Turkish Neurosurgery 2007, Vol: 17, No: 4, 255-259

A Rare Cause of "Foot Drop": Spinal Epidural Brucella Granuloma

Nadir Görülen Bir "Düşük Ayak" Nedeni: Spinal Epidural Brusella Granülomu

Fatih KÖKEŞ[1]
Ahmet ACIDUMAN[2]
Ahmet GÜNAYDIN[3]
Sami KINIKLI[4]

[1,2,3] S.B. Ankara Etlik İhtisas Hospital, Neurosurgery Department, Ankara, Turkey
[4] S.B. Ankara Eğitim Hospital, Infectious Diseases Department, Ankara, Turkey

ABSTRACT

Spinal epidural abscess is rare but serious medical condition which may cause permanent neurological deficits, or even death, if not treated. Staphylococcus aureus is the responsible microorganism in most spinal abscess cases. However, rarely other microorganisms like Brucella may be responsible. Brucellosis, a zoonotic infection endemic in the Mediterranean Region, Middle East; and South and Central America, most frequently involves the reticuloendothelial and musculoskeletal systems. Vertebral involvement is rarely seen; and its differential diagnosis is rather difficult. Spondylodiscitis and rarely spinal abscess or development of a granuloma are the main pathologies. Spinal epidural granulomas due to brucellosis may cause neurologic deficits. In this case presentation, a spinal epidural brucella granuloma causing foot drop is discussed taking other reports into account.

KEY WORDS: Brucellosis, Foot drop, Spinal epidural abscess, Spinal epidural granuloma

ÖZ

Spinal epidural apseler tedavi edilmezlerse kalıcı nörolojik defisitlere, hatta ölüme de neden olabilen nadir durumlardandır. En sık Staphylococcus aureus nedeniyle oluşan spinal apselere neden olabilen etkenlerden birisi de Brucella türleridir. Zoonotik bir enfeksiyon olan ve Akdeniz Bölgesi, Ortadoğu, Güney ve Orta Amerika'da endemik olan brusellozis, en sık retiküloendoteliyal sistemi ve iskelet-kas sistemini tutar. Vertebral tutulum nadiren görülür ve tanısı oldukça zordur. Spondilodiskitis ve nadiren spinal apse ya da granülom gelişimi söz konusudur. Brusellar spinal epidural granülomlar kendilerini, klinik olarak, nörolojik defisitlerle de gösterebilirler. Kliniğimizde düşük ayak nedeniyle opere edilen bir brusellar spinal epidural granülom olgusu literatür ışığında tartışılmıştır.

ANAHTAR SÖZCÜKLER: Brusellozis, Düşük ayak, Spinal epidural apse, Spinal epidural granüloma

Received: 19.03.2007
Accepted: 15.10.2007

"The article was presented as a poster at the 19th Scientific Congress of Turkish Neurosurgical Society, Antalya, Turkey, May 27-31, 2005"

Correspondence address:
Ahmet ACIDUMAN
Phone: +90 312 2239817
E-mail : ahmetaciduman@yahoo.com

INTRODUCTION

Spinal epidural abscess is a rare but serious clinical pathology which may cause irreversible neurological complications or even death (9). They mainly generate an infectious process within the spinal canal and frequently produce pus. It is considered to be acute if the diagnosis is made within two weeks. In chronic cases (cases diagnosed after two weeks) granulation tissue, rather than pus, is seen in the epidural space (12). Abscesses tend to locate in thoracic and lumbar spine, and less frequently, in the cervical region. The most frequent agent is Staphylococcus aureus (9). However, other microorganisms can also be involved, including Candida glabrata, Streptococcus bovis, Aspergillus spp, Mycobacterium tuberculosis, Haemophilus paraphrophilus, Escherichia coli, Salmonella enteritidis, Pseudomonas spp, Nocardia spp, Pneumococcus spp, and Brucella spp (10).

Brucellosis is an infection caused by a non-encapsulated, aerobic, and gram-negative cocobacillus (4,11). It is a zoonotic infection (1) that frequently occurs as an occupational disease. Many of the patients are farmers, shepherds, those working in manufacturing of dairy products, slaughterhouse workers, veterinaries, and laboratory staff (11). Infection is transmitted to humans via unpasteurized milk and dairy products (4,11).

Brucella is a worldwide infection (1), and is endemic in the Mediterranean Region, Middle East, and South and Central America (9,11). The incidence in Turkey is 0.59 per year per 100,000 population (11). Brucellosis is a systemic disease which invades various tissues and organs, especially the reticuloendothelial and musculoskeletal systems (6-8). Symptoms related to the musculoskeletal system are mostly seen, and the most frequently detected complications are peripheral arthritis, sacroiliitis, and rarely spondylodiscitis that can be complicated by epidural abscess (6,10). The diagnosis and treatment of vertebral involvement is rather difficult (6).

A case of vertebral brucella granuloma with foot drop is presented here and the diagnosis and treatment are discussed taking other reports into account.

CASE REPORT

A forty-six year-old female patient occupied with stockbreeding was admitted to the emergency room with pain in both lower extremities that was more prominent in her low back and left extremity. One week before admission to our center, she applied to an emergency room with weakness in her left foot. Since the Rose-Bengal test applied to the patient was (++), triple anti-biotherapy had been started three days before requesting consultation from our neurosurgery clinic. The physical examination of the patient revealed that low back movements were painful and restricted, The straight leg raising (SLR) test was positive at 40° on the left and at 60° on the right side. Foot drop developed on the left side. She had difficulty in standing up straight because of severe pain.

She was admitted to the neurosurgery clinic for evaluation of flattening in the lumbar axis and destruction in L4-5 vertebral plates on lumbosacral x-ray. A lumbosacral Magnetic Resonance Imaging (MRI) was performed; it could be seen hypointensely on T1-weighted (T1-W) MRI sequences that there was soft tissue between the L4-S1 vertebral levels within the anterior epidural fatty tissue plane consistent with discovertebral infectious findings and abscess formation that surrounded and compressed on the thecal sac in the left lateral aspect (Figure 1A). The lesion was seen as a hyperintense mass on T2-weighted (T2-W) images (Figure 1B). Heterogeneous contrast enhancement was observed on the postcontrast sequences (Figure 1C, D). White blood cell (WBC) count was 6500, erythrocyte sedimentation rate (ESR) was 62 mm/h, alkaline phosphates level was 325 (80-290) and Brucella agglutination test (Wright test) was positive at a dilution of 1:1280.

A surgical intervention was planned for excision of the lesion. Left L4 hemi-partial laminectomy, left L5 hemi-laminectomy, flavectomy, and foraminotomy of L5 and S1 nerve roots were performed. The spinal epidural granulomatous lesion, located between the upper margin of the L4 vertebra and sacrum, was removed. Thecal sac and nerve roots were decompressed. The mass lodge was washed with a solution of rifampicin. Histopathological examination of the material revealed a non-specific granulomatous infection.

The patient was referred to the Infectious Diseases Department. She was admitted for the medical treatment of the infection. Her treatment continued with rifampicin P.O. 600 mg/d, doxycycline P.O. 200 mg/d, and ciprofloxacin 1g/d P.O. for three months.

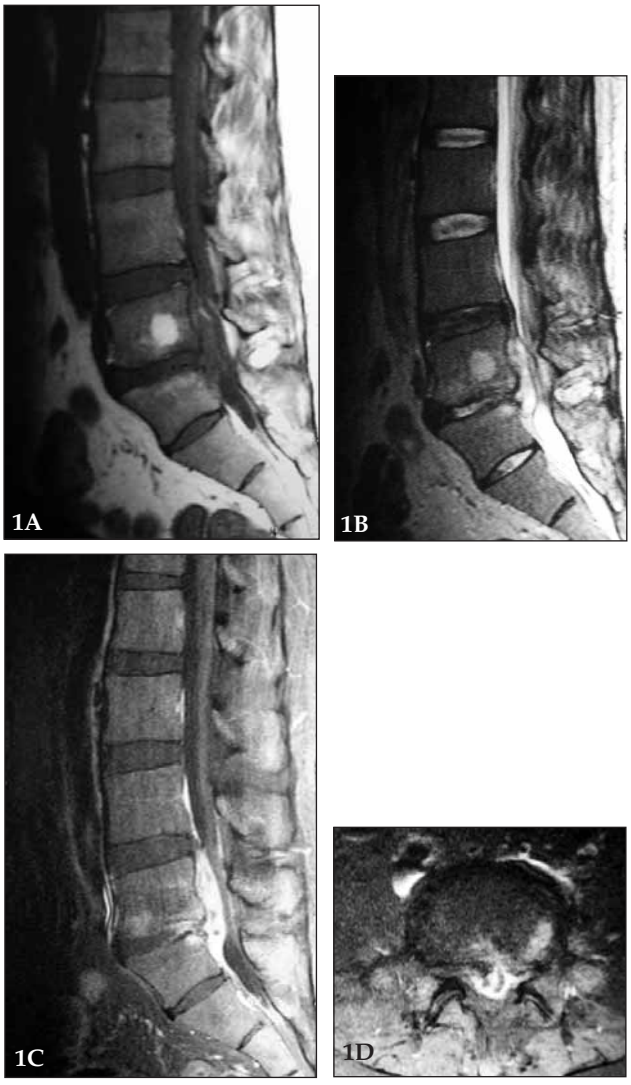

Figure 1: The sagittal T1-W sequence of MRI (**A**) demonstrated a hypo-intense mass in the anterior epidural fat tissue in spinal canal between L4-S1 vertebrae. The sagittal T2-W slice (**B**) of the same lesion was seen as a hyper-intense mass compressing the thecal sac. Decreased height of the L4-L5 intervertebral disc and increased signal intensity in the L4-L5 intervertebral disc were noted. The post-contrast sequences (**C**) revealed a soft tissue enhancing heterogeneously. The linear contrast enhancements in end plates and the posterior part of the L4-L5 disc were observed. The axial slice showed the mass compressing spinal cord on the left lateral side (**D**).

In the control examination of the patient, it was found that SLR was bilaterally unrestricted and the extensor hallucis longus (EHL) and dorsal flexion (DF) muscles had 4/5 strength. WBC count was 8700, ESR was 40 mm/h, the Wright test was positive at a dilution of 1:160, and the alkaline phosphates level was 104 (35-104). On the control lumbar MRI, which was taken three months after the surgery, left L4 and L5 laminectomy defect, and minimal granulation tissue in its neighborhood were observed. No abscess or granuloma were seen (Figure 2A, B).

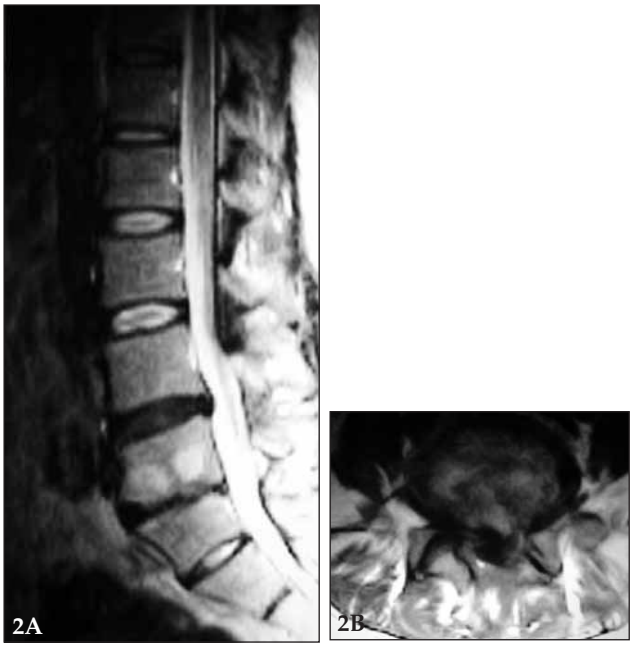

Figure 2: The sagittal T2-W post-operative MRI (**A**) showed neither abscess nor granuloma formation. No contrast enhancement in the end plates and decreased height of L4-L5 intervertebral disc were seen. The hyperintensity seen in the anterior part of the inferior L4 end plate can be explained as type-1 degeneration and shows no contrast enhancement. The post-contrast axial sequence of post-operative MRI (**B**) revealed no contrast enhancement. Minimal granulation tissue can be observed.

DISCUSSION

Brucellosis is a multi-system disease transmitted to humans by consumption of non-pasteurized milk and dairy products from infected cows (4,6,9). There are two forms of spinal brucellosis: focal and diffuse. In the local form, the organism becomes localized in the anterior aspect of the superior end plate, causing a small area of bony destruction. In the diffuse form, the infection spreads throughout the involved vertebra, and to the adjacent vertebrae. Occasionally, granulomatous tissue can develop in the epidural space. The diffuse form and paravertebral or epidural abscess and granulation tissue are more frequently seen in cervical brucellosis than those with lumbar involvement (3). Neurological involvement in brucella infection is seen in 2-5% of

the patients in the form of meningitis, encephalitis, myelitis, radiculoneuritis, brain abscess, demyelinizing syndromes, and meningo-vascular syndromes (8,9). It has been reported that epidural abscesses make up less than 1.5% of neurological complications, and it is generally accompanied by spondylitis (9). The spine is the most frequently involved bony structure in brucellosis (2-53%) (4,5,8). Brucella spondylitis was first described by Kulowski and Vinke (11,13), and is known as one of the most serious complications of brucellosis (11). Brucella spondylitis involves the lumbar region most frequently (4,7) and it may also be seen in the thoracic and cervical regions (1,6). Mousa et al (5) have found a close relationship between brucella spondylitis and central nervous system complications of brucellosis, and have reported neurological complications in 74% of the patients with chronic active or late spondylitis. Myelitis or myelopathies may develop during the course of disease due either to its compressing or inflammatory effect.

Although plain x-rays, bone scintigraphy and computerized tomography (CT) are used for the diagnosis of brucellosis (6), the most advanced diagnostic radiological tool is MRI (4,7,10). MRI is useful in revealing the spondylodiscitis, medulla, root compression and surrounding anatomical structures. Epidural abscess is seen as a mass lesion within the spinal canal and outside to the spinal cord, hyperintense on T2-W, and slightly hypointense on T1-W MRI. Abscess is better delineated with contrast enhancement with Gd-DTPA. There is no particular finding in MRI for brucellar spinal infections (2). In our case, increased signal of L4 -L5 intervertebral disc space and linear contrast enhancement in the end plates made us consider a discovertebral infection. The soft mass lesion in anterior epidural fat tissue was thought that the lesion would be an accompanying abscess formation. On the other hand, developing abscess formation in early stage without involvement of vertebral body should be taken into consideration.

Recognition of spinal epidural abscesses is difficult because of the various nonspecific presentations (5). A high index of suspicion is necessary for the diagnosis of spinal brucellosis (3). Hemograms and ESR are not useful indicators for the diagnosis of brucellosis or spondylitis (5). Diagnosing brucellosis is done via serological tests of blood and cerebrospinal fluid (CSF) (5,10); and serological tests in chronic brucellosis are more sensitive compared to blood and bone marrow cultures. Therefore, performing the Wright test in endemic areas for brucellosis is of great importance (6). A single sample is considered positive at a dilution of 1:160 or more, but repeating the test is advised (11).

Pyogenic osteomyelitis caused by other bacteria (usually staphylococci), tuberculosis, intervertebral disc herniation, actinomycosis, and metastatic lesions, as well as primary tumors of the reticuloendothelial system (such as multiple and solitary myelomas) must be considered in the differential diagnosis (11).

Spinal brucellosis may mimic other diseases affecting the spine, particularly tuberculosis (TB). The most frequent area of involvement in TB is the mid and lower thoracic spine, with more severe destruction of the affected vertebral bodies. Patients with TB are usually younger. The proliferative changes that appear in the healing stage of brucellosis are not seen in TB. Osteoporosis is more marked in TB of the spine, where angulation deformity is also frequent, but this is extremely rare in spinal brucellosis (11).

Brucellosis rarely mimics a herniated disc with acute radicular symptoms. Pain with palpation and local tenderness are the most common but nonspecific findings (2). It should be included in the differential diagnosis of localized back pain, or radiculopathy in patients who live in countries where the disease is endemic (3).

Although some authors have advocated that pharmacological therapy is sufficient for spinal epidural abscesses caused by brucella, surgical intervention is mainstay of treatment in cases presenting with neurological deficits. Reversing the developed neurological deficit, or preventing the developing neurological deficit is crucial. Antimicrobial treatment of brucellosis should be initiated immediately following surgery, or if already started, should be continued after the surgery. It has been reported that continuing the treatment for 12-24 weeks is valuable (5,11).

CONCLUSION

When investigating the etiology of a serious neurological deficit such as foot drop in a country like Turkey, located in the Mediterranean Region and

one of the endemic areas of brucellosis, brucellar epidural abscess or granuloma should be kept in mind, and brucella agglutination tests and MRI should be performed in addition to obtaining a detailed medical history.

Acknowledgement

We thank Dr. Süreyya Boyacıgil for her valuable contribution regarding the neuroradiological evaluation.

REFERENCES

1. Başaranoğlu M, Mert A, Tabak F, Kanberoğlu K, Aktuğlu Y. A case of cervical brucella spondylitis with paravertebral abscess and neurological deficits. Scand J Infect Dis 1999;31:214-5.
2. Görgülü A, Albayrak BS, Görgülü E, Tural Ö, Karaaslan T, Oyar O, Yılmaz M. Spinal epidural abscess due to brucella. Surg Neurol 2006;66:141-7.
3. Güzey FK, Emel E, Sel B, Bas S, Özkan N, Karabulut C, Solak Ö, Esenyel M. Cervical spinal brucellosis causing epidural and paravertebral abscesses and spinal cord compression: A case report. Spine J 2007;7:240-4.
4. Kılıç T, Özer AF, Özgen S, Pamir MN. Brucellar spondylitis mimicking lumbar disc herniation; case report. Paraplegia 1995;33:167-9.
5. Mousa AM, Bahar RH, Araj GF, Koshy TS, Muhtaseb SA, Al-Mudallal DS, Marafie AA. Neurological complications of brucella spondylitis. Acta Neurol Scand 1990;81:16-23.
6. Namiduru M, Karaoğlan İ, Gürsoy S, Bayazıt N, Şirikçi A. Brucellosis of the spine: evaluation of the clinical, laboratory, and radiological findings of 14 patients. Rheumatol Int 2004;24:125-9.
7. Özateş M, Özkan Ü, Bükte Y, Ceviz A, Sarı I, Simşek M. Lumbar epidural brucellar abscess causing nerve root compression. Spinal Cord 1999;37(6):448-9.
8. Özerbil ÖM, Ural O, Topatan Hİ, Erongun U. Lumbar spinal root compression caused by brucella granuloma. Spine Feb 15; 1998;23(4):491-3.
9. Papaioannides D, Giotis C, Korantzopoulos P, Akritidis N. Brucellar spinal epidural abscess. Am Fam Physician May 15; 2003;67(10):2071-2.
10. Pina MA, Ara JR, Modrego PJ, Juyol MC, Capablo JL. Brucellar spinal epidural abscess. Eur J Neurol 1999;6:87-9.
11. Tekkök İH, Berker M, Özcan OE, Özgen T, Akalın E. Brucellosis of the spine. Neurosurgery 1993;33:838-44.
12. Watson T, Benson D. Nondiscogenic back and leg pain. In: Youmans JR, ed. Neurological Surgery, Vol. 4, third ed. Philadelphia: W.B.Saunders, 1990; 2629-63.
13. Yıldız B. Brucella spondilitinde epidural apse ve granülasyon dokusu formasyonu; olgu bildirisi (Epidural abscess and granulations tissue formation in brucella spondylitis: A case report). Türk Nöroşirürji Dergisi 1994;4:49-51 (Turkish).