Pseudoseptic arthritis in a patient with Behçet's disease

Sir,

Behçet's disease (BD) is a chronic, inflammatory disease characterized by oral and genital aphtae and iritis; additional clinical features include skin, joint, neurologic, vascular and gastrointestinal involvement (1-2). Here we describe a male suffering from BD who recently presented with bilateral gonarthritis with Baker's cyst, whose diagnosis resulted difficult, because the clinical picture mimicked septic arthritis.

In September 2005 an Algerian 40-year old man affected by BD was admitted to the Emergency Department with a three day history of acute left calf pain and swelling associated with severe difficulty in weight bearing. His past medical history was relevant for recurrent oral aphthae and genital ulcers. HLA typing was positive for HLA-B51 antigen, pathergy test was also positive, he was in treatment with colchicin (1 mg/day). Physical examination revealed fever of 37.8 °C, erythema, warmth and swelling of the left calf, and mild erythema of the knee. Laboratory investigation showed elevated C-reactive protein 82.8 mg/l (normal 0-5 mg/l) and mild leukocytosis (12.740 /mm^3). Ultrasonography excluded deep venous thrombosis and showed a large, ruptured popliteal cyst with fluid collection within the fascial compartments and gastrocnemius muscle, a small effusion in the suprapatellar pouch was also observed. Then an US guided aspiration of the cyst was made and 40 ml of white and cloudy synovial fluid were obtained. Microscopic analysis showed 83.000 white blood cells (WBC) /mm^3 with marked neuthrophilia (95%); gram-stained smear and analysis for crystals were negative. Culture of both synovial fluid and blood was negative. Treatment with i.v. ceftriaxone (2 g/day) and ciprofloxacin (200 mg bid) was started since septic arthritis was suspected. After 5 days a sudden, severe swelling of the right calf appeared; this was also warm and erythematous. For the second time a deep venous thrombosis was ruled out and a massive not ruptured Baker's cyst was diagnosed by ultrasonography (fig. 1). A new arthrocentesis was made with aspiration of 80 ml of cloudy synovial fluid. Microscopic analysis showed 95.000 WBC/mm^3 with marked neuthrophilia (97%); both gram-stained smear and analysis for crystals were negative. Pseudoseptic arthritis due to BD was diagnosed. The patient was treated with colchicin (2 mg/day), cyclosporin (150 mg/day) and steroid (6-methyl-prednisolone 12 mg/day). In a few days the arthritis improved and fever disappeared.

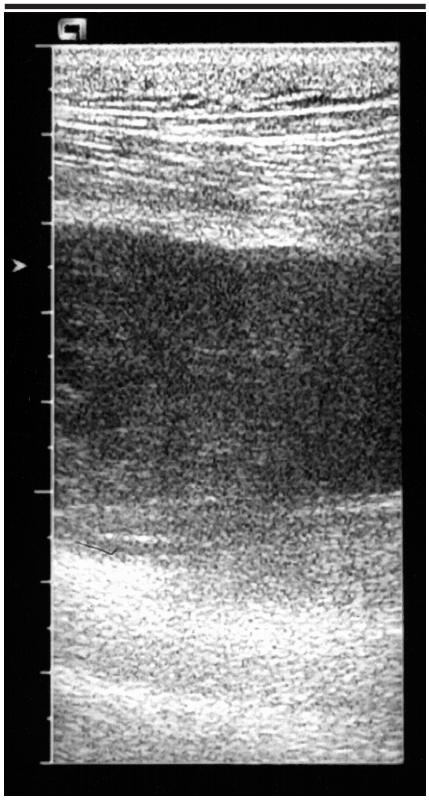

Fig. 1. Baker's cyst. Large hypoechoic effusion is immediately deep to the medial head of the gastrocnemius.

Pseudoseptic arthritis is defined as extremely inflammatory arthritis not due to bacterial infection (3). Several diseases are reported to cause pseudoseptic arthritis: rheumatoid arthritis, reactive arthritis, gout and pseudogout (3-4). Although some authors reported elevated cell count (up to 39.960 WBC/mm^3) in synovial fluid of patientes with BD (1, 5), to our knowledge no cases of pseudoseptic arthritis in BD have been previously reported. The very high leukocyte count in synovial fluid of both knees is an intringuing aspect because it parallels with other typical features of BD such as pathergy test, acne-like skin lesions and the increased neutrophil migration demonstrated by Senn's skin window (6).

These neutrophil-rich manifestations of BD overlap with those of autoinflammatory diseases. BD could be included among the autoinflammatory diseases (7); some clinical features comprising pseudoseptic arthritis here described recall a rare and inherited autoinflammatory disease, named PAPA syndrome, characterized by pyogenic sterile arthritis, pyoderma gangrenosum and acne. Neutrophil-rich sterile joint effusion represents a typical feature of PAPA syndrome as well as pathergy reaction (8). It is well known that autoinflammatory diseases share some clinical features; but, while the cutaneous manifestations of PAPA syndrome have alredy been reported in BD (2, 9-10), this is the first case describing the shared joint manifestation: pseudoseptic arthritis.

This case report shows that BD may be a cause of pseudoseptic arthritis.

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