Letters to the Editor

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Recurrent tenosynovitis in Sweet’s syndrome

Sir, An uncommon association of inflammatory polyarthritis is acute febrile neutrophilic dermatosis or Sweet’s syndrome. This has the four cardinal features of fever, peripheral neutrophilic leucocytosis, painful plaque-forming inflammatory papules and a diffuse dermal neutrophilic infiltrate without vasculitis [1, 2]. There may be ocular, central nervous system, pulmonary, hepatic and renal involvement as well as oro-genital ulceration. Up to 62% of cases [3, 4] have musculoskeletal manifestations including myalgia, arthralgia and arthritis. Of the cases of arthritis described in the literature [4–6], there are no reports of associated tenosynovitis. The cause of Sweet’s syndrome is unknown but it has an increased incidence in a number of conditions, particularly infections, haematological malignancies and solid tumours, inflammatory bowel disease, other autoimmune diseases and with certain drugs [3, 4].

We report a case of recurrent tenosynovitis in Sweet’s syndrome, in a middle-aged woman who presented in the city where the first eight cases were originally described by Robert Douglas Sweet in 1964.

A 50-yr-old Caucasian barmaid was referred to the Dermatology Clinic, with a history of two episodes of painful skin lesions with an associated polyarthritis over the preceding 3 months. Each attack was characterized by the sudden onset of myalgia in the upper arms, thighs and calves, followed 24 h later, by arthralgia in her shoulders, knees and ankles with the development of swelling in her wrists and ankles, after 3 days. The rash appeared 1 week after the onset of her symptoms, initially as discrete, erythematous macules over her shoulders, the extensor aspect of her arms, thighs and shins. Blisters gradually formed in the centre of the macules, with fresh lesions continuing to develop for approximately 7 days and the rash lasting about 2 weeks. There were no subcutaneous nodules. The joint symptoms lasted 10 days. Her only regular medication was oestradiol which she had been taking for at least 2 yr. She smoked 20 cigarettes per day. She had had no symptoms suggestive of infection prior to the onset of each attack. Her past medical history was unremarkable apart from a Billroth II gastrectomy performed at the age of 39 yr, for recurrent peptic ulceration.

On examination during the second attack, she was febrile with a temperature of 38°C. She had a capsulitis of the right shoulder with painful, restricted movements. Over the extensor surface of the left wrist there was a tender swelling, consistent with an extensor tenosynovitis. There was redness, tenderness and swelling over the left tibialis posterior tendon. Fading, erythematous macules were present over the extensor surface of the upper arms and thighs. In addition there were discrete, tender, inflammatory papules and pseudovesicles over the upper arms (Fig. 1), thighs and shins. There was no oral or genital mucosal ulceration. She had no eye inflammation. The remainder of the examination was normal.

Investigations showed a total white cell count of $12.4 \times 10^9/l$ with a neutrophil count of $9.3 \times 10^9/l$. ESR was 38 mm/h. Serological tests for parvovirus, enterovirus and coxsackie virus were negative. ANF, RF, pANCA and cANCA were negative. Full blood count,

![Fig. 1. Upper arm showing inflammatory papules and pseudovesicles typical of Sweet’s syndrome.](https://academic.oup.com/rheumatology/article-abstract/41/9/1067/1784036)
liver function tests, uric acid, complement levels, immunoglobulins and protein electrophoresis were normal. Bence Jones protein was negative. Urinalysis was normal. No abnormality of the involved joints was demonstrated on plain X-ray. Joint scan showed minimally increased uptake in the right knee and right foot but the appearances were not suggestive of synovitis. Ultrasound scan of the left wrist was in keeping with a hyperaemic tenosynovitis of the extensor tendons (Fig. 2). Fluid was present around the left tibialis posterior tendon. Biopsy of one of the skin lesions was typical of Sweet’s syndrome with a mid-dermal dense inflammatory infiltrate consisting of large numbers of polymorphs. There was endothelial swelling but no true vasculitis. The papillary dermis was grossly oedematous with early vesicle formation. Chest X-ray and ultrasound scans of the abdomen and pelvis were normal.

Treatment was commenced with dapsone in a dose of 100 mg per day. However, she continued to have further episodes of skin eruptions with myalgia, arthralgia and tenosynovitis approximately every 2 months, her last attack being associated with tenosynovitis of the left tibialis posterior tendon and the flexor tendon of the right index finger. Colchicine was added to the dapsone as the patient was reluctant to take corticosteroids.

Sweet’s syndrome forms part of a spectrum of neutrophilic dermatoses which include pyoderma gangrenosum, subcorneal pustular dermatosis and erythema elevatum diutinum [7] and may occur in conjunction with erythema nodosum [8]. It is thought to be a hypersensitivity reaction to a bacterial, viral or tumour antigen. T-lymphocyte activation by cytokines and haemopoietic growth factors may be involved [3]. Studies of neutrophil function have not shown a consistent abnormality [4]. In the context of an acute arthritis, it is frequently confused clinically with a cutaneous vasculitis. Behcet’s disease is another important differential diagnosis. Most cases respond to corticosteroids. Other previously reported treatments include indomethacin, naproxen, sulphaspyridine, potassium iodide, clofazimine, dapsone, isotretinoin, methotrexate, chlorambucil cyclophosphamide, cyclosporin, interferon \( \alpha_2 \) and pulse methylprednisolone [3].

This patient demonstrates many of the manifestations of Sweet’s syndrome with a recurrent, self-limiting, painful, blistering rash occurring in a middle-aged female in association with a fever, myalgia and a migratory, asymmetrical, polyarticular, large joint arthritis. The histology of affected skin, a peripheral neutrophilic leucocytosis and an elevated ESR are also typical. However, a significant clinical feature of her episodes has been tenosynovitis at multiple sites. This has not previously been included in descriptions of classical Sweet’s syndrome, although tenosynovitis is recognized in the bowel bypass syndrome [9]. Histological findings in this condition are the same as in Sweet’s syndrome. Bowel-associated dermatosis–arthritis syndrome has been used to describe the Sweet’s syndrome which is found in bowel bypass syndrome and other bowel conditions, including ulcerative colitis, Crohn’s disease and following partial gastrectomy [10], including Billroth II gastrectomy for peptic ulceration [11].

We have not been able to identify any obvious underlying association with this patient’s Sweet’s syndrome. We considered the possible role of her Billroth II gastrectomy but post-operatively she remained well, with no obvious enteropathy. The onset of her current symptoms occurred 11 yr after her surgery, making any causative link unlikely. The syndrome has been reported in a patient on an oral contraceptive containing ethinyloestradiol [12]. Our patient had been on this preparation for several years prior to the development of the syndrome, again making this an unlikely association. Initial investigations have not unearthed a haematological malignancy or occult carcinoma. It is likely that this patient’s Sweet’s syndrome is idiopathic.

A review of the current literature, including Medline and Embase searches, has failed to reveal reports of any similar cases.

We report this case to highlight the previously undescribed feature of tenosynovitis occurring in association with an unusual cause of inflammatory arthritis, Sweet’s syndrome.

A. M. E. Brown, M. G. Davies, P. Hickling
Departments of Rheumatology and Dermatology, Derriford Hospital, Plymouth, Devon PL6 8DH, UK
Accepted 7 March 2002
 Correspondence to: A. M. E. Brown, Royal Devon and Exeter Hospital, Barrack Road, Exeter EX2 5DW, UK.

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Back pain

SIR, Mounce [1] is to be congratulated on her concise editorial on the complex subject of back pain. I am grateful for the opportunity to make some comments.

The article referred to low back pain and should have had that as its title. It made no reference to neck pain (with or without referral to the interscapular areas) or to pains arising from the thoracic spine.

Benefit payments have actually declined in the UK since 1995. Thus the number of working-age recipients of UK invalidity or incapacity benefits for back incapacities (irrespective of site in the spine) fell from 381,000 in 1995 to 308,000 in 2000 (Fig. 1).

Red flags remain the bible of spinal screening, but they are not infallible. Symptomatic osteoporosis is not always generalized, is not associated with constitutional symptoms and may not be picked up on blood tests. Vitamin D deficiency is frequently seen in the presence of an elevated parathyroid hormone concentration but with normal calcium and alkaline phosphatase levels in north-west London. Metabolic bone disease is, however, the largest single diagnostic group (other than mechanical/degenerative back pain) seen in rheumatology back clinics, at least where there is a strong ethnic minority population [2]. In our large study of 667 consecutive referrals to a large rheumatology service in north-west London between 1994 and 1996, 51 of the patients (8%) had identifiable causes of low back pain. The largest subgroup of 15 patients had metabolic bone disease [2].

The term ‘sciatic pain’ was not defined by Mounce. Many prefer the use of ‘leg pain’ to describe patterns of referral into the leg [3, 4], as they would differentiate between the pains of sciatic root irritation, femoral root irritation and referred pain into the leg. In our series of 538 patients with mechanical/degenerative low back pain, 144 patients had pain referred below the knee without neurological signs and 74 had neurological signs, only 35 of which related to nerve compression confirmed with imaging [2]. Our experience did not confirm Mounce’s view that the most common cause of ‘sciatica’ is nerve compression. I accept that pain from sciatic roots may arise from chemical rather than mechanical causes. Our patients with radiating leg pain were significantly more disabled (Roland score, \( P < 0.001 \)) and depressed (modified Zung score, \( P < 0.05 \)) than those without radiating leg pain [2].

I entirely support the view that ‘person management’ is vital in facilitating adjustment and compliance and that rheumatologists are poorly trained in helping those for whom the medical model of disease is unhelpful [5].

‘Medical interventions appear to have little effect on work resumption . . .’ needs clarification. The subject has been reviewed authoritatively by the Faculty of Occupational Medicine, who stated [6]: There is moderate evidence that, for the patient who is having difficulty returning to normal activities at 4–12 weeks, changing the focus from purely symptomatic treatment to ‘back school’ type of rehabilitation programme can produce faster return to work, less chronic disability and less sickness absence. There is no clear evidence on the optimum content or intensity of such packages, but there is generally consistent evidence on certain basic elements—education, reassurance and advice, exercise, pain management, vocational rehabilitation in an occupational setting and rehabilitation.

The principles of rehabilitating individuals back into work have been reviewed recently and are appropriate for those with back pain [7, 8].

It is important to recognize that ‘pain management’ and ‘rehabilitation’ are converging, with the realization that pain management is facilitated by assisting the patient to focus on goals, e.g. improved leisure or returning to work. I entirely endorse Mounce’s recognition of the value of cognitive behavioural therapy (CBT), but it is the incorporation of CBT with education, exercise and vocational rehabilitation that many now believe is the way ahead for those disabled by low back pain.

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Hydrotherapy has had and has a rationale

Sir, Reilly and Bird’s [1] review of promising results of hydrotherapy such as swimming is tempered with scientific scepticism. Does this scepticism apply to all scientists?

By 1653 Olof Rudbeck in Uppsala, according to the Finnish physiologist Tigerstedt’s translations (Latin original at British Medical Association library in London) into Swedish [2], had found ligation of Asellio’s valvular lymphatics to induce swelling of the (lymph) glands and other tissues from which they stem. Familiar with Harvey’s circulation of blood from arteries to veins through invisible connections [2], Rudbeck found that hepatic lymph was filtered by some mechanism from blood, but he considered the function of lymph as yet to be shrouded in haze.

That haze had been dispelled by the time Ernest Starling at Guy’s Hospital clarified that hydrostatic and colloid osmotic pressure differences across capillary walls regulate exchange of fluid between plasma and connective tissues [3]. Starling found dropsy to consist in the accumulation of lymph, his term for the interstitial fluid filtered from blood [4: iii]. He had sought in vain for evidence for the absorption of ‘proteid’ by blood and concluded that escaping protein not used up by tissue elements must be collected by the lymphatics and restored to the blood by the thoracic or right lymphatic ducts. He found no lymph flow in resting limbs [4: iii]. Acknowledging his contemporary Tigerstedt’s contributions, Starling found that activity of muscle is followed by arterial dilatation, more plentiful blood supply and increased leakage of lymph and supply of protein to the cells. He found that hypertrophy involving not only growth of individual cells, but also multiplication of cells, can be brought about by increased supply of nutritive material, especially protein [4: i].

By 1900 it was known that (i) increased leakage from capillaries not compensated by lymphatic drainage, enhanced by movement, results in oedematous swelling of tissues, and that (ii) work increases local blood flow and leakage. Both have a bearing on hydrotherapy.

(i) Fisher [5] may have been the first to study lymphatics of (rabbit) joints. Potassium iodide absorption, enhanced by movement, may have taken place in synovial capillaries or lymphatics, or both. A colloid (colloidal silver) after 5 h of free movement, stained a synovial mesh of lymphatics, and also popliteal and other glands.
In the treatment of arthritic swelling (non-infective and non-tuberculous) Fisher found that a combination of rest and skilfully applied but not excessive movement may prevent ankylosis. In Fisher’s camera lucida drawings synovial bulges seem to be separated by a mesh, slightly wider than rabbit arteriolar–venular loops, of collecting lymphatics. Normal villi lack lymphatics. The relationship between the microvascular loops, the parallel vessels (deep arteries?) in photographs of Hunter’s specimens, and lymphatics may not have been clarified; this might elucidate the mechanism of lymph flow increase at change of joint angle linked to synovial fluid (SF) pressure, and the influence of arterial pulsation on lymph flow.

According to a review of lymphatic function, initial lymphatics, often provided with leaflets that function as one-way valves, are anchored to the interstitium by filaments. It seems widely accepted that traction by the filaments upon increase of interstitial fluid volume opens the leaflets and results in filling of initial lymphatics. The latter are considered to empty into collecting valvular lymphatics by intermittent external compression and pressure change on movement, massage, arterial pulsation, etc. Submersion of a limb in water increases pressure on and in it. Constant pressure in head-out-of-water immersion decreased lymph flow in spite of high interstitial pressure. Intermittent compression of sheep hooves increased lymph flow, but flow increase per compression fell when the interval between cuff inflations was reduced to below 8 s. Active movement, and passive motion at a frequency of 10–100 per min, increased lymph flow, etc. Fast walking did not increase lymph flow more than normal everyday activity. The possibility that lymph from positive tissue pressure compartments such as the kidney and liver (swollen joints?) may be propelled into low-pressure ones could not be excluded. The review might be useful for those who plan hydro- and physiotherapy.

(ii) In 1743 William Hunter found it hard to believe that blood vessels could withstand the high pressures at loading of avascular joint cartilage, so he was not surprised by the large number of vessels in the soft (synovial) articular membrane. He could ‘not help observing that the Distribution of Blood-vessels to ... Cartilages ... seems calculated for obviating great Inconveniences’. He did not know if the vessels serve for nourishing (cartilage) only, or if they also ‘pour out a dewy (synovial) Fluid’. Both deductions were correct.

Recent data indicate that loading of cartilage increases the rate of processes involved in cartilage resistance to pressure, that may include protein synthesis. Many of these processes require energy. Normally glycolysing cartilage might, at low SF glucose, start to oxidize fuel, possibly including amino acids. In active muscle, Starling found energy demands to be met by vasodilatatory increase of capillary pressure and increased escape of (fluid and) protein. The conclusion agrees with a novel two-pore theory.

To sum up, (i) compression that increases tissue pressure and lymph flow occurs in any hydrotherapy that involves intermittent vertical movement of limbs in water. Swimming adds to this the exercise of muscles. The crawl and backstroke (at a slow pace?) ought to change pressures in forearms and hands more than breaststroke, and the former might spare neck (and low back?) joints. Effects on lymph flow of swimming, cheap and liked by patients, ought to be quantified. (ii) In many types of exercise, the cartilage of many joints is loaded more than in swimming. Does swimming minimize secondary vasodilatation in the synovium?

British–Scandinavian co-operation might possibly benefit rheumatoid patients even more than it does today if attention were also paid to physiological research in this part of the world. Hunter has priority on lymphatic ligation-induced swelling, but this is, according to rather reliable Finnish and Swedish parish registers, also a homage to one of my many forefathers. Hydrotherapy theory might have evoked more interest had Starling not, because of bad timing, lost a Nobel prize for which he was twice a candidate.

J. AHLQVIST

Sibbvik, FIN-25830 Västanfjärd, Finland

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A possible novel mechanism of opportunistic infection in systemic lupus erythematosus, based on a case of toxoplasmic encephalopathy

Sir, Opportunistic infection is common in patients with systemic lupus erythematosus (SLE). In some patients, it is difficult to distinguish between the effect of infection and exacerbation of SLE because both can produce similar symptoms. Toxoplasma infection (toxoplasmosis) is generally benign in healthy persons, with a tendency to chronic latency. However, activation of toxoplasmosis in patients with human immunodeficiency virus (HIV) infection or organ transplantation may have serious consequences. There have been many reports of toxoplasmosis in SLE patients, with conditions such as cerebritis and pericarditis mimicking SLE manifestations. In addition, seropositivity for *Toxoplasma gondii* is more common among SLE patients than control individuals.

In September 1999, a 24-yr-old woman was admitted to our hospital with fever, arthralgia, lymphopenia and proteinuria (3 g/day). She also showed several immune abnormalities [antinuclear antibody (ANA) >1280, normal (n) <40; anti-DNA antibody >400 IU/ml, n <10.0; CH50 15.1 U/ml, n = 30–40]. The CRP level was 1.3 mg/dl (n <0.3) and the ESR was 80 mm/h. SLE was diagnosed and prednisolone therapy was started from 60 mg/day. This was effective for her SLE-related symptoms and laboratory abnormalities. There were no abnormalities on her brain computed tomography (CT) at admission. After discharge, the steroid dose was gradually tapered to 12.5 mg/day. In September 2000, she was re-admitted to hospital with a right homonymous hemianopsia, transient unconsciousness, and right hemiparesis. Brain CT revealed a giant mass in the left cerebral hemisphere (Fig. 1a). On admission, exacerbation of SLE was not suggested by immunologic findings (anti-DNA antibody <2.0 IU/ml, CH50 40.2 U/ml). Her CRP level and ESR were 16.3 mg/dl and 101 mm/h, respectively. Serum IgG antibodies, but not IgM antibodies, for *Toxoplasma* were positive (IgG 1200 IU/ml, n <5; IgM 0.1 IU/ml, n <0.7), while HIV infection was negative. The tumour was removed and the histological examination revealed an inflammatory mass with necrosis and infiltration of lymphocytes (mainly B cells) and histiocytes (Fig. 1b). She was diagnosed as having toxoplasmic encephalopathy, probably due to recurrence of chronic latent infection, and was treated with anti-*Toxoplasma* agents (sulphamonomethoxine and sulphadoxine-pyrimethamine). The steroid dose was increased to 60 mg/day in order to prevent an increase of intracranial pressure though her SLE activity was stable. Her symptoms subsequently improved, so anti-*Toxoplasma* agents were stopped and her steroid dose was tapered to 40 mg/day. She is now being followed as an outpatient without further exacerbation of SLE and/or toxoplasmosis.

In our patient, toxoplasmosis occurred when her SLE activity and steroid dosage were relatively low. Similarly, a previous case of toxoplasmic encephalitis also occurred when SLE was inactive. Furthermore, it has been reported that a high titre of *Toxoplasma* antibody is not correlated with any of the parameters used to monitor SLE, including ANA and anti-DNA antibody, or with the prior treatment. Recent evidence about the relationship between opportunistic infections and HIV infection is interesting with regard to these issues. Several reports have indicated that an increase of CD4+ T cells following intensive antiretroviral therapy (HAART) can induce the development of symptoms related to opportunistic infection.

Fig. 1. (a) CT findings. An arrow indicates the toxoplasmic tumour. (b) Haematoxylin–eosin and (c) anti-*Toxoplasma* antibody stainings of the inflammatory cerebral tumour (original magnification ×200).
such as retinitis in certain HIV-infected patients and that steroids may be effective for suppressing such symptoms [8, 9]. This may result from normalization of the immune response by a recovery of memory CD4+ T-cell numbers and activity against opportunistic pathogens. The CD4+/CD8+ T-cell ratio (CD4/CD8 ratio) generally decreases in patients with SLE as compared with normal individuals and the ratio increases with amelioration of the disease [10]. Although the ratio in our patient was decreased at the onset of SLE (CD4/CD8 ratio 0.13, n = 0.6–2.9), it was higher when toxoplasmosis occurred (0.44), reflecting the lower activity of SLE. The importance of immunosuppressive therapy, including steroids, has been noted with regard to the induction of SLE-mediated opportunistic infection [1]. In addition, the findings obtained in HIV-infected patients treated with HAART indicate that normalization of immunity in patients who have inactive SLE and are receiving relatively low doses of steroids and/or immunosuppressants may contribute to the manifestation of opportunistic infections, as the immune system of SLE patients may originally show hyper-responsiveness to non-self as well as self-antigens [11]. A similar course to that observed in our toxoplasmosis patient seems to empirically occur in SLE patients with other opportunistic infections such as cytomegalovirus infection [12]. Investigation of similar cases, especially SLE patients showing a severe decrease of the CD4/CD8 ratio like our patient, is required to clarify this novel concept of the relationship between SLE and opportunistic infections.


Department of Medicine, Department of Neurosurgery and Department of Pathology, Juntendo University Izu-Nagaoka Hospital, Izu-Nagaoka-cho, Tagata-gun, Shizuoka 410-2295 and Department of Internal Medicine and Rheumatology, Juntendo University School of Medicine, 2-1-1 Hongo, Bunkyo-ku, Tokyo 113, Japan

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Correspondence to: I. Sekigawa.


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Do patients receive appropriate information and treatment following bone mineral density measurements?

SIR, There is no policy for osteoporosis screening in the UK. It is generally recommended that bone density measurements be targeted towards high-risk groups [1] since it is not appropriate or effective to treat on the basis of clinical risk factors alone [2]. We offer an open access bone densitometry service measuring lumbar spine and total hip bone mineral density (BMD) using dual-energy X-ray absorptiometry (DXA). Patients are also asked to complete a risk-assessment questionnaire. Using information from the referral form, BMD measurement and patient questionnaire an individualized report is issued by a metabolic bone physician giving an interpretation of the BMD result and advice regarding management and follow-up. In approximately 10% of cases an appointment in the metabolic bone clinic is offered for detailed assessment on the basis of clinical need. In the remainder, the patient is advised to obtain the result of their scan from the referring physician. In cases where the referral is made from another hospital speciality, a copy of the BMD report is also sent to the patient’s GP.

Our objective was to evaluate the information given to those patients who did not attend the metabolic bone clinic and identify whether they received appropriate treatment and lifestyle advice.

We retrospectively identified 295 patients who had undergone BMD scanning over a 2-month period a year earlier. Patients were sent a covering explanatory letter, questionnaire and pre-paid envelope. Consent was implied if patients responded. Non-responders were sent a reminder at 1 month. The actual report form presents a graphical and tabular summary of the BMD results with free text comments and for the purposes of this audit we classified BMD results on the basis of the lower Z score from the two measured sites as: normal (Z > 1); borderline (Z = 1 to −2); and low (Z < −2). This simplified classification enabled comparison with the patient’s recollection of the results using kappa statistics.

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Two hundred and forty-three (82%) patients responded. 218 patient responses were suitable for analysis. One hundred and twenty-six patients (58%) had been referred by their GP, of these 118 (94%) had received their results, 112 (95%) via GP, four (3%) by a hospital doctor, three (2%) at the time of their scan. Of the remaining patients referred by a hospital consultant, 75 (82%) had received their results, 50 (67%) by hospital physician, 22 (29%) by GP, three (4%) by post. In total, 87% had received their results, 71% via their GP. Of the 189 patients who had received their results 20 (11%) were unable to recall the results given.

The comparison between the patients’ understanding of their results and the actual BMD, resulted in a kappa score of 0.43 (95% CI 0.31–0.55).

Medication for the prevention or treatment of osteoporosis was commenced in 72 patients, this was appropriate in all cases. However, 17 patients did not receive treatment when this had been advised and of these seven had also not received their results (kappa score of 0.83, 95% CI 0.75–0.90). Eighty-two percent of patients were still taking treatment at 1 yr.

One hundred and eleven (51%) reports advised lifestyle advice as being appropriate. Only 36% recorded receiving some form of lifestyle advice, and only 30% of smokers recalled receiving advice to stop smoking.

Our questionnaire-based study involved 218 patients attending for DXA scan over a 2-month period. We found that 87% of our study population had received their results, mainly through their GPs and that, of these, 64% correctly recalled their result. Patient demographics, understanding of results and treatment implementation did not vary depending on who had been responsible for delivering their results.

A limitation of this project was the difficulty in classifying BMD measurements to compare with the patient’s perceptions of their result. Our report forms take both T and Z scores into account in recommending treatment, together with the influence of risk factors acting independently of BMD such as family history of fractures. Therefore, it is difficult to categorize patients into simple groupings and we would not have expected to find more than a moderate agreement between BMD and the patient’s understanding of the measurement. Generally, treatment was commenced appropriately in the 75% of those in whom it had been advised. In cases where treatment was started without specific advice this was mainly preventative in nature in patients with osteopenia and therefore appropriate. We were particularly encouraged to find that 82% of patients who had started treatment were still taking it 1 yr later. This is in contrast to previous studies, which suggest that persistence with osteoporosis therapy, particularly HRT, is very poor [3].

We found that many patients with lifestyle risk factors for osteoporosis had not been advised about these, or did not recall receiving this advice. This may be due in part to the wording on the report forms. As a result of this audit we now report lifestyle risk factors and advice to modify these in a more consistent manner and with greater emphasis.

To our knowledge this is the first audit of an open access bone densitometry report system. We specifically evaluated patients’ understanding of their results, the treatment started and lifestyle advice received. We related this to the information given on the report forms. This has enabled us to assess the efficacy of an open access system and has highlighted areas where improvements could be made.

In conclusion, we believe the report form we have developed appears to be an effective way of delivering results of DXA scans to patients. As a result of this, treatment is in general being commenced appropriately and continued for at least 1 yr.

R. Kilding, R. Eastell, N. Peel

The Osteoporosis Centre, Northern General Hospital, Herries Road, Sheffield S5 7AU, UK
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Correspondence to: N. Peel.


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Bilateral recurrent focal myositis of gastrocnemius muscles after BCG vaccination

SIR, Focal myositis (FM) is a rare, benign, self-limiting inflammation of the skeletal muscle, of unknown aetiology, initially described by Heffner et al. [1] in 1977. It is typically characterized by a localized, painful swelling of the soft tissues without a systemic involvement, frequently mimicking a sarcoma or thrombophlebitic disease [2]. Infectious aetiology, traumatic or ischaemic events have been suggested to explain the focal nature of the muscle injury in predisposed subjects. The muscle MRI and muscle serum enzyme evaluation are useful tools for diagnosis, which must be confirmed by muscle biopsy examination [2–4]. We describe one case of a young man with recurrent episodes of bilateral localized FM of the gastrocnemius muscle after a BCG vaccination.

A healthy 22-yr-old man, 3 weeks after a BCG vaccination, developed a sudden pronounced exertional weakness, pain and swelling of the calves with referred increased serum level of CK and erythrocyte sedimentation rate (ESR). The patient was treated with non-steroidal anti-inflammatory drugs and methylprednisolone (8 mg/day).
Recovery was complete within a month and the patient stopped drug therapy. One year later, he experienced a further episode of acute calf pain. The EMG showed myopathic changes in the gastrocnemius muscle, bilaterally. Laboratory investigation showed a polyclonal hyper-γ-globulinaemia, increased CK serum level (350 IU/l; normal value 20–170) and circulating immunocomplexes, and ESR (27 mm/h). The patient was treated with methylprednisolone (4 mg/day) with a progressive clinical amelioration in a few weeks. The dosage of the drug was tapered and stopped within 1 yr. At the time, laboratory tests were normal.

At 27 yr of age, the patient was admitted to our hospital for walking difficulty, bilateral calf swelling and pain. He denied a history of external trauma and had not had any constitutional symptoms such as fever, malaise or rashes. There was no personal or familial history of connective tissue disorders. Clinical examination showed a tender enlarging mass of the postero-lateral region of the calves, more pronounced on the left side. The subcutaneous tissue did not appear to be involved in the lesion; no regional lymph node enlargement was found. Neurological examination showed difficulty in walking on tip-toe. Laboratory investigation showed an increased level of: CK (2813 IU/l; normal value 20–170); aldolase (20 U/l; normal value 1.0–10); SGOT (52 U/l; normal value 5–40); SGPT (70 U/l; normal value 5–40). The ESR was increased (26 mm/h). The Tyne test was strongly positive. The following tests were negative or normal: electrophoresis and renal function, rheumatoid factor, antinuclear, anti-Scl 70, anti-SSA, anti-SSB, anti-Rnp, anti-Jo-1, anti-DNA and anti-Sm antibodies. Serology for trichinosis, echinococcosys, cysticercosis, toxoplasmosis, Lyme borreliosis, hepatitis and common viral infections were all negative. The chest radiograph was normal. Electromyographic examination only showed myopathic changes with slight denervation in the gastrocnemius muscle, bilaterally. Muscle MRI in the upper and lower limbs showed a focal lesion, with an increased signal on T2 weighted images, confined to the gastrocnemii (Fig. 1). A biopsy of the right lateral gemellus muscle was performed. Histological examination revealed a severe variation in fibre size with clusters of hypotrophic degenerating fibres and some hypertrophied fibres. Six to eight percent of nuclear centralization, some pale necrotic fibres, often undergoing phagocytosis, and several endomysial inflammatory cells, sometimes forming slight perivascular infiltrates, were observed. In addition, there was a slight increase in the endomysial connective tissue (Figs 2 and 3). Using a NADH-TR stain, some fibres showed areas devoid of activity. A combined treatment with methylprednisolone (8 mg/day) and methotrexate (10 mg/week i.m.) was started, followed by a complete regression of the mass lesion and normalization of inflammation indexes and CK level, within a few months. Two years later, clinical and laboratory data were normal. A further MRI examination showed residual gastrocnemius lesions and the EMG showed myopathic changes. At that time, drug...
therapy was stopped, and the clinical and serological remission persisted 6 months later.

In our patient the clinical and histological features, the muscle MRI and the EMG were consistent with the diagnosis of FM. The clinical course usually presents a high rate of spontaneous regression, but focal recurrence in other muscles or a progression to polymyositis have been reported [2, 3, 5–8]. Such evolution can be suggested by the early increase in serum level of the muscle enzymes as well as the acute phase reactants [2–8]. In the present case, the bio-humoral abnormalities were associated with a particular recurrent form, always confined to the gastrocnemii. Although the majority of previously reported cases were carefully screened for an infectious cause, the aetiology of FM remains unknown.

A study using the polymerase chain reaction failed to find any viral agents [9]. Recently, a case of FM caused by *Campylobacter* infection has been reported [10], and a possible infectious trigger such as *Borrelia burgdorferi* has also been described [11]. The BCG vaccination has been suggested as a possible trigger for dermatomyositis [12, 13], but not for FM. In the reported cases [12, 13], muscle symptoms began some weeks after vaccination. In the present case, there may also be a significant temporal relationship between the vaccination and the onset of symptoms. Therefore, our work could support the hypothesis that the BCG vaccination is a possible trigger for FM.

S. MANGANELLI, R. DE STEFANO, A. MALANDRINI1, E. SELVI, E. FRATI, S. GAMBELLI1, R. MARCOLONGO
Institute of Rheumatology and 1Institute of Neurologic Sciences, University of Siena, 53100 Siena, Italy

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Correspondence to: S. Manganelli.