Undifferentiated Seronegative Spondyloarthropathy in Females

Sir—I read with great interest the paper by Uppal et al. [1] dealing with differences in the clinical profile of unclassifiable seronegative spondyloarthropathy between male and female patients in Northern India.

The Indian group has the great merit of having been among the first, in the early 1980s, to draw attention to the high frequency of forms of seronegative spondyloarthropathy (SpA) failing to meet criteria for the definite categories of the group [2, 3]. Subsequently, the concept of unclassifiable or undifferentiated spondyloarthropathy (uSpA) was widely accepted [4–6], and criteria for the classification and diagnosis of all the forms of SpA were suggested [7, 8].

I have ever been impressed by the low frequency of uSpA in females with respect to males in Northern India. In a previous paper, Malavija and Mehra [9] reported a male to female ratio of 16:1. The rarity of uSpA in females prompted the authors to collect a series of consecutive female patients to be compared with the more frequent male patients. The numbers of females were 17 in 1990 [9] and 25 in 1995 [1].

In Italy, uSpA is approximately as frequent in females as in males. As in Northern India, proper epidemiological studies on SpA are lacking. Owing to the absence of diagnostic criteria, uSpA has been ignored by the epidemiological studies performed in the past all over the world. Every time my group has studied a series of consecutive patients with uSpA, seen in rheumatic disease units in Pisa and Bologna, a low male to female ratio has been found. In the first study, we decided to follow up prospectively all consecutive child patients with the seronegative enthesopathy and arthropathy (SEA) syndrome without sacroiliac joint and spine involvement, which is a form of uSpA [10]. Seven boys and seven girls were seen in the period 1983–1990. The second study was undertaken in 1989, when we examined all consecutive adult patients with SpA seen in a 9-month period for the European Spondyloarthropathy Study Group work on preliminary classification criteria for the whole group of SpA [8]. A total of 109 patients with uSpA were seen in the study period in the seven participating European departments of rheumatology. Of these, 64.2% were males. The 1.8:1 male to female ratio was the same as that found in the 34 patients in our department.

Finally, more recently, we decided to study all consecutive patients with late-onset uSpA [11]. Twenty-three patients (12 females and 11 males) were seen in the 5 yr study period.

Boyer and co-workers [12, 13] have performed excellent epidemiological studies in populations of Alaskan Eskimos. uSpA, the most frequent form of SpA in Eskimos, is as frequent in men as in women.

There seems to be no conclusive explanation for the lower frequency of uSpA in Northern Indian compared to Italian and Alaskan Eskimo females. Both Pisa and Bologna rheumatic disease units are tertiary referral centres, like the Indian authors’ centre in New Delhi [14]. The clinical experience in uSpA of the two study groups is similar. The Indian authors suggested that the delay in diagnosis in females, due to the milder form of the disease and lower index of suspicion, was the reason for the high male to female ratio of uSpA in their country [1]. An alternative explanation was the less access or recourse to medical facilities of Indian women with respect to men. The same reasons could explain the different frequency of uSpA in Indian and Italian female patients. If the first explanation is the correct one, genetic and racial differences may play an important role [15].

I hope that proper epidemiological studies comparing the frequency and severity of uSpA in different populations, including Indians and southern Europeans, will be performed in the future.

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10. Olivieri I, Foto M, Ruji GP, Gemignani G, Giustarini S, Pasero G. Low frequency of axial involvement in Caucasian pediatric patients with seronegative enthesopathy and

Lymphatic Function in Inflammatory Arthritis

Sir—We read with interest the case, reported in a letter by McRorie et al. [1], of a woman with congenital lower limb lymphatic hypoplasia in whom the development of RA resulted in the onset of lymphoedema in one leg and the exacerbation of lymphoedema in the other leg.

We have compared lymphatic drainage in the upper limbs of patients suffering from inflammatory arthritis with and without co-existent lymphoedema and found an impairment of lymph drainage only in the oedematous group [2]. As there was no reduction in lymph drainage in the patients with inflammatory arthritis, and no oedema, we conclude that inflammatory arthritis alone does not directly impair lymph drainage. We have therefore proposed that in patients with inflammatory arthritis and lymphoedema, there must be an additional unrelated factor impairing lymphatic function which primarily places them at risk of developing lymphoedema [2]. The case described by McRorie et al. [1] illustrates and supports this hypothesis well: the onset of RA resulted in a critical increase in local capillary filtration to an extent that could not be accommodated by the hypoplastic abnormal lymphatics, resulting in the onset of oedema in one leg and its exacerbation in the other.

Unfortunately, in the clinic, it is not possible to predict which patients with inflammatory arthritis are at risk of developing lymphoedema, unless lymphatic function is already known to be abnormal. Unlike the case described by McRorie et al., others describe the persistence of oedema after the resolution of arthritis (reviewed in [3]), suggesting that a prolonged increase in load (i.e. increased capillary filtration) through abnormal lymph vessels may induce further damage and lead to permanent oedema. To avoid this, we suggest prompt anti-inflammatory treatment to reduce capillary filtration (and hence the load on the lymphatics) beneath the oedema-inducing threshold, thereby preventing further damage to lymphatics, reversing oedema and preserving function in the affected limb.

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Flatfeet in Pregnancy

Sir—We report two cases of flatfeet developing for the first time during pregnancy; this caused significant and lasting disability in one patient.

A 35-yr-old woman presented in the third trimester of her second pregnancy with an acute onset of flatfeet. Her weight gain was ~3 stones, she had marked peripheral oedema and consequently had walked barefoot as she had found shoes uncomfortable. Her main symptoms were pain along the medial aspect of her feet on weightbearing and ill-fitting shoes (her shoe size increased from 39 to 41). Her symptoms were so severe that she had to use a wheelchair for several weeks. The patient had had several miscarriages (all more than a year before this pregnancy) and hyperemesis in her first pregnancy. Her cousin had also had flatfeet in pregnancy. At the time of writing, her pain had subsided into an intermittent ache and although her shoe size had returned to normal, she was only comfortable in supportive shoes, such as trainers.

On examination (9 months post-partum), she had significant loss of the medial arch of her foot. The rest of her joints were normal, with no hypermobility. Her pain slowly improved with physiotherapy, insoles and wearing supportive shoes; however, her flatfeet still persist. An MRI showed normal tibialis posterior tendons.

Our first patient put us in touch with a 38-yr-old woman who had developed pain in her feet on weightbearing at 30 weeks gestation. Her weight gain was over 4 stones. After delivery, she noticed that the shape of her feet had changed and she was no longer able to wear her old shoes. Her symptoms gradually improved, although 3 yr later her feet were not back to normal. This was her first successful pregnancy: she had had numerous miscarriages (ranging from 6 to 26 weeks gestation), cause unknown. Her most recent had occurred 3 months prior to this pregnancy and had been a 20-week twin pregnancy. She had suffered long-standing back ache and in 1981 had had bilateral osteotomies for bunions. On examination (3 yr post-partum), she had bilateral flatfeet and no evidence of joint laxity.

A Medline search from 1966 to the present day did not reveal any case reports of flatfeet in pregnancy. However, the fact that our first patient knew two other cases personally suggests that the condition may be quite common. The problem may often be so minor