

Hypermobility and fibromyalgia frequency in childhood familial Mediterranean fever

Sirs,

Familial Mediterranean Fever (FMF) is a hereditary disease inherited autosomal recessively and characterized by fever and episodes of polyserositis (1). Symptoms of musculoskeletal system are present in 40-70% of patients with FMF (1-3). Leg pain after exertion is an important clinical finding. This finding is accepted as a minor criterion according to the criteria of FMF described in 1997 (4). Some studies revealed that hypermobility (HM) and fibromyalgia (FM) syndromes can cause extremity pain in children (5,6). The sole study focusing on the relationship between FMF and FM was conducted by Langevitz *et al.* (7) in an adult population, but there is no data about the presence of HM and FM in children with FMF.

The aim of this study was to assess the presence HM and FM among the musculoskeletal findings of FMF and to determine the relationship between these findings and articular manifestations of FMF.

1. FMF group: This group consisted of 108 patients who were diagnosed as having FMF according to the diagnostic criteria of Livneh (4). Clinical findings were determined according to the follow-up records of the patients. Articular involvement in the positive group was defined as patients having joint pain for more than 3 months and at least one episode of arthritis during their disease course.

2. Diseased control group: This group consisted of 105 patients who were admitted to pediatric out-patient clinics with the complaint of joint pain. Patients with upper and lower extremity pain were evaluated with regard to the presence of joint pain for at least 3 months and the exclusion of a primary or secondary rheumatic disease that can be the cause of arthritis or arthralgia.

3. Healthy control group: 103 school children without any complaints were studied.

In order to diagnose HM and FM, the Beighton scoring system and 1990 ACR diagnosis criteria were used, respectively (8, 9). For hypermobility, a 6/9 positive score and for fibromyalgia the presence of 11/18 tender points were accepted as diagnostic. All questionnaires and tests that were applied to the study groups were performed by the same investigator (MT). This investigator was not informed about the diagnosis of the patients. Data obtained from the study groups are summarized in Table I.

The incidence of HM in children diagnosed with FMF was significantly higher than that in the healthy children ($\chi^2 = 23.21$; $p < 0.001$), but the incidence was similar to the diseased control group. According to gender, 19 (40.5%) of 47 males with FMF and 31 (50.8%) of 61 females with FMF had HM. The rate of HM in FMF was found to be higher in girls but this difference was not statistically significant. In 77 patients with FMF (71.2%) articular involvement was present. Forty-four (57%) of these patients had arthralgia whereas 33 (43%) had both arthritis and arthralgia. No relationship was found between joint findings in FMF and HM ($\chi^2 = 0.34$; $p > 0.05$). The incidence of HM in the diseased group was significantly higher than that in the healthy control group ($\chi^2 = 19.8$; $p < 0.001$).

Fibromyalgia was detected in 2 (1.8%) of 108 FMF patients, one boy and one girl. Both had joint involvement. There was no significant difference in the incidence of FM between patients with FMF and healthy children ($p = 0.26$). This incidence was significantly higher in the diseased group than in the other two groups ($\chi^2 = 12.49$; $p = 0.00019$).

On the basis of these data, we can conclude that HM may be a finding in FMF, but is not directly related to articular involvement. The frequency of HM was similar between the disease control group and patients with FMF. HM was not a specific finding of FMF. In this study, the rate of FM in children with FMF was found to be 1.8%, which was not statistically different from

the rate of FM in healthy children. Therefore it can be said that FM is not a common disorder encountered in childhood FMF.

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Table I. Demographic characteristics, hypermobility and fibromyalgia frequency in all study groups.

Study group	n	F/M ratio	Mean age at investigation	Hypermobility frequency	Fibromyalgia frequency
Familial Mediterranean fever	108	61/47	10.3 ± 3.4 yrs.	50/108 (46.3%)	2/108 (1.8%)
Diseased control group	105	57/48	8.4 ± 2.9 yrs.	49/105 (46.6%)	12/105 (11.4%)
Healthy children	103	43/60	8.7 ± 2.9 yrs.	16/103 (15.5%)	0
Total	316	161/155			