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THE ASSOCIATION OF SOFT-TISSUE RHEUMATISM AND HYPERMOBILITY

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SUMMARY

Soft-tissue rheumatism (STR—tendinitis, bursitis, fasciitis and fibromyalgia) accounts for up to 25% of referrals to rheumatologists. The estimated prevalence of generalized hypermobility in the adult population is 5–15%. There have previously been suggestions that hypermobile individuals may be predisposed to soft-tissue trauma and subsequent musculoskeletal pain. This study was designed to examine the mobility status and physical activity level in consecutive rheumatology clinic attendees with a primary diagnosis of STR. Of 82 patients up to age 70 yr with STR, 29 (35%) met criteria for generalized hypermobility. Hypermobile compared to non-hypermobile individuals reported significantly more previous episodes of STR (90% *vs* 51%, $P < 0.01$), and more recurrent episodes of STR at a single site (69% *vs* 38%, $P < 0.001$). Although we were unable to show any difference in the time spent carrying out physical activity between the two groups, the hypermobile patients were performing significantly more repetitive activities. When specific anatomical sites of STR were analysed, small joints (elbows, hands and feet) currently affected with STR were more likely to show localized hypermobility than if those joints were asymptomatic. These findings suggest that hypermobility may be a factor in the development of STR. Repetitive activity may be a contributing factor towards STR in some hypermobile individuals.

KEY WORDS: Hypermobility, Soft-tissue rheumatism, Fibromyalgia.

SOFT-TISSUE rheumatism (STR, e.g. tendinitis, bursitis, fasciitis and regional pain syndromes) accounts for up to 25% of new referrals to rheumatologists [1]. The causation of STR is not clearly understood, but is probably multifactorial. We have previously reported that hypermobile rheumatology patients are more likely to be diagnosed with STR than other forms of arthropathy [2]. An association between hypermobility and STR was noted as early as 1967. In their original description of the hypermobility syndrome, Kirk *et al.* [3] reported several cases of supraspinatous tendinitis, bicipital tendinitis, medial and lateral epicondylitis, and Achilles tendinitis in patients with generalized hypermobility. Subsequently, the existence of an association between hypermobility and STR has been suggested on the basis of anecdotal reports of symptoms experienced by patients with hypermobility. However, most published reports on hypermobility have been studies describing the demographic characteristics of hypermobile individuals in various populations [4–10]. It has been established that hypermobility is more frequent in females [4, 5, 8, 9, 11–16] and decreases with age [4, 6, 7–9, 11, 12, 16]. In North American and European Caucasian adults, both population-based studies and studies from rheumatology clinics have reported the prevalence of generalized hypermobility to be between 5 and 15% [2, 12, 14]. Similarly, 13% of Israeli schoolchildren were reported to be hypermobile [10]. In contrast, Al-Rawi *et al.* [15] reported higher prevalence rates among Iraqi university studies between the ages of 20 and 24 yr: 39% in females and 25% in males. In a community-based study of West Africans aged 6–66 yr, selected for musculoskeletal pain, Birrell *et al.* [16] reported a 43% prevalence of hypermobility. Thus, sex, age and ethnic background appear to influence the prevalence of hypermobility.

The consequences of having lax ligaments are largely unknown. The purpose of the present study was to examine the mobility status and physical activity level in patients with STR.

PATIENTS AND METHODS

Consecutive clinic attendees, both newly referred and follow-up patients at a community-based rheumatology practice, were assessed for the presence of STR, i.e. tendinitis, bursitis or fibromyalgia (FM). The study was conducted over a 6 month period. Inclusion criteria required that patients be between 16 and 70 yr of age, and agree to participate by providing written informed consent. Only patients with an inflammatory or degenerative arthritis as the primary rheumatology diagnosis were excluded. Patients with a possible inherited connective tissue disease were not specifically excluded. No patients were taking medications known to enhance joint laxity. Patients were assessed by three different individuals: the treating rheumatologist and two other rheumatologists blind to the rheumatological diagnosis.

Demographic data including age, sex, racial group, primary and any other rheumatological diagnosis were obtained by the treating rheumatologist. A second assessor, blind to these data, obtained a musculoskeletal history, which included a history of previous tendinitis or bursitis, defined as a diagnosis made by a physician, resulting in treatment with either medications, local measures such as physiotherapy or a local steroid injection. A previous history of single or multiple sites of pain or widespread pain as defined by the American College of Rheumatology (ACR) criteria for FM [17], as well as a history of joint dislocations and fractures, were recorded. Upper limb complaints

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suggestive of thoracic outlet symptoms, including aching of upper limbs and hand paraesthesiae when the arms were elevated, were recorded. Raynaud's phenomenon, defined as a triphasic colour change when the extremities were exposed to cold, was also noted. The patients were asked whether they considered themselves presently to be, or previously to have been, double-jointed or extremely flexible. This was recorded as positive if the patients said yes to either of the following questions: (1) Can (could) you press your thumb forwards to touch your forearm? (2) Can (could) you do the splits? An obstetrical history was taken from the female patients, particularly with respect to pregnancy-related complications, including preterm labour, premature delivery, fetal wastage and peripartum haemorrhagic complications.

The daily physical activity level for the previous 3 months was recorded as the percentage of time, both at work and leisure, spent sedentary or up and about. Patients reported the number of hours they were exposed to either repetitive activity or prolonged immobility. Repetitive activity was recorded as present if at least half of normal working time required the patient to perform repetitive activity involving either the upper or lower limbs. Forceful activity was considered to be present if activities, either at work or at leisure, required an action perceived by the patient to require strenuous effort and if the activities were performed regularly throughout a usual week. Sporting activities were recorded, as well as the average number of hours spent per week in the last 3 months in sporting activities.

Joint mobility was scored by a third evaluator, blind to the data obtained by the previous two assessors. Sites of mobility were scored clinically according to the Beighton definition (Table I) [4] and the Bulbena definition (Table II) [18]. This latter scoring system was used to supplement the Beighton criteria since it is a more comprehensive scale, including sites such as shoulders, hips, ankles and feet, which are not included in the Beighton criteria, but which are likely to be clinically important. Patients were classified as having generalized hypermobility if they fulfilled four or more of the nine Beighton criteria, or four or more of the 10 Bulbena criteria for males and five or more for females. In the event that a site could not be examined because of pain, then that particular joint was designated as hypermobile if the corresponding contralateral joint was hypermobile. However, this presumption was not used in determining overall mobility status by the Beighton or Bulbena criteria. It was used in the analysis linking localized hypermobile joints to a specific STR site. The tender point count according to the ACR criteria for FM was recorded [17]. Patients were assessed for the presence of thoracic outlet symptoms according to the following criteria: aching of upper limbs and hand paraesthesiae, with loss of pulse when the arms were abducted to 90°, the shoulders externally rotated to the maximum range of motion and the head was rotated to the opposite shoulder. Both the development of symptoms and loss of the radial pulse were required for a clinical diagnosis of thoracic outlet symptoms.

Statistical analysis for categorical variables was performed using the χ^2 test, and when small numbers were present (any cell of five or less) Fisher's exact test was used. Continuous variables were contrasted with a two-sided unpaired Student's *t*-test.

TABLE I
Beighton modification of the Carter and Wilkinson criteria for hypermobility*†

1. Passive dorsiflexion of the little finger beyond 90°
2. Passive apposition of the thumb to the flexor aspects of the forearm
3. Hyperextension of the elbow beyond 10°
4. Hyperextension of the knee beyond 10°
5. Forward flexion of the trunk, with knees straight, so that the palms of the hands rest easily on the floor

*One point is allocated for the ability to perform a manoeuvre at each site (criteria 1–4 can score up to two each as both right and left sides may be involved). The range is from 0 to 9. Hypermobility is defined as a total score of four or more.

†Derived from ref. [4].

TABLE II
Bulbena criteria for hypermobility*

Upper arm
1. Thumb: Passive apposition of the thumb to the flexor aspect of the forearm at <21 mm
2. Metacarpophalangeal: With the palm of the hand resting on the table, the passive dorsiflexion of the fifth finger is $\geqslant 90°$
3. Elbow hyperextension: Passive hyperextension of the elbow $\geqslant 10°$
4. External shoulder rotation: With the upper arm touching the body, and the elbow fixed at 90°, the forearm is taken in external rotation to $>85°$ of the sagittal plane (shoulder line)

Lower extremities—supine position
5. Hip abduction: Passive hip abduction $\geqslant 85°$
6. Patellar hypermobility: Holding with one hand the proximal end of the tibia, the patella can be moved well to the sides with the other hand
7. Ankle and feet hypermobility: An excess range of passive dorsiflexion of the ankle and eversion of the foot can be produced
8. Metarsophalangeal: Dorsal flexion of the toe over the diaphysis of the first metarsal is $\geqslant 90°$

Lower extremities—prone position
9. Knee hyperflexion: Knee flexion allows the heel to make contact with the buttock
10. Ecchymosis: Appearance of ecchymoses after hardly noticed, minimal traumatism

Hypermobility scoring system: using one point per item (overall range 0–10); males classified as hypermobile by a score >4 and females classified as hypermobile by a score >5. The non-dominant side is scored.

*Derived from ref. [18].

RESULTS

Twenty-nine (35%) of the 82 patients with STR fulfilled criteria for generalized hypermobility according to the Beighton and/or the Bulbena criteria. If those who reported having been double-jointed previ-

ously were included, then 42 (51%) of the patients in this study were currently or had been hypermobile. However, for the purpose of the study, only those currently hypermobile were classified in the hypermobile group. No patient was diagnosed with an inherited connective tissue disease known to induce joint laxity.

Demographic data and clinical characteristics of the patients meeting the Beighton and/or Bulbena criteria (hypermobiles) and those not meeting these criteria (non-hypermobiles) are shown in Table III. Hypermobile individuals were younger than non-hypermobiles with a mean age of 44 yr *vs* 52 yr, respectively ($P < 0.01$). There were 27 female patients (93%) in the hypermobile group and 45 (85%) in the non-hypermobile group.

Hypermobile individuals were more likely to have experienced at least one prior episode of STR than the non-hypermobile group (90% *vs* 51%, $P < 0.001$) (Table III). Recurrent episodes of STR at the same site were also reported more frequently in hypermobile patients (72% *vs* 26%, $P < 0.001$). Of those who complained of recurrent episodes of STR, 20 (69%) of the hypermobile group reported previous symptoms at multiple anatomical sites, whereas only 20 (38%) of the non-hypermobile group had experienced previous pain at multiple sites ($P < 0.01$). Thoracic outlet symptoms and symptoms suggestive of Raynaud's phenomenon tended to be more frequent in the hypermobile patients, but did not reach statistical significance. FM occurred with equal frequency in the two groups. There were no differences between the two groups in the frequency of past fractures or dislocations, and no differences in pregnancy-related problems among the female study subjects (data not shown).

Physical activities, including daily activity levels, exposure to repetitive activities, prolonged immobility and sporting activities during the 3 months prior to the onset of the present episode of STR are shown in Table IV. Repetitive activities included work in industry on a service line and office work at a keyboard. Slightly more patients in the hypermobile group reported exposure to repetitive activities (48% *vs* 21%, $P < 0.05$) and sporting activities (85% *vs* 64%, $P < 0.04$). There was no difference between the groups in the time spent on sporting activities. No patient was involved in competitive sport, and no patient was following a rigorous sport training programme. No patient was diagnosed with repetitive strain syndrome. The groups did not differ in the percentage of time spent either sedentary or up and about. There was no difference between the groups in the numbers performing tasks requiring prolonged immobility or forceful activities.

The frequency of hypermobile joints for those with or without STR at a given site is reported in Table V. Two groups are defined by the presence or absence of current STR, and the mobility status of individual anatomical sites is examined. The first group constitutes the total number of sites currently affected by STR. The second group was formed by pooling all the specific anatomical sites examined that were not currently affected by STR and recording the mobility status at each specific site. Localized hypermobility was present equally at the shoulder, hip and knee,

TABLE III
Characteristics of 82 patients with soft-tissue rheumatism grouped according to mobility status*

Characteristic	Hypermobile ($n = 29$)	Non-hypermobile ($n = 53$)	P
Age, mean yr (S.D.)	44 (14)	53 (11)	<0.01
Female sex	27 (93)	45 (85)	n.s.
Ethnic origin			
Caucasian	27	50	n.s.
Asian	2	3	n.s.
STR			
Any previous episode	26 (90)	27 (51)	<0.001
Recurrent at 1 site	21 (72)	14 (26)	<0.0001
Recurrent at $\geqslant 1$ site	20 (69)	20 (38)	<0.01
TOS	9 (31)	13 (25)	n.s.
Raynaud's phenomenon	7 (24)	8 (15)	n.s.
FMS by criteria	8 (28)	14 (26)	n.s.
Mobility score			
Beighton, mean (S.D.)	5.0 (2.0)	1.1 (1.1)	<0.0001
Bulbena, mean (S.D.)	6.2 (2.1)	2.3 (1.2)	<0.0001

n.s., not significant; DJ, double-jointedness; STR, soft-tissue rheumatism; TOS, thoracic outlet syndrome; FMS, fibromyalgia syndrome.

*Except where otherwise indicated, values are the number (%).

TABLE IV
Daily physical activities of 82 patients with soft-tissue rheumatism, grouped according to mobility status*

	Hypermobile ($n = 29$)	Non-hypermobile ($n = 53$)
Level of activity		
Sedentary, % of day	42	50
Up and about, % of day	58	50
Type of activity		
Repetitive activities, *n* (%)	14 (48)	11 (21)
Prolonged immobility, *n* (%)	11 (38)	17 (32)
Forceful activities, *n* (%)	15 (52)	20 (38)
Sporting activities, *n* (%)	25 (85)	34 (64)

*No statistically significant differences noted.

TABLE V
The frequency of hypermobile joints for those with or without STR at a given site

Anatomical site	STR present, hypermobile (%)*	STR absent, hypermobile (%)*
Shoulder	41	38
Elbow	75	37
Hand	50	31
Hip	9	10
Knee	56	62
Feet	67	21†

STR, soft-tissue rheumatism. Excludes patients with FMS who did not have a particular site of STR. Some patients had more than one site of current STR.

*Percentage of all individual sites examined.

†$P > 0.01$.

whether or not STR was currently present at that site. However, localized hypermobility was present more often in the small joints (elbow, hands and feet) if that site was currently affected with STR (75% *vs* 37%, 50% *vs* 31% and 67% *vs* 21%, respectively). These results reach significance only for the feet.

DISCUSSION

These results suggest that hypermobility was a common finding in this group of patients with STR. Thirty-five per cent of the patients satisfied the Beighton and/or Bulbena criteria for generalized hypermobility. This is most likely an underestimation in that mobility declines with age, and the present criteria for hypermobility do not allow for age adjustment. Thus, some who were hypermobile in the past may no longer fulfil the criteria for hypermobility. The significantly younger age of those with hypermobility probably reflects the loss of mobility with age. Despite this presumed underestimation, these results indicate an increased frequency of hypermobility in patients presenting with a primary complaint of STR to a rheumatologist, as compared to the established prevalence of generalized hypermobility of between 5 and 15% [2, 12, 14].

In patients with STR, those fulfilling the criteria for generalized hypermobility were more likely to have antecedent STR, either recurrent at the same site or at multiple sites. Since the hypermobile group was younger than the non-hypermobile group, they had less time to develop recurrent episodes of STR. Thus, this age bias would tend to make it more difficult to demonstrate this association, and strengthens the observation of the association of hypermobility with previous recurrent STR. Grahame *et al.* [13], reporting on a similar group of 80 patients selected for non-inflammatory rheumatic complaints, also showed a statistically significant increase in history of recurrent ligamentous injuries in their hypermobile group as compared to their non-hypermobile group. In contrast, Birrell *et al.* [16] was unable to show an association between joint pains and hypermobility in individuals screened in a West African community.

Although these results do not show any difference in the time spent carrying out physical activity between the two groups, hypermobile patients were more likely to be exposed to repetitive activities and sporting activities. Particular physical activities may be a factor in the development of STR in certain hypermobile individuals. The present study also explored more closely the relationship between the mobility status at individual anatomical sites and the presence of STR. Our findings suggest that there may be an association between STR complaints and hypermobility at small joint sites such as the hands, feet and elbows, but this was not observed in larger joints.

The concept of localized hypermobility as a factor in the development of STR, although recognized intuitively and reported anecdotally [19–22], may be more clinically relevant than an assessment of overall hypermobility, particularly in the context of a patient presenting with a specific site of STR. An important consideration in labelling an individual with generalized hypermobility is that the presence or absence of hypermobility neither reflects the degree of mobility nor the particular distribution of joints involved with hypermobility. It is understandable that a patient with hip and knee hypermobility may not be comparable to one with finger hypermobility. Furthermore, Larsson *et al.* [9] have reported that pauciarticular hypermobility is more common than generalized hypermobility, affecting 79% of females and 59% of males in a study of factory workers in Sweden. A correlation with back pain and hypermobility of the spine in industrial workers has recently been shown in Swedish workers [23]. In a further study of 660 musicians, Larsson *et al.* [24] concluded that hypermobility of fingers, thumbs and elbows undergoing repetitive activities was an asset, but hypermobility of joints required to provide support, such as knees and spine, was a liability. These individuals were, however, pre-selected on the basis of their musical accomplishments and may not resemble individuals in the general population.

FM patients were included in this study as these patients complain of both widespread pain and local pain at the sites of tendon insertions, i.e. the trochanteric region at the thigh, anserine bursal region at the knee and the medial epicondyle of the elbow. Gedalia *et al.* [10] reported a strong association of joint hypermobility and FM in schoolchildren. We have reported a higher frequency of FM as a primary rheumatological diagnosis in a general rheumatology population selected for hypermobility [2]. Eight of 22 (36%) FM patients in the present study were hypermobile, suggesting an increased frequency of lax ligaments in individuals with FM compared to the population.

In summary, the high rate of generalized hypermobility observed in patients selected for STR as compared to that observed in the general rheumatology population, as well as the recurrent nature of the STR in hypermobile patients, suggest that hypermobility may be an important factor in the development of STR complaints. Although the development of STR is probably multifactorial, the substantial number of sites affected by STR, and observed to be hypermobile suggests that this association is of clinical importance. Repetitive activity may be a contributing factor in some hypermobile individuals. These findings suggest that hypermobility may be at least one risk factor in some individuals in the development of STR complaints.

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