



DELAYED BRUCELLAR SPONDYLITIS IN A PATIENT WITH A HISTORY OF SPINAL INSTRUMENTATION

SPİNAL ENSTRÜMANTASYON ÖYKÜSÜ OLAN BİR HASTADA GEÇ TANI KONULAN BRUSELLA SPONDİLİTİ

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SUMMARY

Objective: To present and discuss some of the difficulties in the diagnosis of brucellar spondylitis.

Case Report: We report a patient with lower back pain and pain in both hips, misdiagnosed with a delayed infection after spinal instrumentation, who actually suffered from brucellar spondylitis. Diagnosis was established by analysis of the history and a compatible clinical picture, together with a standard tube agglutination titer of $\geq 1/160$ of antibodies for brucellosis. The patient was treated with a combination of oral doxycycline and ciprofloxacin.

Conclusion: An early diagnosis of brucellar spondylitis can often be difficult. In endemic regions, as in Turkey, brucellar spondylitis should always be considered in the differential diagnosis of older patients with back pain and constitutional symptoms. An early diagnosis will help to prevent the development of more severe complications such as spinal cord compression.

Key Words: Brucellosis; diagnosis; spondylitis; spinal instrumentation.

Level of evidence: Case report, Level IV

ÖZET

Amaç: Bu çalışmada brusellar spondilit teşhisinde karşılaşılan güçlüklerin tartışması amaçlanmıştır.

Vaka Sunumu: Her iki kalça ve bel ağrısı şikayeti ile spinal enstrümantasyon sonrası geçikmiş enfeksiyon teşhisi konan, ancak aslında brucellar spondilit olan bir olgu sunulmuştur. Hastaya brusellosis için standart tüp aglutinasyon testi titresinin $\geq 1/160$ olmasının yanı sıra hikayesi ve uyumlu klinik tablo ile teşhis konulmuştur. Hasta oral doksisisiklin ve ciprofloksasin kombinasyonu ile tedavi edilmiştir.

Tartışma: Brucellar spondilitisin erken teşhisi zor olabilir. Bu vakada olduğu gibi endemik bölgelerde, yapısal semptomları ve sırt ağrısı olan hastaların ayırıcı tanılarında brucellar spondilitis mutlaka göz önünde bulundurulmalıdır. Erken teşhis, spinal kord kompresyonu gibi daha ciddi komplikasyonların gelişmesini de önleyecektir.

Anahtar Kelimeler: Brucellosis, diagnosis, spondilitis, spinal enstrümantasyon.

Kanıt Düzeyi: Olgu sunumu, Düzey IV

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INTRODUCTION

Brucellosis is a systemic infection caused by a facultative intracellular, non-encapsulated, non-motile, gram-negative coccobacillus. Brucellosis is a zoonosis, and can occur in all regions of the world¹⁴. The intracellular location of the bacteria protects them from some of the basic mechanisms of the host's immune system and from antimicrobial therapy. Over 500,000 cases of brucellosis are reported yearly to the World Health Organization from 100 countries. *Brucella* can infect multiple organs and tissues, and consequently there are a variety of clinical manifestations of brucellosis^{10,14}.

Osteoarticular disease is the most common complication of brucellosis and has been described in 10–85% of patients^{7,10,14}. The spectrum of bone and joint lesions includes arthritis, bursitis, tenosynovitis, sacroiliitis, spondylitis, and osteomyelitis¹⁴. Spondylitis is an osteoarticular focal complication of brucellosis that often results in residual damage and usually involves the lumbar spine. It may be difficult to diagnose and can be complicated by neurological or vascular conditions¹⁰. The major obstacle to establishing a clinical diagnosis of spondylitis early in the course of brucellosis is the nonspecific nature of the signs and symptoms at presentation, such as spinal pain, fever, and constitutional symptoms^{1,7}. Additionally, brucellar spondylitis tends to affect older individuals, who may display evidence of general osteoarticular degeneration unrelated to *Brucella*.

In the present study, a patient with lower back pain and pain in both hips is reported, who was misdiagnosed as having a delayed infection after spinal instrumentation but who actually suffered from brucellar spondylitis.

CASE REPORT

A 42-year-old male patient was admitted to our outpatient clinic with lower back pain and bilateral hip pain spreading to the posterior part of both lower extremities. His occupation was as a heavy industry worker. He had a history of discectomy and transpedicular screw stabilization at the L5–S1 level, due to recurrent disc herniation and segmental instability 2.5 years previously (Fig. 1). The patient

also had a postoperative wound infection and was re-operated on for debridement. Although the patient had no complaints referable to the spine since then, for the last month he had suffered lower back pain and pain spreading to the bilateral hips, especially during motion. His body temperature was 36.5°C. The straight-leg-raise test was positive bilaterally, reproducing leg symptoms at approximately 45°. There were no sensory deficits or loss of motor function. All hematological and blood chemistry values, except the erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP), were normal. The ESR was 66 mm/h and the CRP was 93.9 mg/dl. Plain radiograms, contrast-enhanced computed tomography (CT) scans and magnetic resonance imaging (MRI) of the lumbar spine revealed edema in the vertebral disc space of L5–S1 and the vertebral bodies of L5 and S1, and irregularity at the end plates (Fig. 2). No abnormality was detected of the pedicle screws inserted previously. The patient was diagnosed with spondylodiscitis of unknown etiology and hospitalized for further evaluation.



Figure-1. X-ray imaging after the initial operation revealing abnormal angulation and segmental instability at the L5–S1 disc level.



Figure-2. X-ray imaging after the second operation showing the pedicle screws in a normal position.

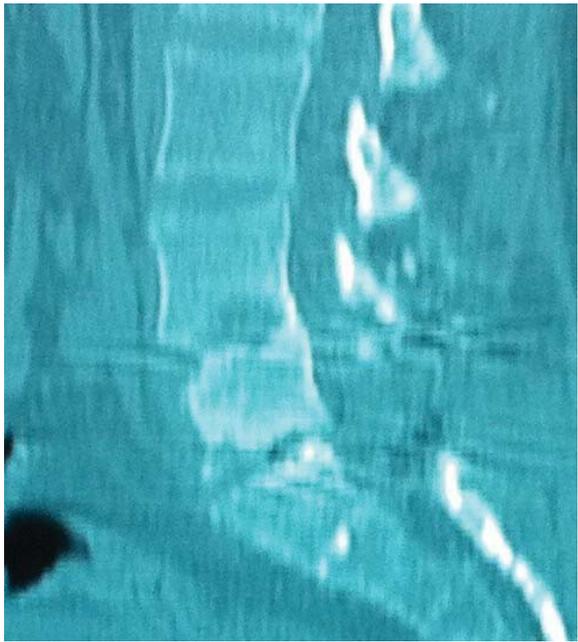


Figure-3. Sagittal CT reconstruction revealing narrowing at the posterior parts of the L4-5 and L5-S1 disc levels, increased sclerosis and contour irregularities of the L4-5 and L5-S1 vertebral endplates, increased sclerosis of the L5 vertebral corpus, widening of the L4-5 disc space, and loss of height of the posterior part of the L4 vertebra.



Figure-4. Lateral X-ray imaging revealing a malposition of one of the L5 pedicle screws.

A CT-guided biopsy of the disc space was performed to exclude delayed spinal infection related to the implantation, and antibiotic treatment with teicoplanin (800 mg per day), metronidazole (200 mg per day), and ciprofloxacin (800 mg per day) was administered. Although no infectious agent was isolated in the biopsy sample, the serum CRP level was 46.1 mg/dl on the seventh day of the antibiotic therapy. As the control plain radiogram revealed malposition of a pedicle screw, the patient was admitted to surgery (Fig. 3,4). The pedicle instrumentation device was removed while the posterior fusion was protected.

Microbiological samples were obtained from the holes of the removed screws and from the disc space. All screws were sent for microbiological examination but no infectious agent was isolated. However, no decrease in serum CRP level was observed after one month of antibiotic treatment.

A detailed clinical history of the patient revealed that the elder brother of the patient who lived in the same house had been hospitalized for the

treatment of an osteoarticular *Brucella* infection of the left hip joint six months previously. On suspicion of brucellar spondylitis, a blood sample was taken for serological tests for *Brucella*. Standard tube agglutination testing yielded positive results for *Brucella* antibodies (titers, 1/1640). Specific treatment for infection with *Brucella spp.* using doxycycline (100 mg every 12 h) and ciprofloxacin was initiated. The patient's complaints relieved after three weeks and completely disappeared after two months. Clinical signs and symptoms of instability were not present, which suggested that posterior lumbar fusion had occurred and so re-instrumentation was not necessary. The patient was then discharged with no back or leg pain, and followed up. The patient follow-up at one year showed favorable developments, with no relapse of symptoms (Fig. 5).



Figure-5. Lumbar MRI showing normal intervertebral disc spaces at the L4–5 and L5–S1 disc levels, and normal height of the L4 and L5 vertebral corpus. Note there is a minimal heterogeneity of signal intensities.

DISCUSSION

Brucellosis is an endemic zoonotic disease, especially in the Middle East and Mediterranean regions, which can involve many organs and tissues⁵. Human infection of workers (for example farmers, shepherds, slaughterhouse workers and veterinarians) results from contact with an infected animal or by ingestion of infected milk or dairy products^{5,10}. Osteoarticular involvement is the most common complication and includes spondylitis, sacroiliitis, and arthritis. Spondylitis is the most prevalent clinical form in adults, varying from 8% to 45.6%^{10,14}. The lumbar segment is the most commonly affected region in brucellar spondylitis, as seen for our patient.

A high index of suspicion is necessary for the diagnosis of spinal brucellosis. Clinical and radiological diagnoses may be difficult, since brucellar spondylitis may resemble many diseases that affect the spine, such as tuberculosis, pyogenic osteomyelitis, intervertebral disc herniation, and malignancy^{4,6}. A delayed diagnosis leads to increased morbidity. In our case, the delay in diagnosis was one month. The patient's back pain was initially attributed to a delayed infection after spinal instrumentation; however, back pain is the leading symptom in most patients with infectious spondylitis¹². A detailed initial history would have revealed that his brother was also diagnosed with brucellosis. Furthermore, brucellosis should have been considered sooner, since it is endemic in our country⁵.

A definitive diagnosis of brucellosis is made when *Brucella* is isolated from blood, bone marrow, or other tissues. Despite the use of Rose-Bengal as a screening test, the standard tube agglutination (STA) test ($\geq 1/160$) has a high sensitivity and specificity. Therefore, performing the STA test in endemic areas for brucellosis is very important^{5,10}. For our patient, brucellosis was diagnosed using the positive result of the STA test (1/1640). However, microbiological samples obtained by means of CT-guided biopsy of the disc space and also during surgical intervention showed no infectious agent.

Delayed infections after spinal surgery, especially in cases that have received instrumentation, have been reported in a few cases^{2,8}. Local pain in the operation region with a generally normal body

temperature and normal vital findings, occurring months or even years after the spinal surgery, are the characteristic signs of delayed spinal infection related to instrumentation¹³. Elevated ESR and CRP serum levels are the most common laboratory findings in delayed infections¹³. In the present case, all hematological and blood chemistry values were normal, except ESR and CRP. Two different theories have been proposed for delayed infections after spinal instrumentation. The first theory is that bacteria of the normal skin flora with low virulency insemminate to the operation site at the time of operation, and after a few months or even years, the infection appears. The second theory is that the infectious agents are spread to the operation region by a hematogenous route^{2,9}. In the present case, the first theory may be valid, as the patient had a postoperative wound infection and was re-operated on for debridement. However, it was finally understood that *Brucella* migrated to the previous operation region by a hematogenous route.

The choice of treatment is generally medical for brucellar spondylitis. However, surgical treatment is indicated in cases with neurological deficit due to medullar and root compression. The need for surgical treatment exists more often in patients with cervical involvement, followed by thoracic involvement. Surgical or percutaneous drainage can be used to treat paravertebral or epidural abscess. In medical treatment, the most widely used antibiotic combinations consist of streptomycin, doxycycline, and rifampicin. The antibiotic treatment must be of a longer duration for spondylitis than for systemic brucellosis without spondylitis^{3,11}. In our study, the initial antibiotic treatment consisted of doxycycline and ciprofloxacin.

CONCLUSION

An early diagnosis of brucellar spondylitis can often be difficult. Particularly in endemic regions, as in the case of Turkey, brucellar spondylitis should always be considered in the differential diagnosis of older patients with back pain and constitutional symptoms. An early diagnosis will prevent the development of more severe complications such as spinal cord compression.

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