Letter to the Editor

Successful response to infliximab in a patient with undifferentiated spondyloarthropathy coexisting with polyarteritis nodosa-like cutaneous vasculitis

Sirs,

The search for an effective treatment for patients with active and severe spondyloarthropathy is an important issue for rheumatologists (1). Recently, Brandt *et al.* showed the efficacy of the monoclonal anti-TNF-alpha antibody infliximab in patients with active and severe undifferentiated spondyloarthropathy (uSpA) (2). With this report we would like to demonstrate the efficacy of infliximab therapy in a patient with polyarteritis nodosa-like cutaneous vasculitis in the setting of uSpA.

In 1998, a 45-year-old man presented at the outpatient Rheumatology Clinic because of arthritis of the right knee. He also complained of a 6-month-history of early morning back pain and stiffness that was relieved by exercise, and a 7-year history of episodes of bilateral inflammation at the insertion of the Achilles tendon. The patient denied a family history of spondyloarthropathy, psoriasis or inflammatory bowel disease. He did not recall episodes of urethritis, conjunctivitis, uveitis or diarrhea. On physical examination reduced lumbar spine motion, right knee arthritis and bilateral Achilles tendinitis were observed. Arthrocentesis of his right knee yielded an inflammatory synovial fluid (4,250 leukocytes/mm^3 with 75% polymorphonuclear cells and 25% mononuclear cells) with normal glucose levels, absence of micro-crystals and negative cultures. ESR and C reactive protein were 51 mm/1st hour and 22 mg/L (normal < 5), respectively. Rheumatoid factor, antinuclear antibodies and HLA-B27 were negative. Plain pelvis radiograph did not confirm the presence of sacroiliitis. However, plain radiographs of the calcaneus showed bilateral spur formation and erosions (Fig. 1).

A diagnosis of uSpA was established (3). Treatment with indomethacin (150 mg/day) and sulphasalazine (3 g/day) was prescribed without success. Due to this methotrexate (17.5 mg/week) was added, but only partial improvement was obtained. Two years after the diagnosis of uSpA he developed palpable purpuric skin lesions on his legs. Biopsy of the cutaneous lesions disclosed a necrotizing arteritis involving small and medium-sized arteries with fibrinoid necrosis and inflammatory infiltrate in the artery wall. At that time, laboratory parameters including hepatic and renal function tests, rheumatoid factor, ANA, anti-DNA, C3, C4, anticardiolipin antibodies, cryoglobulins, and ANCA were negative or normal. Chest radiograph and abdominal ultrasonography were also normal.

A diagnosis of polyarteritis nodosa-like vasculitis was made and treatment with methotrexate (15 mg/week) plus infliximab (5 mg/Kg) was commenced. With this therapy rapid improvement of both cutaneous lesions and clinical symptoms of spondyloarthropathy was achieved. Eighteen months after the onset of this combination therapy, the patient remains asymptomatic on treatment with methotrexate (15 mg/week) and infliximab (5 mg/kg every 2 months).

Although the efficacy of anti-TNF-alpha therapy in ankylosing spondylitis and psoriatic arthritis has been proved, more studies are needed to confirm the efficacy of this therapy for other spondyloarthropathy subsets (4). We have recently reported a successful response to infliximab therapy in a patient with refractory mononeuritis multiplex due to rheumatoid vasculitis (5). A wide spectrum of vasculitic syndromes can involve the skin. Polyarteritis nodosa can be localized to the skin and subjacent tissues or, more commonly, can involve multiple organ systems. The presence of purpura might have led to a diagnosis of microscopic polyangiitis, as this vasculitis affects vessels smaller than arteries and causes purpura, while classic polyarteritis nodosa generally affects arteries in the skin causing nodules.

The presence of vasculitis prompted us to consider the use of infliximab in this patient with uSpA. The dramatic improvement of both the rheumatic manifestations and vasculitic complications observed in our case supports the potential use of anti-TNF-alpha therapy in uSpA patients with severe complications refractory to classic therapies.

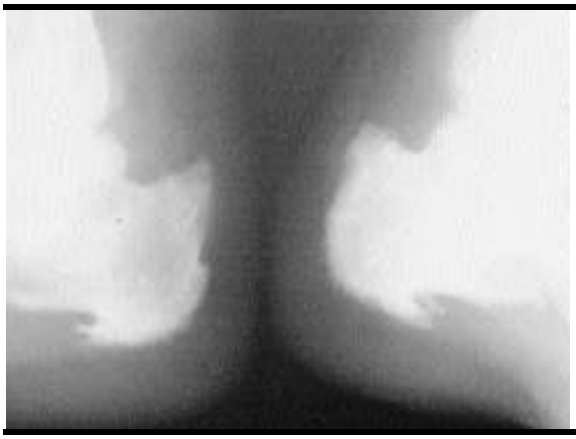

Fig. 1. Advanced changes in enthesopatic lesions: erosions and calcaneal spur.

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