



Case Report

Spinal Epidural Abscess of Brucella Mimicking Lumbar Disc Herniation: Case Report and Review of the Literature

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Abstract

A 37 years old woman admitted to neurosurgery clinic, with progressive nerve root compression symptoms for three months. She had no benefit from some medications and physical therapy courses which were performed due to diagnosis of protruded lumbar disc herniation. At the initial evaluation, she had difficulty on standing up and walking. A cyst 2x3 cm in size was removed from L5-S1 level with surgical operation. Brucella was isolated from the pus culture of the cyst content. The patient was completely recovered with postoperative doxycycline and co-trimoxazole therapy. Review of the published reports have suggested that spinal epidural abscess of brucella has predominantly seen in lumbar region and particularly among the patients over 30 years old. Although alone antimicrobial treatment has been increasingly recommended by some authors, surgical operation plus postoperative antibiotic treatment seems to be very effective for short term recovery. Brucella seroagglutination tests may be very helpful in differential diagnosis.

Keywords: Brucella, Lumbar disc herniation, Abscess

Lumbar Disk Herniasyonunu Taklit Eden Brusella Spinal Epidural Disk Absesi: Olgu Sunumu ve Literatür Derlemesi

Özet

Otuzyedi yaşında bir bayan hasta üç aydır devam eden ilerleyici sinir kök bası semptomları ile nöroşirurji kliniğine başvurdu. Hasta, protrüze disk herniasyonu tanısı nedeniyle aldığı bazı ilaç tedavileri ve fizik tedavi programlarından fayda görmemişti. İlk değerlendirmede hasta ayakta durmakta ve yürümekte zorlanıyordu. Cerrahi operasyonla L5-S1 seviyesindeki 2x3 cm boyutundaki kist çıkarıldı. Kist içeriğinden elde edilen pü kültüründe brusella üredi. Hasta, postoperatif doksisiklin ve ko-trimoksazol tedavisi ile tamamen iyileşti. Yayınlanmış olgular gözden geçirildiğinde, brusella nedenli spinal epidural abselerin çoğunlukla lumbar bölgede ve özellikle 30 yaş üzeri kişilerde görüldüğü anlaşılmaktadır. Bazı otörler tarafından gittikçe artan şekilde tek başına antimikrobiyal tedavinin önerilmesine rağmen, kısa zamanda iyileşim için cerrahi müdahale ve antimikrobiyal tedavi kombinasyonu daha etkin olarak görülmektedir. Brusella seroagglütinasyon testleri ayırıcı tanıca oldukça yardımcı olacaktır.

Anahtar Kelimeler: Brusella, Lumbar disk herniasyonu, abse

INTRODUCTION

Brucella is a Gram-negative coccobacillary bacteria that causes zoonotic infections

worldwide. The organism generally spreads to human after consuming unpasteurized dairy products from infected animals. It is typically an intracellular

pathogen localized in granulomatous nodules of reticuloendothelial system organs, including the liver, spleen, lymph nodes, bone marrow, and kidneys⁽²⁰⁾.

The onset of brucellosis is insidious with arthralgia, fever, sweat, and weight loss. Although it may involve many organs, osteoarticular manifestation is seen in over half of the diagnosed patients⁽¹⁾. However, it is envisaged that at least 50% cases of brucellosis are undiagnosed⁽⁷⁾.

Abscess formation is rarely seen during brucella infection. Details of spinal-epidural localization are very limited in the literature. In this paper, we report an epidural brucella abscess located in the lumbar spinal canal, which was formerly misdiagnosed as lumbar disk herniation due to severe nerve root compression symptoms. Additionally, we reviewed similar cases, which were particularly published in the recent decade, to have an overview for brucellar spinal epidural abscess (SEA).

CASE PRESENTATION

A 37-year-old woman admitted to neurosurgery clinic of Malatya State Hospital with sciatic pain radiating into the left leg, formication, and difficulty to straighten up and walk. She reported that her complaints presented 3 months ago had progressively increased despite usage of many analgesics and myorelaxants. She also had a physical therapy course after a disk protrusion image was observed at L5–S1 level on computerized tomography (CT) scan that was performed 1 month ago. There was no history of trauma or constitutional degeneration that could explain the present clinical manifestation. As the clinical manifestation worsened, the physical therapy and rehabilitation specialist suggested obtaining neurosurgical consultation.

During physical examination, the patient was positioned in a right-lateral oblique posture so that she was could not stand upright. Her vital signs were normal. She

localized a painful area on the lower back corresponding to the lumbosacral vertebrae, which had branched to the postero-medial gluteal region, posterior-tibial area, and plantar face of the left foot. Lumbar spine was severely limited in dorsal and lateral motions, and paravertebral muscles were contracted. Patellar reflex was normal bilaterally but Achilles tendon reflex was diminished (+2) on the left side. Hypesthesia was determined in the S1 dermatome particularly for pinprick and light touch sensations. Significant muscle power loss (3/5) was determined in the left foot plantar flexors. Straight leg elevation test was positive at 30° for bilateral legs.

Hematological and biochemical analyses of the blood were normal. C-reactive protein (CRP) was moderately elevated and erythrocyte sedimentation rate (ESR) was 52 mm/h. Plain radiographs of the lumbosacral spine yielded no significant result. In contrast to the initial evaluation report, which indicated protruded lumbar disk herniation, further evaluation using magnetic resonance (MR) images revealed a possible abscess formation, narrowing in the disk space, and endplate edema on both side of the L5–S1 vertebrae (Figure 1, 2). Some additional tests including blood culture and Brucella analysis was performed, and some tumor markers were further studied. Only, brucella lam agglutination test yielded a positive result, and tube agglutination with and without mercaptoethanol was positive for 1/80 dilutions.

The patient underwent L5 hemilaminectomy and discectomy surgery under spinal anesthesia. During the operation, a dirty-brown abscess about 2 × 3 cm in diameter was visualized at the L5-S1 disks' posterior localization. It was compressing the techal sac and left S1 nerve dorsolaterally. Approximately, 1 ml yellowish light-brown pus was aspirated with a syringe and sent to the laboratory for microbiological and histopathological

analyses. Small gram-negative coccobacillary bacteria and many neutrophils were observed during microscopic examination. After 24 hours of aerobic incubation, small gray-white colony growth was observed in the culture plate. Further analyses confirmed that the isolate were *Brucella* spp.

The patient recovered without sequel following 8 weeks of doxycycline and cotrimoxazole therapy. Consequently, she did not report any complaint related with surgery and brucellosis, and no further complication was observed during the control visit after 4 months of surgery. However, the patient refused to perform CT or MR imaging.

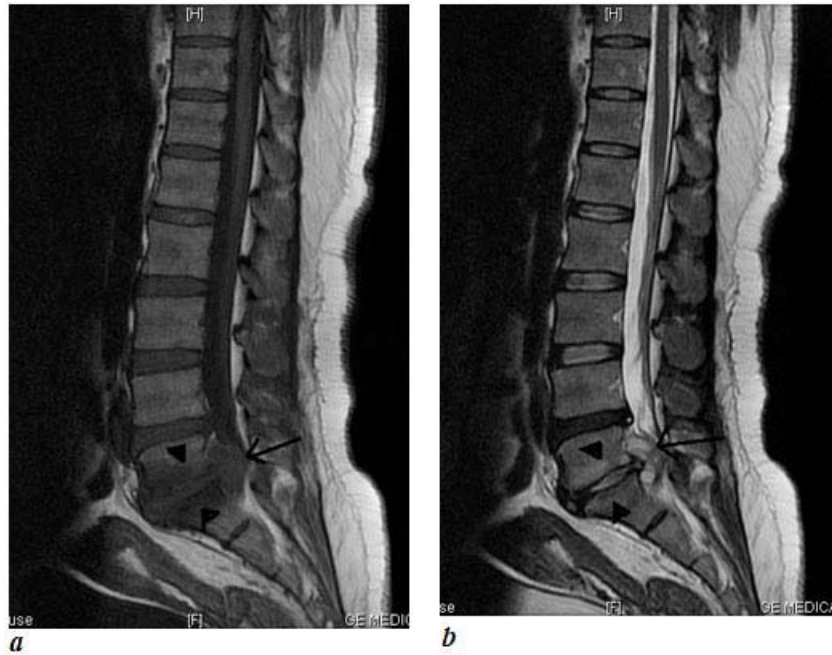


Figure 1: Sagittal T1-weighted (a) and T2-weighted (b) MR images reveals marked endplate edema on both side of L5-S1 intervertebral (arrowhead) disc, and intrathecal thick walled cystic mass (arrows)

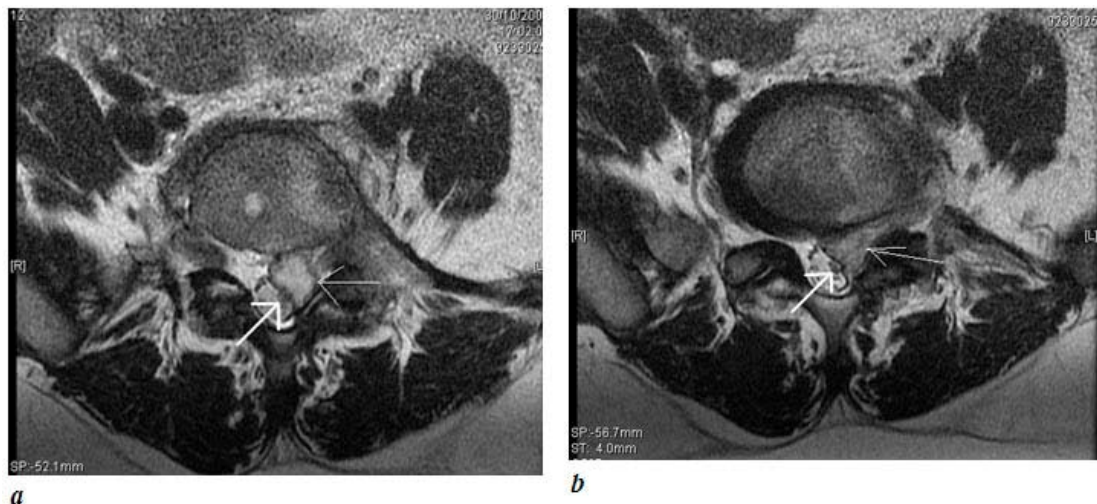


Figure 2: Serial axial T2-weighted images (a, b) through the L5-S1 intervertebral disc confirms intrathecal thick walled cystic mass (thin arrows) and shows compression of left S1 neural root (thick arrows)

DISCUSSION

Brucella is an endemic infection in the Middle East and Mediterranean region. Although it is a controlled disease in many industrialized countries, brucellosis remains a significant public health concern for Turkey. The Health Ministry reported that almost 19,000 Turkish people suffered from brucellosis in 2004 (attack rate: 25.67 per 100,000 population), of whom 6,772 required hospital treatment and 21 died⁽¹⁷⁾. However, it is estimated that the exact number of annual brucella cases was around 50,000-100,000, if all the unreported and subclinical cases were considered⁽⁷⁾.

As it is a very rare complication of brucellosis, there is no adequate data about the incidence of brucellar SEA even in endemic populations. Nevertheless, regarding published reports, it can be seen that the vast majority of cases of brucellar SEA have been reported from Turkey, Spain, and Greece⁽¹⁶⁾.

Clinical symptoms of brucellar lumbar SEA may vary from mild sciatica to severe complaints such as paraplegia, urinary and fecal incontinence, and vertebral fracture^(2,3,19). Although many patients have completely recovered after the therapy, loss of irreversible neuromuscular function and even death has been reported⁽⁴⁾. Clinical manifestation of spinal brucella abscess may depend on both size and localization of the lesion. Like the present case, close localization of a small cyst to a nerve trace may be symptomatic earlier. In addition, inflammation and edema in surrounding tissues may aggravate the symptoms due to compression as well as local tenderness, which have been reported uniformly from all brucellar SEA patients. Unlike the present case, many authors reported co-presentation of brucellar SEA with classic brucellosis symptoms. Fevers, chills, and generalized pain in association with neural deficits were the major complaints of these

patients^(8,18). However, our patient presented only symptoms of progressive nerve root compression. In a detailed interview, she reported an imprecise fever and sometimes fatigue for 1 year, which might be suggestive of chronic brucellosis.

There is no specific patient population for brucellosis. Cetinkaya et al.⁽⁵⁾ reported that any statistical difference was not observed based on the patients' gender and age groups according to their prevalence of brucella sero-positivity. Correspondingly, we did not detect predominance of any gender among the published cases. In contrast, vast majority of the reported brucellar SEA patients were over 30 years old (mean varied between 35–41 years)^(2,8,9,12,13,18,19). However, very few instances of teenagers and a 6-year-old child have been reported⁽¹⁴⁾.

Including our patient, more than 80% of the brucellar SEA were localized on the lumbar vertebrae, particularly L4, L5, and S1 region. Cervical and thoracic involvement were almost equally reported from patients of which C4, C5, and C6, and T7, T8, and T9 localizations were most common.

There is very limited number of laboratory tests for the diagnosis of brucellar SEAs. Hematologic and biochemical analysis of blood frequently yielded no positive result. However, high neutrophil levels may indicate a pyogenic spinal abscess rather than that of *Brucella* etiology. A mild increase in CRP and ESR were reported in majority of the cases, including the present patient. Although brucellosis progresses with recurrent bacteremic episodes, positive blood culture have been reported in very few brucellar SEA cases^(8,9,13,18,19). This is possibly due to the limitation of bacteria in cyst fluid during chronic infection progress. Nevertheless, in addition to the present case, many patients have brucellar SEA diagnosed in postoperative stage, after histopathological examinations and bacterial culture of the

cyst content. However, maximum half of the cyst culture was reported to be positive in the published cases⁽¹⁸⁾. On the other hand, since seroagglutination tests of brucella were uniformly positive in almost all cases, we can suggest that lam and tube agglutinations have been the most valuable preliminary analyses, which can indicate the causative agent.

Plain radiographs of the spinal column cannot be helpful in most cases, unless significant bone degeneration is present. Nevertheless, they may be useful for differential diagnosis of spinal Brucella abscess from degenerative spondylolisthesis, metastatic tumors, or other solid lesions. Discography or CT-discography is reported to be very useful to demonstrate the contrast communicating from the nucleus pulposus into the cyst. However, MR is considered as a superior imaging method that can show not only the nature of the cystic lesion but also its relationship to the thecal sac⁽¹⁰⁾.

The treatment of brucellar SEA includes combination of surgical cyst excision and antimicrobial therapy. Selection of the most appropriate treatment method depends on the abscess' localization and size, and patients' neurologic condition at the time of admission (18). Recently, antimicrobial treatment without surgery, which has been increasingly suggested by authors, using double or triple combination of rifampin, doxycycline, and cotrimoxazole for up to 1 year, if the clinical symptoms are not so severe^(2,8,18). In fact, drug therapy may be more beneficial for patients who cannot tolerate surgery. However, sudden neurologic deterioration was reported in 2 out of 4 patients who were treated with antibiotics; consequently, they underwent surgery⁽¹⁸⁾. Additionally, the favorable therapy period is still uncertain, and almost 30% of adequately-treated patients were stated to develop relapses within the 6-month period^(2,15). Furthermore, relatively longer time may be required for medically-treated patients to

improve neurologic functions. Correspondingly, Bingol et al⁽²⁾ have reported that her patient was able to walk and had menstruated after 5 months and 1 year, respectively, with antimicrobial therapy.

Different surgical approaches are used for cyst extraction, including laminectomy, discectomy, and corpectomy. Like the present case, many patients reported complete resolution of the complaints within a few weeks after the operation, and a relatively shorter duration of antimicrobial usage (6–8 weeks) may be required in the patients who underwent surgery^(9-11,19). Additionally, any brucellar relapse which is common in medically-treated SEA patients, has not been reported after the surgery plus antibiotics treatment. Since recognition of this uncommon entity is very difficult and requires a substantial index of suspicion, successful diagnosis is essential before the onset of a long-term antimicrobial therapy. On the other hand, many patients are diagnosed in postoperative stages after histopathological and/or microbiological analyses. Furthermore, biopsy or surgery is essential in any case to rule out other lesions when the diagnosis is indefinite despite imaging and clinical studies⁽⁶⁾. Therefore, surgery can be regarded as not only a therapeutic intervention but also a diagnostic tool.

Parallel to the widespread use of advanced medical methods, brucellar SEA cases have been increasingly diagnosed in the recent years. However, the pathophysiology of brucellar cyst formation in the epidural area is still unclear. Furthermore, we do not know exactly why it tends to involve the lumbar vertebrae rather than the other parts of the spinal column. Additionally, although Brucella affects almost all age groups, there is no answer as to why the vast majority of patients are over 30 years old.

CONCLUSION

In this paper, we reported the case of a patient with brucellar SEA, who was

formerly suggested for surgery after the diagnosis of a lumbar disk herniation. Although CT and MR could not identify the lesion during initial evaluations, positive seroagglutination tests and postoperative microbiological culture helped identify the pathogen. Since brucellosis and lumbar disk herniation are highly prevalent diseases in our country and their clinical presentation is somehow intersected, it may be better for physicians to consider both diseases in patients presenting symptoms of back pain and nerve root compression.

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