ≥ 50 mm on a 100-mm pain visual analog scale (VAS). Patients were excluded if they could not complete the exercises, had a previous sleep study, had used pain or sleep medications within one week of the study, or had co-morbidity illnesses that could affect sleep or pain. Ten girls of mean age 16.2 ± 0.65 SD years (range = 15.1-17.4 years) completed the study. Age- and sex-matched controls were selected from healthy asymptomatic controls who had participated in previous studies of adolescent sleep. This study was approved by the Institutional Review Board for human subjects research. All participants and legal guardians provided appropriate informed consent/assent.

**Study Design**

This was a single-center, preintervention, and postintervention observational study. Self-report measures were used to assess pain intensity, functional disability, and subjective sleep quality. Actigraphy, PSG, and the Multiple Sleep Latency Test were used to evaluate objective sleep quality. Each subject was compared to herself before and after treatment (Figure 1). Baseline PSG data were also compared to age- and sex-matched controls.

**Intervention**

Treatment was similar to that previously described\(^{35}\) and included six hours of one-to-one physical and occupational therapy daily, emphasizing intense aerobic training and desensitization. Treatment can be outpatient or residential, depending on the family’s travel needs and insurance dictates, but the intervention is identical for both groups. Patients saw a psychologist for cognitive behavioral therapy, received sleep hygiene counseling, and participated in art and music therapy. Treatment duration depends on individual needs, but typically lasts four weeks. Similar programs have been shown to improve pain and functioning in children with chronic pain.\(^{35-37}\) Our own preliminary outcomes data for patients with total body pain or JPFS show significant improvement: pain was resolved in 58%, and 75% report no or minimal disability within one year of treatment.

**Subjective Measures**

**Pain VAS**

A 100-mm VAS was used to assess the intensity of patients’ usual pain in the previous two weeks. The Pain VAS\(^{38}\) has established reliability and validity.\(^{39,40}\) Healthy adolescents report pain scores < 3 on the VAS,\(^{9}\) significantly less than adolescents with chronic pain.\(^{9,10,41}\)

**Functional Disability Inventory**

The Functional Disability Inventory (FDI)\(^{2}\) measures the effect of pain on daily activities. FDI scores range from 0-60, with higher scores indicating greater impairment. Healthy children generally score ≤ 5,\(^{41}\) whereas average scores for pediatric pain patients range from 17.2 to 26.3.\(^{2,42,43}\) In patients with recurrent abdominal pain, coefficient alpha was 0.89 and 3-month test-retest reliability was 0.60.\(^{2}\)

**Sleep and Energy Visual Analog Scales**

Patients used a 100-mm Sleep VAS and a 100-mm Energy VAS to rate the severity of subjective sleep and energy difficulties, respectively, in the previous two weeks.

**School Sleep Habits Survey**

The School Sleep Habits Survey is a reliable and validated scale that assesses sleep/wake patterns and daytime functioning.\(^{44-46}\) It includes four scales: a sleepiness scale measuring daytime somnolence; a sleep/wake behavior problems scale assessing the frequency of erratic sleep behaviors (i.e., trouble falling asleep); a depressive mood scale\(^{47}\) measuring the frequency of depressive symptoms; and a morningness/eveningness questionnaire\(^{48}\) assessing circadian preference.

**Objective Measures**

**Actigraphy**

Actigraphy captures sleep patterns over longer periods of time and is well validated in measuring treatment outcomes.\(^{49-51}\) Subjects wore the Actiwatch 16/64 (Respironics, Inc., Bend, OR) on their nondominant wrist for 7-14 days before and during each overnight PSG, removing it only when bathing. This Actiwatch has been validated against PSG.\(^{52}\) Using a 10-min immobility threshold and low wake threshold as previously described,\(^{52}\) activity counts from each subject for
each night at home were analyzed and compared. Patients also kept sleep diaries, which have been recommended for use with actigraphy to improve accuracy in total sleep time (TST) and sleep efficiency,\(^3\) for one week before and after treatment. Actigraphy variables included TST, sleep efficiency, sleep latency, and wake after sleep onset.

**Polysomnography**

Subjects reported to the sleep laboratory at approximately 7 PM Lights out occurred at the subject’s usual weekday bedtime. PSG recordings began between 9 PM and 11 PM and were completed by 7 AM. The following parameters were recorded (Rembrandt, Embla, Broomfield, CO): EEG (C3/A2, C4/A1, O1/A2, O2/A1), electrooculogram (left and right), submental electromyogram (EMG), tibial EMG, modified lead two electrocardiogram, chest and abdominal wall motion by respiratory inductance plethysmography (SensorMedics, Yorba Linda, CA), airflow by nasal pressure (Pro-Tech Services, Inc, Mukilteo, WA) and three-pronged thermistor (Pro-Tech Services, Inc, Mukilteo, WA); end-tidal partial pressure of carbon dioxide by capnography (Novametrix 7000; Novametrix, Wallingford, CT), arterial oxygen saturation (Novametrix 7000 or Masimo, Irvine, CA), oximeter pulse waveform, and digital video. Data were collected in 30-sec epochs. PSG variables included TST, sleep efficiency, sleep latency, arousal/awakening index, sleep staging, and ADS.

Each study was deidentified and scored, according to American Academy of Sleep Medicine (AASM) guidelines,\(^4\) by a board-certified sleep physician (LJB) in a blinded fashion. Time in each sleep stage was expressed as a percentage of TST. ADS was expressed both in minutes and as a percentage of total SWS (Stage N3). The same observer scored each study to eliminate inter-observer variability. Intraobserver reliability was assessed by assigning each patient and control a new identification number and rescoring all studies in a blinded fashion. Original and rescored studies were compared by calculating the Cronbach alpha coefficient of reliability and intraclass correlation.

**Multiple Sleep Latency Test**

The Multiple Sleep Latency Test\(^5\) evaluates daytime somnolence by providing opportunities for sleep at 2-h intervals throughout the day and measuring the time to sleep onset. Sleep onset in less than 5 minutes reflects pathological sleepiness, whereas 10-20 minutes is normal.\(^6\)

**Statistical Analysis**

Primary outcomes (pain, disability, and sleep quality) were analyzed by paired \(t\)-test. The mean change in each variable pretreatment and posttreatment and its standard deviation were computed. One-sample Kolmogorov-Smirnov tests of normal distribution found no evidence that our variables were not normally distributed; therefore, Pearson correlations were calculated to assess the relationship between ADS and pain intensity/duration, subjective sleep difficulty, and functional disability. One-sample \(t\)-tests were used to compare our sample to literature norms for all measures except PSG. PSG for subjects and controls were compared using parametric \(t\)-tests. SPSS version 17.0 (SPSS Inc., Chicago, IL, release date: August 23, 2008) was used for all data analyses.

**RESULTS**

A total of 14 patients consented to participate. Two participants withdrew consent before completing any studies, and one could not sleep in the laboratory. The fourth was withdrawn due to behavioral issues that prevented completion of the program. A final total of 10 Caucasian females with a mean age of 16.2 years and average symptom duration of 48.6 ± 50.7 SD months completed this study. Seven of the patients completed treatment as outpatients, and the remaining three were residential.

**Pain and Disability**

Pain and disability pretreatment and posttreatment are presented in Table 1. At baseline, patients reported high usual pain intensity and severe functional disability.\(^1\) After treatment, pain (\(P = 0.000\)) and disability (\(P = 0.004\)) improved significantly. Half of the subjects’ pain scores decreased to ≤ 4, including three subjects with pain scores of 0 after treat-
ment, and 8 of 10 patients had FDI scores within the normal range (≤ 5).

**Subjective Sleep Difficulties**

**Sleep VAS and Energy VAS**

At baseline, patients endorsed considerable sleep and energy difficulties. Both sleep (P = 0.008) and energy (P = 0.001) improved significantly after treatment (Table 1).

**School Sleep Habits Survey**

School Sleep Habits Survey scores pretreatment and posttreatment are presented in Table 1. At baseline, circadian preference was significantly shifted to eveningness compared with healthy female adolescents (P = 0.018). Patients had significantly higher scores on the Sleepiness Scale (P = 0.003) but significantly lower scores on the Sleep-Wake Behavior Problems Scale (P = 0.001) compared with healthy adolescents.

After treatment, circadian preference shifted significantly toward morningness (P = 0.014). No significant changes were observed in other School Sleep Habits Survey subscales, although differences in Sleep-Wake Behavior Problems Scale scores approached significance (P = 0.056) (Table 1).

**Actigraphy**

Complete preactigraphy and postactigraphy data were available for only 6 subjects due to technical difficulties. Individual nights that appeared anomalous were eliminated from analysis. For example, one subject displayed significantly different sleep patterns on December 24th (Christmas Eve): therefore, this night was excluded.

At baseline, compared with normative data, patients had significantly poorer sleep efficiency (P = 0.003) and more wake after sleep onset (P = 0.025), but similar TST (P = 0.131) and sleep latency (P = 0.148). None of these variables changed significantly after treatment (Table 2).

**Polysomnography**

At baseline, patients had significantly more arousals and awakenings than controls (P = 0.049; Table 3), suggesting sleep fragmentation. However, there were no significant differences between patients and controls for TST, sleep efficiency, sleep latency, or sleep staging (Table 3). None of these variables changed significantly after treatment (Table 2).

**Alpha-Delta Sleep**

All patients displayed ADS at baseline, averaging 60.6 ± 35.8 SD minutes and encompassing 70.7 ± 32.0 SD% of total SWS, significantly more than controls both in terms of both minutes (P = 0.005) and percentage of total SWS (P = 0.002) (Table 3). There was no significant correlation between ADS and pain intensity or duration, subjective sleep difficulty, or functional disability. Neither the amount nor the percentage of ADS changed after treatment (Table 2). Intraobserver reliability for scoring ADS was excellent, with a reliability coefficient of 0.98 and intraclass correlation of 0.97 (95% confidence interval: 0.87, 0.99).

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**Table 2—Objective sleep measures pretreatment versus posttreatment**

<table>
<thead>
<tr>
<th>Measure</th>
<th>n</th>
<th>Baseline Mean (SD)</th>
<th>Posttreatment Mean (SD)</th>
<th>Paired samples t-test</th>
<th>Significance (two-tailed)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Actigraphy</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>TST(min)</td>
<td>6</td>
<td>396.1 (69.2)</td>
<td>404.3 (33.7)</td>
<td>0.778</td>
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<td>Sleep efficiency (%)</td>
<td>5</td>
<td>69.6 (6.2)</td>
<td>71.3 (7.1)</td>
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<tr>
<td>WASO (min)</td>
<td>6</td>
<td>109.1 (42.2)</td>
<td>107.3 (57.8)</td>
<td>0.906</td>
<td></td>
</tr>
<tr>
<td>Sleep latency (min)</td>
<td>6</td>
<td>35.1 (21.2)</td>
<td>23.9 (9.2)</td>
<td>0.284</td>
<td></td>
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<tr>
<td>PSG</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>TST (min)</td>
<td>10</td>
<td>397.2 (76.6)</td>
<td>409.2 (37.6)</td>
<td>0.637</td>
<td></td>
</tr>
<tr>
<td>Sleep efficiency (%)</td>
<td>10</td>
<td>78.9 (13.9)</td>
<td>79.2 (8.5)</td>
<td>0.957</td>
<td></td>
</tr>
<tr>
<td>Sleep latency (min)</td>
<td>10</td>
<td>42.4 (28.7)</td>
<td>46.4 (25.6)</td>
<td>0.735</td>
<td></td>
</tr>
<tr>
<td>N1 sleep (%)</td>
<td>10</td>
<td>10.5 (10.0)</td>
<td>10.0 (5.1)</td>
<td>0.849</td>
<td></td>
</tr>
<tr>
<td>N2 sleep (%)</td>
<td>10</td>
<td>49.4 (8.5)</td>
<td>52.9 (8.5)</td>
<td>0.401</td>
<td></td>
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<tr>
<td>N3 sleep (%)</td>
<td>10</td>
<td>20.3 (5.7)</td>
<td>17.7 (5.4)</td>
<td>0.084</td>
<td></td>
</tr>
<tr>
<td>REM sleep (%)</td>
<td>10</td>
<td>19.7 (3.4)</td>
<td>19.4 (6.0)</td>
<td>0.900</td>
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<tr>
<td>Arousal-awakens index</td>
<td>10</td>
<td>15.21 (4.74)</td>
<td>17.28 (6.94)</td>
<td>0.304</td>
<td></td>
</tr>
<tr>
<td>ADS (min)</td>
<td>10</td>
<td>60.6 (35.8)</td>
<td>56.1 (28.7)</td>
<td>0.556</td>
<td></td>
</tr>
<tr>
<td>ADS (% SWS)</td>
<td>10</td>
<td>70.7 (32.0)</td>
<td>74.8 (29.6)</td>
<td>0.157</td>
<td></td>
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<tr>
<td>MSLT</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
</tr>
<tr>
<td>Sleep onset latency (min)</td>
<td>10</td>
<td>14.47 (3.58)</td>
<td>15.14 (3.73)</td>
<td>0.539</td>
<td></td>
</tr>
</tbody>
</table>

ADS, alpha-delta sleep; MSLT, Multiple Sleep Latency Test; N1 sleep, Stage 1 nonrapid eye movement sleep; N2 sleep, Stage 2 nonrapid eye movement sleep; N3 sleep, Stage 3 nonrapid eye movement sleep; PSG, polysomnography; REM, rapid eye movement; SD, standard deviation; SWS, slow wave sleep; TST, total sleep time; WASO, wake after sleep onset.
TABLE 3—PSG data for patients at baseline versus healthy control patients

<table>
<thead>
<tr>
<th>Measure</th>
<th>n</th>
<th>Patients</th>
<th>Controls</th>
<th>Significance (two-tailed)</th>
</tr>
</thead>
<tbody>
<tr>
<td>TST (min)</td>
<td>10</td>
<td>397.2 (76.6)</td>
<td>453.5 (98.5)</td>
<td>0.172</td>
</tr>
<tr>
<td>Sleep efficiency (%)</td>
<td>10</td>
<td>78.9 (13.9)</td>
<td>84.0 (7.6)</td>
<td>0.326</td>
</tr>
<tr>
<td>Sleep latency (min)</td>
<td>10</td>
<td>42.4 (28.7)</td>
<td>34.1 (32.0)</td>
<td>0.549</td>
</tr>
<tr>
<td>N1 sleep (%)</td>
<td>10</td>
<td>10.54 (10.01)</td>
<td>8.83 (4.68)</td>
<td>0.633</td>
</tr>
<tr>
<td>N2 sleep (%)</td>
<td>10</td>
<td>49.43 (8.45)</td>
<td>53.58 (12.92)</td>
<td>0.408</td>
</tr>
<tr>
<td>N3 sleep (%)</td>
<td>10</td>
<td>20.30 (5.72)</td>
<td>16.73 (7.07)</td>
<td>0.231</td>
</tr>
<tr>
<td>REM sleep (%)</td>
<td>10</td>
<td>25.20 (19.14)</td>
<td>20.86 (5.81)</td>
<td>0.507</td>
</tr>
<tr>
<td>Arousals-awakenings index</td>
<td>10</td>
<td>15.21 (4.74)</td>
<td>11.67 (2.37)</td>
<td>0.049</td>
</tr>
<tr>
<td>ADS (min)</td>
<td>10</td>
<td>60.4 (36.2)</td>
<td>16.7 (18.9)</td>
<td>0.005</td>
</tr>
<tr>
<td>ADS (% of total SWS)</td>
<td>10</td>
<td>70.3 (32.9)</td>
<td>21.9 (23.1)</td>
<td>0.002</td>
</tr>
</tbody>
</table>

ADS, alpha-delta sleep; N1 sleep, Stage 1 nonrapid eye movement sleep; N2 sleep, Stage 2 nonrapid eye movement sleep; N3 sleep, Stage 3 nonrapid eye movement sleep; REM, rapid eye movement; SD, standard deviation; TST, total sleep time.

**Multiple Sleep Latency Test**

At baseline, mean sleep onset latency was normal\(^{15}\) (14.47 ± 3.58 SD minutes, range = 8.9-20.0 minutes). There was no significant change after treatment (Table 3).

**DISCUSSION**

Pain, disability, and subjective sleep quality improved significantly in our adolescents with JPFS following completion of a multidisciplinary pain treatment program that included intensive exercise therapy. As expected, patients with pain had significantly more ADS than controls at baseline; however, neither ADS nor other objective measurements of sleep quality changed after treatment. In other words, patients’ perceptions of sleep improved even though objective sleep quality did not. We were unable to demonstrate any relationship between ADS and pain intensity or subjective sleep quality, at least in the short term.

ADS was first described in 1973\(^{16}\) and has since been established as common in individuals with fibromyalgia. An early study\(^{16}\) found considerable ADS in adults with fibromyalgia, and these findings have been replicated by other studies.\(^{37,58}\) Similar to our results, several other studies have found significantly more ADS and subjective sleep difficulties in adults with fibromyalgia compared with healthy controls.\(^{11,14,15,59}\) Although Horne and Shackell\(^{60}\) did not find a significant difference in ADS between adults with fibromyalgia and healthy controls, one of their inclusion criteria was that subjects consider themselves to be good sleepers. Because poor sleep is described by 90% of adults\(^{61}\) and 96% of children and adolescents with fibromyalgia,\(^{6}\) the sample in this particular study may not be representative of the disease population.

Although ADS has been less well studied in children, it also appears to be associated with JPFS. Roizenblatt and colleagues\(^{31}\) found significantly more ADS in both children and mothers with fibromyalgia than in healthy controls, similar to our results. Also consistent with our findings, they observed significantly poorer sleep efficiency and more arousals in children with JPFS than in controls. ADS was significantly associated with the number of tender points and inversely related to pain threshold in both children and mothers with fibromyalgia, a correlation we expected to find but did not observe. In our study, pain improved significantly after treatment but the amount of ADS did not change, suggesting these constructs may not be causally related. Though further study is clearly indicated, our results suggest that the etiologies of pain and ADS may be independent in JPFS.

In addition to its prevalence in fibromyalgia, ADS has been associated with pain, mood disturbances, and subjective reports of unrefreshing sleep.\(^{14,16,17,57,62}\) However, the direction of causality between pain and ADS remains unclear. Some studies suggest that pain is caused by ADS, whereas others seem to indicate the reverse. Depriving healthy adults of SWS via auditory stimulation induced musculoskeletal symptoms of fibromyalgia, including muscle aching, stiffness, and tenderness.\(^{63}\) These results were replicated in a later study\(^{64}\) and are further supported by findings of increased morning pain intensity in patients with fibromyalgia who have significant ADS overnight.\(^{57}\) Conversely, Drewes and colleagues\(^{65}\) induced ADS in healthy adults by applying painful stimuli to muscles during SWS. Collectively, these findings suggest a bidirectional interaction between pain and ADS\(^{22}\); however, this relationship is not supported by our finding that ADS persisted after pain diminished, nor by a previous study of adults with fibromyalgia that found no correlation between ADS and symptom severity.\(^{66}\) Additionally, another recent study of adults with fibromyalgia found that neither sleep duration nor time awake after sleep onset significantly predicted pain intensity.\(^{57}\)

The efficacy of exercise therapy in treating fibromyalgia is well documented. It has been shown to improve sleep in adult patients,\(^{25}\) although whether it affects sleep architecture is unknown. In healthy children and adolescents, exercise and physical fitness are associated with increased SWS and better sleep quality.\(^{20,50}\) It has been suggested that poor physical fitness in fibromyalgia\(^{69}\) contributes to both pain and SWS disturbances,\(^{69}\) and that ADS may interfere with the restorative role of sleep.\(^{14,16}\) Evidence that exercise promotes and preserves SWS\(^{32,70}\) has led to the suggestion that physical fitness may protect against ADS.\(^{16}\) In support of this hypothesis, Moldofsky and colleagues\(^{16}\) found that sedentary but otherwise healthy adults de-
veloped musculoskeletal and mood symptoms of fibromyalgia when deprived of SWS, whereas those who exercised regularly did not. Our findings do not support the idea that exercise therapy for pain improves sleep quality by reducing ADS. ADS was not correlated with subjective sleep difficulty and persisted in the absence of reports of unrefreshing sleep. The prevalence of ADS in our sample was high at baseline and, despite significant improvements in subjective sleep quality and physical fitness, did not change after treatment.

It is worth emphasizing that ADS is not specific to fibromyalgia. ADS has been observed in patients with chronic fatigue syndrome, as well as in healthy individuals. It is possible that, rather than pain causing ADS (or vice versa), some other factor, such as genetics, predisposes some individuals to chronic pain, ADS, or both. The existence of a genetic component to fibromyalgia has been suggested, but susceptibility to ADS might be separately heritable. In healthy individuals, Scheuler and colleagues found some evidence for a familial component to ADS, but further research is needed.

The statistical power of this study is limited by its small sample size. Actigraphy data from several subjects were excluded from analysis due to technical difficulties, further decreasing the sample size for actigraphy variables. Additionally, only PSG data were collected from controls. Although we compared our sample to healthy literature controls for other variables, an optimal study design would include collecting control subject data for all measures. It is also important to acknowledge that the intervention described consists of multiple components, including sleep hygiene counseling and cognitive behavioral therapy, in addition to intensive exercise. Although intensive aerobic training constitutes most daily treatment-related activity, it is likely that other aspects of treatment contributed to our patients’ recovery. Therefore, we cannot say with certainty that exercise alone is responsible for the improvements we observed in pain, disability, and subjective sleep quality. Finally, our sample consisted of a very specific population, Caucasian female adolescents, which may limit the generalizability of our findings. Although the overrepresentation of girls is consistent with the 4:1 female-to-male ratio among adolescents with amplified musculoskeletal pain and JPFs is most often diagnosed in white adolescent girls, it is important to determine whether sex and/or race influence outcomes.

This study provides an important foundation for understanding the relationship between pain and sleep in adolescents with JPFs. We have demonstrated that sleep difficulties, including ADS, are common in these patients, and that intensive treatment, including exercise therapy, results in markedly improved pain and self-reported sleep difficulties. However, there were no objective improvements in sleep, particularly sleep fragmentation and ADS. Our results do not suggest causal relationships between ADS and pain or subjective sleep quality, although further study is clearly indicated. Future research should include a larger, more diverse sample and incorporate additional longitudinal points to address the possibility that ADS is slower to resolve than pain symptoms. It may be that EEG abnormalities, including ADS, require more time to resolve than the one to two months between measurements in this study. Future studies might also consider other factors, such as genetics, that may predispose patients to both chronic pain and ADS.

REFERENCES

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DISCLOSURE STATEMENT
This was not an industry supported study. The authors have indicated no financial conflicts of interest.

ABBREVIATIONS
ADS, alpha-delta sleep
DMS, Depressive Mood Scale
FDI, Functional Disability Inventory
JPFS, juvenile primary fibromyalgia syndrome
MEQ, Morningness/Eveningness Questionnaire
MSLT, Multiple Sleep Latency Test
PSG, polysomnography
SLS, Sleepiness Scale
SSHS, School Sleep Habits Survey
SWBPS, Sleep-Wake Behavior Problems Scale
SWS, slow wave sleep
TST, total sleep time
VAS, Visual Analog Scale
WASO, wake after sleep onset
74. Buskila D, Neumann L. Fibromyalgia syndrome (FM) and nonarticular tenderness in relatives of patients with FM. J Rheumatol 1997;24:941-4.