Development and Evaluation of a Cognitive-Behavioral Intervention for Juvenile Fibromyalgia

Pamela J. Degotardi,[1] PʜD, Emily S. Klass,[2] PʜD, Batya S. Rosenberg,[1] PʜD, Diana G. Fox,[1] PsʏD, Kim A. Gallelli,[1] PʜD, and Beth S. Gottlieb,[1] MD, MS

[1]Schneider Children's Hospital and [2]NYU Child Study Center

Objective   To describe the development and test the efficacy of a cognitive-behavioral intervention (CBT) for juvenile fibromyalgia.   **Method**   Sixty-seven children with fibromyalgia and their parents were recruited to participate in an 8-week intervention that included modules of pain management, psychoeducation, sleep hygiene, and activities of daily living. Children were taught techniques of cognitive restructuring, thought stopping, distraction, relaxation, and self-reward. Additionally, they kept daily pain and sleep dairies. Children completed questionnaires of pre- and post-treatment measuring physical status and psychological functioning.   **Results**   Following CBT, children reported significant reductions ($p < .006$) in pain, somatic symptoms, anxiety, and fatigue, as well as improvements in sleep quality. Additionally, children reported improved functional ability and had fewer school absences.   **Conclusion**   Children with fibromyalgia can be taught CBT strategies that help them effectively manage this chronic and disabling musculoskeletal pain disorder.

Key words   cognitive-behavioral intervention; juvenile fibromyalgia; pain management.

Juvenile Primary Fibromyalgia Syndrome (JPFS) is a chronic pain condition of unknown etiology, primarily distinguished by widespread musculoskeletal pain, sleep difficulty, depression, and fatigue (Breau, McGrath, & Ju, 1999; Kimura, 2000; Yunus & Masi, 1985). Children with fibromyalgia have no objective signs of arthritis, normal laboratory tests, and evidence of multiple tenderpoints at characteristic soft-tissue sites. The diagnosis often comes after extensive, unproductive medical examinations, and trials of ineffective treatment. Physicians and families alike are frustrated as the medical examination does not support the high level of pain and functional disability reported by children. Musculoskeletal pain syndromes, including JPFS, account for up to 25% of new referrals to pediatric rheumatologists in the United States (Anthony & Schanberg, 2001). Prevalence rates as high as 6% have been reported (Buskila et al., 1993), and adolescent females are predominantly affected (Kimura, 2000). Presentation is slightly dissimilar from adults in that children experience more pronounced sleep disturbance, particularly sleep latency, and fewer painful tenderpoints (Siegal, Janeway, & Baum, 1998; Vandvik & Foresth, 1994; Yunus & Masi, 1985). Children also show evidence of better long-term prognosis (Buskila et al., 1995; Gedalia, Garcia, Molina, Bradford, & Espinozo, 2000; Mikkelsson, 1999).

Understanding etiology has been elusive and yet is important because of treatment implications. Rheumatologists debate whether fibromyalgia should be classified as a rheumatologic or psychiatric disorder, as well as who is best suited to treat the condition (Crofford & Clauw, 2002; Weinblatt, 2002). This debate between biological and psychological mechanisms is eloquently summarized by Kashikar-Zuck, Graham, Huenefeld, and Powers (2000). A biopsychosocial framework has underpinned recent research examining psychiatric symptoms in adolescents with JPFS (Mikkelsson, Sournader, Piha, & Salminen, 1997) and the interplay

of psychological functioning with sleep disturbance (Roizenblatt et al., 1997; Tayag-Kier et al., 2000) and pain (Schanberg, Keefe, Lefebvre, Kredich, & Gil, 1996, 1998). This biopsychosocial model provides the theoretical rationale for nonpharmacological interventions that aim to treat the combination of pain and psychological symptoms associated with fibromyalgia.

Intervention studies in youth with JPFS are scarce. Walco and Ilowite (1992) administered an open clinical trial of cognitive-behavioral therapy to children with JPFS with good outcome for the five completers. Cyclobenzaprine (Flexeril) was used by Romano (1991) to treat 15 children with JPFS, and 73% responded positively. The paucity of pediatric outcome data led us to examine adult treatment studies in designing an intervention suitable for JPFS. A meta-analysis of 49 treatment outcome studies comparing pharmacological with nonpharmacological [i.e., cognitive-behavioral intervention (CBT) or exercise] treatments for adults with fibromyalgia concluded that the latter treatments were more effective in improving self-reported fibromyalgia symptoms and daily functioning (Rossy et al., 1999).

Extrapolating from the good results in adult fibromyalgia using interdisciplinary therapy (Manon, Goolkasian, & McCain, 1998; Masi & Yunus, 1990; Turk, Okifuji, Sinclair, & Starz, 1998) and the efficacy of cognitive-behaviorally based techniques (Rossy et al., 1999), we developed an outpatient, multimodal 8-week protocol for treating youth diagnosed with JPFS (Klass, Degotardi, Fox, Rosenberg, & Ilowite, 1998). In efforts to make the intervention replicable, to identify elements contributing to therapeutic success, and to reduce error because of variation in treatment (Drotar, 1997), we manualized the treatment. Because of the speculation about the primacy of sleep disturbance, the high levels of pain reported by children, and the prevalence of anxiety and depressive symptoms, we incorporated pertinent intervention strategies addressing pain management, sleep hygiene, anxiety, and stress reduction. By specifying the cognitive-behavioral treatment in a manual format, by applying reliable and valid outcome measures across multiple domains, and with diagnostic certainty provided by our rheumatology colleagues, we attempted to design an effective intervention for JPFS.

Development of Current Intervention for JPFS

During the past 5 years, our pediatric psychology/rheumatology team has assessed and treated over 200 children with JPFS and conducted the following two pilot studies.

Pilot Study 1

Fifteen children (10–18 years) diagnosed with JPFS were randomly assigned to either an experimental group (CBT using a rudimentary version of the JPFS Treatment Manual) or a control group that received therapist attention and training in relaxation therapy. Independent sample t-tests indicated that the eight children in the CBT group showed significant improvement in sleep quality, pain control, and physical functioning, $t(7) = 2.8$, $p < .05$. In contrast, the seven children in the control group showed increasing disability (mean scores on the Functional Disability Index rose from 20.2 to 26.5), were dissatisfied with treatment and were at risk for dropout (Klass et al., 1998). As it was clear that those in the treatment group were uniformly improving while those in the control group were doing poorly, ethically we felt obliged to discontinue the study and offer all children CBT treatment.

Pilot Study 2

The relationship between sleep quality and pain was explored in a group of 40 children (10–18 years) diagnosed with JPFS. Daily sleep diaries and pre- and post-treatment measures on the Pediatric Sleep Disturbance Questionnaire, Fatigue Severity Scale (FSS), sleep quality Visual Analogue Scale (VAS), and fatigue VAS scores were compared using paired sample t-tests. We found that global sleep difficulties, in particular, sleep latency ($M = 37$ min) and nonrestorative sleep, were common and severe. VAS measures of current pain were significantly correlated ($p < .05$) with sleep latency ($r = .4$), sleep disturbance ($r = .5$), and fatigue ($r = .6$). Path analysis indicated that improved sleep was associated with decreased levels of pain. Following CBT treatment, sleep improved, pain and fatigue scores decreased, and children reported less interference in activities of daily life (Fox et al., 1999).

This Investigation

Based on our two pilot studies, revisions were made to the JPFS Treatment Manual. For example, due to the relationship between improvements in the quality of children's sleep and decrease in pain, the sleep module is now introduced first. Strategic family therapy, cognitive-behavioral therapy, and interpersonal psychotherapy techniques are woven throughout. In particular, the manual is moderately structured in that general issues and activities are defined, and word-for-word scripts are not included except for the specific relaxation exercises. The four primary treatment modules in the manual are psychoeducation, sleep improvement, pain management,

and activities of daily living. Briefly, these treatment modules are the following.

Psychoeducation (Week 1)
Information regarding JPFS etiology and treatment was adapted from recent literature (Anthony & Schanberg, 2001; Breau et al., 1999; Buckelew et al., 1998; Roizenblatt et al., 1997). JPFS symptoms, the role of stress in pain perception, the physiology of pain, posture and body mechanics, the relationship between emotional issues and the maintenance of symptoms, and normal sleep cycles were discussed with each child and parent. Arthritis Foundation brochures, "pain" and "fibromyalgia," were given to families.

Sleep Improvement (Weeks 2–3)
Following a discussion to identify problematic sleep patterns, a structured protocol was used to treat sleep difficulties. This involved helping children make changes in sleep hygiene and identify maladaptive cognitions that interfered with sleep. Stimulus control and sleep restriction techniques were used, and children were instructed in relaxation and breathing techniques. Children were instructed to practice relaxation exercises daily, using individualized relaxation tapes to aid home practice. Children were also asked to maintain daily sleep logs to identify problematic patterns of behavior and to map changes in sleep quality. These diaries were used as a source of positive reinforcement, as they clearly delineated improvements in sleep habits. Successes in maintaining behavioral changes and controlling fibromyalgia symptoms were reviewed weekly.

Pain Management (Weeks 4–6)
The pain management protocol included exploring maladaptive cognitions regarding pain; teaching cognitive reframing, thought stopping and distraction techniques; and the use of visual imagery and self-hypnosis for pain reduction. At each session, children were asked to rate their level of pain using a 10-cm VAS adapted from the Pediatric Pain Questionnaire (Varni, Thompson, & Hanson, 1987). Children also kept a daily pain diary to help identify patterns of pain behavior and the association between stress, activity, emotions, and pain. This mapping of pain symptoms and identifying determinants of pain strengthened children's beliefs in their ability to modify pain experiences. Setting behavioral goals and specifying self-rewards helped shape adaptive behavior. Strategies to ensure continuing success and planning for future challenges were discussed. The therapist worked with parents to ensure that illness-encouraging patterns did not perpetuate the cycle of pain complaints (Kimura, 2000).

Activities of Daily Living (Weeks 7–8)
Children were encouraged to make any necessary ergonomic and postural changes and to participate in regular aerobic activities. The importance of stretching, warmups, and increasing muscular strength and flexibility was emphasized. Cognitive restructuring was used to address maladaptive attitudes regarding the relationship between exercise and pain. Modifying computer usage, pacing of activities, time management skills, and interpersonal issues maintaining illness behaviors, such as family structure, and secondary gains from the sick role were also discussed.

Developmental Considerations

Because of the age range in our sample (8–20 years), therapists slightly modified the materials and the issues discussed. For younger children (8–11 years) with JPFS, the psychoeducation module was simplified. The therapist used visual aids to illustrate concepts and simple nontechnical language to explain cognitive-behavioral techniques. The therapist was directive, gave clear, detailed instructions, and reviewed major concepts frequently. Specific issues relevant to this age group included managing fatigue during the school day, carrying book-bags, participating at recess and gym, play dates with friends, and making the transition to middle school. For early adolescents (12–14 years), independence from parents and treatment adherence started to become major issues (Rapoff, 1999). The therapist focused on encouraging treatment adherence through self-monitoring (e.g., diaries), behavioral contracting, and self-reward. Issues pertinent to early adolescents included managing classroom changes, homework, participation in gym or team sports, the increasing importance of peers, dealing with parental conflicts, independently managing symptoms, and the transition to high school. Older adolescents (15–20 years) often expressed more curiosity about disease processes, etiology, and alternative treatments. This intellectual curiosity and skepticism was directed toward the goal of assuming personal responsibility for managing fibromyalgia symptoms and exploring library and Internet resources. Specific issues addressed with this older age group included learning time management strategies to cope with the overwhelming demands of school, extracurricular activities, and part-time work; stress reduction techniques; friendships, dating; and preparing for the transition to college and independent living.

Collaborating with Parents

The purpose of the collaboration was to educate parents in the treatment of JPFS and to teach them how to coach their child in applying cognitive-behavioral strategies. Parents were trained to overcome obstacles to the resumption of normal school and social activities. Because an "illness identity" had already been established for many children, parents were taught to challenge sick-role beliefs while remaining supportive and compassionate. Parents were seen weekly ranging from sitting in on the entire session with their younger children, to having a 5-min review after their older child's treatment.

Study Goals

This study aims to describe the physical and psychological status of children with JPFS and their families. We expect a complex clinical picture that is complicated by family medical and psychiatric issues. Our CBT treatment goals are to enable children with fibromyalgia to (a) cope more effectively with musculoskeletal pain, (b) improve sleep quality, (c) decrease levels of disability, and (d) improve patient defined quality of life. The efficacy of our 8-week CBT program is evaluated using pre- and post-treatment measures completed by children and their parents. Based on our pilot data, we anticipate that CBT will improve children's physical and psychological symptoms.

Method
Participants

Sixty-seven children who met the Yunus and Masi (1985) criteria for JPFS and their parents were recruited from the Pediatric Rheumatology Clinic at Schneider Children's Hospital, NY, over the course of 24 months. According to this criteria, children must (a) report generalized musculoskeletal aching at three or more sites for 3 or more months in the absence of any underlying condition, (b) have a normal physical examination except for expressing pain at 5 of 11 tenderpoints and no pain at control points, (c) have normal laboratory findings, and (d) report 3 of 10 minor criteria for fibromyalgia (poor sleep, fatigue, chronic headaches, irritable bowel, numbness, subjective swelling, chronic anxiety or depression, modulation of pain by physical activity, weather, or stress).

Study participants ranged in age from 8 to 20 years (M = 13.9 years). As expected, there was a preponderance of females (88%). Fifty-two families (78%) were white, and 15 families (22%) were of other ethnicity (African American, Hispanic, or Asian). Using Hollingshead's criteria (1979), socioeconomic status (SES) of patients was middle class (M = 50.3). This skewed population (middle- to upper-class white females) is typical of clinical fibromyalgia populations (Anthony & Schanberg, 2001; Conte, Walco, & Kimura, 2003).

Procedures

All children diagnosed with juvenile fibromyalgia by our pediatric rheumatologists were referred to the Fibromyalgia Treatment Program. Written and verbal explanations of the research were given, and both parents and children signed informed consent/assent forms. Families who consented to participate in the 8-week intervention made an appointment for a psychological evaluation. A complete social, psychological, and medical history of the child was obtained, and families completed packets of standardized instruments measuring the child's current health and functional status, levels of pain and sleep disturbance, and psychological distress. To screen for underlying psychiatric illness, we administered a semistructured diagnostic interview, Schedule for Affective Disorders and Schizophrenia for School-Age Children (Chambers et al., 1985), to parents and children separately. Two children were referred for psychiatric treatment and excluded from further study (one child had schizoaffective disorder and one child had depression with suicide ideation). After this initial medical and psychological evaluation, children began the 8 weeks of CBT treatment, described previously. The therapeutic team met weekly to discuss individual cases and evaluate treatment progress. This procedure was reviewed and approved by the hospital Institutional Review Board.

Measures

Parents reported family demographic information, medical and psychiatric history, and current life stressors. Measures of the mother's current psychological distress were obtained using the Brief Symptom Inventory (BSI). This 52-item self-report instrument is adapted from the Symptom Checklist-90, R measures nine primary psychiatric symptoms (Derogatis, 1983).

Measures of the child's physical status and psychological functioning were administered before the initial psychological evaluation and immediately following the 8-week CBT. Data regarding aspects of the child's physical functioning (perceived pain, sleep quality, fatigue, functional disability, and somatic symptoms) were primarily self-reported. The severity of children's pain was assessed using the VAS of the Pediatric Pain Questionnaire

(Varni et al., 1987). This scale is a 10-cm horizontal VAS anchored by the phrases "no pain" and "pain as bad as it could be." Children were instructed to rate their current level of pain. Higher scores indicated greater pain.

Children's sleep quality was assessed using the Pittsburgh Sleep Quality Index (PSQI) (Buysse, Reynolds, Monk, Berman, & Kupfer, 1989). This self-report measure of sleep disturbance and quality produces seven component scores and a composite global sleep quality score. A score greater than five is indicative of sleep problems. This latter global score was used in data analysis. Children reported their level of fatigue using the FSS. This nine-item Likert scale measured the disabling qualities of fatigue. Higher scores indicated greater levels of fatigue. Validity studies have demonstrated that the FSS is able to identify aspects of fatigue that are unique to chronic illness (Krupp, LaRocca, Muir-Nash, & Steinberg, 1989)[1]. Degree of functional impairment was assessed using the Functional Disability Inventory (FDI). This 15-item scale measured the difficulties experienced in physical or psychosocial functioning due to poor health. Good internal consistency and construct validity have been demonstrated for use of the scale with chronically ill children (Walker & Green, 1991).

Data pertaining to the child's psychological functioning were reported from two sources; the child's self-report of anxiety and somatic symptoms and the parent's report of any internalizing, that is, withdrawn, depressive behaviors. Children reported symptoms of anxiety using the Multidimensional Anxiety Scale (MASC). The 39 items of the MASC utilize a 4-point Likert scale ranging from "never true of me" to "always true of me." The total T-score (above 65 is considered clinically significant) was used in data analyses (March, Parker, Sullivan, Stallings, & Conners, 1997). The number and extent of somatic symptoms was measured by the Children's Somatization Inventory (CSI). Children rated the extent to which they experienced 35 somatic symptoms on a 4-point scale ranging from "not at all" to "a whole lot." Higher scores indicated more somatizing. Parents reported on the social competencies and behavioral problems of their child using the Child Behavior Checklist (Achenbach, 1991). Profiles of internalizing (withdrawn, depressed, or anxious) behaviors were used in the data analysis. Children also reported on aspects of their quality of life and ability to cope with fibromyalgia using the Satisfaction with Abilities and Well-Being

---

[1]Although the FSS and the PSQI have not been validated for children, they are used extensively to assess the sleep quality of children and adolescents referred to the sleep disorder clinic affiliated with our hospital.

Scale (SAWS, Katz, & Alfieri, 1997). This scale has been used in studies of families where the child has a rheumatic condition (Degotardi, Revenson, & Ilowite, 1999) and is a global measure of health-related quality of life. Higher scores are associated with improved quality of life.

Data Analysis

Descriptive statistics confirmed the normal distribution of data. Given the small proportion of males in the sample and the wide age range, a series of analysis of covariance (ANCOVA) were also conducted. These analyses confirmed that there were no gender or age effects, that is, treatment outcomes did not vary according to the age of the child nor were there differential responses of males and females. Therefore, males were retained in all data analyses, and age was treated as a continuous variable. In the following sections, children's physical characteristics relevant to their fibromyalgia are reported. Family history of medical and psychiatric problems is described. To ensure that children who completed treatment and post-test questionnaires did not differ in significant ways from those who did not complete the intervention, chi-square analyses and t-tests were conducted. Next, differences on pre- and postintervention measures of physical and psychological symptoms were explored using paired t-tests. As multiple comparisons were conducted, a Bonferroni correction was used, and only p values less than .006 were reported as significant.

Results
Child and Family Characteristics

At the time of initial evaluation, all children had a minimum of five tenderpoints ($M = 7.5$; range = 5–13) and reported widespread pain ($M = 5.0$, on a VAS of 0–10). Few children (12%) reported they were currently experiencing "no pain." Almost 90% of the sample reported sleep difficulties ($M = 10.1$ on the PSQI), especially difficulty in falling asleep. Parents also reported that their child frequently experienced headaches (67%), severe fatigue (52%), and gastric problems (51%). Eleven children (16%) had a concurrent rheumatologic condition: juvenile rheumatoid arthritis (9%), systemic lupus erythematosus (4%), and raynauds (3%). Children had missed an average of 2.9 days of school during the past month because of illness, and two children had been receiving home tutoring for over 6 months.

Families reported an average of 1.7 current life events in the past year, such as death in the family, divorce of parents, or change of school. Family history was notable for 58% reporting chronic medical

problems, and of these 27% reported a history of rheumatic disease. Additionally, almost a third of the sample reported a family history of psychiatric problems. Mother's reported a high level of psychiatric symptoms on the BSI (Derogatis, 1983). Between 16.7 and 36.7% of mothers exceeded clinically significant cutoffs (T-scores >65) on at least one of the nine symptoms. Most commonly, mothers reported obsessive-compulsive symptoms, anxiety, and paranoid ideation.

Of the 67 children referred for CBT, 44 (66%) completed the entire 8 weeks of treatment protocol and the post-treatment questionnaires. Reasons for attrition included scheduling conflicts (9%), problems with insurance coverage (7%), dissatisfaction with the program (7%), accessibility to the clinic (4%), failure to return posttreatment questionnaires (4%), and need for psychiatric referral (3%). Chi-square analyses and t-tests were used to compare children who completed treatment with those did not complete all aspects of the program. Means for demographic (age, gender, ethnicity, and SES) and disease status variables (tenderpoint count, reported pain, fatigues severity, functional disability, and family history of rheumatic disease and/or psychiatric problems) are summarized in Table I. There were no significant differences found on any of these variables between those who completed CBT and those who did not.

Physical Symptoms

Using paired t-tests, significant differences ($p < .006$) were found between pre and post-intervention measures of *all* physical symptoms as summarized in Table II. Post CBT, children's subjective levels of pain and fatigue decreased as measured by the "pain now" VAS variable and the total FSS score. Additionally, there was an increase from 12 to 24% in the number of children who reported they were currently experiencing "no pain." Children also reported significantly improved sleep quality as measured by the global score on the PSQI. Overall, children's scores on the FDI significantly decreased following the 8-week intervention. Following treatment, all children had returned to school full time, and the average days missed each month had dropped from 2.9 to .8, $t(25) = 2.23$, $p < .05$.

Psychological Symptoms

Again, using paired t-tests, significant differences ($p < .006$) were found between pre and post-intervention measures of *all* psychological symptoms as summarized in Table II. Post CBT, children's self-reported anxiety significantly decreased as measured by the MASC. Children also reported a reduction in the number and severity of somatic symptoms as measured by the CSI. Additionally, parent's report of their child's internalizing behaviors also decreased as measured by the Child Behavior Checklist. Overall, children reported an improvement in their quality of life as shown by the significant increase in scores on the SAWS.

Discussion

This study tested the efficacy of a manualized CBT for the treatment of JPFS. Our pilot data and findings in the

Table I. Comparison of Demographic and Disease Variables for Children who Completed 8 Weeks of Cognitive-Behavioral Treatment and Children who did not Complete Treatment

	Total sample (N = 67)	Completed cognitive-behavioral treatment (N = 44)	Noncompleters (N = 23)	Group difference
Child's characteristics				
Age	13.9 (SD = 2.8)	14.1 (2.8)	13.7 (2.7)	$t(65) = -0.45$, $p = .65$
Gender				
Female	59 (88.1%)	39 (88.6%)	20 (86.9%)	$\chi^2(1) = 0.84$, $p = .56$
Male	8 (11.9%)	5 (11.4%)	3 (13.1%)	
Tenderpoint count	7.5	7.7	7.0	$t(58) = -1.4$, $p = .16$
Reported pain	4.9	5.0	4.6	$t(65) = -.49$, $p = .62$
Fatigue severity	35.7	34.5	37.9	$t(65) = .96$, $p = .34$
Functional disability	16.4	15.5	18.2	$t(65) = 1.1$, $p = .29$
Family characteristics				
Ethnicity				
White	52 (77.6%)	38 (86.4%)	18 (81.8%)	$\chi^2(1) = .63$, $p = .44$
Other	15 (22%)	6 (13.6%)	4 (18.2%)	
Socioeconomic status	50.3	50.1	50.6	$t(65) = .21$, $p = .84$
Rheumatic history	18 (26.9%)	11 (26%)	7 (31.8%)	$\chi^2(1) = .63$, $p = .77$
Psychiatric history	21 (32.3%)	13 (31.7%)	8 (36%)	$\chi^2(1) = .71$, $p = .78$

Table II. Paired Comparisons of Physical and Psychological Status Pre- and Post-Cognitive–Behavioral Treatment Intervention for Children who Completed Treatment (*N* = 44)

Measures	Pretreatment [*M* (*SD*)]	Posttreatment [*M* (*SD*)]	Paired comparisons
Physical symptoms			
Pain now (10-cm Visual Analogue Scale)	5.0 (3.2)	2.6 (2.8)	$t(43) = 4.9, p = .000$
Global Pittsburgh Sleep Quality Index sleep score	10.1 (4.5)	5.6 (3.7)	$t(41) = 6.4, p = .000$
Fatigue Severity Scale	34.3 (14.0)	28.2 (13.2)	$t(42) = 3.1, p = .003$
Functional Disability Inventory	15.6 (9.4)	8.2 (8.3)	$t(43) = 5.1, p = .000$
Psychological symptoms			
Multidimensional Anxiety Scale	51.5 (12.0)	46.0 (10.8)	$t(39) = 3.2, p = .003$
Children's Somatization Inventory	37.1 (15.1)	24.9 (15.9)	$t(42) = 5.3, p = .000$
Internalizing scale (Child Behavior Checklist)	57.9 (8.9)	52.1 (10.7)	$t(34) = 4.0, p = .000$
Quality of life (Satisfaction with Abilities and Well-Being Scale)	43.0 (8.9)	49.2 (12.0)	$t(43) = -2.9, p = .005$

adult literature (Bradley, 1989; Rossy et al., 1999; Turk et al., 1998) led us to expect that the intervention would be successful and, indeed, we saw statistically and clinically significant positive change across the board. The self-reported improvement in physical and psychological functioning was corroborated by our clinical observations where we saw improvement in children's ability to cope with pain and manage symptoms. The dramatic improvement in somatic symptoms is particularly notable. Children not only reported fewer symptoms, but during clinical interviews they also reported feeling more in control of their fibromyalgia after completing CBT. Additionally, parents reported that children were complaining less about pain and had resumed normal school and social activities.

As the presence of a chronic illness can cause children to swerve off their developmental course, quick and efficient treatment is necessary, particularly for JPFS, a diagnosis fraught with ambiguity. When treatment is recommended to patients with JPFS and their families, it is often multidisciplinary treatment programs which, as Anthony and Schanberg (2001) indicate, are often difficult to coordinate, expensive, and unpalatable for families. Indeed, many families in our study had received similar advice from their primary care physicians and were unhappy with this approach. Because we treated only those patients receiving a diagnosis of JPFS at our tertiary care center, a biopsychosocial explanation for the condition was given at that first contact point and successfully set the stage for our CBT. Our therapeutic team worked with insurance companies and streamlined the process for families so that weekly, after-school visits were arranged with one professional. During the initial psychoeducation session, we lowered family's expectations for immediate change while promoting optimism for a good overall outcome. Weekly parent meetings to anticipate and address obstacles to treatment may also have optimized outcome.

Although 24% of our sample reported they were currently pain free, for most children the physical symptoms of JPFS—the pain, fatigue, sleep disturbance, sleep latency, and the other related physical symptoms (including headaches and gastric disturbances)—were reduced following CBT but often did not remit completely. Our treatment instilled optimism for a better quality of life without promising the "magic wand," that is, the absence of symptoms. Instead, children were taught strategies that empowered them to assume control over symptom management. As there are no laboratory markers to define presence, absence, remission, or exacerbation of JPFS, there is no way to medically verify children's report of symptom abatement. However, we suspect that a gradual shedding of illness identity portends for a better adjustment and adoption of a healthy role within the family dynamic.

Essential to children's perception of treatment success, "pain now" scores fell significantly with all children reporting that the pain could be "handled" with minimal interference to normal social and school functioning. Sleep latency and sleep quality were also judged to be significantly improved with children reporting less anxiety associated with delayed sleep and worries about tiredness the next day. A review of children's daily sleep diaries depicts a pattern of progressive improvement over the course of treatment. Fatigue, severe and disabling at first, was judged by children to no longer inhibit their participation in activities, which improved quality of life significantly. Overall, children's reports of general somatic complaints and their scores on anxiety scales lessened. Children's perceptions of their abilities and general well-being all significantly improved both statistically and in clinical report. Parent reports corroborated

these effects. If we consider JPFS to be the "final common pathway" of complex biopsychosocial mechanisms (Kashikar-Zuck et al., 2000), then intervening in multiple domains seems most appropriate as "psychological" changes may be as important as physical improvement in affecting outcome.

This study design does not allow us to link specific parts of the intervention with specific outcomes either in this study or in adult studies that preceded it. We can conjecture that because this treatment addresses many of the putative components and associated symptoms of JPFS, the individual child's relief in his or her perceived area of need mitigates other symptom areas. It is acknowledged that aspects of psychological adjustment may be a risk factor for development of a pain syndrome in childhood (Conte et al., 2003), and it is widely reported in the pediatric literature that psychosocial characteristics help to predict levels of adjustment. Adults with fibromyalgia have significantly higher lifetime prevalence for rates of mood and anxiety disorders and somatic complaints (Epstein et al., 1999; Walker et al., 1997). In children with "widespread pain disorders," Mikkelsson et al. (1997) reported higher total emotional and behavioral scores. It has also been reported that the parents of children with JPFS present with complex pain histories (Schanberg et al., 1997), and we saw this in our study as well. The way in which parents cope with their own pain appears to be instrumentally important in determining the coping styles their children develop. Studies that can begin to untangle that relationship would be helpful in both the prevention and treatment of pain disorders in children. Our study establishes a surface congruence between parental and child pain behavior, based on clinical measurements for both pain and psychological adjustment. Such a direct relationship replicates adult interventions studies and validates the utility of a primary psychological treatment modality for JPFS.

Limitations to our study include potential self-selection bias and our attrition rate (34%), and this may have compromised external validity. We were unable to control for potential self-selection bias, as only families who expressed a willingness to make behavioral changes and attend eight treatment sessions enrolled in the study. Zebracki et al. (2003) addressed the problem of high attrition rate (up to 60%) in pediatric intervention studies and operationalized types of attrition based on the point in time that a patient drops out. We did not systematically track the differences among families who rejected our services either before or after the psychological evaluation, compared with those who dropped out mid-treatment. However, our data show that we did not only analyze potential good responders as most attrition could be attributed to logistical issues, such as insurance or scheduling problems, but also loss of interest in the program. Indeed, several families discontinued treatment as they felt their child's symptoms had improved and the inconvenience of weekly therapy was burdensome. In our sample, those who completed treatment did not significantly differ from the noncompleters in the areas we reviewed, but there may have been relevant areas that we overlooked. For example, we did not look at the role of the age of caregiver, and Zebracki et al. (2003) found younger caregivers are associated with higher attrition rates. Important subgroupings of children with JPFS might emerge from such analyses with possibly different treatment implications.

Additionally, we did not randomize to control groups as our pilot data indicated that "attention control" was ineffective and ethically questionable. Our study design restricted us from addressing the relative efficacy of different treatment modules (e.g., the primacy of the pain management compared with sleep hygiene in improving symptoms) and controlling for the passage of the time. As Mikkelsson (1999) and Buskila et al. (1995) point out, fibromyalgia in children can be of shorter duration than in adults. Where in the "natural course" of the illness, the child began our intervention might be an important predictor of symptom abatement and willingness to engage in treatment. This would be meaningful information for treatment as well. Although it is possible that either regression to the mean or the shorter duration of fibromyalgia in children could account for our findings, the robustness of improvements (both physical and psychological) suggests that our intervention is effective. Follow-up data would best answer outstanding issues concerning the role of our intervention on the natural course of JPFS.

For patients, families and physicians alike, dealing with JPFS is a frustrating process characterized by extensive medical workups and trials of ineffective treatment. Although fibromyalgia as a condition has been mired in controversy, the reality is that these children account for a significant portion of rheumatology practice. To date, there are no empirically validated treatments for JPFS. For families who are willing to engage in a comprehensive treatment that integrates the techniques of several disciplines, CBT is a promising cost-effective intervention to enable children and adolescents with JPFS to return to productive role functioning.

Acknowledgments

References

Achenbach, T. (1991). *Manual for the child behavior checklist/4–18 and 1991 profile*. Burlington: University of Vermont.

Anthony, K. K., & Schanberg, L. E. (2001). Juvenile primary fibromyalgia syndrome. *Current Rheumatology Research*, *3*, 165–171.

Arthritis Foundation. (2002). *The Arthritis foundation's guide to good living with fibromyalgia*. Atlanta, GA: Arthritis Foundation.

Bradley, L. (1989). Cognitive behavioral therapy for primary fibromyalgia. *Journal of Rheumatology*, *16*, 131–136.

Breau, L. M., McGrath, P. J., & Ju, L. H. (1999). Review of juvenile primary fibromyalgia and chronic fatigue syndrome. *Journal of Developmental Behavioral Pediatrics*, *20*, 278–288.

Buskila, D., Neumann, L., Hershman, E., Gedalia, A., Press, J., & Sukenik, S. (1995). Fibromyalgia syndrome in children: Outcome study. *Journal of Rheumatology*, *22*, 525–528.

Buysse, D. J., Reynolds, C. F., Monk, T. H., Berman, S. R., & Kupfer, D. J. (1989). The Pittsburgh Sleep Quality Index: A new instrument for psychiatric practice and research. *Psychiatry Research*, *28*, 193–213.

Chambers, W. J., Puig-Antich, J., Hirsh, M., Paez, P., Ambrosini, P. J., Tabrizi, M. A., et al. (1985). The assessment of affective disorders in children and adolescent by semi-structured interview. *Archives General Psychiatry*, *42*, 89–98.

Conte, P. M., Walco, G. A., & Kimura, Y. (2003). Temperament and stress response in children with juvenile primary fibromyalgia syndrome. *Arthritis and Rheumatism*, *48*, 2923–2930.

Crofford, L. J., & Clauw, D. J. (2002). Fibromyalgia: Where are we a decade after the American College of Rheumatology classification criteria were developed? *Arthritis and Rheumatism*, *46*, 1136–1138.

Degotardi, P. J., Revenson, T. A., & Ilowite, N. T. (1999). Family-level coping in juvenile rheumatoid arthritis: Assessing the utility of a quantitative family interview. *Arthritis Care and Research*, *12*, 314–324.

Derogatis, L. R. (1983). *Brief symptom inventory (BSI): Administration, scoring and procedures manual* (3rd ed.). Minneapolis, MN: National Computer Systems.

Drotar, D. (1997). Intervention research: Pushing back the frontiers of pediatric psychology. *Journal of Pediatric Psychology*, *22*, 593–606.

Fox, D. G., Degotardi, P. J., Klass, E. S., Rosenberg, B., Rothenberg, S., & Gottlieb, B. S. (1999). Sleep in children with fibromyalgia [Abstract]. *Arthritis Care and Research*, *12*, S14.

Gedalia, A., Garcia, C. O., Molina, J. F., Bradford, N. J., & Espinozo, L. R. (2000). Fibromyalgia syndrome: Experience in a pediatric rheumatology clinic. *Clinical and Experimental Rheumatology*, *18*, 415–419.

Hollingshead, A. B. (1979). *Four factor index of social status*. Unpublished manuscript.

Kashikar-Zuck, S., Graham, T. B., Huenefeld, M. D., & Powers, S. W. (2000). A review of biobehavioral research in juvenile primary fibromyalgia syndrome. *Arthritis Care and Research*, *13*, 388–397.

Katz, P. P., & Alfieri, W. S. (1997). Satisfaction with abilities and well-being: Development and validation of a questionnaire for use among persons with rheumatoid arthritis. *Arthritis Care and Research*, *10*, 89–98.

Kimura, Y. (2000). Fibromyaglia syndrome in children and adolescents. *Journal of Musculoskeletal Medicine*, *17*, 142–158.

Klass, E. S., Degotardi, P. J., Fox, D. G., Rosenberg, B., & Ilowite, N. T. (1998). Developing a multi-modal assessment and treatment protocol for children with fibromyalgia [Abstract]. *Arthritis and Rheumatism*, *41*, S249.

Krupp, L. B., LaRocca, N. G., Muir-Nash, J., & Steinberg, A. D. (1989). The fatigue severity scale: Application to patients with multiple sclerosis and systemic lupus erythematosus. *Archives of Neurology*, *46*, 1121–1123.

Manon, L. W., Goolkasian, P., & McCain, G. A. (1998). Evaluation of a multimodal treatment program for fibromyalgia. *Journal of Behavioral Medicine*, *21*, 163–178.

March, J. S., Parker, J., Sullivan, K., Stallings, P., & Conners, K. (1997). The Multidimensional Anxiety Scale for Children (MASC): Factor structure, reliability, and validity. *Journal of American Academy of Child and Adolescent Psychiatry*, *36*, 554–565.

Masi, A. T., & Yunus, M. B. (1990). Fibromyalgia – which is the best treatment? A personalized, comprehensive, ambulatory, patient-involved management program. *Bailliere's Clinical Rheumatology*, *4*, 333–370.

Mikkelsson, M. (1999). One year outcome of preadolescents with fibromyalgia. *Journal of Rheumatology*, *26*, 674–682.

Mikkelsson, M., Sournader, A., Piha, J., & Salminen, J. J. (1997). Psychiatric symptoms in preadolescents with musculoskeletal pain and fibromyalgia. *Pediatrics*, *100*, 220–227.

Rapoff, M. A. (1999). *Adherence to pediatric medical regimens*. New York: Plenum.

Roizenblatt, S., Tufik, S., Goldenberg, J., Pinto, L. R., Hilario, M. O. E., & Feldman. D. (1997). Juvenile fibromyalgia: Clinical and polysomnographic aspects. *Journal of Rheumatology*, *24*, 579–585.

Romano, T. J. (1991). Fibromyalgia in children: Diagnosis and treatment. *West Virginia Medical Journal*, *87*, 112–114.

Rosenberg, A. M. (1990). Analysis of a pediatric rheumatology clinic population. *Journal of Rheumatology*, *17*, 827–830.

Rossy, L. A., Buckelew, S. P., Dorr, N., Hafflund, K. J., Thayer, J. F., McIntosh, M. J., et al. (1999). A meta-analysis of fibromyalgia treatment interventions. *Annals of Behavioral Medicine*, *21*, 180–191.

Saylor, C. F., Finch, A. J., Spirito, A., & Bennett, B. (1995). The Children's Depression Inventory: A systematic evaluation of psychometric properties. *Journal of Consulting Clinical Psychiatry*, *52*, 955–967.

Schanberg, L. E., Keefe, F. J., Lefebvre, J. C., Kredich, D. W., & Gil, K. M. (1996). Pain coping strategies in children with juvenile primary fibromyalgia syndrome: Correlation with pain, physical function, and psychological distress. *Arthritis Care and Research*, *9*, 89–96.

Schanberg, L. E., Keefe, F. J., Lefebvre, J. C., Kredich, D. W., & Gil, K. M. (1998). Social context of pain in children with juvenile primary fibromyalgia syndrome: Parental pain history and family environment. *Clinical Journal of Pain*, *14*, 107–115.

Siegal, D. M., Janeway, D., & Baum, J. (1998). Fibromyalgia syndrome in children and adolescents: Clinical features at presentation and follow-up. *Pediatrics*, *101*, 377–382.

Tayag-Kier, C. E., Keenan, G. F., Sclazi, L. V., Schultz, B., Elliot, J., Zhao, H., et al. (2000). Sleep and periodic limb movement in sleep in juvenile fibromyalgia. *Pediatrics*, *106*, 70–75.

Turk, D. C., Okifuji, A., Sinclair, D., & Starz, T. W. (1998). Interdisciplinary treatment for fibromyalgia syndrome: Clinical and statistical significance. *Arthritis Care and Research*, *11*, 186–195.

Vandvik, I. H., & Foresth, K. O. (1994). A bio-psycho-social evaluation of ten adolescents with fibromyalgia. *Acta Paediatrics*, *83*, 766–771.

Varni, J. W., & Bernstein, B. H. (1991). Evaluation and management of pain in children with rheumatic diseases. *Rheumatic Disease North America*, *17*, 985–1000.

Varni, J. W., Thompson, K. L., & Hanson, V. (1987). The Varni/Thompson Pediatric Pain Questionnaire: Chronic musculoskeletal pain in juvenile rheumatoid arthritis. *Pain*, *28*, 27–28.

Walco, G. A., & Ilowite, N. T. (1992). Cognitive behavioral intervention in juvenile primary fibromyalgia syndrome. *Journal of Rheumatology*, *19*, 1617–1619.

Weinblatt, M. E. (2002). The best of times, the worst of times: Rheumatology 2001. *Arthritis and Rheumatism*, *46*, 567–573.

Yunus, M. B., & Masi, A. T. (1985). Juvenile primary fibromyalgia syndrome: A clinical study of thirty-three patients and matched normal controls. *Arthritis and Rheumatism*, *28*, 138–145.

Zebracki, K., Drotar, D., Kirchner, L., Schluchter, M., Redline, S., Kercsmar, C., et al. (2003). Predicting attrition in a pediatric asthma intervention study. *Journal of Pediatric Psychology*, *28*, 519–528.