

Brucellar arthritis of the knee: A case report with delayed diagnosis

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Brucellar septic arthritis has become extremely rare in Western developed countries since bovine brucellosis has been successfully eradicated. Nevertheless, we should remain vigilant for this disease, as brucellosis is still endemic in many parts of the world.

The authors present the rare manifestation of a brucellar septic arthritis of the knee joint, associated with a lytic lesion of the proximal tibia. The diagnosis of brucellosis was delayed by 11 months because of the microbiological and histological negativity of repeated joint fluid aspirations, bone biopsies and synovial specimens. Eventually Brucella melitensis could only be isolated from one extended culture of synovial fluid. The treatment with a combination antimicrobial therapy of rifampicin and tetracycline was successful, but the lytic lesion needed reconstruction with bone grafts.

A high index of suspicion for brucellosis is needed in any patient coming from an endemic region with a non-specific and chronic arthritis to allow for early diagnosis and treatment.

Keywords: Brucella; brucellosis; arthritis; lytic lesion; knee.

INTRODUCTION

Brucellosis is a zoonotic disease caused by a Gram-negative rod, first described by sir David Bruce in 1887. The disease is transmitted from infected animals, mainly sheep, goats and camels, by inoculation through conjunctivae, cuts and abrasions in the skin or by ingestion of non-pasteurised

milk and infected milk products (7). It is a systemic infectious disease with a broad spectrum of clinical presentation, in which a single swollen and painful joint is frequently observed as osteoarticular manifestation. The most common types of infections are sacroiliitis and arthritis of a peripheral large joint, mainly in the lower extremity (knee, hip or shoulder) (1,7,11), but more recently, infections around prosthetic implants have also been reported (2,8).

CASE REPORT

A 58-year old man, recently immigrated from the south-eastern region of Turkey, presented with a two-year history of mechanical pain with swelling and limited motion of the right knee without any history of trauma. He reported that he had been treated by multiple aspirations of clear viscous fluid with only temporary relief of his symptoms. Our investigation consisted in standard radiographs (fig 1) and magnetic resonance imag-

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Fig. 1. — Antero-posterior (a) and lateral (b) view radiograph showing a lytic lesion in the postero-medial part of the proximal tibia

ing (fig 2) showing an important hydathrosis, synovitis and a lytic lesion of the proximal tibia with semi-liquid density, appearing to be in connection with the insertion of the PCL and measuring $4.5 \times 4.0 \times 3.5$ cm.

Laboratory tests revealed a white blood cell count of 12.6×10^3 /mm³, C-reactive protein of 67 mg/L and erythrocyte sedimentation rate of 37 mm/h. Rheumatoid factor, antistreptolysin-O, antinuclear antibodies and circulating immune complexes all were negative.

At first, we performed a diagnostic arthroscopy, which showed an important and panarthritic nodular synovitis without the presence of chondromatosis (fig 3a) and we performed subtotal synovectomy. When performing an open biopsy of the tibial lesion, we fell into a large hole, filled with a turbid viscous and caseous liquid. When introducing our scope into the hiatus we also could see an important amount of inflammatory tissue (fig 3b).

Histopathologically, chronic synovitis corresponding to that of a rheumatoid arthritis was seen and there was no microbiological growth in the synovectomy specimens, nor in the synovial fluid samples.

In a second session, we performed a stabilisation procedure, filling up the tibial lesion with autologous cancellous bone, mixed with freeze-dried allograft bone (Tutoplast®) together with autologous platelet rich plasma. The patient however never became symptom-free after surgical treat-

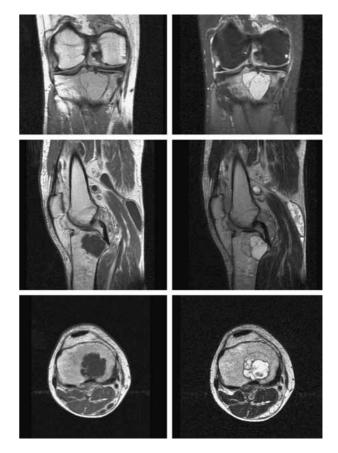


Fig. 2. — MRI-scan showing a generalised synovial hypertrophy with a cystic formation in the proximal tibia measuring $4.5 \times 4.0 \times 3.5$ cm. T_1 -weighted images in the left column and T_2 -weighted images in the right column.

ment and again had several periods of recurrent swelling and pain. Anti-inflammatory drugs were prescribed with some relief of the symptoms. Bone grafts incorporated fairly well radiographically (fig 4), although some osteolysis was seen on CT-scan at the tibial insertion of the PCL (fig 5). Again, several synovial fluid aspirations had to be done for symptomatic hydrarthrosis and all specimen cultures were negative, except for one, on which *Brucella melitensis* was isolated after extended culture.

The diagnosis of brucellosis was confirmed by the Rose-Bengal test (6), which was only positive in one of two repeated tests, with a one week interval. Blood cultures all remained negative as well.

We diagnosed brucellosis with a delay of 11 months, after which the patient was treated with

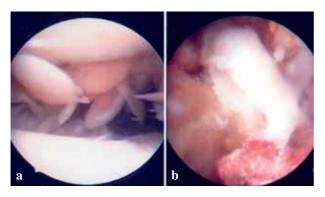


Fig. 3. — a) Arthroscopic view showing panarthritic nodular synovitis, without the presence of chondromatosis; b) Intracystic view with the arthroscope showing a cavity filled with a caseous liquid and surrounded by an important amount of inflammatory tissue.



Fig. 4. — Radiographs of the knee three months after operation: antero-posterior (a) and lateral (b) views showing the tibial cyst filled with bone grafts.

a combination of rifampicin (Rifadine®) and tetracycline (Doxycycline®) orally, with improvement of the joint symptoms within two weeks. The antibiotic therapy was continued for a total duration of 8 weeks with complete resolution of the knee symptoms, normalisation of the inflammatory blood parameters and a good bone-graft ingrowth on CT-scan (fig 6).

DISCUSSION

Brucellosis is still endemic in many parts of the world, especially in the Persian Gulf, Latin America and the Mediterranean countries (1,4,5,7,11). During

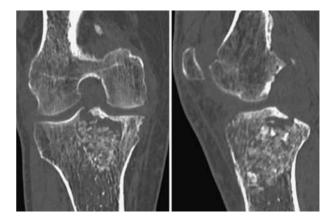


Fig. 5. — Follow-up CT-scan 5 months post-operative before antibiotic therapy was started, showing incorporation of the bone-grafts, although some proximal resorption at the insertion of the PCL is present.

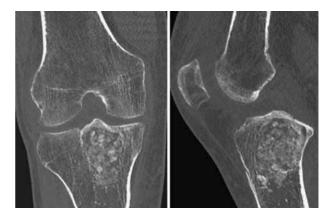


Fig. 6. — Follow-up CT-scan 8 months post-operative after antibiotic therapy, with good graft incorporation and reduction of osteolysis.

the last decade, about 9000 cases yearly were reported to the Turkish Ministry of Health (incidence 14/100.000) (6). In our Western developed countries, we have successfully eradicated bovine brucellosis by eradication programs, using a combination of vaccination, test-and-slaughter, surveillance and abattoir trace back. Pasteurising milk has made the human disease very rare, and it has thus become less well known by the practitioners.

Brucellar septic arthritis results from haematogenous spread to joints and seems to be able to locally extend into the metaphyseal bone with subsequent erosion. To our knowledge, such lytic lesions of the proximal tibia have only been described in a few reports: one case with a delay in diagnosis of more than two years (I2) and some cases due to an osteomyelitis of the proximal tibia (3,7). In publications of large series from endemic regions (1,4,11), the only radiographic appearance was a soft tissue swelling, but a lytic lesion was never reported. Earlier recognition and treatment in endemic regions can presumably avoid local progression.

The diagnosis of brucellosis may commonly be missed because of its rarity, its variable clinical presentation and, in some chronic cases, microbiological and serological negativity (1,11,13). To come to an earlier diagnosis, a high suspicion for *Brucella* infection should exist in any patient coming from an endemic region with a non-specific and chronic arthralgia. Some authors recommend to inoculate the synovial fluid into blood culture bottles (Castaneda bottles) and to alert the laboratory to keep the cultures for at least 3-4 weeks (13). Even with these measures, the sensitivity still is no higher than 60%.

The recommended treatment for brucellar arthritis is non-surgical with a combination therapy of doxycycline with streptomycin or rifampicin for 6 to 8 weeks (9). Recovery is mostly good and a relapse rate of only 2%-3.4% is seen, more frequently in spondylarthritis (4,10). We conclude that the diagnosis of brucellar arthritis in our regions appears to be more difficult than its treatment.

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