Pediatric rheumatology

Fibromyalgia syndrome in young children: onset at age 10 years and younger

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Abstract

Objective

To report our experience of fibromyalgia syndrome (FMS) in young children with onset at age 10 years and younger as compared to older children.

Methods

Clinical and laboratory data were reviewed in all patients that had been diagnosed with FMS between November 1994 and March 2003. Patients with onset above the of age 18 years, and patients with FMS and concomitant rheumatic diseases were excluded from this study. The study population included two groups: group "A", young children with onset at age 10 years and under and group "B", children with onset above 10 years old. A questionnaire was used at follow-up visits or by telephone interview to evaluate the outcome.

Results

There were 148 children with the diagnosis of FMS (based on ACR criteria), of these 46 children in group A and 102 children in group B. The mean age at onset and mean age at diagnosis were 7.5 years and 10 years in group A, and 13.2 years and 14.5 years in B, respectively. The mean interval between the age of onset and the age at diagnosis was 32 months in group A, and 18 months in group B ($p= 0.007$). There was a predominance of female gender and Caucasian ethnicity in both groups. Diffuse aching was reported in all patients in both groups. Stiffness, subjective joint swelling, abdominal pain and initial presentation on wheelchair were found more frequently in group A, compared with group B ($p= 0.03$, 0.001, 0.01, 0.03 respectively). The mean count of tender points at diagnosis was higher in group A, compared with group B (15.3 vs. 14.2, $p = 0.004$). The differences of other clinical features and laboratory tests in both groups were not statistically significant. Thirty-six patients in group A (78%) and 83 in group B (81%) were available for one or more follow-up visits and/or telephone interview. The mean follow-up period was 14 months in group A, and 19 months in group B (p value = 0.3). There was no difference in the type of treatment or outcome in both groups.

Conclusion

FMS in young children of 10 years old and younger is frequently under-recognized. As compared with the older group, stiffness, subjective joint swelling, abdominal pain, initial presentation on wheelchair and a higher mean count of tender points at diagnosis were significantly more common in the younger age group. However, the type of medications used and outcome were similar in both groups. Prospective studies with large patient population are needed to clarify these findings.

Key words

Fibromyalgia syndrome, young children, tender point count, outcome.

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Introduction

Fibromyalgia syndrome (FMS) is a non-articular rheumatic condition that is characterized by diffuse musculoskeletal aching and the presence of multiple tender points at specific soft tissue sites.

The prevalence of this syndrome in the adult general population is about 2% (1), and tends to increase with age with prevalence between the ages 20 - 60 years. The reported prevalence of this syndrome in children varies widely probably reflecting differences in ethnicity, sociocultural background, psychological traits of the population and diverse methodologies that have been used in the published studies (2).

We report herein our experience in children with FMS, focusing on young children with onset at the age of 10 years and younger as compared to older children. A specifically designed questionnaire was used to gather pertinent information. This questionnaire was filled in at follow-up visits or by telephone interview.

Patients and methods

The records of all patients that had been diagnosed with FMS between November 1994 and March 2003 at the pediatric rheumatology clinic of the Children's Hospital of New Orleans were reviewed. The study population was divided into two groups (A and B) according to the age of onset. Group "A" included young children with an onset at age 10 and under, and group "B" included children with onset above 10 years old. Patients with an onset above the age of 18 years, and patients with FMS and concomitant rheumatic diseases were excluded from this study. A specifically designed format (see appendix) was used to gather pertinent information. This included the following variables: Gender, ethnicity, age at onset, duration of the disease before diagnosis, duration of follow-up, family history of FMS, clinical manifestations, laboratory findings, management and outcome. Patients and their parents were questioned concerning the presence of widespread pain or aching and other symptoms including: headaches, sleep disturbance, morning stiffness greater than 15 minutes duration, subjective joint swelling, fatigue, abdominal pain, and family history of FMS. The quality of sleep was assessed by asking the patients and their parents 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 disturbance, while "never", "seldom" or any other replies scored as a negative indication. A similar assessment was performed to determine other signs and symptoms (headaches, subjective joint swelling, fatigue, and abdominal pain and numbness). In all patients, a count of 18 TP was conducted by thumb palpation. In addition, 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 (3). Patients were evaluated for the presence or absence of joint hypermobility, using the criteria of Carter and Wilkinson as modified by Bird *et al*. (4). 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 those patients who could not make it for some of their follow-up visits. Laboratory data was reviewed on all patients. Informed consent was obtained from patients and their parents. The assessment for FMS (including TP counts) and joint hypermobility were performed by the same clinician/observer (AG) who had conducted also the evaluation at initial and follow-up visits. Our physical therapist instructed the patients and their parents about home-exercises, to be done at least a half-hour a day. In addition, they were encouraged to do low-impact exercises such as walking,

Table I. Comparative demographic characteristics of patients in Group A and Group B.

Characteristic	Group A (Onset at age 10 or under)	Group B (Onset above 10 years of age)	*P* value
Gender			
Females	32 (70%)	79 (77%)	0.3
Males	14 (30%)	23 (23%)	
Ethnicity			
Caucasians	35 (76%)	68 (67%)	0.3
African – Americans	10 (22%)	29 (28%)	0.4
Hispanics	1 (2%)	5 (5%)	0.6
Mean age at onset	7.5 years	13.2 years	< 0.001
Range	(3-10 years)	(10.2–18 years)	
Mean age at diagnosis	10.07 years	14.5 years	< 0.001
Range	(6.6 – 15.7 years)	(10.4–19 years)	
Mean interval: onset – diagnosis	2.58 years (32 months)	1.43 years (18 months)	0.008
Range	(0.25- 7.7 years)	(0.25-6.2 years)	
Mean follow-up	14 months	19 months	0.3

Table II. Clinical features of Total group, Group A and Group B.

Clinical feature	Total Group		Group A (Onset at age 10 or under)		Group B (Onset above 10 years of age)		*P* value
	n = 148	(%)	n = 46	(%)	n = 102	(%)	
Generalized aches & pain	148	(100)	46	(100)	102	(100)	-
Headache	118	(80)	36	(78)	82	(80)	0.8
Fatigue / tiredness	37	(25)	13	(28)	24	(23)	0.5
Sleep disturbances	106	(72)	30	(65)	76	(74)	0.3
Stiffness	40	(27)	18	(39)	22	(21)	0.03
Subjective joint swelling	32	(22)	18	(39)	14	(14)	0.001
Abdominal pain	37	(25)	18	(39)	19	(19)	0.01
Numbness	4	(3)	3	(6)	1	(1)	0.08
Raynaud's phenomenon	1	(1)	0	(0)	1	(1)	1
Anxiety	3	(2)	1	(2)	2	(2)	1
Depression	13	(9)	4	(9)	9	(9)	1
Joint hypermobility	31	(21)	8	(17)	23	(23)	0.5
Initial presentation on wheelchair	5	(3)	4	(9)	1	(1)	0.03

swimming, cycling, low-impact aerobics or stretching exercises on a regular basis.

Statistical analysis

Wilcoxon test, Fisher's exact test and Mann Whitney test were used for statistical analysis, using a *p* value of 0.05, level of significance.

Results

Of the 148 children diagnosed as having FMS, 46 young children had onset at 10 years and under (Group A) and 102 children had onset above 10 years of age (Group B). All patients met the currently accepted ACR criteria for the diagnosis of FMS, namely widespread pain in combination with tenderness of 11 or more of 18 specific tender point sites (3). The mean age at onset was 7.5 years (range 3-10 years) in group-A and 13.2 years (range, 10.2 – 18 years) in group-B. The mean age at diagnosis was 10 years (range 6.6-15.7 years) in group-A and 14.5 years (range 10.4-19.8 years) in group-B. The mean interval between the age of onset and the age at diagnosis was 32 months, and 18 months in group-A and B respectively. The difference of this interval between both groups was found to be statistically significant ($p = 0.007$). There was a clear predominance of female gender and Caucasian ethnicity in both groups. (Table I). Eighteen patients (12%) had a positive family history of FMS; 8 (17%) in group-A and 10 (9.8%) in group B ($p = 0.28$).

Diffuse aching was reported in all the patients in both groups. Stiffness, subjective joint swelling, abdominal pain and initial presentation, on wheelchair, were found more frequently in group-A, compared with group-B. These differences were statistically significant ($p = 0.03$, 0.001, 0.01, 0.03 respectively). The mean count of tender points at diagnosis was higher: 15.3 in group A as compared to 14.2 in group B, with a statistically significant difference ($p = 0.004$).

The differences of other clinical features in both groups were not statistically significant (Table II).

Laboratory tests done at the initial visit showed that complete blood count (CBC) was performed in 40 patients in group-A (87%), in 97 patients in group B (95%), and was normal in all of them. Antinuclear antibody (ANA) test (Hep 2 cell substrate) was performed in 39 and 87 patients in group-A and group B respectively. This test was positive (titer > 1:40) in 46 (31%) patients; 17 (44%) of group-A and 29 (33%) of group-B (p value = 0.3186); the ANA profile (including antibodies to SS-A, SS-B, ds-DNA, Sm, RNP, and Scl-70) was negative in all of them.

Cyclobenzaprine (2.5-20 mgs at bedtime) was prescribed in 117 patients at diagnosis, 36 (78%) in group A and 81(80%) in group B ($p = 1.0$). Twenty patients received amitriptyline (10-20 mgs), 3(7%) in group A and 17 in group B (17%) ($p = 0.1$). Thirty-five patients received NSAIDS (mostly naproxen) with or without acetaminophen as needed, 10 (22%) in group-A and 25 patients (25%) in group B ($p = 0.8$).

A total of 119 patients, 36 in group A (78%) and 83 in group-B (81%) were available for one or more follow-up visits and/or telephone interview. Of the 119 patients, only 75 (26 in group A and 49 in group B) were available and

were evaluated at the clinic at the last follow-up visit. The remaining 44 patients (10 in group A and 34 in group B) were interviewed by telephone.

The mean duration of follow-up was 14 months in group A (range 3 months - 3.7 years) and 19 months in group B (range 3 months - 6 years) (p value = 0.3).

The tender point count at the last visits was available in 75 patients (26, 56% in group A and 49, 48% in group B). The mean count of tender points at the last follow-up was 12.6 in group A and 12.5 in group B ($p = 0.7$).

Symptoms had improved in 55 (46%) of the 119 patients: 19 (53%) in group-A and 36 (43%) in group-B, ($p = 0.4$). There was no change in the symptoms of 51 (43%) patients: 13 (36%) in group A and 38 (46%) in group B ($p = 0.4$). Symptoms became worse in 13 (11%) patients: 4 (11%) in group A and 9 (11%) in group B ($p = 1$).

Discussion

FMS is a well-recognized condition in the adult population. During the past years, several reports on FMS in children have been published. However, this is the first report describing the characteristics of this syndrome in young children aged 10 years or younger. In comparison with the older pediatric patients, we found several differences.

The mean interval between the age of onset and the age of diagnosis was significantly longer in the younger group (32 *vs*. 18 months), suggesting that this syndrome in a younger age is frequently under-recognized.

Predominance of female gender and Caucasian ethnicity was found in our FMS study population, as has been reported by others (2, 3, 5, 6). However, regarding these variables, there were no statistically significant differences between the younger and older age groups.

Familial occurrence of FMS has been previously described. Roizenblatt *et al*. (6) reported a positive family history of FMS in 24 out of 34 (71%) children. However, in the present study and in a previous report, we found that the family occurrence of FMS in children was 10 and 12% respectively (6). There was no statistically significant difference in family occurrence between group A and group B.

Comparing both groups, the younger (Group A) and the older (Group B), diffuse aching was found in all patients of both groups. There was no statistically significant difference in the frequency of headache, fatigue and sleep disturbances between the two groups. The frequencies were similar to other published reports (2, 5-9).

Although the above manifestations of the FMS did not differ in either group, the following clinical characteristics: stiffness, subjective joint swelling, abdominal pain, and initial presentation on wheelchair were found more frequently in the younger children (group A). Furthermore, the mean count of tender points at diagnosis was also significantly higher in the younger group. Numerous studies in children and adults showed that FMS patients have lower thresholds for tenderness (9, 11-13). Additionally, others have reported that the number of painful tender points was strongly correlated with distress (including pain, fatigue, sleep disturbances, anxiety, and depression) (14, 15). However, no comparison between age groups was made. Buskila *et al*. (16) assessed 338 healthy schoolchildren with a mean age of 11.5 years (range 9-15 years) for tenderness thresholds and prevalence of FMS. The study population was divided into 3 age groups: 9-10; 11-12; and 13-15. They found no statistically significant difference in the thresholds of tenderness and prevalence of FMS between these age groups. Yunus and Masi (9) studied the frequency of clinical manifestations in 33 pediatric FMS patients. They found no significant difference between the groups aged 15 or younger and 15 and older.

In the present study, we found a high frequency of ANA positivity (31%). However, the ANA profile was negative in all of these ANA positive patients. There was no statistically significant difference between both groups. Similar ANA frequency was found by Smart *et al*. (16) in a group of 66 FMS patients. Nevertheless, in another report Bengtsson *et al*. (18) found no statistically significant difference in the frequency of ANA positivity, comparing a group of 223 patients with a group of 255 controls (blood donors).

According to some reports, the prognosis of FMS in children is better than in adults. Buskila *et al*. studied 21 children and adolescents with FMS, with 7 fulfilling point count criteria only. Re-examination of 15 out of the 21 patients who were available for follow-up revealed that 11 (73%) were no longer fibromyalgic 30 months after the initial assessment, and none of the 7 fulfilling point count only developed FMS (19). Thirty-three out of 45 patients with FMS cohort studied by Siegel *et al*. were available for telephone interview (5). Using a self-rating scale of 1 to 10, in which 1 represented complete disability and 10 indicated no disability, interviews found a mean positive change of 4.8 within one-year follow-up. All patients in this study were prescribed cyclobenzaprine or low-dose tricyclic antidepressant medications, moderate exercise, and analgesics (as needed). Mikkelsson reported a 1-year follow-up on 16 out of 22 Finnish children with FMS and found that only four of them (25%) had persistent fibromyalgia. Three (19%) were pain-free, and the rest had intermittent pain only. No treatment interventions had been provided in this study (20). Calvo *et al*. reported on 22 Spanish adolescents with FMS who were treated with analgesics and cyclobenzaprine. At 48-month follow-up, 15 (68.2%) had no longer fulfilled the FMS criteria (21).

In the present study, we found that 46% of the patients had improved, 43% remained unchanged, and in 11% symptoms became worse. There was no statistically significant difference between the younger and older patients. This study was a retrospective chart review. The authors concur with the possibility of biases, and that the above data should be interpreted cautiously.

In summary, the present study revealed that FMS in young children of 10 years old and under is frequently under-recognized. Stiffness, subjective joint swelling, abdominal pain, initial presentation on wheelchair and a higher mean count of tender points at diag-

nosis were more common in this age group. However, compared with the older group, the outcome was similar. Prospective studies with a larger patient population, and a longer follow-up are needed to clarify these findings.

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APPENDIX

PRIMARY FIBROMYALGIA IN CHILDREN

Date ----------

LAST NAME _____ FIRST NAME _____ SEX _____

RACE _____ DOB _____ LAST VISIT_____

DATE (ONSET) _____ DATE (DIAGNOSIS) _____ AGE AT DIAGNOSIS _____

INTERVAL (ONSET – DIAGNOSIS) _____ PREVOIUS DIAGNOSIS _____

CLINICAL MANIFESTATIONS AT DIAGNOSIS

1. Generalized aches & pains		8. Anxiety	
2. Headaches		9. Depression	
3. Fatigue		10. Abdominal pain	
4. Sleep disturbances		11. Irritable bowel	
5. AM Stiffness (duration)		12. Raynaud's	
6. Subjective joint swelling		13. Sicca symptoms	
7. Numbness			

FAMILY HISTORY _____

_____

SOCIAL HISTORY _____

PHYSICAL EXAMINATION:

1. NUMBER OF TENDER POINTS __/ 18
2. PRESENCE OF JOINT HYPERMOBILITY YES / NO
3. OTHER FINDINGS _____

LABORATORY TESTS

ESR	ANA	RF	TSH
T-3	T-4	PROLACTIN	

TREATMENT

TYPE	DOSE (DURATION)
ANALGESICS	
NSAID's	
FLEXERIL	
ELAVIL	
PHYSICAL THERAPY	
OTHERS (----------)	

ARE YOU TAKING YOUR MEDICINES?

(_____) YES, DAILY (_____) NO (_____) YES, BUT NOT DAILY

OUTCOME

MANIFESTATION	BETTER	WORSE	UNCHANGED
PAIN			
SLEEP DISTUBANCES			
AM STIFFNES			
FATIGUE			

PHYSICAL EXAMINATION:

NUMBER OF TENDER POINTS __/ 18

HOW WOULD YOU SAY YOUR OVERALL SYMPTOMS ARE DOING COMPARED TO YOUR INTIAL PRESENTATION?

(___) A LOT BETTER (___) A LOT WORSE (___) A LITTLE BETTER

(___) A LITTLE (___) NO CHANGE

HOW MANY TIMES HAVE YOU MISSED SCHOOL BECAUSE OF YOUR FIBROMYALGIA?

LAST MONTH _____ LAST 3 MONTHS _____ LAST 6 MONTHS _____