

of peripheral neuropathy and the appearance of central nervous symptoms with coma. The patient condition worsened despite an intravenous bolus of cyclophosphamide (800 mg) and he died with global polyvisceral insufficiency. A post-mortem examination showed severe small and medium-size vessel necrotic vasculitis.

Case 2 was a 60-yr-old woman who had a blood transfusion in 1962 and had a diagnosis of hepatitis C-associated MC in 1992 after she presented with cutaneous purpura of both legs and arthralgia. Slightly elevated liver enzymes, type 1 genotype and a Knodell score of 7 on liver biopsy were present. A type II MC with low C4 (0.03 g/l) and CH50 (45%) levels were found. During the subsequent years, the patient received several treatments, including interferon (three courses), low doses of steroids, plasma exchanges, ribavirin. In July 2000, however, her physical condition worsened, with recurrent purpura, severe leg ulcers, sensitive motor neuropathy, and increased creatininaemia with persistent proteinuria (1 g/24 h). Since plasma exchanges were impossible at that time, and interferon unusable because of severe side-effects, cyclophosphamide was introduced but with no benefit. She then received, after informed consent, two injections (5 mg/kg) of infliximab at J1 and J15. No improvement was observed and this treatment was discontinued. Subsequently, despite corticosteroids and cyclophosphamide reintroduction, her condition deteriorated and she died a year later with severe renal insufficiency and cutaneous symptoms of vasculitis.

MC is the most frequent immune disorder associated with hepatitis C virus (HCV) infection [2], and may present in two clinical forms. The more common is a chronic vasculitis, affecting the skin, joints, peripheral nerves and kidneys, and is characterized by a mononuclear cell perivascular infiltrate [3]. The other type, reminiscent of hepatitis B virus-associated periarteritis nodosa, is an acute, life-threatening polyvisceral vasculitis with both mononuclear cell and neutrophil infiltrates, inducing vessel fibrinoid necrosis. Although both types of vasculitis are generally successfully treated with a combination of interferon, ribavirin, corticosteroids and plasma exchange [3], in some patients this treatment may be insufficient or cannot be fully conducted, thus other therapeutic strategies may be needed. Since we previously observed increase soluble TNF- $\alpha$  receptor concentrations in HCV-associated MC-related vasculitis, suggesting a role for this cytokine in the pathogenesis of this disease [4], we decided to use infliximab. Unfortunately, unlike in the case reported by Bartolucci *et al.*, this treatment appears at least inefficient. Although pathological data are not available, the patient reported by these authors, like our second patient, belongs to the first type of HCV-associated MC vasculitis; after a long-lasting hepatitis C, she had a chronic cutaneous and renal form of the disease and could not be given the classical treatment, but experienced a rapid but transient skin improvement 2 weeks after the first infliximab infusion. We did not observe such a response in our second patient, nor did we observe a favourable response in our first patient, who presented a severe form of HCV MC vasculitis. Noteworthy is the fact that, in this patient, severe periarteritis nodosa followed a chronic form of MC-induced vasculitis, a very uncommon and intriguing finding that led us to question the role of infliximab in such an event. Although infliximab may be efficient in certain types of vasculitis, we think our two observations suggest that caution is needed in using this treatment in hepatitis C-associated vasculitis, and suggest that other therapeutics such as anti-CD20 antibody may be more promising, as recently reported [5].

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### Acute-onset thigh pain

SIR, A 53-yr-old male Caucasian presented with a 12 h history of sudden-onset right hip pain, worse with movement, inability to weight bear and mild symptoms of urinary frequency. There was no history of recent travel, trauma or intravenous drug use. Past history included previous angioplasty for ischaemic heart disease, 56 units/week alcohol intake but previously 98 units/week, and seronegative RA treated with azathioprine and low-dose prednisolone for 8 years with excellent remission.

Examination revealed pyrexia 37.9°C and point tenderness over the right superomedial thigh and groin with absence of swelling or induration. There were no signs of active synovitis. Hip movements were globally restricted. Leucocytes were  $13.5 \times 10^9/l$  with neutrophils  $10.89 \times 10^9/l$  and haemoglobin 107 g/l. ESR was 147 mm/h, CRP 430 mg/l, creatinine kinase (CK) 15 U/l and urea and electrolytes was normal. Midstream urine, blood cultures (BC), and the Heaf test were negative. The right hip X-ray was normal. Isotope bone scanning revealed increased uptake in the inferomedial border of the right femoral neck as well as both knees, ankles, left elbow, left wrist and right shoulder. Subsequent MRI showed an abnormal bone marrow signal from the right femoral neck but no obvious features of osteonecrosis, synovitis or joint effusion. Surrounding soft tissue oedema in the iliopsoas muscle was, however, striking and compatible with presuppurative pyomyositis (Fig. 1).

He was treated empirically for pyomyositis with flucloxacillin 1 g qds intravenously for 7 days then oral maintenance. Clinical findings and symptoms resolved dramatically within 1 week, allowing full mobility. The ESR remained elevated at 133 mm/h but CRP improved to 43.6 mg/l. A repeat MRI 14 days later showed persisting signal disturbance in the right femoral neck, now involving the femoral head with features of early osteonecrosis and no joint effusion. The soft tissue changes in the iliopsoas muscle were significantly improved and largely resolved (Fig. 2).

Two months after presentation the patient had an excision arthroplasty of the right femoral head for the severe pain associated with the osteonecrosis, and he is awaiting right hip replacement. Histology of the excised femoral head, synovium and capsule showed no evidence of a septic inflammatory process and confirmed osteonecrosis of the femoral head. Nine BCs over 2 months, femoral head tissue and muscle aspirate all failed to culture a causative organism.

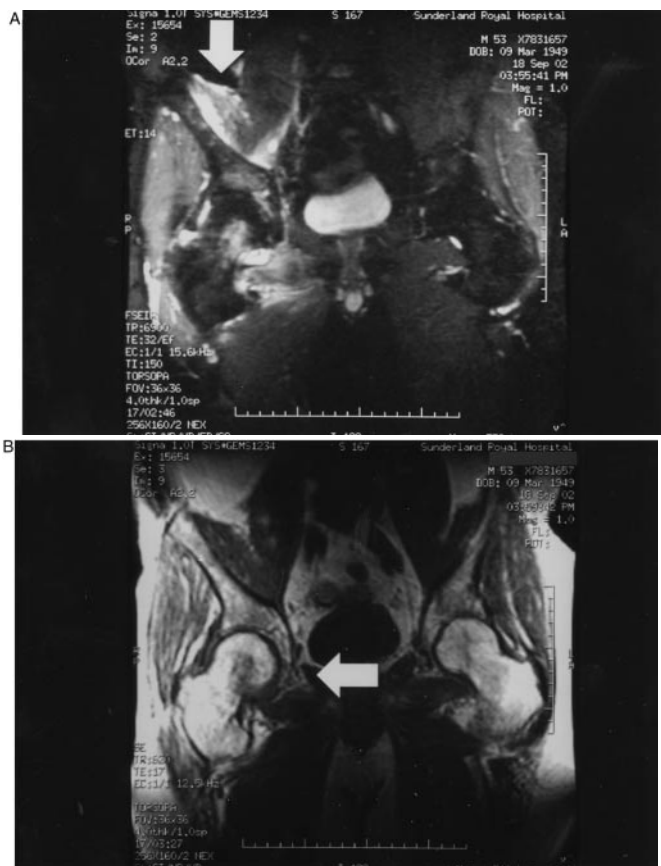


FIG. 1. (A) Coronal MRI demonstrating inflammatory oedema of the iliopsoas muscle (stir sequence). (B) Coronal MRI demonstrating normal femoral head, absence of synovitis of femoral head and abnormal femoral neck (T1 sequence).

Pyomyositis is a primary infection of striated muscle traditionally recognized in tropical climates [1–3], where it is endemic and can account for up to 4% of surgical admissions [1]. Pyomyositis is increasingly reported in non-tropical populations, where the spectrum of clinical presentation, although poorly defined, is recognized to differ from that of tropical pyomyositis [4].

In the tropics pyomyositis is associated with eosinophilia, caused by *Staphylococcus aureus* in more than 90% of cases, rarely affects individuals aged over 55 yr, and predominates in children and adults aged 20–45 yr. It has a good prognosis if diagnosed and treated [3].

In contrast, non-tropical pyomyositis consistently has an absence of eosinophilia, is caused by *S. aureus* in 65% of cases, can often affect older patients, and is associated with debilitating disease [4]. Specifically immunosuppressed states are associated with it, perhaps accounting for the increased reported mortality of 23% [4] and 7.7% [5] vs 0.5–2% in tropical pyomyositis [3].

Mortality increases with delayed diagnosis and the mean delay in diagnosis in the elderly or immunosuppressed states is 38.5 days [4], highlighting the importance of considering this condition in the at-risk population.

Pyomyositis can be divided into two clinical stages relating to abscess formation. The presuppurative stage is characterized by muscle pain, fever, malaise, raised inflammatory markers and leucocytosis. The affected muscle, if superficial, may be tender and have a firm, woody consistency but without a distinct mass [3]. Needle aspiration and culture at this stage does not reveal a causative organism [5]. The suppurative stage is heralded by fluctuance and a purulent collection, typically with a lack of local

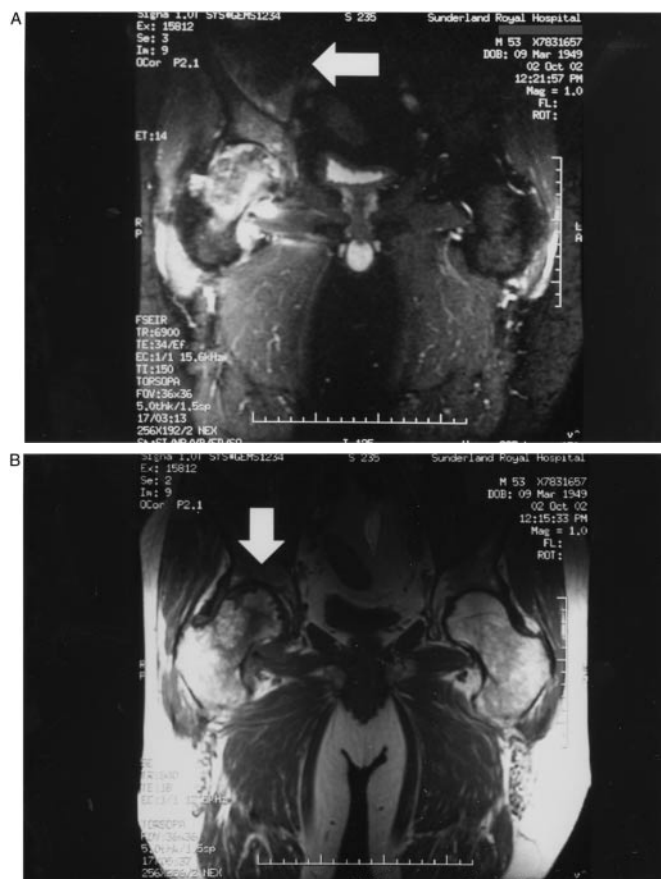


FIG. 2. (A) Coronal MRI demonstrating resolution of the inflammatory oedema of the iliopsoas muscle (stir sequence). (B) Coronal MRI demonstrating osteonecrosis of the right femoral head (T1 sequence).

lymph node involvement. This suppuration may remain covert if a deep muscle is involved, e.g. the iliopsoas [6]. If it is unrecognized, pyomyositis can progress to sepsis and its complications which are sometimes referred to as a third stage. Any skeletal muscle site can be involved but inflammation of a single muscle in the thigh (60%) is typical.

The commonest presentation in a review of 97 patients with non-tropical pyomyositis is a male (75%) aged 43 yr with fever, initial stiffness or inflammation of a single muscle in the thigh, and, if cultured, *S. aureus* is isolated [4]. Additional features include a normal CK and negative BCs.

The pathogenesis of pyomyositis is unknown. Immunosuppressed states are a risk factor but in addition the affected muscle may have localized immunosuppression as *Staphylococcus septicaemia* is rarely associated with muscle abscesses [7]. Preceding viral infections of muscle and trauma have been linked to pyomyositis. Nutritional deficiency, especially of thiamine [8], can result in a biochemical lesion of the pyruvate oxidase system, subsequently lowering skeletal muscle immunocompetence. This case had a history of sustained high alcohol ingestion, which may have led to nutritional deficiencies. Although BCs are typically negative, bacteraemia is a necessary step in pyomyositis and a decreased ability in immunosuppression to combat bacteraemia could facilitate muscle infection.

Treatment consists of appropriate intravenous antibiotics and drainage of abscess if present. There is previously reported experience of complete resolution with antibiotics alone in tropical pyomyositis if therapy is commenced at an early stage [3].

The imaging, histological, microbiological, operation notes and the typical lateness of extramuscular involvement with pyomyositis suggest that the associated femoral head destruction was aseptic. The iliacus inserts into the psoas muscle tendon, which inserts into the femoral neck. Perhaps inflammation at this site interrupted the blood supply to the femoral head. The osteonecrosis of the hip may also be unrelated to the pyomyositis, given the existence of independent risk factors for this condition, although this appears the least likely.

Diagnosis requires clinical suspicion highlighting the importance of considering pyomyositis in the context of immunosuppression and thigh pain. This case illustrates the usefulness of MRI, which is sensitive for detecting subtle fascial and muscle signal changes as well as staging osteonecrosis. MRI criteria for pyomyositis help differentiate other causes of muscle tenderness and swelling, and can be used to monitor therapeutic response [9]. MRI should be the imaging modality of choice in suspected pyomyositis, and in this case it confirmed osteonecrosis, which is previously unreported in association with pyomyositis.

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### Symptoms without pathology

SIR, Peter Croft's wise words [1] are slightly marred by his reference to the American College of Rheumatology criteria for fibromyalgia as diagnostic criteria rather than as classification criteria for reporting purposes [2]. There are no diagnostic criteria, as even the 'tender points' are circular reasoning and self-reported (and in many instances, learned). The sooner we abandon fibromyalgia as a diagnosis and treat the very real physical and psychological symptoms that characterize chronic pain, the better off we and the patients will be [3–6].

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### Reply: Symptoms without pathology

I accept George Ehrlich's gentle censure that the ACR criteria were intended to help standardize reporting and research and not to provide the basis for a clinical diagnosis.

My understanding of Professor Ehrlich's concerns, both from his letter and the referenced articles, is that 'labelling' the real distress of chronic pain sufferers as fibromyalgia has created a false sense of biological, pathological and aetiological certainty, which is not justified by the evidence and has done more harm than good.

I do not feel qualified to enter this particular debate, but, as my editorial indicated, I certainly believe that (i) current evidence suggests that tender points are not uniquely part of a chronic pain syndrome, and (ii) there is little evidence yet that eliciting tender points is a clinically useful exercise. The status of fibromyalgia as a syndrome of chronic pain and high tender point counts is, from this perspective, yet to be established.

More problematic for me is the question of whether getting rid of a label would make the treatment of 'the very real physical and psychological symptoms that characterize chronic pain' any easier or any more detached from the social and cultural influences that surround the experience and treatment of chronic pain in modern society.

In the 1970s, for example, the most widely used label in British general practice for the presenting symptom of low back pain was 'spinal osteoarthritis'. In the next decade or two, British general practitioners were persuaded of the spurious nature of this pathological label for most patients, and they reverted to the currently most frequently favoured label—namely 'low back pain'. However, during those same two decades, work absence and invalidity payments for back symptoms and conditions accelerated exponentially. Taking the pathology out of the label did not convert the problem to an acceptable part of everyday life.

Professor Ehrlich highlights an important problem. More empirical research on the influence of labels on patients' well-being and clinical outcome would be welcome.

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