Rare disease

Brucella arthritis of the knee, 1 year after revision of anterior cruciate ligament reconstruction

S G Papastergiou, N E Koukoulas, P Koumis, D Kyparlis, R Santas

Department of Orthopaedics, Agios Pavlos General Hospital, Thessaloniki, Greece

Correspondence to Nikolaos E Koukoulas, nkoukoulas@yahoo.gr

Summary

Brucellosis is a zoonotic infection with a broad spectrum of clinical manifestations. The authors report the first case in the literature of septic arthritis of the knee 1 year after revision of anterior cruciate ligament reconstruction. *Brucella melitensis* biotype 3 was found in both synovial fluid and blood cultures. The patient was treated initially with arthroscopic debridement. After the diagnosis was confirmed, a second arthroscopic lavage and metal work removal was applied leaving the graft in place. Antimicrobial chemotherapy was prescribed for 3 months. The infection was fully eradicated and the patient is still asymptomatic, 4 years after the treatment.

BACKGROUND

Septic arthritis of the knee after anterior cruciate ligament reconstruction (ACLR) is an infrequent complication.^{1–4} The infecting pathogens most commonly implicated in knee infection after ACLR are staphylococci, streptococci and gram-negative rods.^{1–4} This is the first case reported in the literature of septic arthritis of the knee after ACLR caused by *Brucella melitensis*. Moreover, *Brucella* arthritis presents with special clinical characteristics that are discussed.

CASE PRESENTATION

A 33-year-old Greek man presented with a 1-month history of fever (temperature up to 38°C) and 4 days' inflammation of the right knee. He had undergone ACLR of the suffering knee 2 years ago (synthetic graft) and revision of the reconstruction (bone-patella-tendon-bone autograft) 1 year ago. Both operations were performed in an institution of a different country (Central Europe). Physical examination revealed redness, warmth and effusion of the knee joint without any signs of instability.

INVESTIGATIONS

Radiographic knee examination demonstrated the existence of one metallic screw in the femur and two metallic screws in the tibia (figure 1). Haematological and biochemical evaluation showed leucopenia (WBC=3.6 k/μl), elevated C reactive protein (CRP 7.5 mg/dl), elevated erythrocyte sedimentation rate (ESR 18 mm) and normal procalcitonin (PCT 0.1 ng/ml). Knee arthrocentesis of 150 cc haemorrhagic fluid demonstrated low density of synovial fluid in glucose (2 mg/dl). The patient was admitted to our orthopaedic department and he was initially treated with ampicillin (15 mg/kg/day/intravenous) and cefuroxime (3 g/day/intravenous). On the third day of hospitalisation, the patient underwent arthroscopic lavage and debridement. At arthroscopy, mild inflammation of synovium and intact anterior cruciate ligament graft were found. Further

investigation of the fever revealed positive serological Wright test with a titre of 1/800.

TREATMENT

Based on the Wright test, antibiotic therapy was changed to streptomycin (1 g/day/intramuscular), doxycycline (200 mg/day/po) and rifampicin (600 mg/day/po) for 3 weeks. In elevated fever, two sets of blood cultures were obtained. In the ensuing days, fever progressively decreased and knee pathological findings improved, but serological Wright test was positive in a titre of 1/3200. Both synovial fluid and blood cultures were positive in *B melitensis* biotype 3. Histopathology revealed diffusive inflammatory infiltration of synovium by polymorphonuclear lymphocytes, histiocytes and plasmacytes. Fever during his hospitalisation had an undulant form and the patient was afebrile 5 days after his admission. The patient was discharged after 10 days of hospitalisation under the diagnosis of 'Brucella arthritis of the knee'.

At follow-up, 3 weeks after the admission day, the patient was still afebrile and free of clinical symptoms of knee infection. Haematological and biochemical evaluation revealed leucopenia (WBC=3.9 k/μl), normal CRP (0.3 mg/dl), normal ESR (4 mm at 1st h) but the serological Wright test was still positive with a titre of 1/3200. At that time, a technetium 99m bone scan was performed to rule out any other bone and joint involvement. The scan demonstrated right knee inflammation, with no other osteoarticular manifestation (figure 2). Since the Wright test was still positive, the patient underwent second arthroscopic surgery and metal work removal. The graft was well incorporated into femur and tibia tunnels and left in place. Streptomycin intramuscular injection was discontinued and replaced by oral administration of sulfamethoxazole/trimethoprim (800+160 mg/day/po). This combination of antibiotics (sulfamethoxazole/trimethoprim, doxycycline and rifampicin) was applied for 3 months.

Figure 1 Anteroposterior and lateral radiographs of the knee.

Figure 2 Nuclear scan.

OUTCOME AND FOLLOW-UP

The patient was followed up every month for 6 months, at 9 months and every year after the admission day. There were no clinical signs of knee infection and all laboratory tests for infection were within normal values. Wright test became negative 1 month after the second arthroscopy. Four years after the onset of symptoms, the patient is still asymptomatic.

DISCUSSION

Brucellosis is a zoonotic infection that is transmitted from animals to humans by ingestion of infected food products, direct contact with an infected animal or organism, or inhalation of aerosols. Interestingly, our patient was working in a restaurant as a kitchen assistant. Of the six main species of *Brucella*, *Brucella melitensis* (from sheep) has the highest human pathogenicity. Frequency of brucellosis

varies across nations. The highest incidence is observed in the Mediterranean region, western Asia and parts of Africa and Latin America. Brucellosis is an endemic infection, but with the advent of easy international travel and the mixed population, *Brucella* arthritis of the knee may even present in a non-endemic country.⁵

Brucellosis is a systemic disease with a wide spectrum of clinical presentations. Bone and joint involvement is a frequent complication of brucellosis and the reported incidence ranges from 11% to 56%. Sacroiliac, hip and knee joints are the commonest osteoarticular manifestation.^{6,7} Any joint that has been previously damaged, such as by degenerative changes in elderly people, is more liable to infection.^{7,8}

Nevertheless, there are not many reports of joint arthritis following surgery. In fact, only seven cases of total joint replacement have been reported in the English literature, infected by *Brucella*.⁹ This is the first case of revision (or primary) ACLR reported in the literature to date, which is complicated by *Brucella* arthritis.

Besides the cause (*B melitensis*) of infection, timing of infection is also an interesting feature of our case. Postoperative infections are classified as either acute (<2 weeks), subacute (between 2 weeks and 2 months) or late (>2 months).⁴ The vast majority of the patients have acute or subacute intra-articular knee infections after ACLR. Late infections are difficult to be related to the previous procedures. Nevertheless, Van Tongel *et al*³ reported a case of streptococcal arthritis of the knee 15 months after ACLR. As a result, this is the second case reported in the literature with knee infection presented 12 months postoperatively. Moreover, *Brucella* arthritis after total hip or knee joint replacement has been reported to present late. In the reported cases, the median time from prosthesis implantation to onset of symptoms was 38.7 months (range 2 months to 14 years).⁹

In contrast to septic arthritis caused by pyogenic organisms, joint inflammation seen in patients with *B melitensis* is mild, and erythema of overlying skin is uncommon.⁷ That have been said, clinical diagnosis could be difficult when trying to rely on local symptoms of inflammation. Fever, classically undulant, is the most prevalent symptom that should alert the physician for brucellosis.⁷

After the diagnosis of *Brucella* arthritis had been established, we followed the antibiotic therapy recommended by the World Health Organization.¹⁰ Combination of intramuscular streptomycin (1 g/day for 2–3 weeks) gives fewer relapses whereas trimethoprim-sulfamethoxazole (TMP/SMX) is a popular compound usually used in triple regimens.¹¹

Septic arthritis after primary or revision ACLR is a rare but devastating complication. The infection rate has been reported to range from 0.14% to 1%.^{1–4} If antibiotic therapy and arthroscopic lavage fail to eradicate the infection, a two-stage procedure is required. Graft and metal

work (or other implants used for graft stabilisation) are removed in the first procedure and 6–9 months later, after the infection has been eradicated, a revision procedure is applied.¹² In revision, ACLR surgery potential difficulties are graft selection, bone stock, bone quality, graft stabilisation, postoperative regimen, infection and others.¹³ As a result, the importance of early and accurate diagnosis of septic arthritis following ACLR is mandatory in order to minimise the chances of revision surgery.

Learning points

- ▶ *B melitensis* could be the cause of septic arthritis following ACLR.
- ▶ *Brucella* arthritis presents with special clinical characteristics (late presentation, mild symptoms, undulant fever).
- ▶ *Brucella* arthritis diagnosis requires positive cultures and Wright test.
- ▶ Besides surgical intervention, special antibiotic therapy is mandatory for successful treatment.
- ▶ Since ACLR is a common orthopaedic procedure, the treating physicians should have a high index of suspicion for the condition.

Competing interests None.

Patient consent Obtained.

REFERENCES

1. Fong SY, Tan JL. Septic arthritis after arthroscopic anterior cruciate ligament reconstruction. *Ann Acad Med Singap* 2004;**33**:228–34.
2. Indelli PF, Dillingham M, Fanton G, *et al*. Septic arthritis in postoperative anterior cruciate ligament reconstruction. *Clin Orthop Relat Res* 2002;**398**:182–8.
3. Van Tongel A, Stuyck J, Bellemans J, *et al*. Septic arthritis after arthroscopic anterior cruciate ligament reconstruction: a retrospective analysis of incidence, management and outcome. *Am J Sports Med* 2007;**35**:1059–63.
4. Williams RJ 3rd, Laurencin CT, Warren RF, *et al*. Septic arthritis after arthroscopic anterior cruciate ligament reconstruction. Diagnosis and management. *Am J Sports Med* 1997;**25**:261–7.
5. Corbel MJ. Brucellosis: an overview. *Emerging Infect Dis* 1997;**3**:213–21.
6. Khateeb MI, Araj GF, Majeed SA, *et al*. *Brucella* arthritis: a study of 96 cases in Kuwait. *Ann Rheum Dis* 1990;**49**:994–8.
7. Mousa AR, Muhtaseb SA, Almudallal DS, *et al*. Osteoarticular complications of brucellosis: a study of 169 cases. *Rev Infect Dis* 1987;**9**:531–43.
8. Jacob NR, Rodríguez CG, Binaghi MA, *et al*. Brucellosis complicating chronic non-infectious disorders: diagnostic and therapeutic dilemmas. *J Med Microbiol* 2008;**57**:1161–6.
9. Weil Y, Mattan Y, Liebergall M, *et al*. *Brucella* prosthetic joint infection: a report of 3 cases and a review of the literature. *Clin Infect Dis* 2003;**36**:e81–6.
10. FAO/WHO Expert Committee on Brucellosis. Sixth Report. World Health Organization Tech Rep Ser No. 740. World Health Organization: Geneva, 1986.
11. Mantur BG, Amarnath SK. Brucellosis in India - a review. *J Biosci* 2008;**33**:539–47.
12. Matava MJ, Evans TA, Wright RW, *et al*. Septic arthritis of the knee following anterior cruciate ligament reconstruction: results of a survey of sports medicine fellowship directors. *Arthroscopy* 1998;**14**:717–25.
13. George MS, Dunn WR, Spindler KP. Current concepts review: revision anterior cruciate ligament reconstruction. *Am J Sports Med* 2006;**34**:2026–37.

This pdf has been created automatically from the final edited text and images.

Copyright 2011 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Please cite this article as follows (you will need to access the article online to obtain the date of publication).

Papastergiou SG, Koukoulas NE, Koumis P, Kyparlis D, Santas R. Brucella arthritis of the knee, 1 year after revision of anterior cruciate ligament reconstruction. *BMJ Case Reports* 2011;10.1136/bcr.01.2011.3786, date of publication

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow