



BRUCELLOSIS SPINAL EPIDURAL ABSCESS WITHOUT SPONDYLODISCITIS: CASE REPORT

SPONDİLODİSKİT OLMASIZIN BRUSELLAR SPİNAL EPİDURAL ABSE: OLGU SUNUMU

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SUMMARY

Spinal epidural abscesses (SEAs) were first described in the medical literature in 1761 and represent a severe, generally pyogenic infection of the epidural space requiring emergency neurosurgical intervention to avoid permanent neurological deficits. SEA is primarily a bacterial infection, and the gram-positive bacteria *Staphylococcus aureus* is its most common causative agent. Brucellosis spinal epidural abscesses are a rare but potentially fatal medical entity, and very few cases have been reported so far. All of the reported cases of SEA due to *Brucella* species have been associated with spondylodiscitis. We present a SEA case due to brucellosis without spondylodiscitis. Magnetic resonance imaging clearly showed the lesion, and the blood and purulent material cultures were positive for *Brucella melitensis*. This case report emphasizes that SEA is not just a complication of Brucellosis spondylodiscitis, pure spinal epidural abscess formation can also be seen.

Key Words: Brucellosis, spinal epidural abscess, spondylodiscitis

Level of Evidence: Case report, Level IV

ÖZET

Spinal epidural apse (SEA) tanımı ilk kez 1761 yılında yapılmış ve spinal epidural mesafenin kalıcı nörolojik defisitlere yol açmadan acil nöroşirurjik girişime gereksinim duyan genellikle piyojenik enfeksiyonu olarak tarif edilmiştir. SEA primer olarak bakteriyel bir enfeksiyon olarak kabul edilir ve gram pozitif *Staphylococcus Aereus* en sık görülen etken patojendir. Brusellar spinal epidural apse nadir görülen, fatal seyirli olabilen klinik bir antitedir. Yayınlanmış tüm brusella SEA olgularında spondilodiskit bulgusu mevcuttur. Bu çalışmada spondilodiskitis bulgusu olmayan bir brusella SEA olgusu sunulmaktadır. MRG ile lezyon net olarak tespit edilmiş, kan ve pürülan materyalde Brucellosis Melitensis izole edilmiştir. Bu makale ile SEA'nin brusella spondilodiskitinin sadece bir komplikasyonu değil, doğrudan spinal apse formasyonu şeklinde görülebileceği vurgulanmak istenmiştir.

Anahtar Kelimeler: Brusella, spinal epidural abse, spondilodiskitis

Kanıt Düzeyi: Olgu sunumu, Düzey IV

INTRODUCTION:

An SEA usually presents as a suppurative process localized between the spinal dura mater and the vertebral periosteum within the spinal epidural space. According to Strohecker and Grobovschek, “the problem with spinal epidural abscesses is not treatment, but early diagnosis – before massive neurological symptoms occur.” Almost all reviews and case studies on SEA emphasize this point^{7,16}. According to Heusner’s staging of SEA, following the initial stage of severe back pain associated with fever and local tenderness in the area of the spinal column, the second stage is dominated by signs of spinal irritation¹⁰.

These manifestations include Lasègue’s, Kernig’s, and Lhermitte’s signs, Brudzinski’s reflex, and neck stiffness. Additionally, back pain can radiate into the arms or legs, depending on the cranio-caudal location of the abscess. In Heusner’s third stage, initial neurological deficits are observed, such as weakness of the voluntary musculature or fecal or urinary incontinence. Sensory deficits may also occur. In the fourth stage, muscle weakness progresses to paralysis. The duration of the individual stages varies. Importantly, the transition to the terminal stage with paralysis can occur very quickly. The combination of severe pain near the spine with fever is characteristic and should always be regarded as a warning sign of a possible SEA. An SEA is primarily a bacterial infection, and the gram-positive *Staphylococcus aureus* is the most common causative agent, accounting for about two-thirds of all cases. *Brucella* species are responsible for only 0.1% of cases¹⁷.

The few reported cases of fungal infections have been seen mainly in immuno-compromised patients, and individual cases of SEAs due

to parasites have been reported from certain geographic regions. Spinal brucellosis usually starts in the superior endplate, an area with a rich blood supply, but occasionally the inferior endplate may also be involved. The subsequent progression 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 vertebrae¹.

SEAs due to brucellosis are considered a complication of spondylodiscitis. This case report emphasizes that SEAs are not just a complication of brucellosis spondylodiscitis, but pure spinal epidural abscess formation can also be seen.

CASE REPORT:

A 28-year-old female patient was admitted to our hospital with complaints of difficulty walking, fever and lower back pain. Neurological examination revealed severe weakness (1/5) of the left ankle dorsiflexion. A sagittal lumbar MRI showed an epidural mass posterior to the L4–5 vertebral bodies (Figure-1), and axial plane MRI revealed that the mass filled the left-side foramen at the L4–5 level (Figure-2). There were no signs of spondylodiscitis.

The initial laboratory values were as follows: white blood cell count: 6600/mm³, hemoglobin: 8.7 g/dl, hematocrit: 29.6%, platelet count: 369,000/mm³, erythrocyte sedimentation rate (ESR): 120 mm/h, C-reactive protein (CRP): 8.57 mg/L, and a normal blood biochemistry profile.



Figure-1. Sagittal MRI shows formation of an L4–5 located spinal epidural abscess. There are no signs of spondylodiscitis.

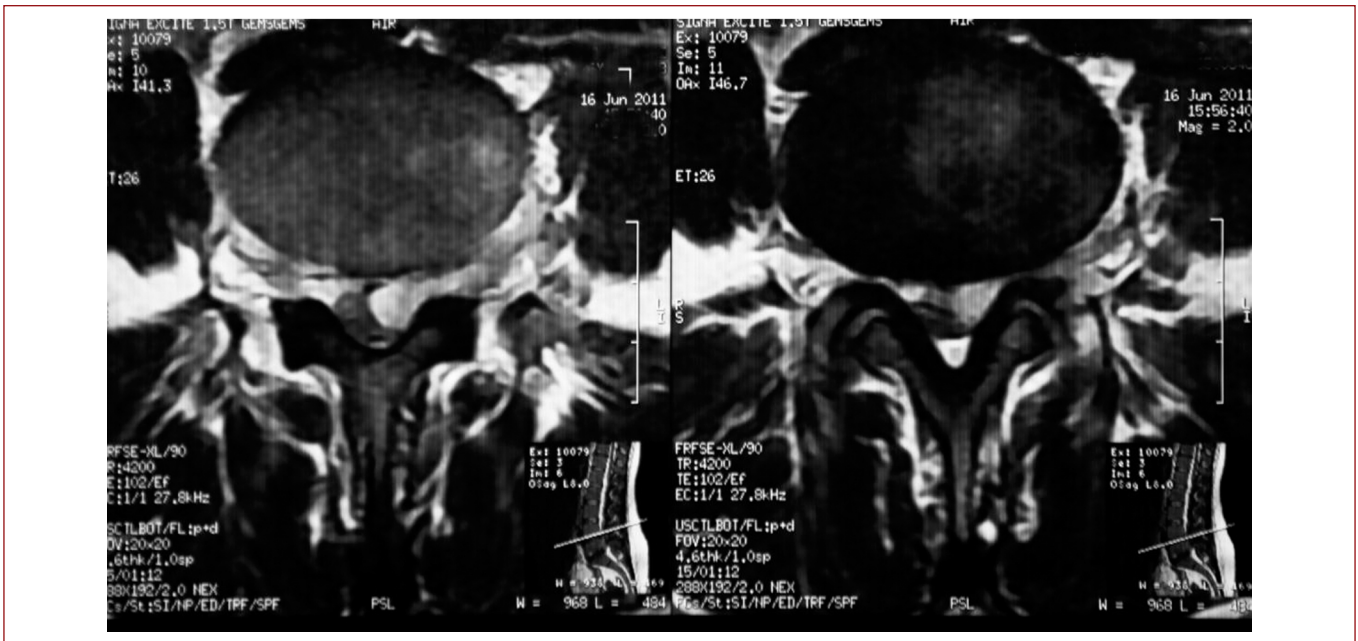


Figure-2. Axial MRI shows a mass filling the left-sided L4–5 foramen.

The patient underwent surgery immediately on diagnosis of non-specific epidural abscess formation. After left-sided L4 hemilaminectomy and flavectomy, we reached the abscess and aspirated a yellowish purulent material. A rapid recovery occurred in the neurological signs and symptoms of the patient after surgery. Broad spectrum antibiotics,

consisting of 1 g IV ceftriaxone BID and 2 g IV ampicilline sulbactam TID were given in the early postoperative period. After receiving the surgical sample and blood cultures results, which revealed the presence of *Brucella melitensis*, and a positive brucellosis sero-agglutination test (titer 1:320), the treatment protocol was changed to a regimen consisting of 200 mg/day doxycycline

and 600 mg/day rifampicin for three months postoperatively.

Written permission was received from the patient to publish this report.

DISCUSSION:

SEAs occur primarily in individuals over 30 years of age. However, previously published case series have reported that younger patients may suffer from SEAs¹⁶. Most patients have some risk factors, such as diabetes mellitus, previous invasive procedures, intravenous drug use, trauma, primary infection focus, alcohol abuse, or immune system disorders.

The combination of severe pain near the spine with fever is characteristic, and should always be regarded as a warning sign of a possible SEA. Back pain is the most common symptom, occurring in 71% of patients, and fever is seen in 66% of patients with an SEA¹⁶. Spinal irritation and signs of initial or progressive neurological deficits have been reported in the international literature to occur in only a fifth to a third of patients.

The manifestations of the different stages of SEA are accompanied by various laboratory findings that reflect severe inflammation, including leukocytosis and an accelerated erythrocyte sedimentation rate. Conventional radiography of the vertebral column is not necessarily reliable for the detection of SEAs, and CT is of limited value in the diagnosis of SEAs.

MRI is able to form multiplanar tomographic images with high contrast among soft tissue structures and without bone artifacts. The use of gadolinium in MRI allows better delineation of SEAs from contiguous structures. No imaging

technique is specific for vertebral brucellosis. The lesions are so heterogeneous that they range from mild erosion of an end plate to severe destructive lesions with large paravertebral masses^{2,15}.

Distinctive MRI features of brucellosis spondylitis include moderately abnormal paraspinal soft tissue without abscess formation, diffuse but exclusively anterior involvement, intact vertebral architecture despite evidence of diffuse vertebral osteomyelitis, disc space involvement, and a lack of gibbus deformity⁽¹²⁾. Typical MRI findings in vertebral osteomyelitis include decreased signal intensity in the disc and adjacent vertebral bodies in T2-weighted images, loss of endplate definition in T1-weighted images, and contrast enhancement of the disc, adjacent vertebral bodies and involvement of paraspinal and paravertebral soft tissues in T1-weighted images. Ring enhancement of paraspinal and epidural processes correlates with abscess formation^{15,8}. Because there was no sign of bony or endplate destruction in this case, the initial diagnosis was a non-specific SEA.

An SEA is primarily a bacterial infection, and *Staphylococcus aureus* is the most common causative agent¹⁶. SEAs due to *Brucella* species are very rare. 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. It is a systemic infection caused by facultative intracellular bacteria of the genus *Brucella* that can involve many organs and tissues^{4,9,11,14}.

Brucellosis may be asymptomatic, with only serological evidence of infection. The manifestations of symptomatic brucellosis may be divided into three forms as follows: acute brucellosis, localized disease, and chronic

brucellosis. Localized brucellosis may occur at any anatomical location, but osteomyelitis is more common than other locations. It involves the vertebrae, especially the lumbosacral region¹³.

Bone involvement is reported in approximately 10% of cases, with the spine, especially the lumbosacral region, most commonly affected. Usually, spinal infection presents as spondylodiscitis with no specific clinical or radiological findings. Neurological complications of brucellosis range from 0–17.8%, with the majority around 2–5%. Spondylitis is a late complication of brucellosis, appearing in 5–10% of cases. Only 15% of spondylitis cases will complicate with an epidural abscess¹⁴.

SEAs due to *Brucella* species are considered complications of brucellosis spondylodiscitis. A high index of suspicion is necessary for diagnosis. Diagnosis depends on the history, physical examination, and blood tests for brucellosis. A positive blood culture is a definite diagnostic tool. Furthermore, culturing surgical or biopsy specimens provides another opportunity to detect the presence of *Brucella* species. After diagnosis, urgent surgical decompression should be performed in cases with moderate to severe neurological deficits, particularly if they are progressive⁴⁻⁶.

Brucellosis is a completely curable infection. Tetracycline is the most common first-line drug. There is no standard therapy protocol for chronic brucellosis, although the first-line antimicrobial regimen tends to be doxycycline and streptomycin (200 mg/day for 12 weeks and 1 g/day for 2–3 weeks, respectively). If there are adverse reactions or contraindications, alternative combinations of antibiotic treatment may be doxycycline and rifampin,

co-trimoxazole and rifampin, ciprofloxacin and rifampin, or ciprofloxacin and streptomycin 1. A combination of a tetracycline and an aminoglycoside remains the most effective regimen because of the synergistic effects. This combination therapy should continue for at least one month, followed by tetracycline and rifampicin for a further 1–2 months³.

Brucellosis spinal epidural abscesses are a very rare condition. An SEA without spondylodiscitis is usually due to a non-specific infection. This case report shows that brucellosis SEAs can be seen without spondylodiscitis. Blood and surgical purulent material should be cultured for *Brucella* species, and serological tests for brucellosis should be evaluated in all cases of SEA.

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