# Efficacy of Cognitive-Behavioral Intervention for Juvenile Primary Fibromyalgia Syndrome

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*ABSTRACT.* ***Objective.*** There are currently no controlled studies of behavioral interventions for juvenile primary fibromyalgia syndrome (JPFM). In this small-sample randomized study, we tested the efficacy of a behavioral intervention, i.e., coping skills training (CST), for the treatment of adolescents with JPFM. Outcomes tested in this study were functional disability, pain intensity, pain-coping efficacy, and depressive symptoms.

***Methods.*** Thirty patients with JPFM were randomly assigned to 8 weeks of either CST or self-monitoring. Adolescents in the CST condition received training in active pain-coping techniques, while those in the self-monitoring condition monitored daily pain intensity and sleep quality with no instructions about behavior change. After posttreatment assessment, subjects were crossed over into the opposite treatment arm for 8 weeks (so that all adolescents eventually received both CST and self-monitoring) and were reassessed at Week 16.

***Results.*** At Week 8, adolescents in both conditions showed significant decrease in depressive symptoms and functional disability. Those who received CST showed significantly greater ability to cope with pain than those in the self-monitoring condition and a trend toward decreased pain intensity. At Week 16, adolescents had significantly lower levels of disability and depressive symptoms compared to baseline, but those who received self-monitoring followed by CST seemed to receive the most benefit.

***Conclusion.*** CST can lead to improved functioning among JPFM patients. Although some of the improvement may be due to increased monitoring and attention, CST provides the specific benefit of improving adolescents' ability to cope with pain. (J Rheumatol 2005;32:1594–602)

*Key Indexing Terms:*
JUVENILE FIBROMYALGIA COGNITIVE BEHAVIOR THERAPY PEDIATRIC PAIN

Fibromyalgia syndrome (FM) is one of the most common rheumatic conditions, forming about 20% of patients seen in rheumatology clinics[1]. The diagnosis and treatment of juvenile primary fibromyalgia syndrome (JPFM) is a significant challenge for pediatric rheumatology clinics. Prevalence estimates of JPFM in community samples range from 1.24% to 6.2% of school-age children, mostly females in the adolescent age range[2-4]. Information from a national registry in the United States indicates that JPFM forms about 7% of new patient diagnoses in a pediatric rheumatology setting[5], but some rheumatology clinics report a much higher percentage of JPFM (up to 45%)[6].

The etiology of JPFM remains unknown and there are no controlled studies of interventions for adolescents with JPFM. Studies have shown that adolescents with JPFM have difficulty with daily functioning, miss a great deal of school, and experience increased emotional distress[7-10]. Teenagers with JPFM report significantly lower levels of health related quality of life than children with other rheumatic diseases such as juvenile rheumatoid arthritis (RA) and lupus[11]. The lack of effective intervention for JPFM symptoms at a time when teenagers are developing critical social skills and preparing for their occupational roles as adults may adversely affect their future psychosocial adjustment; teenagers who are unable to attend school regularly may miss academic preparation necessary for college. Early treatment that enables the adolescent to return to daily activities may reduce the longterm influence of FM.

Most current knowledge about treatment of FM is based on research in adult populations, and little is known about whether these treatments are effective in pediatric populations. Studies show an approximate 30% reduction in pain with pharmacotherapy in adult FM[12,13]. In addition, physical exercise has been found to be beneficial in reducing pain and increasing function among adult patients with FM[14,15]. However, the longterm success of exercise programs clearly depends on a high level of motivation and consistent effort from the patient. In clinical practice, patients' compliance with exercise programs can be quite low, particularly among those who are coping poorly with pain and are psychologically distressed. Psychological approaches, such as

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cognitive-behavioral treatment (CBT), play an important role in the success of multidisciplinary treatment programs but tend to be underutilized[16]. Ideally, when patients have developed strong self-management skills, the likelihood of using regular physical exercise will be greater and longterm pain and symptom relief can be achieved. Development of self-management skills is especially important when FM is diagnosed and when teenagers are beginning to learn critical independent coping skills. Early training in pain-coping skills for teenagers with JPFM may be a step toward prevention of longterm problems associated with chronic pain.

The goal of CBT is to reduce pain and disability and to improve pain-coping efficacy (ability to cope with and manage pain), giving the individual an increased sense of control over pain. Nielson and Jensen[17] reported that a greater sense of control over pain, a belief that one is not necessarily disabled by FM, decreased guarding, use of activity pacing, and other behavioral strategies were associated with favorable treatment outcome in patients with FM. CBT consists of training individuals to take an active role in pain management by using techniques such as activity pacing (e.g., gradually increasing activity levels with frequent rest breaks), muscle relaxation techniques (progressive muscle relaxation), use of calming self-statements (challenging negative thoughts and beliefs about pain), and problem solving (planning ahead for situations that may potentially lead to increased pain). Passive and maladaptive strategies such as reduction of activity and catastrophic thinking (e.g., "I'll never be able to deal with this pain," "my pain is never going to go away") are replaced by more adaptive strategies that increase the person's sense of his or her ability to manage pain. Clinical trials in adults over the past 15 years have established CBT as being efficacious in the treatment of chronic rheumatic conditions including osteoarthritis, RA, and FM[18-21]. These studies found that CBT is effective in reducing psychological and physical disability and increasing pain-coping efficacy. Despite the significant improvement in quality of life and overall well-being, substantial reduction in pain intensity was not consistently found in adult CBT trials. This could be because CBT by itself, without other modalities such as physical exercise, is not sufficient to change pain levels in adults with long-standing pain problems.

Cognitive behavioral approaches have been used with positive results in some studies on the treatment of pediatric pain, primarily in nonrheumatic disorders[22-24]. Two studies have shown promising results in the use of CBT in children with rheumatic conditions. Walco, *et al*[25] describe cognitive behavioral self-regulatory techniques in patients with juvenile RA, and found significant reduction in pain intensity for the children who completed an 8-session CBT protocol. However, this study had a high dropout rate (50%) and no comparison group, making the results difficult to interpret. Walco and Ilowite[26] found preliminary support for the usefulness of CBT for adolescents with JPFM. Specifically, the 5 (out of 7) adolescents who completed the treatment protocol showed reduction in pain and improved functioning. Again, no comparison group was used and the length of treatment varied, based upon when the outcome of pain reduction was achieved. Given the limitations of the designs in these studies, it is not possible to ascertain the specific outcomes most favorably affected by CBT in a pediatric population, the extent of improvement one might expect from CBT, and whether some of the improvement is due to the increased attention from healthcare providers. One of the goals of our study was to determine the effects of a standardized CBT protocol (compared to a self-monitoring control condition) on multiple outcomes, including pain, coping efficacy, functional disability, and mood. Establishing preliminary effect sizes would allow further research using sophisticated clinical trials methodology in patients with JPFM.

We have developed a modified version of a protocol based on adult CBT studies[27,28]. This Coping Skills Training (CST) intervention is a 6-session, individually administered protocol that includes specific training in behavioral pain-coping skills for the adolescent. The content of the CST manual is similar to those used in prior adult and pediatric studies (relaxation training, distraction techniques, calming statements, activity pacing, pleasant activity scheduling, and problem solving). The sessions include developmentally appropriate explanations and training guidelines with examples that are relevant to adolescent life. In addition, we added a parent training component, as in other pediatric studies[25,29], where parents were taught coaching techniques for behavior management strategies in the home. These include suggestions for encouraging the adolescent to manage their pain independently, maintaining normal day to day routines, and guidelines to reduce avoidance of school or social activities. Parents were included in 3 of the 6 sessions. Studies in CBT have found that inclusion of family members can be beneficial in assisting an individual make lifestyle changes and maintain behavior change outside the training sessions[30]. Inclusion of parents is important in pediatric populations, where parents play a more direct role in the child's coping and adjustment to pain.

The primary purpose of our study was to test whether the CST is effective in reducing disability, pain, and depressive symptoms and improving pain-coping efficacy in adolescents with JPFM. The choice of placebo control or comparison conditions was limited because "inert" behavioral conditions (comparable to placebo) are not available. Nevertheless, minimal intervention conditions, which require subject participation but lack specific instructions for behavior change, do exist. For example, self-monitoring is a behavioral technique that requires minimal instruction and no behavioral training in coping strategies (which is the core element of CST). While self-monitoring can lead to

behavior change in patients with chronic health conditions[31-33], we felt that it was the most appropriate comparison group for the CST condition because participants would receive some medical attention but no specific instruction in CST. Subjects in the self-monitoring condition in this study kept a daily record of pain intensity, sleep quality, and medications, but were not provided any instructions for behavior change.

Our primary objective was to test whether there were significantly greater reductions in pain, disability, and depressive symptoms and significantly improved pain-coping efficacy in subjects who received CST compared to the self-monitoring condition. A secondary objective was to test whether there was decreased pain sensitivity (increased tender point threshold) in the CST condition compared to the self-monitoring condition.

In a second phase of the study, we crossed over the subjects to the alternative condition. That is, those who were in the self-monitoring condition were then provided CST, and those who received CST first were asked to self-monitor for the second phase. This study design was chosen for 2 reasons. Ethically, it was felt that all subjects should have an opportunity to receive CST, which is a potentially beneficial intervention. Also, for practical reasons, retention through an 8-week self-monitoring phase followed by no intervention might otherwise have been quite challenging, especially in adolescents subjects with persistent pain.

Outcomes were assessed at baseline (Time 1), at the end of the first 8-week phase of the study (Time 2), and at the end of the second phase of the study (Time 3) at Week 16. We hypothesized that (1) at Time 2, subjects who had received the active treatment (CST) would have significantly lower levels of disability, pain intensity, and depressive symptoms and greater pain-coping efficacy, compared to those who had received self-monitoring only; and (2) at Time 3, all subjects would be significantly improved from their baseline measures, because they had received essentially the same treatments (albeit in reverse order).

## MATERIALS AND METHODS

Subjects were recruited from a pediatric rheumatology clinic in a large midwestern US children's hospital. Enrollment of consecutive eligible subjects occurred over a 2 year period (2001-2003). The clinic sees roughly 25 new cases that meet criteria for JPFM each year. Of about 50 new patients seen over the course of 2 years, 44 were assessed for eligibility and 30 were eventually enrolled for study (Figure 1).

*Inclusion criteria.* Subjects between the ages of 13 and 17 years were eligible for the study. Subjects were included if they met the JPFM diagnostic criteria suggested by Yunus and Masi[34]: generalized musculoskeletal pain at 3 or more sites for 3 or more months in the absence of underlying conditions, severe pain in at least 5 tender point sites upon palpation (using standardized dolorimetry), and other associated symptoms such as sleep disturbance, fatigue and abdominal pain. All patients were taking medications as part of their standard medical care in the rheumatology clinic; subjects were required to be stabilized on medication for at least 4 weeks prior to enrollment, so that effects of the behavioral treatment on outcome measures would not be confounded with medication effects.

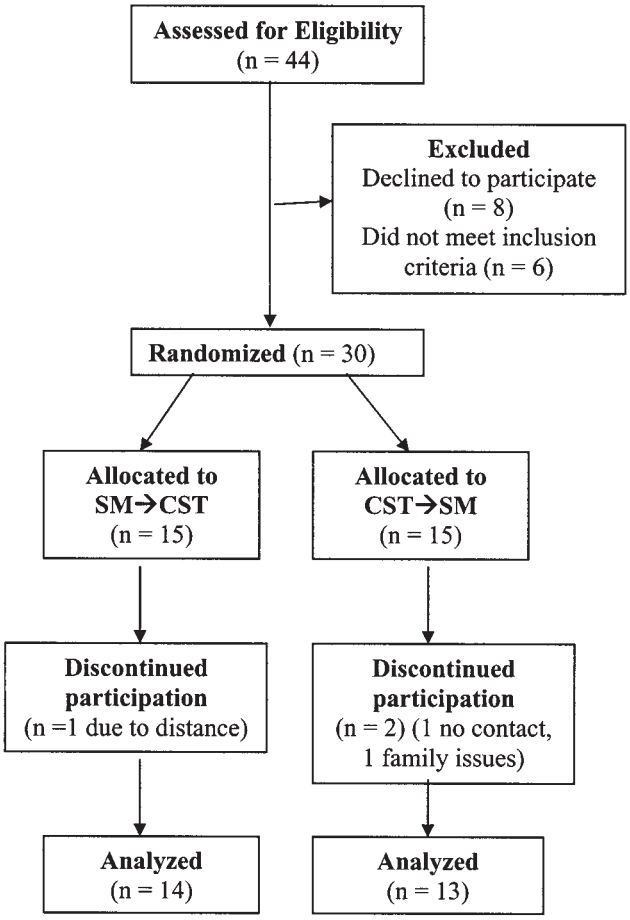


*Figure 1.* Participants' progress through the study; 90% of participants attended all sessions and completed the protocol. SM: self-monitoring condition, CST: coping skills training.

Additional inclusion criteria included average pain level of at least 3 (mild pain) over the preceding 2 weeks on a 10 cm visual analog scale (VAS), and functional disability score greater than 7 (mild disability) on the Functional Disability Inventory (FDI). The cutoff scores for average pain and functional disability were based on pediatric pain studies[8,10,35] and were used to ensure that subjects had at least mild pain intensity and disability levels before they began the behavioral intervention.

*Exclusion criteria.* Participants were excluded if they had a comorbid rheumatic disease such as juvenile RA, or documented developmental delay, or impairment such as autism or mental retardation. Adolescents who met criteria for major depressive disorder [based upon clinical interview and T score > 80 on the Children's Depression Inventory (CDI)[36]] were also excluded, because their immediate needs were for pharmacological treatment of depression, and they were not as likely to benefit from a behavioral pain-coping skills program that required active participation.

Written informed consent was obtained from parents of the subjects and written and verbal assent was obtained from the adolescents. This study protocol was approved by the hospital institutional review board. To reduce attrition and enhance compliance, we provided parents with reimbursement for transportation expenses to bring the adolescents for weekly sessions, and token incentives to the adolescents for daily diaries and each completed evaluation.

*Randomization.* A computer generated pseudo-random number list was used. Due to the small sample size in this preliminary study, a simple ran-

domization technique was used with a 1:1 allocation ratio for the 30 subjects as a single block (so that 15 subjects were assigned to each of the 2 treatment conditions). After confirming that a subject met eligibility criteria, a research assistant who was blind to the objectives of the study enrolled the subject and opened a sealed envelope with the subject's study identification number. The principal investigator then contacted the postdoctoral fellow to begin CST (if the subject was assigned to CST first) or mailed the subject a set of diaries with instructions for completion of diaries (if they were assigned to self-monitoring first).

The 2 treatment conditions are shown below:

| | Phase 1 | Phase 2 |
|---|---|---|
| Treatment Condition 1 (SM→ CST) | Self-monitoring (SM) | Coping skills training (CST) |
| Treatment Condition 2 (CST → SM) | Coping skills training (CST) | Self-monitoring (SM) |

Behavioral intervention

*Coping skills training.* CST was administered by a doctoral level pediatric psychology resident or psychology fellow trained by the principal investigator in the standardized administration of the treatment protocol using the manual developed for this study. The 8-week protocol consisted of 4 weekly individual training sessions, followed by 2 biweekly sessions alternated with 2 telephone check-ins with the therapist. The adolescent was seen for one-on-one sessions with the therapist. One or both parents were included in Sessions 1, 5, and 6. The content of the sessions included explaining the rationale for behavioral techniques; training the adolescent in muscle relaxation techniques to reduce muscle tension and assist with sleep; distraction and activity pacing techniques to cope with pain flares and gradually increase involvement in activities; cognitive techniques to deal with negative thoughts and mood difficulties; and finally, problem-solving to anticipate and plan for difficult or stressful situations and improve sleep hygiene. Examples from the adolescents' daily lives involving home, school, and peer related situations were used to illustrate use of these techniques. Home practice was assigned for each of these techniques at the end of each training session. Parents were provided guidelines to encourage active coping behavior, discourage passive or maladaptive coping, assist adolescents in managing their pain independently, and assess progress. Each session was audio-taped and the principal investigator reviewed each tape to ensure that the content was delivered accurately. The principal investigator also met with the therapist once a week to give feedback and ensure treatment integrity.

*Self-monitoring.* Subjects assigned to self-monitoring were provided with weekly diaries for 8 weeks in which they merely recorded their average pain level for each day (on a 0–10 VAS), sleep quality for each night on a 1–3 scale (good, not so good, bad), and their pain medications. Diaries were collected either by mail or in person after 4 weeks and 8 weeks.

Measures

*Primary outcomes. Functional Disability Inventory.* The FDI is a 15-item global measure of the effect of illness on physical and psychosocial functioning in children and adolescents. The items assess the ability to perform daily activities in home, school, and recreational settings (e.g., doing chores at home, activities in gym class, going out with friends). Subjects rate how much difficulty they have performing each of the activities on a 5-point Likert scale (0 = no trouble to 4 = impossible). The total score indicates the extent of disability, with higher scores indicating greater disability. Research has shown that healthy children's average score is 3.50 on the FDI[35]. Pediatric patients with juvenile FM score an average of 22.06[10]. Internal consistency of the items is high ($\alpha = 0.90$) and test-retest reliability is also high ($r = 0.80$). Construct validity, concurrent validity, and sensitivity to change have also been documented[35].

*Visual analog scale.* The VAS is a 10 cm horizontal line with the words "no pain" and "worst pain" anchored at the 2 ends. Each participant marked average pain intensity and highest pain intensity over the preceding 2 weeks on separate VAS lines. The VAS is a widely used measure of pain intensity, and has been found to be suitable for use in older children and adolescents[37,38]. We have found[39] that children and adolescents seen at a pediatric pain clinic report an average VAS pain intensity rating of 5.62.

*Children's Depression Inventory.* The CDI is a 27-item, norm-referenced self-report measure that assesses symptoms of depression in children and adolescents[36]. The CDI has well established psychometric properties and is one of the most frequently used measures of childhood depressive symptoms. Items on the scale measure symptoms of negative mood, interpersonal problems, ineffectiveness, anhedonia, and negative self-esteem. The responses are scored on a 3-point scale (0–2), with 2 representing a severe form of a depressive symptom and 0 indicating the absence of that symptom. Raw scores are converted to T scores (mean = 50, SD = 10) that are based upon age and sex based norms. A T score below 50 is considered within the normal range. In our previous research, we found that mildly elevated symptoms of depression (T score 55–65) are not uncommon among children and adolescents with chronic pain[39].

*Pain-coping efficacy.* Three items on the Pain Coping Questionnaire (PCQ; described in detail below) assess overall pain-coping efficacy (i.e., subjects' perceived ability to manage and cope with pain). Scores on the pain-coping efficacy scale range from 1 to 5, a higher score indicating greater pain-coping efficacy.

*Secondary outcomes. Pain Coping Questionnaire.* The PCQ is a 39-item questionnaire that assesses a child or adolescent's typical style of coping with pain. The subjects are asked to rate on a 5-point scale (1 = never, 5 = very often) how much they use a particular strategy in response to hurting or being in pain for a few hours or days. The measure assesses 8 individual pain-coping strategies: information-seeking (e.g., trying to find out more about their pain problem), problem-solving (figuring out what to do about it), seeking social support (talking to someone about how it feels), positive self-statements (telling oneself it is not so bad), behavioral distraction (doing something fun), cognitive distraction (trying not to think about it), externalizing (getting mad and throwing/hitting something), and internalizing/catastrophizing (worrying about being in pain). These individual coping strategies can be collapsed into 3 broader (higher order) scales: approach, distraction, and emotion focused avoidance. Approach and distraction (characterized by direct attempts to deal with pain and emotional distress) are considered adaptive, whereas emotion focused avoidance (characterized by a tendency to avoid regulating negative feelings when in pain) is considered maladaptive. The PCQ was developed for use in children and adolescents and has demonstrated good internal consistency ($\alpha = 0.79$–$0.89$). The structure of the subscales and higher order scales is supported by factor analysis[40].

*Tender point examination.* The examination included the following tender point sites bilaterally: low cervical (at C5–C7); second rib (at costochondral junctions); lateral epicondyle (2 cm distal to the epicondyles); knee (medial fat pad proximal to the joint line); occiput (at the suboccipital muscle insertions); trapezius (midpoint of the upper border); supraspinatus (above the scapular spine near the medial border); gluteal (upper outer quadrant of buttocks in anterior fold of muscle); and greater trochanter (posterior to the trochanteric prominence). Two "control" sites, middle of the forehead and right thumbnail, were also assessed. Pressure was applied to each site at a rate of 1 kg/cm$^2$ per second to a maximum of 4 kg/cm$^2$. Subjects were asked to indicate when the sensation of pressure became painful, at which time the pressure was immediately discontinued and the dolorimeter reading recorded. Total tender point count and average pressure-pain threshold for each subject were calculated.

Assessment periods.

*Screening and baseline assessment.* Chart reviews were conducted by the principal investigator and attending rheumatologist to determine if potential subjects met entry criteria for the study. If so, subjects were scheduled

by a research assistant for an initial screening and baseline evaluation (Time 1) after medications were stabilized. At the evaluation, the principal investigator explained the study to the adolescent and parents, obtained consent, and then conducted a semistructured clinical interview to obtain complete background information and history. After the initial interview, subjects responded to the primary and secondary outcome assessment questionnaires and underwent tender point examinations (using dolorimetry).

*Posttreatment assessment (Time 2).* Administration of questionnaires and tender point examinations were repeated at the end of Week 8 (Time 2); that is, after subjects completed the first phase (8 weeks of CST or self-monitoring) of the study. A research assistant who was blind to the study objectives and to the subjects' treatment assignment administered the self-report measures. The rheumatologist or occupational therapist who conducted the tender point assessments was blind to the subjects' treatment assignment.

After this assessment, the subjects entered the second, crossover phase of the study. Those who had completed the CST treatment were provided diaries and instructions for self-monitoring for the final 8 weeks of the study. Subjects who were assigned to self-monitoring for the first phase began the CST component of the study.

*Final assessment (Time 3).* A final assessment took place at the end of Week 16 (Time 3) and consisted of administration of the same outcome measures and tender point examination as in the previous assessments.

*Power calculations.* Although there have been no studies in the treatment of JPFM with adequate sample sizes to estimate effect sizes, we calculated approximate effect sizes based upon a published CBT study for recurrent abdominal pain in children[35], another disorder that is associated with a high level of disability. Improvement in functional disability is of primary importance in demonstrating treatment efficacy: therefore, we used the estimated effect size from the FDI. With an estimated effect size of 0.79 for the FDI, the power to detect within-group differences (Time 1 to Time 2) was 0.98, and the power to detect differences between the 2 groups at Time 2 (n = 15 in each group) was 0.69.

*Statistical methods.* First, descriptive statistics for the 2 treatment conditions at each assessment timepoint were computed (Table 1). To ascertain whether subjects assigned to the 2 conditions showed any pretreatment differences, t tests were carried out comparing the 2 groups on primary outcome measures (FDI, VAS, and CDI) at baseline. The relationship between coping (approach, distraction, and emotion focused avoidance), pain coping efficacy, and primary outcome measures was also assessed using Pearson product-moment correlations (Table 2). Bonferroni corrections were utilized to assess the significance of t tests and Pearson correlations to account for multiple testing.

To test our primary hypothesis, we utilized a repeated measures analysis of variance (ANOVA) to assess whether subjects improved on outcome measures from baseline to Time 2 (within-subjects effects) and whether there was a significant difference between those who received CST versus those who received self-monitoring (between-group effects). The next analysis tested our second set of hypotheses, i.e., whether subjects significantly improved over the course of the 16-week trial (within-subjects effects) and whether there was any difference between the 2 conditions (self-monitoring to CST or CST to self-monitoring) based upon sequence of treatment delivery. To achieve this, we analyzed the change in scores over time, taking into account the sequence of treatment delivery (self-monitoring to CST or CST to self-monitoring) using a Proc Mixed model in SAS, version 8.2. A random coefficient model was used because this approach allows subjects to have their own initial starting point. All analyses were based upon intent to treat according to group assignment, using the last available value carried forward for missing data.

In addition to tests of statistical significance, effect sizes for changes in primary outcome measures from Time 1 to Time 2, Time 2 to Time 3, and overall effect size (Time 1 to Time 3) were computed (Table 3). Descriptions of small (0.2–0.4), medium (0.5–0.8), and large (> 0.8) effect sizes were based upon guidelines suggested by Cohen[41].

## RESULTS

*Sample characteristics.* The final sample consisted of 30 adolescent girls diagnosed with JPFM, ranging in age from 13 to 17 years (median 15.83, SD 1.26). The majority were Caucasian (n = 28) and the remaining were African American (n = 2). Although adolescent males were also eligible to participate, there were none identified that met inclusion criteria for the study. Subjects were enrolled in grades ranging from 7th to the 12th grade. Over half the parents (54% of mothers and 59% of fathers) had a college education or an advanced degree, and few had a high school diploma or less (23% of mothers and 26% of fathers).

Information from the initial assessment revealed that the majority of adolescents reported experiencing widespread pain for over 2 years (63.33%), and the remainder (36.67%) reported having had pain from 6 months to 2 years. All were taking medications to manage their symptoms, the majority taking a combination of antidepressant medication and an analgesic or nonsteroidal antiinflammatory drug (NSAID). Specifically, 50% of subjects were taking a low dose tricyclic antidepressant medication and 23.3% a selective-serotonin reuptake inhibitor. Eighty percent of subjects were taking a NSAID, 26.6% were using a non-opioid analgesic, and 6.6% were taking a skeletal muscle relaxant.

The attrition rate in the study was very low, with 90% of participants attending all sessions and completing the protocol (Figure 1).

*Baseline assessment.* Descriptive data showed that the adolescents were experiencing moderate to high levels of pain

*Table 1.* Mean (SD) scores on disability, pain intensity, tender points, depressive symptoms, and coping efficacy.

| | SM → CST Group | | | CST→ SM Group | | |
|---|---|---|---|---|---|---|
| | Time 1 | Time 2 | Time 3 | Time 1 | Time 2 | Time 3 |
| Functional disability (0–60) | 21.87 (9.47) | 16.64 (8.30) | 9.62 (6.31) | 21.00 (8.84) | 15.07 (9.08) | 16.36 (8.82) |
| Average pain (0–10) | 5.30 (1.05) | 5.92 (2.04) | 4.45 (1.95) | 5.71 (2.15) | 4.40 (1.91) | 4.90 (1.94) |
| Highest pain (0–10) | 8.03 (1.19) | 7.76 (1.90) | 6.24 (2.14) | 8.29 (1.81) | 6.60 (2.53) | 7.33 (2.01) |
| Tender point count (0–18) | 16.45 (1.51) | 14.69 (3.54) | 15.08 (3.17) | 14.50 (3.24) | 11.86 (6.69) | 12.38 (6.95) |
| Tender point threshold (0–4) | 2.57 (0.40) | 2.73 (0.56) | 2.76 (0.44) | 2.58 (0.44) | 2.90 (0.81) | 2.84 (0.83) |
| Depressive symptoms (T score) | 56.07 (12.42) | 48.46 (12.89) | 42.08 (9.81) | 55.47 (14.08) | 49.57 (17.60) | 50.86 (15.47) |
| Coping efficacy (1–5) | 2.84 (0.59) | 3.14 (0.43) | 3.62 (0.71) | 3.09 (0.62) | 3.71 (0.68) | 3.55 (0.67) |

SM: self-monitoring, CST: coping skills training.

*Table 2.* Pearson correlations between coping, pain, functional disability, and depressive symptoms. Values in parentheses show significance.

| | Approach | Distraction | Emotion Focused Coping | Pain-Coping Efficacy | Pain Intensity (average VAS) | Functional Disability | Depressive Symptoms |
|---|---|---|---|---|---|---|---|
| Approach | — | — | — | — | — | — | — |
| Distraction | 0.38 (0.038) | — | — | — | — | — | — |
| Emotion focused coping | 0.22 (0.255) | –0.16 (0.387) | — | — | — | — | — |
| Pain-coping efficacy | 0.05 (0.811) | 0.06 (0.749) | –0.45 (0.012) | — | — | — | — |
| Pain intensity (average VAS) | 0.32 (0.088) | –0.15 (0.432) | 0.74* (0.000) | –0.33 (0.076) | — | — | — |
| Functional disability | 0.28 (0.135) | 0.06 (0.764) | 0.58* (0.001) | –0.57* (0.001) | 0.60* (0.000) | — | — |
| Depressive symptoms | 0.00 (0.998) | –0.11 (0.582) | 0.64* (0.000) | –0.50 (0.005) | 0.31 (0.097) | 0.42 (0.021) | — |

* Statistically significant after Bonferroni correction (< 0.002). VAS: visual analog scale.

*Table 3.* Effect sizes (ES) for functional disability, average pain intensity, depressive symptoms, and pain-coping efficacy by treatment condition. Positive ES values indicate improvement, negative values indicate worsening.

| | SM→ CST Group | | | CST→ SM Group | | |
|---|---|---|---|---|---|---|
| | Time 1 to 2 | Time 2 to 3 | Overall ES (Time 1 to 3) | Time 1 to 2 | Time 2 to 3 | Overall ES (Time 1 to 3) |
| Functional disability | 0.55 | 0.84 | 1.29 | 0.67 | –0.14 | 0.52 |
| Average pain | –0.59 | 0.72 | 0.81 | 0.61 | –0.26 | 0.39 |
| Depressive symptoms | 0.61 | 0.50 | 1.13 | 0.42 | –0.07 | 0.33 |
| Coping efficacy | 0.51 | 1.12 | 1.32 | 1.0 | –0.23 | 0.74 |

(average VAS mean 5.50, SD 1.68; highest VAS mean 8.16, SD 1.51) and difficulty with daily activities (mean FDI 21.43, SD 9.01). Overall, subjects' mean rating of how often they could do something to cope with/manage their pain fell between "hardly ever" and "sometimes" (pain-coping efficacy mean 2.96, SD 0.61). As found in prior research in pediatric chronic pain[39], subjects had mildly elevated symptoms of depression (CDI mean T score 55.77, SD 13.05). T tests for independent means comparing the 2 groups at baseline showed no significant pretreatment differences on any measure.

Table 2 summarizes the correlation between coping, pain, functional disability, and depressive symptoms. As might be expected, higher pain intensity levels were significantly correlated with greater functional disability. With respect to coping strategies, emotion focused avoidance (worrying, getting mad) was significantly correlated with 3 of the outcome measures, with greater use of emotion focused avoidance being associated with higher pain intensity, greater functional disability, and more depressive symptoms. On the other hand, higher levels of pain-coping efficacy (ability to manage and control pain) were significantly related to lower levels of disability.

*Hypothesis 1.* Results of repeated measures ANOVA showed there was significant within-subjects improvement in functional disability and depressive symptoms at the end of 8 weeks ($F = 7.33$, $p < 0.02$ and $F = 17.58$, $p < 0.001$), with no significant differences between the CST and self-monitoring conditions. With respect to average pain levels, the interaction term (Group × Time) approached significance ($F = 3.37$, $p = 0.07$), and post-hoc univariate t tests showed that the CST group had significantly lower pain levels at Time 2 than the self-monitoring group ($t = –2.03$, $p < 0.05$). Results for pain-coping efficacy scores showed that there were significant main effects for Group as well as Time ($F = 5.07$, $p < 0.05$ and $F = 11.95$, $p < 0.01$). Although both groups showed significant increases in pain-coping efficacy, the CST group showed greater improvement than the self-monitoring group (effect sizes 1.0 and 0.51, respectively). There was no significant change in either tender point count or pressure-pain threshold in either group.

*Hypothesis 2.* Results showed that at the end of the 16-week trial, functional disability and depressive symptoms were significantly reduced within subjects ($F = 10.85$ and $F = 10.79$; $p < 0.001$) and pain coping efficacy was significantly increased ($F = 8.48$, $p = 0.002$). The reduction in average pain intensity was not significant. Although there was an overall reduction in functional disability, we found that the extent of improvement was significantly greater among subjects who received the SM to CST sequence as compared to those that received CST to SM ($p < 0.05$). Stated differently, the group that followed the SM to CST sequence achieved a 56.01% reduction in functional disability at the end of 16 weeks, as compared to the group that followed the CST to SM sequence, who achieved a 22.09% reduction in functional disability. There was also a corresponding trend toward a sequence effect for depressive symptoms, with the SM to CST group showing greater reduction in depressive symptoms than the CST to SM group, but the difference between groups was not significant ($p = 0.08$) at the final

evaluation. No significant sequence effect was found between the groups for pain measures, tender point count, or tender point threshold.

With respect to effect sizes (Table 3), we found large effect sizes for the 4 primary outcome measures (functional disability, pain, depressive symptoms, and coping efficacy) by the end of the 16-week trial, ranging from 0.81 to 1.32 in the SM to CST condition. In contrast, the effect sizes for the CST to SM condition showed low to moderate effect sizes for the 4 primary outcome measures, ranging from 0.33 to 0.74. The strongest treatment effects were found for functional disability and pain coping efficacy in both conditions.

## DISCUSSION

This is one of the first controlled investigations into the efficacy of cognitive behavioral therapy for juvenile fibromyalgia. The findings of this preliminary study suggest that CBT may enhance outcomes in adolescent patients receiving traditional medical care for juvenile FM. The behavioral intervention in this study was very well accepted by the adolescents and their parents and we had an excellent retention rate (90%). It is likely that involvement of parents (most of whom were fairly well educated) in some of the treatment sessions played a role in the retention of subjects for this 16-week trial.

Baseline assessment replicated previous research showing that adolescents with JPFM typically experienced a long duration of pain (often over 2 years) before being seen at a rheumatology clinic. Even after being stabilized on medications, subjects were still experiencing difficulty with daily activities and moderate to high levels of pain. A significant relationship was found between maladaptive coping (emotion focused avoidance) and higher levels of pain, disability, and depressive symptoms. The goal of the CST intervention was to specifically address difficulties with coping and associated disability and distress experienced by adolescents with JPFM, by training in pain-coping strategies. Introducing parents to guidelines for behavioral management was also a component of the treatment.

The primary comparison was the adolescents' report of functional disability, pain, depressive symptoms, and pain-coping efficacy at the end of 8 weeks, when subjects had either completed a CST protocol delivered in a standardized fashion by a trained therapist, or were self-monitored using daily diaries with no instructions for behavior change. As expected, subjects who received CST showed significantly greater confidence (coping efficacy) in dealing with pain compared to the group that only self-monitored. Interestingly, subjects in both the conditions (CST and self-monitoring) showed significant reduction in functional disability and depressive symptoms. One might speculate that being enrolled in a treatment study and receiving close monitoring (by the subjects themselves and by healthcare staff) had a positive nonspecific effect on the subjects' overall sense of well-being. This shows that training in coping strategies had the overall effect of improving function and mood, with the added specific benefit of increasing the adolescents' sense of control over their pain. Studies indicate that an increased sense of control over pain may be an important mechanism for improved treatment outcome in patients with FM[17], and our study showed that CST can indeed change the adolescents' views about their ability to control and manage pain. With increased confidence in their self-management skills, one might speculate that these adolescents would be much more likely to participate actively and put consistent effort into treatments (such as regular physical exercise) that could lead to further improvement.

Pain reduction was not statistically significant, but there was a trend toward greater pain reduction in those who were trained in coping skills. This finding is not consistent with an early pediatric study that found significant pain reduction in a small number of adolescents with FM after they received CBT[26], but the study had no control group and subjects were treated until they achieved the outcome of pain reduction. Therefore, it is unclear whether there were factors other than the CBT itself that may have played a role in pain reduction. On the other hand, our finding is similar to controlled clinical trials in adult pain populations that have not consistently found that cognitive-behavioral treatment results directly in decreased pain per se[21]. Rather, the improvements seem to be primarily in the areas of reducing disability and improving mood and a perception of control over pain. As discussed earlier, perhaps these changes occur prior to full engagement in other modalities (such as exercise) that may then provide marked reduction in pain levels.

Our study had a crossover component and the evaluation at the end of 16 weeks comparing the 2 conditions revealed some interesting findings. Subjects in both conditions improved significantly from their baseline assessment, showing significantly lower levels of disability and depressive symptoms, and greater pain-coping efficacy. While we hypothesized that the 2 conditions would lead to a similar degree of improvement in both groups at the end of the trial, we found an unanticipated sequence effect on treatment outcome. Specifically, we found that the group that first self-monitored and then received training in pain coping skills showed a significantly greater reduction in functional disability and a strong trend toward even lower levels of depressive symptoms compared to the group that received CST and then self-monitoring. Calculations revealed large overall effect sizes (0.81–1.32) showing improvement in all 4 primary outcomes (functional disability, pain, depressive symptoms, and coping efficacy) in the subjects that self-monitored prior to CST. In contrast, outcomes for the CST followed by self-monitoring showed low to moderate effect sizes (0.33–0.74) for improvement at the end of the 16-week trial. In keeping with the observation that self-monitoring appears to have an overall beneficial effect on well-being, it

is possible that self-monitoring plays a strong anticipatory or "priming" effect when subjects expect that the self-monitoring will be followed by CST. It should be noted that all subjects who were asked to self-monitor for the first 8 weeks were aware that they would receive the CST at the end of the self-monitoring period. An alternative explanation is that a self-monitoring "run-in" phase might lead to better adherence to the CST program, thereby enhancing the effects of CST. One interesting finding was that substantial improvements in coping efficacy were seen only after training in coping skills (regardless of the sequence of treatment; Table 2). This is consistent with the findings showing that the CST leads to specific improvement in perception of control over pain.

The findings of this preliminary controlled study have important clinical implications. Self-management approaches including the use of self-monitoring and CST were well received by adolescent patients with JPFM when framed in the context of multidisciplinary care. Pediatric rheumatologists should keep in mind that training in self-management strategies primarily influenced adolescents' sense of their ability to control and manage pain and led to reduction in functional disability. Therefore, adolescent patients with JPFM who are having trouble coping with pain and day to day activities may be good candidates for behavioral treatment.

There were a number of limitations of this study; formal assessments of treatment integrity (ratings of audiotaped sessions by blind raters) and documentation of treatment adherence (i.e., how consistently subjects practiced coping skills) were not collected. Also, due to the relatively small sample size, we were limited in the number of variables that could be tested. Age effects may be tested in future studies; it would be useful to know whether older versus younger adolescents benefit more from the CST intervention. The pharmacological aspects of treatment were not tightly controlled in this study. Although low dose antidepressant medication is considered the treatment of choice for FM, no pediatric trial has tested the efficacy of antidepressants in juvenile FM. With a larger sample size and increased power, it would be possible to control and test the effects of medication. Further, activity levels of the participants were not systematically assessed during this study. Future research should evaluate whether subjects make any changes to their overall physical activity level as a consequence of better ability to cope with pain and reduced disability. In addition, whether pain and tender point sensitivity can be reduced by adding physical exercise based interventions should be tested.

It is clear that self-monitoring of sleep and pain levels by itself did have some beneficial effects, but the reasons for this remain unknown. Other than making the subject more aware of day to day changes in symptoms, it is possible that being in a treatment study and receiving attention from healthcare professionals had a positive effect on well-being. Future research should take this effect into account and include an "attention" group that receives the same amount of therapist contact time but without active training in pain coping skills. This would help determine whether CST is more effective than therapist attention in reducing pain, improving coping skills, and reducing disability and depressive symptoms.

It is our intent to conduct a larger controlled trial that will compare pharmacological and behavioral treatments by themselves and the combined effects of both. The goal would be to develop the most effective, evidence based treatment for adolescents with JPFM by testing combinations of medication and behavioral and exercise treatments.

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