Brucellar Spine Infection
—Four Case Reports—

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Abstract

Brucellosis can be difficult to diagnose because of the nonspecific and variable clinical picture. This systemic disease is still an important public health problem in the Mediterranean Basin. These four cases of brucellar spine infection originated from rural areas around Aydın, Turkey. The systemic complaints of fever, profuse sweating, malaise, polyarthromyalgia, and weight loss indicated the final diagnosis of brucellosis and presumptive diagnoses were made based on agglutination testing for brucellosis. Computed tomography and magnetic resonance imaging indicated signs of bone infection and soft tissue involvement. Good outcomes were obtained with prolonged treatment with anti-brucellar drugs including streptomycin, rifampicin, and tetracycline. Early diagnosis is important and prompt antibrucellar chemotherapy is effective in most cases, but prolonged follow up is necessary in all patients with spinal brucellosis.

Key words: Brucella, spine, back pain, spondylitis

Introduction

Brucellar spine infection is still a common public health problem in many parts of the world including the Middle East, South America, and the Mediterranean Basin.10–12,16,17,20) The causative agents of brucellosis are hosted by domestic animals: Brucella melitensis in goats, B. abortus in cattle, B. suis in swine, and B. canis in dogs.6,20,24) The organism is transmitted to human beings by consuming contaminated milk products or handling infected animals and may persist for many years.6) Osteoarticular involvement of the spine is a well-known complication of brucellosis and the spine is the most common localization.1,20,24,27) Clinically, back pain is the most common complaint in patients with spinal involvement due to invasion of the richly innervated periosteum. Although involvement of the spine may have widely variable and nonspecific manifestations, more widespread use of current diagnostic studies such as computed tomography (CT) and magnetic resonance (MR) imaging has allowed more accurate and frequent detection of spinal brucellosis.6,10,12,13,17,22) In this study, we present our experience of spinal brucellosis in four cases treated in our institution over a period of 5 years.

Case Reports

Case 1: A 68-year-old male farmer with a history of low back pain persisting for 6 months presented with restriction of spinal movements and weight loss. Physical examination found tenderness over the spinal processes at the L-5 and S-1 levels and paraspinal muscle contraction. Neurological examination revealed that the Lasegue’s test was bilaterally positive at 70 degrees. Laboratory investigations showed erythrocyte sedimentation rate (ESR) of 82 mm/hr, white blood cell (WBC) count of 6.6 × 10⁹ g/l, and hemoglobin of 11.0 g/dl. The Rose-Bengal test for brucellosis was positive and the Wright test confirmed this finding with a titer of 1/640. Radiography of the lumbosacral region revealed multiple sclerotic destructive lesions at the L-5 body and narrowing at the L5-S1 space (Fig. 1). CT showed gross destruction of the L-5 and S-1 vertebral bodies as well as a soft-tissue mass surrounding the L-5 body. The patient was treated with streptomycin (1 g/day intramuscularly [i.m.]) for 30 days combined with tetracycline (2 g/day per os [p.o.]) and rifampicin (600 mg/day p.o.) for 3 months. Follow-up examination 5 years later...
showed the patient had mild persistent low back pain and MR imaging of the lumbar spine showed discovertebral involvement at the L4-5 space and L5–S1 disc changes compatible with degenerative disease. However, ESR was still 46 mm/hr and the Wright test was positive for Brucella at 1/160.

**Case 2**: A 59-year-old male farmer had a history of low back pain beginning 7 days earlier which radiated from the buttock to the heel in his right leg, and was typically aggravated by sneezing. He also complained of fever and sweating at night. The patient’s medical history included consumption of fresh white cheese using unpasteurized milk. Physical examination found tenderness over the spinal processes of the lumbar region and bilateral paraspinal muscle contraction. Lumbar spinal movements were restricted and painful. Neurological examination revealed that the Lasegue’s test was positive at 40 and 60 degrees on the right and left sides, respectively, and hypesthesia was present in the right S-1 dermatome. Laboratory investigations showed ESR of 48 mm/hr, WBC count of $9.5 \times 10^9$ g/l, and hemoglobin of 14.0 g/dl. The Rose-Bengal test for brucellosis was positive and the Wright test confirmed this finding with a high titer of 1/1280. Radiography of the lumbosacral region was normal except for straightening of the spine. CT showed disc bulging at the L4-5 space and mild erosive changes of the lower cartilage end-plate of the L-4 vertebral body. MR imaging of the lumbar spine showed discovertebral involvement at the L4-5 space, indicating active infectious spondylodiscitis (Fig. 2). The patient was treated with streptomycin (1 g/day i.m.) for 14 days combined with tetracycline (2 g/day p.o.) and rifampicin (600 mg/day p.o.) for 3 months. The patient made a complete recovery. This case was previously described in this journal.\textsuperscript{27} 

**Case 3**: A 32-year-old male worker presented with progressive back pain on the left side and right knee joint pain over a 5-day period and subsequent inability to walk. He also complained of profuse sweating at night and fever. He habitually consumed unpasteurized milk and fresh cheese. Physical examination found tenderness over the spinal process of the L-5 vertebra. Lumbar spinal movements were restricted and painful. Neurological examination revealed that the Lasegue’s test was positive at 60 degrees on the left side. Laboratory investigations found ESR of 16 mm/hr, WBC count of $8.9 \times 10^9$ g/l, and hemoglobin of 14.0 g/dl. The Rose-Bengal test for brucellosis was positive and the Wright test confirmed this finding. Radiography of the lumbosacral region and CT found no abnormalities. The patient was treated with tetracycline (2 g/day p.o.) combined with rifampicin (600 mg/day p.o.). He made a complete recovery within 2 weeks, and the course of tetracycline and rifampicin was
Case 4: A 43-year-old man was admitted to our hospital with low back pain radiating to the lower extremities. He had a history of febrile illness and loss of appetite 2 years before admission. Five months after the start of the febrile illness, the patient started to complain of backache and spinal movements were restricted and painful. His medical history included consumption of fresh white cheese using unpasteurized milk. Laboratory examinations showed ESR of 45 mm/hr, WBC count of $6.9 \times 10^9\ g/l$, and hemoglobin of 13.2 g/dl. Radiography of the lumbosacral region found no abnormalities. CT showed disc herniation at the L4-5 space and destruction of bone with surrounding sclerosis. MR imaging revealed discovertebral involvement at the L4-5 space and obliteration of the subarachnoid space with disc tissue. Bone scintigraphy also showed involvement of the sacroiliac joints (Fig. 3). The Rose-Bengal test for brucellosis was positive and the Wright test confirmed this finding with a titer of 1/320. He responded well to a course of streptomycin (1 g/day i.m.) for 20 days combined with rifampicin (600 mg/day p.o.) and tetracycline (2 g/day p.o.) for 3 months. The ESR and agglutination test for Brucella returned to normal. However, the patient's symptoms continued to progress and neurological examination revealed positive Laségue's sign at 60 and 70 degrees in the right and left legs, respectively, and reduced sensory function in the right leg. Repeat MR imaging of the lumbar spine showed considerable healing with resolution of neural compression (Fig. 4). A surgical procedure consisting of laminectomy and discectomy were performed, and the patient made a full recovery. Histological examination revealed degenerated disc tissue consisting of fibroblasts and cartilage cells, degenerated intervertebral disc consisting of collagenous tissue and fibroblasts. HE stain, $\times 200$. 

Fig. 3 Case 4. Bone scintigram demonstrating increased isotope uptake in the sacroiliac joints and the right acetabulum.

Fig. 4 Case 4. (A) Sagittal $T_1$-weighted magnetic resonance (MR) image revealing discovertebral involvement at the L4-5 space and healing of the bone with obliteration of the subarachnoid space with disc tissue. (B) Sagittal $T_2$-weighted MR image indicating the same changes.

Fig. 5 Case 4. Photomicrograph demonstrating degenerated intervertebral disc consisting of collagenous tissue and fibroblasts. HE stain, $\times 200$. 

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but no evidence of inflammatory response (Fig. 5).

### Discussion

Brucellosis is a disorder of worldwide distribution, but occurs relatively frequently in Mediterranean countries in Europe.④, ⑥, ⑧, ⑩, ⑫, ⑬, ⑭ These four adult cases of spinal brucellosis originated from or regularly visited rural areas around Aydın, Turkey, where the disease is endemic (Table 1). The incidence of brucellosis has increased in Turkey during recent years because eradication efforts have not succeeded, especially in goats and sheep.⑩, ⑬ B. melitensis is the most infectious and pathogenic agent and is responsible for almost all of the human disease in Turkey.⑨, ⑱, ⑲ Brucella microorganisms are transmitted to man from animals by ingestion of unpasteurized dairy products or by direct contact with infected animals.⑬ The disease frequently presents with non-specific clinical manifestations such as fever, profuse sweating, malaise, polyarthromyalgia, and weight loss.③, ⑩, ⑲ The diagnosis is established by isolation of Brucella microorganisms from blood samples, but this is very difficult because bacteremia is intermittent. The presence of the following criteria have been suggested for diagnosis: history of exposure to the infectious disease; compatible signs and symptoms; Brucella agglutination exceeding 1/100; and rapid improvement after anti-brucellar antibiotic therapy.②, ⑦, ⑬ In clinical practice, the systemic complaints indicate the final diagnosis of brucellosis and a presumptive diagnosis can be made based on agglutination testing for brucellosis. Radicular pain may simulate lumbar disc herniation as seen in our Cases 2 and 4.

Complications due to involvement of the osteoarticular system are very common and cause a high morbidity in brucellosis.⑩, ⑬ Brucellar involvement of the spine can be focal or diffuse and occurs in 10% to 50% of patients with chronic brucellosis.①, ⑬ Spinal involvement in brucellosis was first described in 1932.⑯ The lumbar sacral region is most frequently involved in spondylodiscitis, followed by the thoracic and cervical segments.①, ②, ③, ⑦, ⑨, ⑱, ⑲, ⑳, ⑳ Brucellar spondylodiscitis is frequently characterized by involvement of the cartilage endplate, an area with a rich blood supply, in the early stages of the disease.②, ③, ⑦, ⑬, ⑲ Spinal extradural or paravertebral abscess formation is an extremely rare presentation of Brucella involvement.⑤ Localized spinal pain is the earliest sign of brucellar spondylitis, and 10% to 43% of patients have some degree of neurological deficit.⑦, ⑩ The disease exhibits non-specific symptomatology and clinical manifestations, and thus is likely misdiagnosed in some cases as tuberculosis of the spine (Pott’s disease) or intervertebral disc herniation because the diagnosis was not established until the disease is at an advanced stage.④, ⑩, ⑫, ⑬, ⑱, ⑲, ⑳, ⑳ Therefore, tuberculosis spondylitis should be differentiated from brucellosis.⑳ The most frequent area of involvement in tuberculosis is the thoracic spine, with more severe destructive change such as angulation deformity.⑲, ⑳ Paraspinal abscess is another feature of tuberculosis, occurring approximately in 50% of cases, whereas the frequency of paraspinal abscess complicating spinal brucellosis is only 13%.⑳ Spinal brucellosis can be detected by radiography, CT, and MR imaging. In addition, bone scintigraphy can detect areas of bone remodeling in spinal brucellosis. Nevertheless, the earliest signs are not pathognomonic and the diagnostic radiological findings due to Brucella may only appear at an

### Table 1 Clinical data of four patients with spinal brucellosis

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age (yrs)</th>
<th>Symptoms</th>
<th>Vertebral level</th>
<th>Brucella antibody titer</th>
<th>Radiological findings</th>
<th>Antibiotic treatment</th>
<th>Surgical intervention</th>
<th>Evolution</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>68/M</td>
<td>backache, weight loss</td>
<td>L5–S1</td>
<td>1/640</td>
<td>spondylitis, spondylodiscitis</td>
<td>Str + Tet + Rif</td>
<td>–</td>
<td>partially recovered</td>
</tr>
<tr>
<td>2</td>
<td>59/M</td>
<td>backache, sciatica, sustained fever, profuse sweating</td>
<td>L4–5</td>
<td>1/1280</td>
<td>spondylitis, spondylodiscitis</td>
<td>Str + Tet + Rif</td>
<td>–</td>
<td>recovered</td>
</tr>
<tr>
<td>3</td>
<td>32/M</td>
<td>backache, arthralgia, profuse sweating, intermittent fever, paraparesis</td>
<td>L–5</td>
<td>NS</td>
<td>normal</td>
<td>Tet + Rif</td>
<td>–</td>
<td>recovered</td>
</tr>
<tr>
<td>4</td>
<td>43/M</td>
<td>lumbosacral pain, arthralgia, relapsing fever</td>
<td>L4–5</td>
<td>1/320</td>
<td>spondylodiscitis, sacro-ilitis</td>
<td>Str + Tet + Rif</td>
<td>+</td>
<td>recovered</td>
</tr>
</tbody>
</table>

NS: not stated, Rif: rifampicin, Str: streptomycin, Tet: tetracycline.
advanced stage, as in our patients.\textsuperscript{5,19} CT demonstrates details of bone destruction adjacent to the involved disc and the cartilage end plate region.\textsuperscript{7} MR imaging is more sensitive to the presence of soft tissue swelling and entrapment of spinal cord and nerve roots.\textsuperscript{6,7,10,12,13,17} MR imaging shows decreased signal intensity in the intervertebral disc, whereas the adjacent vertebral bodies are hypointense on T\textsubscript{1}-weighted images and hyperintense on T\textsubscript{2}-weighted images.\textsuperscript{6,10,12,13,17,27} Brucellar discitis is frequently seen in spinal brucellosis, but may recover spontaneously.\textsuperscript{7} The clinical picture of radiculopathy caused by disc herniation due to Brucella may occur, as seen in two of our patients.

The mainstay of treatment for brucellosis is antibiotic treatment, but no standard regimen is available and various effective combinations have been suggested. The best results have been obtained with different agents such as streptomycin, rifampicin, tetracycline, doxycycline, and ofloxacin.\textsuperscript{3,7,10,12,13,17,24,27} The eradication of infection from the bone is difficult and relapse may occur especially if only short-term treatment is given.\textsuperscript{3,7,8,24} Therefore, long-term anti-brucellar antibiotic treatment should be prescribed immediately after the diagnosis.\textsuperscript{3,7,8,24} Brucella antibody titers have been recommended to assess the therapeutic efficacy and resolution of the disease.\textsuperscript{3,7–9} As a rule, surgical intervention is not generally necessary but laminectomy and/or discectomy procedures may be indicated if localized infection causes extradural compression of the spinal cord or nerve roots or prolonged antibiotic treatment results in no response as occurred in our Case 4.\textsuperscript{5,9}

In conclusion, the spinal form of brucellosis should be considered in the differential diagnosis of any patient with back and radicular pain, and arthromyalgia with a history of contact with brucellosis in endemic areas. Early correct diagnosis and an interdisciplinary approach are vital since brucellosis is an aggressive disease which requires immediate treatment. The infection is curable, and only anti-brucellar antibiotic therapy is effective in most cases.

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References


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