Fibromyalgia syndrome: Experience in a pediatric rheumatology clinic

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Abstract

Objective

To report our experience of fibromyalgia syndrome (FMS) in pediatric rheumatology clinic settings.

Methods

Clinical and laboratory data were reviewed in all patients with FMS between March 1992 and March 1996. Patients with FMS and an underlying rheumatic disease were excluded from the study. At presentation and follow-up visits, all patients had a tender points (TP) count that was conducted by thumb palpation. Both the children and their parents were questioned concerning the presence of widespread pain or aching. All the patients fulfilled the ACR criteria for the diagnosis of primary FMS. All children were evaluated by a protocol that included relevant information on FMS. Telephone survey questionnaires were used for patients who missed some of their follow-up visits.

Results

There were 59 children (47 F and 12 M) diagnosed with primary FMS. The mean age at onset was 13.7 years, and the mean age at diagnosis was 15.5 years. The mean duration of follow-up was 18.3 months. Diffuse aching was reported in 57 patients (97%), headaches in 45 (76%), and sleep disturbances in 41 (69%). Less common were stiffness in 17 (29%), subjective joint swelling in 14 (24%), fatigue in 12 (20%), abdominal pain in 10 (17%), and joint hypermobility and depression in 8 (14%) and 4 (7%) patients, respectively. The mean ESR was 15 mm/h, RF was negative in all patients, and ANA was positive (mean titer 1:160) in 17 patients. The mean initial TP count was 14.6. Nine patients were not available for follow-up. There were 50 patients available for follow-up and survey analysis, and of these 30 (60%) had improved, while 18 (36%) remained unchanged, and 2 (4%) became worse when compared with initial presentation. At the end of study follow-up, 37 patients (74%) were still taking medication (20 of them daily). Out of 25 patients whose TP counts were available at the end of follow-up, the mean TP dropped from 14.12 to 12.04 ($p = 0.09$) for the total group, and 14.05 to 10.84 ($p < 0.01$) for the patients who had improved. 22 out of 30 patients in the improved group and 7 out of 20 in the unchanged or worse group had continued active exercise programs ($p < 0.001$).

Conclusion

The clinical spectrum of FMS in children is similar to that of adults but with better outcomes. The TP count correlates with clinical status only in patients who had improved. Active exercise programs seem to correlate with better outcomes. Prospective and larger patient population studies, and a longer follow-up of children with FMS are needed to clarify these findings.

Key words

Fibromyalgia syndrome, joint hypermobility, tender points, antinuclear antibodies.

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Introduction

Fibromyalgia syndrome (FMS) is a well recognized chronic condition associated with diffuse aching, pain, or stiffness in the muscles or joints and tenderness on multiple tender points (TP) detectable on examination, often, accompanied by characteristic sleep disturbances (1, 2). This syndrome is now established as a recognizable clinical entity in adults and children (3). During the last decade, FMS has been extensively studied in adults, but reports on children in clinical settings are lacking. In the past we have assessed the prevalence and outcome of FMS in normal school children (4, 5). This retrospective study describes our experience (diagnosis, management and outcome) of FMS in a pediatric rheumatology clinic.

Patients and methods

Clinical and laboratory data were reviewed in all patients with FMS at the pediatric rheumatology clinic at Children's Hospital of New Orleans, between March 1992 and March 1996. There were 59 children (47 F and 12 M) diagnosed with primary FMS whose medical records data was available. Nine patients had moved without providing their new address or telephone number. These patients could not be reached despite repeated attempts and therefore were not available for follow-up. Informed consent was obtained from patients and their parents.

In all patients a count of 18 TP was conducted by thumb palpation. Both the children and their parents were questioned concerning the presence of widespread pain or aching. The patients were considered to have FMS if they fulfilled the currently accepted ACR criteria for the diagnosis of primary FMS, namely widespread pain in combination with tenderness of 11 or more of 18 specific TP sites (6). In addition, all children were evaluated by a protocol with relevant information on FMS besides diffuse aching and TP. This included headaches, sleep disturbances, morning stiffness with a duration of more than 15 minutes, subjective joint swelling, fatigue, abdominal pain, and a family history of FMS.

The quality of sleep was assessed by asking the patients to indicate the frequency with which they awakened tired or unrefreshed according to the answer list of 'always', 'usually', 'often', 'seldom', or 'never'. 'Always', 'usually', and 'often' were scored as a positive indication of sleep disturbances, while 'never', 'seldom' or any other replies were scored as a negative indication. A similar assessment was performed to determine other signs and symptoms (headaches, subjective joint swelling, fatigue, and abdominal pain). Patients were evaluated for the presence or absence of joint hypermobility, using the criteria of Carter and Wilkinson as modified by Bird *et al.* (7). Patients were considered to be depressed only if they were evaluated and managed by a psychiatrist or psychologist for depression.

In follow-up visits, patients and their parents were asked to define the status of their FMS symptoms as improved, unchanged or worse compared to that of their initial presentation. In addition, they were asked if they were still taking medication daily, occasionally, or not at all. Telephone survey questionnaires were used for 25 of the 59 patients who could not make it for some of their follow-up visits. Laboratory data was reviewed on all patients. Clinical assessment of FMS (including TP counts) and joint hypermobility was performed by the same observer (AG) who had conducted the initial examination as well as the examination in follow-up visits. Our physical therapist instructs the patients and their parents in at-home exercises, to be done for at least a half hour a day. In addition, they were encouraged to do low-impact exercises such as walking, swimming, biking, low-impact aerobics or stretching exercises.

Statistical analysis

Student's t-test and chi-square analysis were used for statistical analysis, using the 0.05 level of significance.

Results

There were 59 patients included in the study, with a mean age at onset of 13.7 years, and a mean age at diagnosis of 15.5 years. The mean duration of follow-up was 18.3 months. Forty-two (71%) of our patients were Caucasian; 15 were

African-American (25%); and 3 were Hispanic (4%) (Table I). A family history of FMS was found in 6 patients (10%); 5 mothers and 1 sister. Diffuse aching was reported in 57 patients (97%), headaches in 45 (76%), and sleep disturbances in 41 (69%). Less common were stiffness in 17 (29%), subjective joint swelling in 14 (24%), fatigue in 12 (20%), abdominal pain in 10 (17%), and joint hypermobility and depression in 8 (14%), and 4 (7%) patients, respectively (Table II). The mean initial TP count was 14.6. There were 25 patients with available TP counts at the end of follow-up (Table III), and of these, the mean TP dropped from 14.1 to 12.0 ($p = 0.09$) for the total group, and from 14.5 to 10.8 ($p < 0.01$) for the patients who had improved.

There were 50 patients available for follow-up and survey analysis, and of these 30 (60%) had improved while 18 (36%) remained unchanged and 2 (4%) worsened when compared with the initial presentation. Twenty-two out of 30 patients in the improved group and 7 out of 20 in the unchanged/worse group had continued active exercise programs ($p < 0.001$) (Table IV). All 50 patients received 5-20 mg of Flexeril (cyclobenzaprine) at bedtime beginning at the initial visit. During the follow-up period this medication was switched to 10-20 mg of Elavil (amitriptyline) in 11 patients, and to Prozac in 1 due to lack of response. In addition, 32 patients received NSAIDs (mostly naproxan) with or without acetaminophen as needed. Thirty-seven patients (74%) were still taking medications (20 of them daily) at the end of the study follow-up.

Laboratory tests

Complete blood count (CBC), antinuclear antibody (ANA), and rheumatoid factor (RF) evaluations were performed in all 59 patients at the initial visit. The erythrocyte sedimentation rate (ESR) was available in 42 patients. All patients showed a normal CBC and a mean ESR of 15 mm/h. RF was negative in all patients. The ANA test (HEp-2 cell substrate, titer > 1:40) was positive in 17 patients (29%) (mean titer 1:160). During the follow-up period, none of the patients with positive ANA developed any signs or symptoms of an underlying rheumatic disease, especially systemic lupus erythematosus (SLE). Prolactin serum levels were measured in 11 patients and found to be in the normal range in all of them (Table V).

Table I. Demographic and characteristics of 59 children with fibromyalgia syndrome.

Gender	Females	47
	Males	12
Ethnicity	Caucasians	42
	African-Americans	15
	Hispanics	2
Mean age at onset (range)		13.7 years (5-17)
Mean age at diagnosis (range)		15.5 years (7-19)
Mean follow-up (range)		18.3 months (3-65)

Table II. Clinical manifestations in 59 children with fibromyalgia syndrome.

Manifestations	No. of patients (%)
Generalized aches	57 (97%)
Headaches	45 (76%)
Sleep disturbances	41 (70%)
Stiffness	17 (30%)
Subjective joint swelling	14 (24%)
Fatigue	12 (20%)
Abdominal pain	10 (17%)
Joint hypermobility	8 (14%)
Depression	4 (7%)

Table III. Initial and final (last visit) tender point count in 25 children with fibromyalgia syndrome.

Condition	No. of pts.	Tender points initial visit	Tender points end of F/U	p value
Improved	19/25	14.5	10.8	< 0.01
Worse/ No change	6/25	14	15.4	0.32
Total	25/25	14.1	12.0	0.09

Table IV. Active exercise program and outcomes in 50 children with fibromyalgia syndrome.

Condition (n)	n with (+) Exercise program	n with (-) Exercise program	p value
Improved (30)	22	8	0.001
No change (18)	7	11	0.001
Worse (2)	0	2	-
Total (50)	29	21	-

Table V. Laboratory characteristics in 59 children with fibromyalgia syndrome.

Laboratory test	Mean (range)	Number of patients analyzed
Sedimentation rate	15 (3-48 mm/h)	42/59
ANA titer (+ in 17 patients)	1:160 (Neg-1:320)	59/59
Prolactin	normal	11/59
Rheumatoid factor	negative	59/59

Discussion

FMS is more common in young adult females (8). Although a study by Buskila *et al.* (4) found a frequency of 6.2% among Israeli schoolchildren, the prevalence of FMS in children is not well documented. According to a recent report (9), FMS is the third most frequent diagnosis among referrals to a pediatric rheumatology clinic. The mean age at onset in our patient population was 13.7

years, slightly higher than in the study by Siegal *et al*. (9) FMS is much more common in females; 80% of our patients were females and 20% males, in agreement with other adult and pediatric studies (9, 10). Although the familial occurrence of FMS has been reported with great frequency (11), we found a positive history of FMS in only 6 patients (10%), mostly mothers (5/6 patients). Referrals to our center are drawn from a population comprised of 60% African-Americans. However, 42 (71%) of our patients with FM were Caucasian and 15 were African-American (25%). These data suggest that FMS is significantly less common among African-American children. Although referral bias or cultural differences are the most important factors among the other possibilities that can explain these differences, we believe it is unlikely to be the case in our study.

We found that many of the clinical manifestations in our patients with FMS, especially diffuse aching, sleep disturbances, abdominal pain and depression, were similar in frequency to those of adult patients with FMS (6, 12). In our experience, children with FMS tend to have more headaches and are less stiff in the morning. In an earlier study in schoolchildren using validated criteria for joint hypermobility, we found that 13% of 338 children tested had joint hypermobility, and 6.2% had FMS (13). 81% of children with FMS in this study also had joint hypermobility, an association reported previously in adults (14). In the present study, which was carried out in a pediatric rheumatology clinic setting, we found that only 14% of the FMS patients had joint hypermobility, compared to Siegal *et al.* (9) who found that in a similar setting 40% of their FMS patients had joint hypermobility. The association of these two disorders is not fully understood; however, it can be speculated that peripheral microtrauma factors in joint hypermobility may trigger widespread pain and tenderness through a central pain mechanism with peripheral modulation (15). It is suggested by Siegal *et al.* (9) that pain experienced by patients with joint hypermobility might provoke sleep disturbances, and with time lead to FMS symptoms. In addition, we have suggested that FMS may overlap with other musculoskeletal syndromes in children (7). The mean initial TP count in our patients in the present study was 14.6 and dropped to 12.02 ($p = 0.09$) for the total group and to 10.84 ($p < 0.01$) in those patients who had improved. These data suggest that the TP count might serve as a useful monitoring tool in children with FMS. This study was a retrospective chart review and the authors concur with the possibility of referral bias; therefore the above data must be interpreted cautiously. Additionally, to date there is no data in the literature to suggest that the TP count is of value in monitoring patients with FMS.

The goal of current treatment in FMS is to ease pain, promote sleep and above all to help individuals cope with pain. This can be achieved by a low dose of antidepressants (such as amitriptyline), taken at bedtime. Low dose tricyclics seem to counteract the disturbing hyperarousal mechanisms in FMS by promoting deeper sleep. Our experience showed that muscle relaxants (such as cyclobenzaprine) may have the same effect when used in children and may be preferred as the first choice. Only 25% of our patients had to be switched to low-dose antidepressants due to lack of response to muscle relaxants. We found that analgesics such as acetaminophen or NSAIDs may be helpful at times when used in addition to muscle relaxants or antidepressants. We did not use narcotic analgesics due to the addictive potential they possess.

We educated our patients (under a physical therapist's guidance) to exercise, and encouraged them to maintain low-impact type exercises (such as stretching exercises, low-impact aerobics, walking, biking, and swimming) in order to restore and maintain muscle tone and improve their cardiovascular fitness. Educational programs help the patients and their families understand the disorder and to learn how to cope with it, and improve compliance. Our present study showed that exercise programs correlate with better outcomes. Since psychological factors may play a part in children with FMS, a cognitive - behavioral intervention has been shown to be effective in reducing pain and facilitating improved function in these patients (16). Five of our patients had therapy with relaxation techniques (such as biofeedback), of whom only 2 improved. The small number of patients who participated in these sessions makes it difficult to draw any conclusions.

Little is known about the natural history and long-term prognosis of FMS in children, and prospective longitudinal studies are lacking. Several studies (17, 18) in adult patients with FMS suggest a poor outcome. The course tends to be more chronic, i.e., chronic continuous or chronic polycyclic. Three- to 4-year follow-ups showed that 60-85% of the patients continue to manifest significant aching, and almost all took medication regularly to control symptoms. A minority of patients, especially those with chronic emotional disorders such as anxiety, depression, and maladaptive behaviors, might become disabled and require care at multidisciplinary pain or rehabilitation centers (19). In one reported retrospective chart review study in children with FMS (20), 17 out of 28 (61%) failed to improve during the follow-up period (range 1 - 48 months). In contrast to the findings of poor outcome, the present study showed that 60% of the patients had improved while 36% remained unchanged, and only 4% worsened within the mean follow-up period of 18.3 months. Although this study is in agreement with the study in school-children with FMS (5) and a recent report by Siegal *et al*. (9) that have demonstrated a better prognosis, the mean follow-up period was slightly shorter.

In summary, the present study demonstrates that the clinical spectrum of FMS in children is similar to that of adults but with better outcomes. The TP count correlates with the clinical status only in patients who improved, and active exercise programs seem to be associated with better outcomes. Prospective and larger patient population studies and a longer follow-up period for children with FMS are needed to clarify these findings.

References

1. SMYTHE HA: Nonarticular rheumatism and psychogenic musculoskeletal syndromes. *In:* MCCARTY DJ (Ed.): *Arthritis and Allied Conditions: A Textbook of Rheumatology*. Philadelphia, Lea and Febiger, 1989: 1241-54.
2. CAMPBELL SM, CLARK S, TINDALL ES, FOR-

CHAND ME, BENNETT RM: Clinical characteristics of fibrositis. A "blinded" controlled study of symptoms and tender points. *Arthritis Rheum* 1983; 26: 817-24.

3. YUNUS MB, MASI AT: Juvenile primary fibromyalgia syndrome. A clinical study of thirty-three patients and matched normal controls. *Arthritis Rheum* 1985; 28: 138-45.
4. BUSKILA D, PRESS J, GEDALIA A, *et al.*: Assessment of nonarticular tenderness and prevalence of fibromyalgia in children. *J Rheumatol* 1993; 20: 368-70.
5. BUSKILA D, NEUMANN L, HERSHMAN E, GEDALIA A, PRESS J, SUKENIK S: Fibromyalgia syndrome in children - An outcome study. *J Rheumatol* 1995; 22: 525-8.
6. WOLFE F, SMYTHE HA, YUNUS MB, *et al.*: The American College of Rheumatolgy 1990 Criteria for the classification of fibromyalgia. Report of multicenter criteria committee. *Arthritis Rheum* 1990; 33: 160-72.
7. GEDALIA A, PRESS J, BUSKILA D: Diffuse musculoskeletal pain syndromes in pediatric practice. *J Clin Rheumatol* 1996; 2: 325-30.
8. CASSIDY JT: Progress in diagnosing and understanding chronic pain syndromes in children. *Curr Opin Rheumatol* 1994; 6: 544-6.
9. SIEGAL DM, JANEWAY D, BAUM J: Fibromyalgia syndrome in children and adolescents: Clinical features at presentation and status at follow-up. *Pediatrics* 1998; 101: 377-82.
10. FELSON DT, GOLDENBERG DL: The natural history of fibromyalgia. *Arthritis Rheum* 1986; 29: 1522-6.
11. ROIZENBLATT S, TUFIC S, GOLDENBERG J, *et al.*: Juvenile fibromyalgia: Clinical and polysomnographic aspects. *J Rheumatol* 1997; 24: 579-85.
12. SIMMS R: Muscle studies in fibromyalgia syndrome. *J Musculoskel Pain* 1994; 2: 117-23.
13. GEDALIA A, PRESS J, KLEIN M, *et al.*: Joint hypermobility and fibromyalgia in schoolchildren. *Ann Rheum Dis* 1993; 52: 494-6.
14. GOLDMAN JA: Hypermobility and deconditioning: Important links to fibromyalgia/fibrositis. *South Med J* 1991; 84: 1192-6.
15. YUNUS MB: Towards a model of pathophysiology of fibromyalgia: Aberrant central pain mechanism with peripheral modulation. *J Rheumatol* 1992; 19: 846-50.
16. WALCO GA, ILOWITE NT: Cognitive-behavioural intervention for juvenile primary fibromyalgia syndrome. *J Rheumatol* 1992; 19: 1617-9.
17. FELSON DT, GOLDENBERG DL: The natural history of fibromyalgia. *Arthritis Rheum* 1986; 29: 1522-6.
18. NORREGAARD J, BULLOW PM, PRESCOTT E, JACOBSEN S, DANNESKIOLD-SAMSOE B: Preliminary results of a 4-year follow-up study in fibromyalgia. *J Musculokeletal Pain* 1993; 1: 159-63.
19. GOLDENBERG DL, BRODEUR CA, SMITH S, *et al.*: Prognostic factors in fibromyalgia. *Arthritis Rheum* 1992; 35: S113.
20. MALLESON PN, AL-MATAR M, PETTY RE: Idiopathic musculoskeletal pain syndromes in children. *J Rheumatol* 1992; 19: 1786-9.