

Brucellosis as a Primary Cause of Flexor Tenosynovitis on the Hand: Case Report and Literature Review

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Introduction

Brucellosis is primarily a disease of animals but is often transmitted to man either by drinking raw milk; by direct contact through the broken skin or the conjunctiva; or by the inhalation of infected dust [1]. Currently *Brucella melitensis* remains the principal cause of human brucellosis worldwide [2, 3]. Brucellosis is a zoonosis that usually affects the musculoskeletal system. Vertebral bodies, particularly in the lumbar region, are the bones more frequently involved [4]. Osteoarticular involvement in the hand and wrist is not common, and development of secondary tenosynovitis in the

course of a brucellar infection is even more unusual [5, 6]. There are no previous references to flexor tenosynovitis without osteoarticular involvement caused by *Brucella*. As far as we know, this is the first reported case of a primary brucellar tenosynovitis.

Case report

A 34-year-old woman, who used to work in a slaughterhouse in contact with cows and goats, presented with a two year history of progressive painful swelling in the volar aspect of her right wrist extending distally to the thumb and little finger. Wrist motion was 42°/38° compared to 80°/70° on the contralateral side, thumb interphalangeal motion was 0°/45° compared to 0°/90° on the contralateral side and little finger proximal interphalangeal proximal joint motion was 0°/30° versus 0°/100° on the contralateral side. She presented paresthesias in median nerve territory. Plain X-ray and CT scan showed no bony infection, and blood tests were normal. MRI showed a great effusion surrounding the flexor tendons mixed with granules in certain areas (Fig. 1).

With the diagnosis of granulomatous tenosynovitis, distal forearm and wrist were approached volarly in order to perform a biopsy-synovectomy. Median nerve was first identified, and found to be grossly displaced laterally and volarly. Ulnar nerve and artery were also identified and protected. Numerous rice-shaped bodies were found in a notably thickened synovial sheath (Fig. 2). The approach was then extended distally up to the thumb and little finger (Fig. 3). Complete synovectomy and plication of the remaining pulleys of the affected fingers in order to avoid bowstringing were performed. The flexor retinaculum was opened through its ulnar border during the procedure, and finally reconstructed in an elongated position in order to maintain its pulley effect and avoid displacement of

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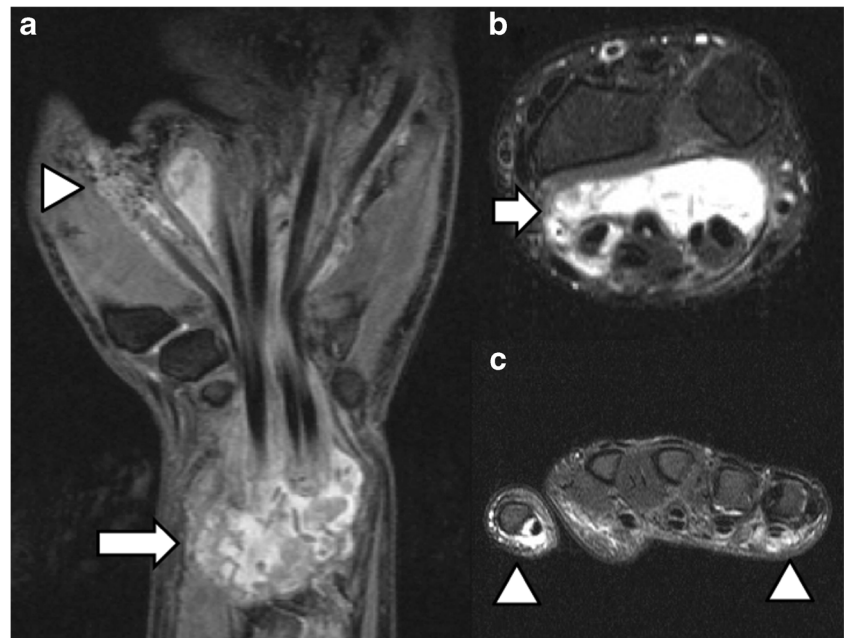
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Fig. 1 Coronal (a), axial wrist (b) and axial hand (c) T1-weighted MRI images showing great effusion mixed with granules surrounding the flexor tendons at the wrist (arrows), and extending distally to the thumb and little finger fingers (arrowheads)



the little and ring finger flexors [7]. Samples were sent for laboratory and histological examination.

Histology showed nonspecific chronic synovitis with fibrinoid component. Diagnosis of brucellosis was done based on positive serology (Wright's agglutination test $>1/320$, and positive enzyme linked immunosorbent assay (ELISA) serology) and *Brucella melitensis* grew on chocolate agar. Antibiotic therapy was started as soon as the serological result was confirmed. Doxycycline was given for 8 weeks, together with streptomycin for the first 2 weeks. The patient regained full and painless range of motion in about 3 months.

Discussion

Brucellosis remains a significant problem for many developing countries. Infection is particularly likely in young men

whose work brings them into contact with brucella-infected cattle, especially farmers and veterinary surgeons [4]. The patient was a young woman who worked in a slaughterhouse in direct contact with goats and sheep, where she could have been inoculated while handling brucella-infected meat.

Although it can present as a systemic febrile illness and then suspected, diagnosed and treated, it can also present with a variety of focal complications or become a chronic problem, especially if mistreated. It has been reported that such chronic cases may remain nearly asymptomatic and have a disproportionately low level of inflammation [4, 6, 8].

The patient had complete recovery of the disease with surgery and treatment with specific antibiotics for 10 weeks. She achieved an excellent functional outcome in her wrist and hand. Seal et al. [4] in 1974 also reported a case of a farmer affected from an arthritis of the wrist caused by *Brucella*. During the operation, similar as us, they found a thickened synovium and a yellowish granulation tissue. They removed

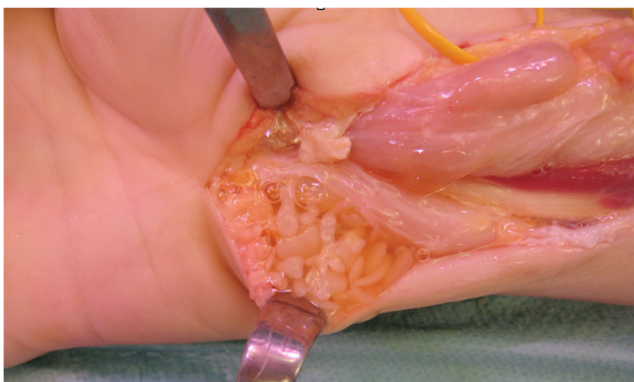


Fig. 2 Intra-operative photograph showing multiple rice bodies contained within the synovial sheath

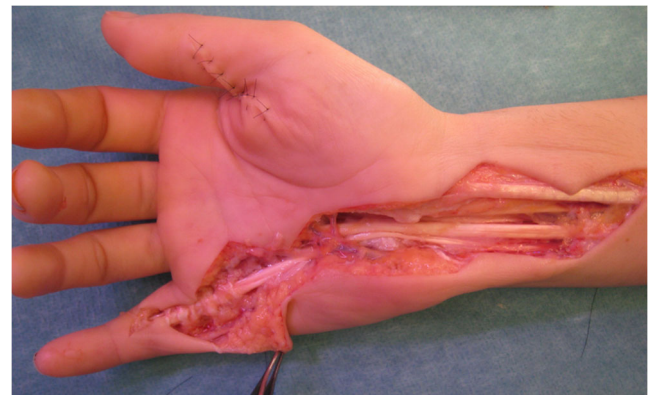


Fig. 3 Intra-operative photograph showing the distal approach extension to the little finger, and the sutured approach to the thumb

the granulation tissue and hypertrophied synovium and their patient was treated with tetracycline for five months, together with streptomycin for 6 weeks. Unlike us, in the radiographic explorations they found subchondral erosions in several carpal bones and signs of osteomyelitis of the hamate.

Pappas et al. [8] published in 2005 three cases of carpal tunnel syndrome (CTS) arising in the course of infection by *Brucella melitensis*, and responding to specific antibiotic treatment during 6 weeks. According to them various pathogenic mechanisms can be proposed for the appearance of CTS in the course of brucellosis. In our case, the carpal tunnel was opened and closed in an elongated way [7] because the concurrent flexor tenosynovitis in the wrist could result in median nerve compression [8]. Brucellosis should be included in the differential diagnosis of granulomatous tenosynovitis of the flexor tendons in endemic areas, even when osteoarticular involvement is not present.

Conflict of interest The authors, their immediate family, and any research foundation with which they are affiliated have not received any financial payments or other benefits from any commercial entity related to the subject of this article.

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