



Case report

First case of cervical epidural abscess caused by brucellosis in Saudi Arabia: A case report and literature review

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ABSTRACT

Spinal epidural abscess (SEA) is an extremely rare and disastrous musculoskeletal complication of spondylodiscitis and sacroiliitis secondary to brucellosis infection. Few case reports and retrospective studies have been published reporting patients with epidural abscesses mainly lumbar, and thoracic while cervical abscess are rare. Management of spinal epidural abscess due to *Brucella* species is not standard and remains controversial. To the best of our knowledge this is the first case of brucellar cervical epidural abscess reported in Saudi Arabia. In this paper, we review the literature and report a case of a 67-year-old Saudi gentleman, who presented with fever and back pain. Further evaluation revealed cervical epidural abscess and a positive serology for brucellosis. Following 6 months of antibiotic therapy alone against brucellosis, he showed significant clinical and radiological improvement. Spinal epidural abscess caused by brucellosis is a rare condition, difficult to diagnose, and can be complicated by disastrous neurological or vascular complication if left untreated. Brucellosis must be considered as a possible cause of spinal epidural abscess in patients from endemic area. Hence, early detection and initiation of appropriate medical treatment alone is crucial in preventing permanent neurological complication, and possibly avoid surgical intervention.

Background

Human brucellosis is one of the most common widespread zoonotic infections worldwide with more than 500,000 new cases reported annually [1,2]. Brucellosis remains endemic in Saudi Arabia with an incidence rate of 12.80 per 100,000 cases and 4062 reported cases in 2016 compared to 3661 cases in 2012 based on a Saudi national registry of MOH [3]. Despite its control in many developed countries, it constitutes a major public health problem and economic burden in Saudi Arabia [4]. Brucellosis is a systemic disease caused by bacteria of the genus *Brucella*, a gram-negative aerobic coccobacillus. *Brucella-melitensis* is the most common species responsible for the most invasive, and severe diseases worldwide followed by *brucella abortus*, *brucella suis* and *brucella canis* [7]. Humans are infected by three primary sources either by direct contact with meat or tissue of infected animals, consumption of unpasteurized dairy products from infected animals or laboratory exposure to *Brucella* isolates [5]. Brucellosis is a systemic infection with a wide range of clinical spectrum of symptoms and signs; it can involve any organs system. Osteoarticular manifestation is the most common complication and usually is presented as arthritis, sacroiliitis, spondylitis or osteomyelitis. [6,7]. It can rarely cause spinal epidural

abscess, which is difficult to diagnose, may be complicated by potentially disastrous neurological and vascular complication if left untreated [8]. Management of spinal epidural abscess is controversial. However, several cases of successful treatment with antibiotic alone have been reported in particular in patients with stable neurological condition. In our case, patient had no neurological deficit and was successfully treated with antibiotic for 6 months with significant clinical and radiological improvement. Brucellar spinal epidural abscess's recovery with medical treatment can be achieved without the need for surgical approaches and its complications, especially if detected and treated early. Herein, we review the literature and describe a case of brucellar spinal epidural abscess in which the patient achieved a complete recovery with only medical treatment.

Case presentation

A 67-year-old Saudi gentleman with a medical history of diabetes mellitus, hypothyroidism, vitiligo, benign prostatic hypertrophy and a prior history of brucellosis, presented to our emergency department with four-month history of fever, night sweats, loss of appetite, back pain, anorexia, fatigue, nausea and vomiting. He also reported contact

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with camels and ingestion of unpasteurized milk in the past. One month prior to admission, he had a motor vehicle accident for which he was diagnosed with compression fractures at thoracolumbar region involving T12 – L3. On clinical examination, he looked comfortable but in mild pain with a temperature of 38.2 °C; pulse rate of 114 beats per minutes; blood pressure 116/68 mmHg; and respiratory rate of 21 breaths per minute. Pertinent findings on physical exam are local tenderness over cervical and lumbar vertebrae and hepatomegaly. No neurological deficit or meningeal signs were noted.

Laboratory investigation showed white blood cell count $3.79 \times 10^9/L$; hemoglobin 11.10 g/dL; platelet count $156 \times 10^9/L$; ESR 55 mm/H; CRP 152 mg/L; alanine transaminase 59 U/I; aspartate transaminase 88 U/I; alkaline phosphatase 97 U/I; gamma GT 95 U/I; total bilirubin 0.7 mg/dL; albumin 30 g/L; total protein 72 g/L; creatinine 1.3 mg/dL; urea 6 mmol/L. Brucella serology using enzyme-linked immunosorbent assay was positive for IgG (17.9) and negative for IgM with positive agglutination titer of 1/ 2560. Blood cultures were negative. Magnetic resonance imaging of the cervical spine showed evidence of spondylodiscitis at C5–C7 with posteriorly located intraspinal epidural collection measuring $1.8 \times 2.2 \times 4$ cm and extending from C5 to C7 causing moderate spinal stenosis without spinal cord compression (Fig. 1).

Given his clinical presentation, prior history of brucellosis, raw milk ingestion, camel contact and a positive brucella serology test, he was diagnosed as a case of acute brucellosis and he was started on doxycycline and an aminoglycoside. Later, his magnetic resonance imaging of cervical spine showed spinal epidural abscess and rifampicin was added as a third agent. Few days following treatment, all his symptoms recovered with exception to his cervical back pain which was gradually improving.

Patient's condition was discussed with spine surgeon to evaluate the need for surgical intervention and it was decided to continue on medical treatment alone, as there was no neurological deficit.

During his regular follow-ups in ID clinic, his back pain started to

improve gradually, his brucella agglutination titer and inflammatory markers were trending down slowly and eventually normalized. Following 6 month of therapy, his repeated magnetic resonance imaging (MRI) showed interval improvement in epidural collection in C5–C7 and at that point his antibiotics were stopped after completing six months of therapy (Fig. 2).

The patient continued to follow up in ID clinic for 1 year and during that time, he remained symptoms free with a normal brucella agglutination titer.

Discussion

Osteoarticular complications secondary to brucellosis like sacroiliitis, spondylitis, peripheral arthritis and osteomyelitis are the most common and frequent of the disease. However, brucellar spinal epidural abscess is a rare complication, which is usually due to spondylitis [9,10].

To the best of our knowledge, no case of spinal epidural abscess associated with brucellosis has been reported in Saudi Arabia before. Moreover, Literature review using PubMed/MEDLINE (1950–2017) yielded 52 adult cases of brucellar epidural abscess published in the English literature to date, in which complete recovery was achieved with medical treatment only in 18 cases. The rest were treated with surgical approach combined with medical treatment. Baseline demographics, risk factors, involved location, antibiotic regimen and duration are shown in (Table 1).

We identified 19 patients, 10 males (58.8%) and 7 females (41.7%), with a mean age of 49.5 years at the time of diagnosis (range, 31–70 years). The most epidemic countries were Spain (47%), Turkey (23.5%) followed by Korea, Tunisia, Greece, Italy and Saudi Arabia. Unpasteurized milk ingestion (23.5%) was the most common risk factor. The frequency of spinal involvement was seen at Lumbar (40.9%), cervical (31.8%), thoracic (13.6%), sacral (13.6%), respectively. *B. melitensis* was the most common isolated specie (18.1%).

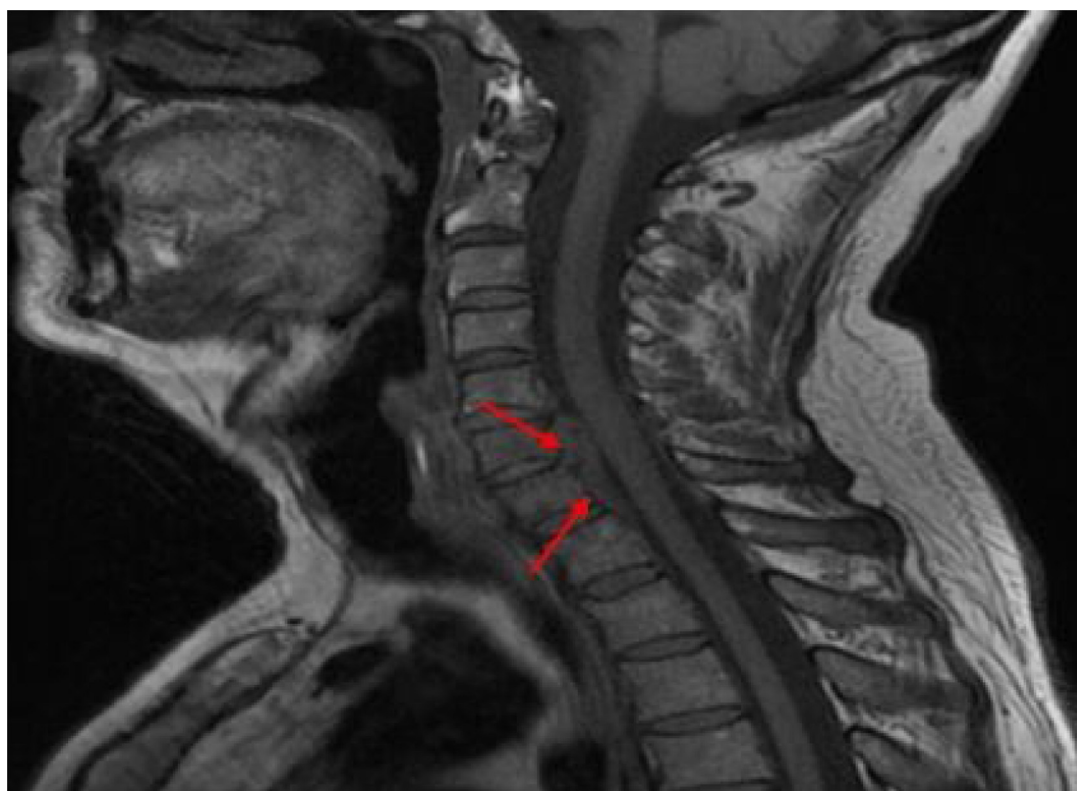


Fig. 1. Sagittal cervical T1 weighted magnetic resonance imaging (MRI) showing epidural collection along the posterior aspect of C5–C7 vertebral bodies causing moderate spinal stenosis.

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