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CASE REPORT / *OLGU SUNUMU*

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 low back pain and pain in both hip, misdiagnosed as having a delayed infection after spinal instrumentation who actually suffered from brucellar spondylitis. Diagnosis was established by history and a compatible clinical picture together with a standard tube agglutination titer of ≥1/160 of antibodies for brucellosis. The patient was treated with oral doxycycline and ciprofloxacin combination.
Conclusion: An early diagnosis of brucellar spondylitis can often be difficult. In endemic regions, as in the case of our country, 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 ≥1/160 olmasının yanı sıra hikayesi ve uyumlu klinik tablo ile teşhis konulmuştur. Hasta oral doksisiklin 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

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 world (14). The intracellular location of the bacteria protects it 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 osteomyelitis (14). 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 conditions (10). The major obstacle to establishing a clinical diagnosis of spondylitis early in the course of brucellosis, is the nonspecific nature of the signs

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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.

At the present study, a patient with low back pain and pain in both hip, misdiagnosed as having a delayed infection after spinal instrumentation who actually suffered from brucellar spondylitis was reported.

CASE REPORT

A 42-year-old male patient was admitted to our outpatient clinic with low back pain and bilateral hip pain spreading to posterior part of both lower extremities. His occupation was being an heavy industry worker. He had a history of discectomy and transpedicular screw stabilization at level L5-S1, because of recurrent disc herniation and segmental instability 2.5 years ago (Fig. 1). Also, the patient had a postoperative wound infection and re-operated for debridement. Although the patient had no complaints referable to the spine since then, for the last month he suffered low back pain and the pain spreading to 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 degrees. There was no sensory deficit and no loss of motor function. All hematological and blood chemistry values except the erythrocyte sedimentation rate (ESR) and C-reactive protein (CRP) were normal; ESR was 66 mm/h and CRP was 93.9 mg/dl. Plain radiograms, contrast-enhanced computed tomography (CT) scan and magnetic resonance imaging (MRI) of the lumbar spine revealed edema at vertebral disc space of L5-S1 and vertebral bodies of L5 and S1, and irregularity at the end plates (Fig. 2). No abnormality was detected in pedicule screws inserted previously. The patient was diagnosed with spondylodiscitis of unknown etiology and hospitalized for further evaluation.

CT-guided biopsy of the disk space was performed to exclude delayed spinal infection related with implantation and antibiotic treatment with Teicoplanin (800 mg per day), Metronidazole (200 mg per day), Ciprofloxacin (800 mg per day) was administrated. Although an infectious agent was not isolated in the biopsy sample, serum CRP level was 46.1 mg/dl on the 7th day of the antibiotic therapy. As the control plain radiogram revealed malposition in a pedicle screw the patient was admitted to the operation room (Fig. 3,4). Pedicle instrumentation device removed while posterior fusion was protected. Microbiological samples were obtained from the holes of removed screws and from

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

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

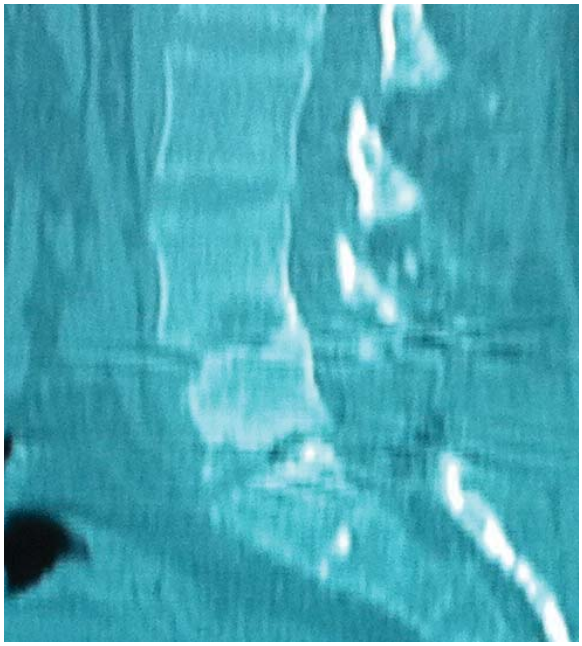

Figure – 3. Sagittal CT reconstruction reveals narrowing at posterior parts of L4-L5 and L5-S1 disc levels, increased sclerosis and contour irregularities of L4-L5 and L5-S1 vertebrae end-plates, increased sclerosis of L5 vertebral corpus, widening at L4-L5 disc space and loss of height of posterior part of L4 vertebra.

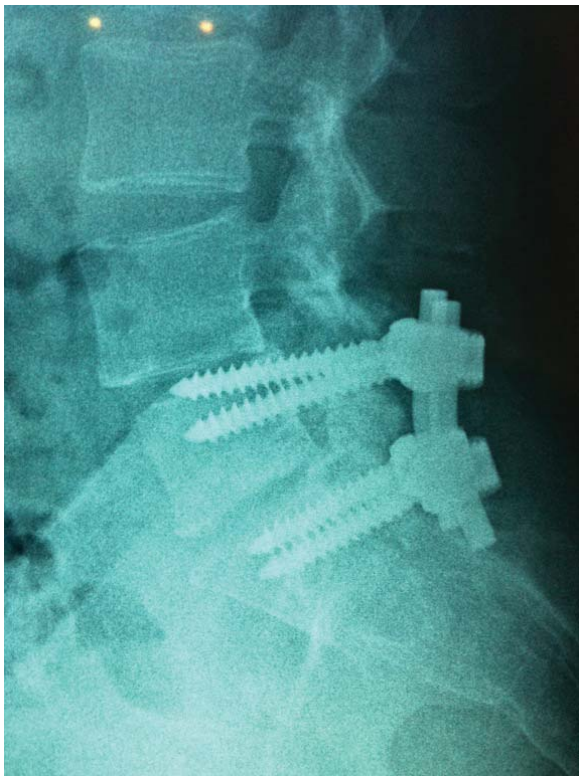

Figure – 4. Lateral x-ray imaging reveals a malposition of one of the L5 pedicle screws.

the disc space. All screws were sent for microbiological examination but an infectious agent was not isolated. Besides, no decrease in serum CRP level was observed after one month of antibiotic treatment.

Detailed clinical history of the patient revealed that elder brother of the patient living in the same house was hospitalized for treatment of osteoarticular Brucella infection of left hip joint 6 months ago. On suspicion of brucellar spondylitis, blood sample was taken for serological tests for Brucella. Standard tube agglutination testing yielded positive results for antibodies to Brucella (titers, 1/1640). Specific treatment for infection with Brucella spp. with 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 suggests that posterior lumbar fusion occurred and re-instrumentation was not necessary. Then, the patient was discharge with no back or leg pain and taken under follow-up. Patient follow-up at one year has shown a favorable evolution, with no relapse of symptoms (Fig. 5).

Figure – 5. Lumbar MR imaging showing normal intervertebral disc spaces at L4-L5 and L5-S1 disc levels and normal height of 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, and can involve many organs and tissues (5). 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 its most prevalent clinical form in adults and varies from 8% to 45.6% (10,14). The lumbar segment is the most commonly affected region in brucellar spondylitis as seen in our patient.

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). Delayed diagnosis leads to increased morbidity. In our case, the delay in diagnosis was 1 month. His back pain was initially attributed to delayed infection after spinal instrumentation; however, back pain is the leading symptom in most patients with infectious spondylitis (12). A detailed initial history would have revealed that his brother was diagnosed with brucellosis. Furthermore, brucellosis should have been considered since it is endemic in our country (5).

A certain 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 (≥1/160) has high sensitivity and specificity. Therefore, performing the STA test in endemic areas for brucellosis is very important (5,10). In our patient, brucellosis was diagnosed using the positive result in STA test (1/1640). However, microbiological samples obtained by means of CT-guided biopsy of the disk space and also during the surgical intervention revealed no isolation of an infectious agent.

Delayed infections after spinal surgery, especially in cases that performed instrumentation, have been reported in few cases (2,8). Local pain in operation region, generally normal body temperature and normal vital findings, occurring months or even years later than spinal surgery are the characteristic signs of delayed spinal infection related with instrumentation (13). Elevated serum levels of ESR and CRP are the most common laboratory findings in delayed infections (13). In the present case, all hematological and blood chemistry values except ESR and CRP were normal. Two different theories have been proposed in delayed infections after spinal instrumentation. The first theory depends on the idea that low virulent bacteria of normal skin flora inseminate 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 operation region via hematogenious route (2,9). In the present case, the first theory may be valid as the patient had a postoperative wound infection and re-operated for debridement. However, it was finally understood that Brucellar agents migrated to previous operation region via hematogenious route.

The choice of treatment is generally medical in brucellar spondylitis. However, surgical treatment is indicated in case of neurological deficits 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 duration of antibiotic treatment must be longer in spondylitis than in systemic brucellosis without spondylitis (3,11). In our study, 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 our country, 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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