

EXTENDED REPORTS

Joint hypermobility and fibromyalgia in schoolchildren

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Abstract

Objectives—To test the hypothesis that joint hypermobility may play a part in the pathogenesis of pain in fibromyalgia, schoolchildren were examined for the coexistence of joint hypermobility and fibromyalgia.

Methods—The study group consisted of 338 children (179 boys, 159 girls; mean age 11.5 years, range 9–15 years) from one public school in Beer-Sheva, Israel. In the assessment of joint hypermobility, the criteria devised by Carter and Bird were used. Any child who met at least three of five criteria was considered to have joint hypermobility. Children were considered to have fibromyalgia if they fulfilled the 1990 American College of Rheumatology criteria for the diagnosis of fibromyalgia, namely, widespread pain in combination with tenderness of 11 or more of the 18 specific tender point sites. The blind assessments of joint hypermobility (by AG) and fibromyalgia (by DB) were carried out independently.

Results—Of the 338 children 43 (13%) were found to have joint hypermobility and 21 (6%) fibromyalgia; 17 (81%) of the 21 with fibromyalgia had joint hypermobility and 17 (40%) of the 43 with joint hypermobility had fibromyalgia. Using χ^2 statistical analysis, joint hypermobility and fibromyalgia were found to be highly associated.

Conclusions—This study suggests that there is a strong association between joint hypermobility and fibromyalgia in schoolchildren. It is possible that joint hypermobility may play a part in the pathogenesis of pain in fibromyalgia. More studies are needed to establish the clinical significance of this observation.

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It is well established that joint hypermobility may predispose children to the development of joint pain.^{1,2} This disorder is an extreme variation of normal joint mobility with no underlying connective tissue disease.³ The mechanism by which joint symptoms develop in this disorder is not well understood, though excessive motion or inappropriate physical

activities that hyperextend the joint capsule and other soft tissue constituents may cause injury.⁴ Fibromyalgia is a syndrome of diffuse muscle and joint pain most common in adults.^{5,6} Although it has been reported to cause diffuse musculoskeletal pain and fatigue in children, its prevalence is not well documented.⁷ The purpose of this study was to examine schoolchildren for the coexistence of the two clinical disorders (joint hypermobility and fibromyalgia) to test the hypothesis that joint hypermobility may play a part in the pathogenesis of pain in fibromyalgia.

Subjects and methods

The study group consisted of 338 schoolchildren aged 9–15 years in one of the public schools in Beer-Sheva, Israel. There were 179 (53%) boys and 159 (47%) girls (mean age 11.5 years, range 9–15 years).

After informed consent had been obtained, two experienced rheumatologists independently blindly assessed joint hypermobility (AG) and fibromyalgia (DB). This was performed simultaneously in two separate rooms. All children were evaluated before noon.

In the assessment of joint hypermobility, the following criteria devised by Carter and Wilkinson,⁸ with modifications by Bird *et al*,⁹ were used: (a) hyperextension of the fingers so that they lie parallel to the forearms; (b) apposition of the thumbs to the surface of the forearms; (c) hyperextension of the elbows $>10^\circ$; (d) hyperextension of the knees $>10^\circ$; and (e) flexion of the trunk with the knees straight and touching the palms of the hands to the floor.

A child who met at least three of the five criteria was considered to have joint hypermobility.

In the assessment of fibromyalgia, children and parents were questioned about widespread musculoskeletal pain and aching. A point count of 18 tender points was conducted in all children. The children were considered to have fibromyalgia if they fulfilled the currently accepted American College of Rheumatology (ACR) criteria for the classification and diagnosis of fibromyalgia, namely widespread pain in combination with tenderness of 11 or more of the 18 specific point sites.¹⁰ Digital palpation was conducted with an approximate force of 4 kg. For a tender point to be 'positive'

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Table 1 Prevalence of joint hypermobility (JH) and fibromyalgia (FM) in 338 schoolchildren

| | No (%) with JH | No (%) with FM |
|---------------|----------------|----------------|
| Boys (n=179) | 20 (11) | 7 (4) |
| Girls (n=159) | 23 (14) | 14 (9) |
| Total (n=338) | 43 (13) | 21 (6) |

The difference in the prevalence of JH and FM between the two sexes is not statistically significant.

the subject must state that the palpation is painful. Finally, the data were analysed using χ^2 statistical analysis.

Results

Joint hypermobility was detected in 43 (13%) of the 338 children. The prevalence of joint hypermobility (table 1) was 14% in girls (23 children) and 11% in boys (20 children) with no statistical differences between them ($\chi^2=0.8$; $p=0.5$).

Fibromyalgia was diagnosed in 21 (6%) of the 338 children (table 1). The frequency of fibromyalgia among boys of 4% (seven boys) was not significantly different ($\chi^2=3.0$; $p=0.1$) from that of 9% in girls (14 girls).

Seventeen (81%) of the 21 children with fibromyalgia had joint hypermobility. Seventeen (40%) of the 43 children with joint hypermobility had fibromyalgia *v* 4 (1%) of 295 children without joint hypermobility (table 2). The association between joint hypermobility and fibromyalgia was statistically significant ($\chi^2=93$; $p<0.0001$). Seven children, in whom 11 of 18 point sites were tender, neither reported pain nor had any of the other features such as fatigue or irritable bowel syndrome, and therefore did not fulfil the ACR criteria for the diagnosis of fibromyalgia. Three of these seven children had joint hypermobility, however. Of the 21 children with fibromyalgia, 13 had a history of recurrent episodes of leg pain, four had episodes of leg pain and low back pain, two had recurrent headaches, and two had recurrent episodes of abdominal pain and headaches. Of these children, only one had a history of morning stiffness. Evaluation carried out by their family doctor showed no underlying organic diseases, though a diagnosis of growing pains was made in 10 of them. Sleep disturbances and psychological factors were not assessed and no data on these parameters are available. More details on the assessment of the non-articular tenderness in these children using a Chatillon dolorimeter have been described elsewhere.¹¹ Briefly, it was shown that in general the boys had a lower degree of tenderness than the girls. The children with fibromyalgia had lower thresholds for tenderness at control and tender points than the subjects without fibromyalgia.

Table 2 Relation between joint hypermobility (JH) and fibromyalgia (FM) in 338 schoolchildren

| | No with JH | No without JH | Total |
|---------------|------------|---------------|-------|
| No with FM | 17* | 4 | 21 |
| No without FM | 26 | 291 | 317 |
| Total | 43 | 295 | 338 |

The association between JH and FM is highly statistically significant ($\chi^2=93$; $p<0.0001$).

Discussion

It has previously been clearly shown that significant numbers of children with joint hypermobility have articular symptoms.² The pathogenesis in which pain develops in joint hypermobility is not well understood, though it may be speculated that it might be related to microtrauma due to misuse or overuse, or both. Another pain disorder sometimes diagnosed in children is fibromyalgia.⁷ This is a chronic syndrome of musculoskeletal pain, the aetiology of which is unknown. The pathogenesis of fibromyalgia is not clear; however, sleep disturbances in stage 4 non-rapid eye movement sleep, muscle hypoxia, generalised muscle deconditioning, and mechanical stress have been suggested to play a part.⁵ The model of pathophysiology described by Yunus¹² may explain the mechanism of fibromyalgia among children with joint hypermobility, as peripheral trauma factors in joint hypermobility may initially cause localised joint pain, which would then cause neuroendocrine dysfunction through central nervous system plasticity, leading to widespread pain and tenderness.

The present study shows a significant association between joint hypermobility and fibromyalgia. This may explain in part the mechanism by which pain develops in fibromyalgia. The association of these two disorders is not well understood. Cognitive-behavioural intervention in children with fibromyalgia has been shown to be effective in reducing pain and facilitating improved functioning.¹³ This observation suggests that psychological factors may also play a part in this disorder. It should be stressed that in any child who presents with diffuse musculoskeletal pain, joint hypermobility and fibromyalgia should both be considered in the differential diagnosis.

Although the two disorders will benefit from drugs and exercises for symptomatic pain, the paediatrician needs to assure the anxious and fearful parents that these are benign and not crippling phenomena. Most interesting are the seven healthy children with more than 11 tender points but no widespread pain. Whether these subjects are more likely to develop pain syndromes in the next few years remains to be seen.

In summary, the prevalence of fibromyalgia in schoolchildren was 6% and 40% among children with joint hypermobility. The association between joint hypermobility and fibromyalgia was highly significant. More studies are needed to establish the clinical significance of this observation.

- Gedalia A, Person D A, Brewer E J, *et al*. Joint hypermobility in juvenile episodic arthritis/arthritis. *J Pediatr* 1985; **107**: 873-6.
- Gedalia A, Press J. Articular symptoms in hypermobile schoolchildren. A prospective study. *J Pediatr* 1991; **119**: 944-6.
- Kirk J A, Ansell B M, Bywaters E G L. The hypermobility syndrome: musculoskeletal complaints associated with generalized joint hypermobility. *Ann Rheum Dis* 1967; **26**: 419-25.
- Lewkonja R M. Hypermobility of joints. *Arch Dis Child* 1987; **62**: 1-2.
- Smythe H A. Nonarticular rheumatism and psychogenic musculoskeletal syndrome. In: McCarty D J, ed. *Arthritis and allied conditions*. Philadelphia: Lea and Febiger, 1989: 1241-54.

- 6 Goldenberg D L. Diagnostic and therapeutic challenges of fibromyalgia. *Hosp Pract* 1989; 24: 39–52.
- 7 Yunus M B, Masi A T. Juvenile primary fibromyalgia syndrome. A clinical study of thirty three patients and matched normal controls. *Arthritis Rheum* 1985; 28: 138–45.
- 8 Carter C, Wilkinson J. Persistent joint laxity and congenital dislocation of the hip. *J Bone Joint Surg [Br]* 1964; 46: 40–5.
- 9 Bird H A, Brodie D A, Wright V. Quantification of joint laxity. *Rheumatol Rehabil* 1979; 18: 161–4.
- 10 Wolfe F, Smythe H A, Yunus M B, *et al.* The American College of Rheumatology 1990 criteria for the classification of fibromyalgia: report of the multicenter criteria committee. *Arthritis Rheum* 1990; 33: 160–72.
- 11 Buskila D, Press J, Gedalia A, *et al.* Assessment of non-articular pain tenderness and prevalence of fibromyalgia in children. *J Rheumatol* 1993; 20: 368–70.
- 12 Yunus M B. Towards a model of pathophysiology of fibromyalgia: aberrant central pain mechanisms with peripheral modulation. *J Rheumatol* 1992; 19: 846–50.
- 13 Walco G A, Ilowite N T. Cognitive-behavioral intervention for juvenile primary fibromyalgia syndrome. *J Rheumatol* 1992; 19: 1617–9.