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## **Case Report**

# Cervical brucellar spondylitis causing incomplete limb paralysis

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#### **Abstract**

Brucellosis, a zoonosis with worldwide distribution, is a systemic infection caused by bacteria of the genus *Brucella*. Meanwhile, brucellosis often causes complications, such as osteoarticular involvement, and spondylitis is the most prevalent and important clinical form. Here, is a case of cervical brucellar spondylitis causing incomplete limb paralysis in a middle-aged male. The diagnosis was based on clinical history, and supported by *Brucella* serology and magnetic resonance imaging. Quadruple antibacterial treatment continued for four weeks. In this case, the epidural abscess causing spinal cord compression resolved without surgery. In addition, the patient had recovered from most of the neurologic deficits.

**Keywords:** Brucellosis. Cervical spondylitis. Epidural abscess.

#### INTRODUCTION

Brucellosis, a zoonosis that has spread to many countries, is caused by facultative intracellular bacteria of the genus Brucella. Brucella species are transmitted by means of direct contact with infected animals or consumption of products from infected animals<sup>1</sup>. Brucella is also endemic in the Mediterranean Region, Middle East, and parts of Africa and Latin America<sup>2</sup>. Each year, half a million new cases are reported worldwide, but according to the World Health Organization, this number is greatly underestimated. At the same time, the situation is also not optimistic in China and the incidence is reported to be 0.59 per year per 100,000 population<sup>3</sup>. Brucellosis affects the whole body, including various human organs, the musculoskeletal system and the reticuloendothelial system4. Symptoms related to the musculoskeletal system are commonly seen, and the most frequently detected complications are peripheral arthritis, sacroiliitis, and spondylitis<sup>5</sup>. Up to now, the diagnosis and treatment of vertebral involvement are rather difficult. Brucellosis spondylitis has rarely been reported in China. Furthermore, rare information about cervical brucellar spondylitis causing incomplete limb paralysis has already

Corresponding Author: Qiang Zhang. e-mail:765926411@qq.com Orcid: 0000-0003-3020-228X Received 15 June 2018 Accepted 13 October 2018 been reported. Cervical brucellar spondylitis is a rare and serious clinical presentation which may cause irreversible complications. A case of cervical brucellar spondylitis causing incomplete limb paralysis is presented in this report and the diagnosis and treatment are discussed referring to other reports.

#### **CASE REPORT**

A 47-year-old middle-aged man was admitted to the Infectious Diseases Department of our hospital because of incomplete limb paralysis. The patient was employed as a breeder in a livestock farm. Moreover, he usually consumed dairy products without disinfection or sterilization. Two weeks ago, the patient developed symptoms of sweating, fever and fatigue. At the local hospital, the laboratory conducted the Rose-Bengal test and the result was positive (+++), and the erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were elevated. During his hospitalization, the patient gradually experienced limb weakness and then came to our hospital for further treatment.

Physical examination of the patient revealed pain and restriction during cervical movements. He had difficulty in standing up straight because of incomplete paralysis and decreased limb muscle strength (3-). His body temperature was 37.5°C. Initial laboratory values were as follows: white blood cell count: 7,600/mm³, hemoglobin: 13.5 g/dl, platelet count: 237,000/mm³, ESR: 86 mm/h, CRP: 55 mg/L, and a normal blood biochemistry profile. The Rose-Bengal test applied to

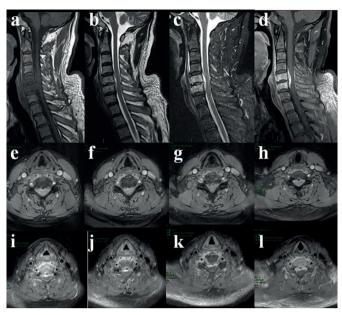
the patient was positive (+++) and *Brucella* agglutination test (Wright test) was positive at a titer of 1:320. However, *Brucella* species were not isolated from repeat blood cultures. Repeat acid-fast bacilli sputum stains and culture for tuberculosis yielded negative results. Cervical enhanced magnetic resonance imaging (MRI) showed C5–6 vertebral body and intervertebral disc destruction, posterior epidural abscess in the spinal canal, low signal on T1WI, high signal on T2WI and pressure-fat T2WI, marked enhancement after contrast infusion, and significant pressure on the spinal cord (**Figure 1**). Meanwhile, thoracic and lumbar MRI showed no obvious abnormalities.

The patient was diagnosed as having cervical brucellar spondylitis with epidural abscess causing incomplete limb paralysis. Quadruple anti-biotherapy had been started with oral doxycycline 200 mg/day and rifampicin 600 mg/day, intravenous drip levofloxacin 500 mg/day and cefotaxime sodium 6.75 g/day. Antibacterial treatment was continued for four weeks. A surgical intervention was planned for excision of the lesion and reduction of epidural abscess. However, there was an obvious improvement in local symptoms and signs. He started to experience an improvement in limb muscle strength (4+). Also, in the control examination of the patient, it was found that the white blood cell count was 6,500/mm<sup>3</sup>, ESR was 29 mm/h, and CRP was 10.6 mg/L. The Rose-Bengal test was (+) and Wright test yielded a titer of 1:160. The size of the posterior epidural abscess in the spinal canal had reduced earlier. T1WI showed a slightly lower signal. T2WI and pressed fat T2WI showed a mixed high signal (Figure 2). His treatment was continued with oral doxycycline 200 mg/day and rifampicin 600 mg/day for three months after discharge.

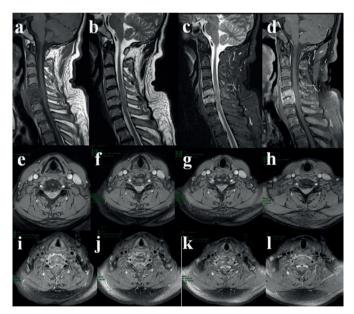
At the 1-year follow-up visit, he started to experience an improvement in limb muscle strength (5+). Also, in the control examination of the patient, it was found that the white blood cell count was 5,600/mm³, the ESR was 7 mm/h and CRP was 6.7 mg/L. The Rose-Bengal test was (+) and Wright test yielded a titer of 1:80. The cervical enhanced MRI showed that the C5–6 vertebral body and intervertebral disc destruction was normalized, the posterior epidural abscess in the spinal canal had disappeared, and no obvious compression was seen in the spinal cord (**Figure 3**). Thoracic and lumbar MRI showed no obvious abnormalities. The patient had recovered from most of the neurologic deficits.

#### **DISCUSSION**

Brucellosis is a multisystemic disease transmitted to humans by consumption of unpasteurized milk and dairy products from infected cows. There are two forms of spinal brucellosis: focal and diffuse. In the focal form, the organism becomes localized in the anterior aspect of the superior endplate, causing bony destruction in a small area. In the diffuse form, the infection spreads throughout the involved vertebra, and to the adjacent vertebrae. The spine is the most frequently involved bony structure in brucellosis (2–53%)<sup>3</sup>. *Brucella* spondylitis was first described by Kulowski and Vinke, and is known as one of the most serious complications of brucellosis<sup>6</sup>. *Brucella* spondylitis involves the lumbar region most frequently and

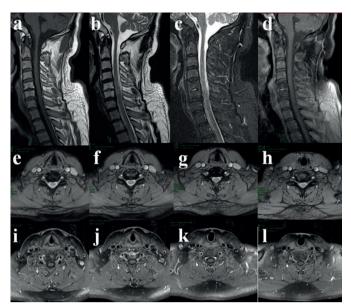


**FIGURE 1:** Cervical enhanced MRI showed C5-6 vertebral body and intervertebral disc destruction, the posterior epidural abscess in the spinal canal, low signal on T1WI, high signal on T2WI and pressure-fat T2WI, marked enhancement after enhancement (**a-d**), and significant pressure on the spinal cord (**e-I**).



**FIGURE 2:** Cervical enhanced MRI showed destruction of C5-6 vertebral bodies and intervertebral discs (a-d). The posterior epidural abscess in the spinal canal was reduced earlier. T1WI showed a slightly lower signal. T2WI and pressed fat T2WI showed a mixed high signal, strengthened, and reduced spinal cord pressure (e-I).

it may also be seen in the thoracic and cervical regions. Ulu-Kilic A et al. evaluated the efficacy and tolerability of antibiotic regimens and optimal duration of therapy in complicated and uncomplicated forms of spinal brucellosis for an update on treatment options for spinal brucellosis<sup>7</sup>. It was found that there were no significant therapeutic differences and the results were similar regarding the complicated and uncomplicated



**FIGURE 3:** Cervical enhanced MRI showed that the C5-6 vertebral body and intervertebral disc destruction basically returned to normal (a-d), the posterior epidural abscess disappeared in the spinal canal, and no obvious compression was seen in the spinal cord (e-l).

groups. Meantime, medical management is usually the basis for treatment, alone or in combination with surgery. Guerado E et al. considered a surgical approach, either by endoscopy or as an open surgery if clinical evolution is unsatisfactory biopsy is needed and no micro-organism has been isolated, and also whenever a root, spinal cord or dural compression is seen on MRI. Spinal instability or severe deformities are also clear indications for surgical treatment<sup>8</sup>.

Treatment of an epidural abscess usually consists of prompt surgical drainage and antimicrobial therapy. Nonsurgical treatment should be reserved for the few patients who carry considerable surgical risk and for those who have no neural tissue damage or significant compression or who have complete paralysis lasting for longer than 3 days. Brucellar epidural abscess occurs primarily during the subacute-chronic phase of the disease process, and *Brucella* responds well to antibiotics, which should be administered in the required doses and continued until complete recovery9. There is no consensus on the selection of appropriate antibiotics or the treatment duration in patients with brucellar spondylitis and epidural abscess. However, it is generally accepted that short-term monotherapy is associated with a high relapse rate (30%)10. It is recommended that antibiotic therapy should be maintained from 6 weeks to 1 year<sup>11</sup>. Although it has been managed with surgery in previous reports, we treated our patient without surgical therapy because he had no aggravating neurologic deficits due to the spinal cord compression caused by the epidural abscess.

The principal mode of treatment of brucellar spondylitis is conservative, namely, immobilization of the affected area and antimicrobial therapy. An open biopsy of the cervical vertebra body or epidural abscess should be indicated when the differential diagnosis of spondylitis cannot be made by the conventional diagnostic methods. In the vast majority of reports, surgery for decompression of the spinal cord or nerve roots is essential in cases with neurological deficits. However, antimicrobial chemotherapy is essential in the presence of a good response which may lead to the avoidance of surgery.

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**Conflict of Interest:** The authors declare that there is no conflict of interest.

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