The Treatment of Juvenile Fibromyalgia with an Intensive Physical and Psychosocial Program

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Objective To assess the short-term and 1-year outcomes of children with fibromyalgia treated with intensive physical and occupational therapy (PT/OT) and psychotherapy.

Study design Children with fibromyalgia seen at a tertiary care hospital were treated with 5-6 hours of intensive PT/OT daily and at least 4 hours of psychosocial services weekly. All medications used for fibromyalgia were discontinued. Children underwent standardized testing, including a visual analog scale for pain; the Bruininks-Oseretsky Test of Motor Performance, Second Edition; the Bruce treadmill protocol; the Functional Disability Inventory; the Pain Stages of Change Questionnaire, adolescent version; and the Pediatric Quality of Life Inventory, Teen Report, at 3 time points: at program entry, at the end of the intensive program, and 1 year after the end of the program.

Results Sixty-four children (median age, 16 years; 95% Caucasian; 94% female; median duration of symptoms, 21 months) were studied. The mean pain score decreased significantly from program entry to the end of the program (from 66 of 100 to 25 of 100; $P = .001$). At the 1-year follow-up, 33% reported no pain. All measures of function on the Bruininks-Oseretsky Test of Motor Performance, Second Edition improved significantly and remained at that level or continued to improve over the subsequent year. The mean Bruce treadmill protocol time first increased from 588 seconds to 801 seconds ($P < .001$) and then dropped to 750 seconds ($P = .005$), which is at the 90th percentile for age and sex. All Pain Stages of Change Questionnaire, adolescent version subset scores improved significantly initially and were stable or improved at 1 year, as did the Pediatric Quality of Life Inventory, Teen Report total score.

Conclusion Children with fibromyalgia can be successfully treated without medications with a very intensive PT/OT and psychotherapy program. They have significantly improved pain and function by subject report and objective measures of function. *(J Pediatr 2015;167:731-7)*.

Fibromyalgia is one of the most common amplified pain syndromes in children, occurring in 2%-6% of the pediatric population.[1-5] It is defined as widespread pain lasting at least 3 months and, depending on criteria, associated with between 5 and 11 of 18 trigger points, along with other somatic complaints, such as irritable bowel syndrome, fatigue, unrestorative sleep, and chronic headache.[6,7] Girls predominate at a rate of approximately 4:1, and the disorder seems to affect Caucasians disproportionately.[8] The etiology is unknown, but some of the factors associated with adults with fibromyalgia include depression,[5,9] low pain threshold,[2] cortisol dysregulation,[10,11] and ischemia.[12-14] Adult fibromyalgia criteria are applied in diagnosing these children, and as such other potential etiologies need to be ruled out before a diagnosis can be made. No criteria have been established for the diagnosis of fibromyalgia in children.

Treatment of childhood fibromyalgia has remained elusive, with the major focus on cognitive behavioral therapy[15-17] and aerobic training.[18,19] Studies of long-term outcomes have reported persistent pain in more than 90% of affected children, and sleep disturbance was found in more than 90% of 33 children with fibromyalgia surveyed 2.6 years after diagnosis.[20,21] In a large cohort of children with fibromyalgia, more than 80% had persistent symptoms into adulthood, and, compared with controls, had more pain, anxiety, and medical visits, along with decreased physical function at 5.9 years after diagnosis.[22] In adults, evidence-based guidelines stress cognitive behavioral therapy and aerobic training.[23] Medications have little role in treating children, and systematic reviews in adults are not encouraging.[24-27]

We have had short- and long-term success in treating children with complex regional pain syndrome with a very intensive physical and occupational therapy (PT/OT) program along with psychological counseling,[28] which has been replicated by others.[29,30] Although complex regional pain syndrome is a different pain diagnosis, children with this form of amplified pain are also treated in our program,

BOT-2	Bruininks-Oseretsky Test of Motor Performance, Second Edition
FDI	Functional Disability Inventory
PedsQL	Pediatric Quality of Life Inventory
PSOCQ-A	Pain Stages of Change Questionnaire, adolescent version
PT/OT	Physical and occupational therapy
VAS	Visual analog scale

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and we have studied them previously. We also have reported short-term functional outcomes in a small group of children with fibromyalgia who underwent sleep studies before and after participating in the intensive PT/OT program with excellent results.[18]

The objective of the present study was to compare a cohort of patients with fibromyalgia on various objective and subjective measures at 3 time points: at admission to our program, discharge from the program, and 1 year after completion of the program.

Methods

The study protocol was reviewed and approved by our hospital's Committee for the Protection of Human Subjects. All parents provided consent, and subjects provided assent before study participation. The source population for the study was children and adolescents aged 13-18 years with a diagnosis of primary juvenile fibromyalgia treated in the inpatient or day hospital amplified musculoskeletal pain program between September 2008 and May 2011. The subjects were a convenience sample enrolled without regard to pain duration, pain severity, or previous therapy. Children for whom this was not the initial participation in the amplified musculoskeletal pain program, as well as those whom required modification of the program because of another medical condition, such as cerebral palsy or fragile bones, were excluded. One patient who consented to participate was discharged from the program before completing any therapy owing to high-level contact precautions (methicillin-resistant *Staphylococcus aureus* carrier). All subjects fulfilled the American College of Rheumatology's 2010 fibromyalgia criteria.[31]

Intervention

Before participating in our program, the children were encouraged to do aerobics and, if they had allodynia, to desensitize. We frequently prescribed a formal home exercise program and local physical therapy, although adherence was not formally measured. All pain medications and medications given for fibromyalgia, such as analgesics, antiepileptics, antidepressants, and sleep medications, were discontinued. If the home exercise program was not successful, then the dose of PT/OT was increased. It is these children who are included in this report.

The children were treated either as day hospital patients or inpatients. All children received individualized 1-on-1 therapy for 5-6 hours a day, with the focus on quickly reestablishing normal function, along with maximizing aerobic conditioning. Activities typically included timed activities (eg, animal walks, stepping in/out of a tub, running up and down stairs, stepping and squatting activity), scooter boards, treadmill, elliptical, stairs, long-distance community ambulation, strengthening and endurance activities, and dance or other video game activities. Treatment goals were set high and quickly advanced as the child progressed through the requirements to a higher level of function and exercise. Children with allodynia received multiple courses of desensitization, including rubbing, local and total body vibration, constant light touch or compression, temperature and noise desensitization, fanning, and exposure to multiple different textures. Desensitization was often incorporated into exercises when possible. Children who experienced pain with eating were often required to eat a minimum of 7 meals and snacks per day.

The duration of therapy for each child was individually determined by the treatment team based on physical functioning goals obtained, rate of improvement, and judgment regarding the child's ability to sustain and further improve on these functional goals in the home environment without formal physical therapy.

Psychosocial support included both 1-on-1 and group sessions with a psychologist for both cognitive and behavioral therapy-based intervention, as well as support for coping during PT/OT sessions, as indicated. Art therapy and music therapy were also included, for a minimum of 4 hours per week of psychosocial support. In addition, parent group sessions were held weekly, and family or parent sessions were added when indicated.

Objective

The objective of this study was to evaluate long-term functional and psychosocial outcomes of the patients completing our intensive program.

Outcome Measures

Bruce Treadmill Protocol[32]: This test consists of walking on a treadmill until the subject is unable to continue walking or running owing to exhaustion or pain. The test increases in speed and incline every 3 minutes until the subject cannot continue or he or she completes the test time of 21 minutes. The patient is allowed to hold the handrail(s) of the treadmill if he or she chooses. The test results can be compared with age- and sex-matched norms.

Bruininks-Oseretsky Test of Motor Performance, Second Edition (BOT-2)[33]: This test measures gross and fine motor function, as well as balance and coordination. It consists of 8 subtests: fine motor control, fine motor integration, manual dexterity, bilateral coordination, balance, running speed and agility, upper limb coordination, and strength. The scores on these subtests are summed to calculate a total composite score, which is then interpreted based on age- and sex-matched norms. For each subtest, higher scores represent better performance.

Functional Disability Inventory (FDI)[34]: This self-report measure asks the subject to rate how much physical "trouble" he or she experiences related to pain when attempting to complete various functional activities. Responses include "no trouble," "a little trouble," "some trouble," "a lot of trouble," and "impossible." These ratings are given a number equivalent. These numbers are summed to arrive at a final score ranging from 0 to 60,