Spinal Epidural Abscess Caused by Brucellosis
—Two Case Reports—

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Abstract

Brucellosis is still an important public health problem in the Mediterranean countries, including Turkey, and is most probably underdiagnosed or underreported. Two rare cases of extradural brucellar granuloma causing spinal cord compression in the thoracic and cervical regions were identified by magnetic resonance imaging. The abscesses were totally excised surgically. Medical treatment was given immediately after diagnosis, but delayed neurosurgical intervention resulted in partial neurological recovery in one patient although the other showed complete neurological recovery. Abscess formation may cause myelopathy due to extradural compression of the spinal cord. Failure to correct this condition may cause irreversible impairment of motor functions. Magnetic resonance imaging should be performed periodically in patients with brucellosis and suspected vertebral involvement to monitor for epidural granuloma formation and prevent extradural cord compression.

Key words: Brucella, cord compression, epidural abscess, spinal involvement

Introduction

Brucellosis is a zoonosis with a worldwide distribution caused by contact with, or inhalation or consumption of meat or products from infected cattle, sheep, or goats. Brucellosis is a systemic infection caused by facultative intracellular bacteria of the genus Brucella that can involve many organs and tissues.21) Osteoarticular involvement is the most common complication of brucellosis and affects 20% to 85% of patients.11,10) The spectrum of bone and joint lesions includes arthritis, bursitis, tenosynovitis, sacroilitis, spondylitis, and osteomyelitis.6,17) The frequency of spondylitis in brucellosis ranges from 2% to 53%.4,6,11) The most common location is the lumbar spine followed by the cervical and thoracic spine. Spondylodiscitis is the most prevalent and important clinical form of osteoarticular involvement in adults with brucellar infection. Spinal cord compression due to brucellar epidural abscess is very rare, with only 18 previously reported cases.2,3,5,7,10,16,18)

Here we describe two cases of brucellar epidural abscess presenting with neurological deficits and discuss the treatment.

Case Reports

Case 1: A 24-year-old woman was referred to our hospital under a diagnosis of brucellosis and paraplegia. She had been admitted to another hospital with the main complaints of fever and dorsal pain 1 month previously. The diagnosis of brucellosis had been established by a positive rose bengal radioactive test and the Brucella seroagglutination test (titer of 1:320). Treatment with doxycycline, rifampin, and trimethoprim-sulfamethoxazole was begun in this center. Three weeks after the beginning of the treatment, the patient began to complain of difficulty in walking and urinary incontinence. She was then referred to our hospital.

On admission, physical examination found no abnormality, but neurological examination revealed paraplegia, loss of sphincter control, impaired sensation, and loss of all deep tendon reflexes below the T-8 level. There were no other abnormalities of the central nervous system. T1-weighted magnetic resonance (MR) imaging with gadolinium revealed increased signal intensity in the T-6 vertebral body and an enhanced hyperintense epidural mass lesion...
Fig. 1 Case 1. A: Sagittal T1-weighted magnetic resonance (MR) images with gadolinium showing narrowing of the T6-7 intervertebral disc space and an enhanced hyperintense lesion, with hypointense center, continuous at this level with the vertebral bodies, affecting T-6 more than T-7, obliterating the epidural space and causing significant spinal cord compression. These features are consistent with abscess formation. B: Axial T1-weighted MR images with gadolinium showing obliteration of the left centrolateral epidural space, extension to the paravertebral space, and significant cord compression.

Located anteriorly at the T-6 and T-7 levels (Fig. 1). The Brucella seroagglutination test was positive (titer of 1:640). Three consecutive blood cultures were negative.

The patient underwent surgery immediately after admission. A left paramedian transpedicular approach was performed to avoid retraction of the spinal cord. The left pedicles of T-6 and T-7 were drilled. The thick, rough capsule of the abscess was exposed anteriorly on the left. A yellowish purulent material with a total volume of 0.5 cm³ was drained and the capsule of the abscess was dissected easily from the posterior aspect of the vertebral bodies. The posterior longitudinal ligament was intact. The abscess had no relationship with the vertebral body or disc space. Histological examination of the lesion revealed edematous granulation tissue formation (Fig. 2).

Treatment with doxycycline, rifampin, and trimethoprim-sulfamethoxazole was continued for 3 months. The brucellosis was resolved but neurological recovery was only partial.

Case 2: A 42-year-old woman presented with a 3-month history of neck pain, and progressive weakness and pain in the upper extremities. She had a history of eating cheese made from raw cows’ milk. General physical examination was normal. Neurological examination revealed mild quadriplegia, positive Babinski signs and clonus bilaterally, and increased deep tendon reflexes in the lower limbs, but no sensory or sphincter disturbances.

Laboratory test results were normal except for the erythrocyte sedimentation rate (80 mm/hr). The Brucella seroagglutination test was positive (titer of 1/320, Wright agglutination test). Blood cultures were negative. T1-weighted MR imaging with gadolinium revealed a hyperintense lesion with a
Fig. 2 Case 1. Photomicrograph showing edematous granulation tissue with polymorphonuclear cells. HE stain, ×100.

Fig. 3 Case 2. Sagittal T1-weighted magnetic resonance image with gadolinium showing narrowing of the C4-5 intervertebral disc and a hyperintense lesion with hypointense necrotic center, peripheral enhancement at the C3–5 levels, and involvement of the C-5 vertebral body. The lesion extends rostrally through the posterior longitudinal ligament and obliterates the epidural space. The spinal cord is compressed significantly and displaced posteriorly.

Discussion

Spinal brucellosis usually starts in the superior end plate, an area with a rich blood supply, but occasionally the inferior endplate may also be involved. The subsequent progress of the infection depends on the size of the initial inoculum, the virulence of the organism, and the immunity of the host, so the infection may either regress and resolve, or progress to involve the entire vertebral body and disc space, and subsequently the adjacent vertebra.9)

MR imaging is important in the diagnosis, assessment, and management of patients with spondylitis. However, MR imaging findings are not characteristic for brucellar spondylitis, and all infectious spondylitis cases show similar MR imaging findings. T1-weighted MR imaging typically shows low signal intensity in the vertebral body that reflects increased extracellular fluid within the marrow. Contrast medium causes enhancement of the affected bone. T2-weighted MR imaging shows high signal intensity in the vertebral body. Two forms of spinal brucellosis are known, focal and diffuse.1) Focal vertebral brucellar osteomyelitis is defined as focal areas of abnormal signal intensity usually localized in the anterior aspect of an end plate of a vertebra at the discovertebral junction. Diffuse vertebral brucellar osteomyelitis is defined as diffuse abnormal signal intensity in the adjacent vertebra and the intervening disc. In our first case, decreased signal intensity on the T1-weighted images and diffuse contrast enhancement on the T1-weighted images with gadolinium in the T-6 hypointense necrotic center and peripheral enhancement at the C3–5 levels. The spinal cord was compressed significantly and displaced posteriorly (Fig. 3).

A right anterior cervical approach was performed. A yellowish purulent material was drained from the prevertebral area under the anterior longitudinal ligament. Total corpectomy of C-4 and C-5 was performed with a high speed drill. Granulomatous tissue, which compressed the cervical cord, was dissected from the epidural space after corpectomy. At surgery, we observed that the posterior longitudinal ligament was intact. Bone graft harvested from the iliac crest was used for C3–6 fusion and an anterior cervical plate system used for stabilization. Histological examination of the lesion revealed edematous granulation tissue formation.

She reported pain relief immediately after surgery. She was given 200 mg doxycycline and 600 mg rifampin daily for 12 weeks. Complete neurological recovery was found at the follow-up examination.

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Table 1   Cases of spinal cord compression due to brucellosis in the literature

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Author (Year)</th>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
<th>Neurological findings</th>
<th>Neuroimaging investigation</th>
<th>Duration of symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>Çeviker et al. (1989)</td>
<td>48</td>
<td>M</td>
<td>T-5</td>
<td>paraplegia</td>
<td>myelography</td>
<td>2 mos</td>
</tr>
<tr>
<td>2</td>
<td>Sharma et al. (1990)</td>
<td>26</td>
<td>M</td>
<td>T-10</td>
<td>paraparesis</td>
<td>myelography</td>
<td>1 mo</td>
</tr>
<tr>
<td>3</td>
<td>Mousa et al. (1990)</td>
<td>?</td>
<td>?</td>
<td>C3–4</td>
<td>quadriplegia</td>
<td>CT</td>
<td>?</td>
</tr>
<tr>
<td>4</td>
<td>?</td>
<td>?</td>
<td>?</td>
<td>C5–6</td>
<td>quadriplegia</td>
<td>CT</td>
<td>?</td>
</tr>
<tr>
<td>5</td>
<td>?</td>
<td>?</td>
<td>?</td>
<td>T-7, T-12 cervical region</td>
<td>paraplegia</td>
<td>CT</td>
<td>?</td>
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<tr>
<td>13</td>
<td>Coºskun et al. (1998)</td>
<td>44</td>
<td>M</td>
<td>T10–11</td>
<td>paraplegia</td>
<td>MR imaging</td>
<td>1 mo</td>
</tr>
<tr>
<td>15</td>
<td>Pina et al. (2001)</td>
<td>35</td>
<td>F</td>
<td>C5–6</td>
<td>quadriplegia</td>
<td>MR imaging</td>
<td>3 wks</td>
</tr>
<tr>
<td>16</td>
<td>?</td>
<td>75</td>
<td>M</td>
<td>C4–5</td>
<td>quadriplegia</td>
<td>MR imaging</td>
<td>3 mos</td>
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<td>17</td>
<td>?</td>
<td>53</td>
<td>M</td>
<td>C5–6</td>
<td>normal</td>
<td>MR imaging</td>
<td>2 mos</td>
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<tr>
<td>18</td>
<td>?</td>
<td>51</td>
<td>F</td>
<td>C6–7</td>
<td>paraparesis</td>
<td>CT</td>
<td>1 mo</td>
</tr>
</tbody>
</table>

CT: computed tomography, MR: magnetic resonance.

vertebra corpus indicated diffuse vertebral brucellar osteomyelitis. Although end plate involvement is the most common feature of the spinal involvement, the end plates were apparently normal in this case. Involvement of the superior end plate of the C-5 vertebra corpus and abscess formation at the paravertebral location were observed in the second case. In our opinion, only superior endplate involvement in the second case indicated focal vertebral brucellar osteomyelitis.

The most common cause of spinal epidural abscess is Staphylococcus aureus. Hematogenous spread is the main route to the epidural space. Spinal epidural abscess usually manifests as excruciating pain localized over the spine and tenderness to percussion, followed by radicular symptoms with subsequent distal cord findings, bowel/bladder disturbance, abdominal distension, and weakness progressing to paraplegia. Epidural abscess associated with brucellosis is rare. Myelopathy and radiculopathy caused by compression of the spine by extradural granuloma may occur in brucellosis. Nerve root compression due to spondylitis or extradural granuloma is uncommon but known in the lumbar region. Myelopathy may also occur as a result of cord compression caused by extradural granuloma in the cervical and thoracic regions. Previous cases of extradural spinal cord compression caused by brucellosis also occurred in the cervical (11 cases) and thoracic (7 cases) regions (Table 1). Cord compression signs were common in all reported cases before radiological evaluation. In our first case, the epidural abscess might have been continuous with the infected vertebra corpus. Dorsal pain was the main presenting complaint of the patient and brucellosis was identified by serological tests, but MR imaging was not obtained until the onset of the progressive neurological deficit. Therefore, neuroimaging evaluation of patients with spinal brucellosis associated with intractable localized pain or paravertebral tenderness is very important for the early diagnosis of epidural abscess formation.

We observed superior end plate involvement in the second case, which is typical for spinal involvement caused by brucellosis. Both diffuse and focal types of brucellar osteomyelitis may cause epidural abscesses. If vertebral involvement associated with brucellosis is suspected, MR imaging must be performed periodically to monitor the possibility of epidural abscess formation.

References


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