Case Report / *Olgu Sunumu*

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Isolated Cervical Spondylodiscitis Due to Brucellosis: A Case Report

Bruselloza Bağlı İzole Servikal Spondilodiskit: Bir Olgu Sunumu

Onur URAL, Şua SÜMER, Nazlım AKTUĞ DEMİR*, Nebahat DİKİCİ, Vildan FIRAT**

Selcuk University Faculty of Medicine, Department of Clinical Microbiology and Infectious Diseases, Konya, Turkey

** Adıyaman State Hospital, Department of Clinical Microbiology and Infectious Diseases, Adıyaman, Turkey*

*** Erzincan State Hospital, Department of Clinical Microbiology and Infectious Diseases, Erzincan, Turkey*

Summary

Brucellosis is a zoonotic disease, which has the potential to affect all organs and systems. It most commonly involves the musculoskeletal system and may result in sacroiliitis, spondylitis, spondylodiscitis, arthritis, osteomyelitis, tenosynovitis or bursitis. Lumbosacral region is the most commonly affected in cases of spinal complications. Involvement of the cervical region is very rare, however, when it does occur, the prognosis is worse than when it affects the lumbar region and more neurological damage is observed. Brucellosis should always be considered in the differential diagnosis of patients presenting with musculoskeletal complaints in regions where brucellosis is endemic. We report an unusual case of brucellar spondylodiscitis involving the cervical region and discuss this with reference to the recent literature. *Turk J Phys Med Rehab 2013;59:256-9.*

Key Words: Brucellosis, discitis, drug treatment

Özet

Bruselloz tüm organ ve sistemleri tutabilme özelliğine sahip zoonotik bir hastalıktır. Brusellozda en sık kas iskelet sistemi tutulumu izlenir. Bu tutulum sonucu bruselloza bağlı sakroileit, spondilit, spondilodiskit, artrit, osteomyelit, tenosinovit veya bursit gelişebilir. Lumbosakral bölge, spinal tutulumlar içinde en sık tutulan bölge olup özellikle servikal bölge tutulumu çok nadir görülür. Bruselloza bağlı servikal tutulumda prognoz lomber tutuluma göre daha kötü olup, daha fazla nörolojik hasar izlenmektedir. Brusellozun endemik olduğu bölgelerde, kas iskelet sistemi şikayetleri ile başvuran hastaların ayırıcı tanısında bruselloz unutulmaması gereken bir hastalıktır. Bu yazıda nadir görülen bir vaka olan servikal tutulum gösteren bruselloz olgusu sunularak literatür eşliğinde tartışıldı. *Türk Fiz Tıp Rehab Derg 2013;59:256-9*.

Anahtar Kelimeler: Bruselloz, diskitis, ilaç tedavisi

Introduction

Brucellosis is a zoonotic disease which is endemic in our country and can potentially involve all organs and systems, though it most commonly affects the musculoskeletal system. As a result of brucella infection, sacroiliitis, spondylitis, spondylodiscitis, arthritis, osteomyelitis, tenosynovitis or bursitis may develop (1,2). Lumbosacral region is the most commonly affected region in case of spinal complications (1,3,4). With appropriate treatment, recovery is observed in 60-90% of patients (5). Brucellosis should be considered in the differential diagnosis of patients who present with musculoskeletal complaints in regions where brucellosis is endemic.

In this article, a case of brucellosis with a cervical involvement is presented and discussed in the light of the literature.

Case

A fifty-one-year-old female patient presented to the Physical Medicine and Rehabilitation Clinic with neck pain radiating down the left arm. The patient was pre-diagnosed with cervical discopathy and an image accordant with spondylodiscitis was detected on cervical magnetic resonance imaging (MRI). Therefore, the patient was transferred to our clinic with a pre-diagnosis of brucellosis. Her medical and family history revealed no contagious disease or unusual features. Her body

Address for Correspondence:/Yazışma Adresi: Şua Sümer MD, Selcuk University Faculty of Medicine, Department of Clinical Microbiology and Infectious Diseases, Konya, Turkey
Phone: +90 332 241 50 00 E-mail: suasumer@gmail.com

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temperature was 36.6°C, her pulse was 80/min and blood pressure was 110/70 mmHg. Her abdomen had a bow-like appearance and the liver was palpable with 1-2 cm in the abdominal examination. In the examination performed, neck movements were restricted in all directions. Muscle power of the deltoid muscle of the left arm was 3/5 and biceps muscle of the left arm was 4/5. Other muscle powers and sense examinations were normal. In evaluation of deep tendon reflexes; reflex for the left biceps was decreased and other reflexes were normal. Neck stiffness, Kernig, Brudzinski and bilateral Babinski signs were negative.

Laboratory tests were as follows: hemoglobin level: 9.8 g/dl (11-16.5 g/dl), leukocyte count: 6.300/mm^3 (49% lymphocyte, 49% polymorphonuclear leukocyte, 2% monocyte) (4.000-10.000/mm^3), platelet count: 270.000/mm^3 (150.000-400.000/mm^3), erythrocyte sedimentation rate (ESR): 70 mm/h (0-20 mm/h), C-reactive protein (CRP) level: 85 mg/L (0-5 mg/L). Biochemical test results were normal. Rheumatoid factor (RF), antinuclear antibody (ANA) and anti-dsDNA were negative. Standard tube agglutination test showed a positive reading in 1/5120 titration. On abdominal ultrasonography, liver size was found to be increased (16 cm) while the spleen and other organs were normal. Cervical MRI scans revealed spondylodiscitis at the C4-5 level (Figure 1 and 2).

The patient had no fever during her follow-up assessments and Brucella melitensis was detected in her blood cultures which were taken at the time of hospitalization. Doxycycline (2x100 mg/day) and streptomycin (1x1 g/day) treatment was started with the diagnosis of Brucellosis. Diclofenac sodium and paracetamol were given for pain relief. After twenty-one days of treatment, a decrease in pain and recovery of movement of the neck and the arm were reported. Laboratory tests were as follows: ESR: 35 mm/h and CRP: 15 mg/L. Streptomycin was discontinued after 3 weeks of administration and the treatment was continued with doxycycline. Treatment for spondylodiscitis was applied for 12 weeks in total including streptomycin and doxycycline treatment for three weeks and single doxycycline treatment for nine weeks. Laboratory parameters returned to normal limits completely at the end of 12 weeks of treatment (ESR: 15 mm/h, CRP level: 3.4 mg/L) and the clinical sings recovered. Partial recovery was also monitored on cervical MRI (Image 3). As radiological recovery may be later, radiological follow-up was approved.

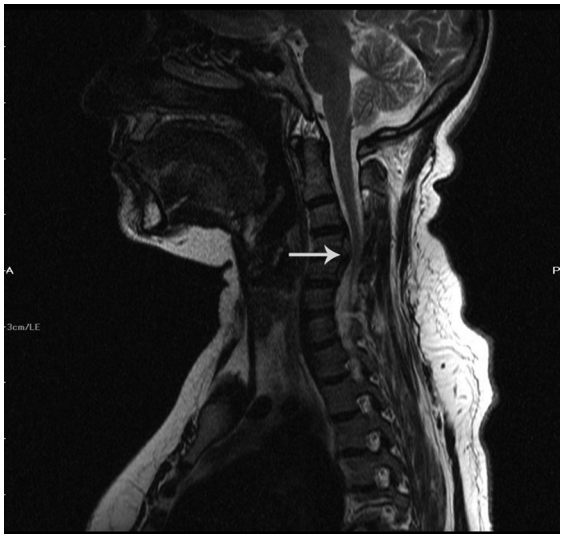

Figure 1. Sagittal T2 weighted image demonstrating cervical kyphosis associated with soft tissues (arrow) filling the epidural space.

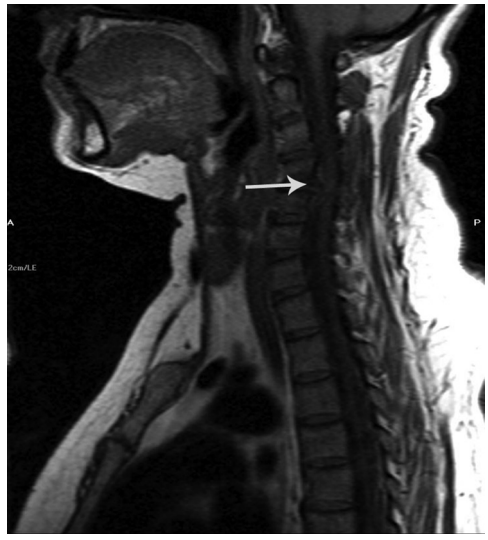

Figure 2. Sagittal T1 weighted image obtained after administration of intravenous contrast material revealing enhancement of the corpus especially at the endplate of vertebrae C4-5 secondary to dyscitis. Marked thickening of the soft tissues (arrow) in the epidural space is also seen.

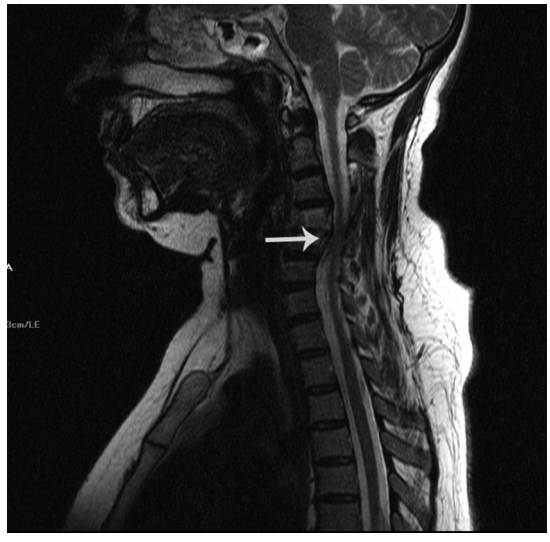

Figure 3. Sagittal T2 weighted image demonstrated the progression of inflammatory enhancement in the vertebral corpus and disruption of the disk posteriorly, with near partial resolution of the soft tissue thickening (arrow) in the epidural space on control MR imaging after therapy.

Discussion

Brucellosis is still a major problem in developing countries. Infection may result from consumption of unpasteurized milk products, being involved in stock breeding and occupational contact (2,6,7). However, the contamination pathway cannot be always detected from a patient's medical history (2,7). In a study by Ural et al. (7), who evaluated 86 cases of brucellosis, no contamination pathway could be detected in 40.8% of cases. Similarly, no contamination pathway was detected in 40.6% and 41.7% of cases in studies by Kartal et al. (8) and Aydemir et al. (9), respectively. Our case had no history of contamination.

Brucellosis usually presents with non-specific symptoms such as fever, chill, night sweating, asthenia and arthralgia. Complaints relating to the affected region may be observed as organ or system irregularities. In case of musculoskeletal involvement, muscle, joint and bone pain, and sometimes neurological symptoms, such as power loss, paresthesia, and paraparesis can be observed (1,2). Bal et al. (10) stated that lumbar, leg and joint pain are usually the first complaints reported by patients. Nas et al. (11) reported a case of brucellosis-induced cervical spondylodiscitis, in which the presenting complaint has been neck pain. The presenting complaint in our case was pain in the neck spreading to the left arm.

The main physical examination findings of brucellosis have been reported to be fever, hepatomegaly, splenomegaly and, less frequently, lymphadenopathy. Additionally, specific physical examination findings included organ and system based irregularities (1,2). In our case, hepatomegaly, limited head movements due to cervical involvement, left arm weakness and flexion limitation were detected on physical examination.

Osteoarticular complications in brucellosis have been reported in 10-80% of cases. As a result of this involvement, sacroiliitis, spondylitis, spondylodiscitis, arthritis, osteomyelitis, tenosynovitis or bursitis may develop (1,2,12,13). The lumbosacral region is the most frequently involved region, while the thoracic and cervical regions are less likely to be affected (5,14,15,16). Involvement of the cervical region is particularly rare (1,3,4). In a study by Özön et al. (4) in which cases of spondylitis and sacroiliitis due to Brucella infection were compared, cervical spondylitis was reported as the least frequent form. In studies by Bodur et al. (17), Kurtaran et al. (18) and Zormpala et al. (19), the rate of isolated cervical spondylitis were reported to be 7.7%, 4% and 1.2-2.1%, respectively. However, isolated cervical spondylodiscitis due to brucellosis was detected in our case.

Prognosis in cases with cervical involvement due to brucellosis is worse than in cases involving the lumbar region, and more neurological damage is observed (19). In a review by Turgut et al. (20), it was reported that in cases with localized spinal involvement neurological deficit rates were observed to be varying between 10% and 43%. In our case, weakness in the left arm and flexion limitation were present.

Diagnosis of spondylodiscitis due to brucellosis is difficult because of non-specific symptoms and findings. Serological tests are the most frequently used methods in the diagnosis. The diagnosis is established by detection of a titer of 1/160 and over in a standard agglutination test or a four-fold increase in the titer of a patient for whom brucellosis is considered likely but who has a standard agglutination test result of under 1/160. In routine laboratory test, brucella standard tube agglutination test has been studied up to 1/640 titration (1,2,21). Since a positivity over 1/640 was detected in our patient's brucella agglutination test, the test was prolonged. A positive brucellosis standard tube agglutination test result in our case was detected in a 1/5120 titration.

Definitive diagnosis of brucellosis is established by noting the reproduction of Brucella bacteria in cultures taken from appropriate samples (1,2). Culture of the Brucella bacterium is not always possible, but blood cultures should always be taken even if the patient does not have fever (22). In a study by Yılmaz et al. (22), Brucella bacteria were detected in blood culture in 32% of cases of spondylodiscitis due to brucellosis. In our case, B. melitensis was found reproducing in blood cultures which were taken in a feverless period.

The most sensitive imaging method that can be used in the evaluation of musculoskeletal involvement due to brucellosis is MRI. It is used for early diagnosis and in follow-up of treatment response. Lesions may be detected after approximately three months via direct imaging. Therefore, direct imaging is not very sensitive for evaluating spinal involvement (1,3,13). As MRI is the most sensitive method available for the evaluation of soft tissues, it is used as a guide for evaluation of abscesses and spinal cord complications which may occur in cases of spinal infection. Via MRI, it is possible to observe recovery after six weeks of treatment. Contrast-enhanced MRI should be preferred to monitor recovery of lesions in the treatment follow-up. Radiological recovery may be later in patients whose appropriate treatment period is completed, therefore, radiological monitoring should be suggested when required (3). Our case showed spondylodiscitis in C4-5 on her cervical MRI and recovery and repair of the cervical lesion was assessed by a follow-up MRI.

A combination of doxycycline and aminoglycoside is frequently used in cases of brucellosis which progress with musculoskeletal involvement. Treatment periods range between 6 and 12 weeks in brucellosis cases with spondylitis or spondylodiscitis (1,5,21). Through recommendations of WHO, aminoglycoside treatment is applied for 2 to 3 weeks and the treatment continues with doxycyclin for an appropriate period. Previous studies have shown that use of streptomycin longer than 2 to 3 weeks does not increase the response to the treatment, rather increases the side effect rates (hearing loss, dizziness) (23,24). Treatment should be continued until the ESR decreases to normal levels and radiological recovery is maintained. Clinical recovery is observed in 60-90% of brucellosis cases with spondylitis using this combined treatment. The treatment period should be at least 3-6 months in cases where an abscess is recorded (1,5,21). As cervical spondylodiscitis was detected in our case, a combination treatment including doxycycline and streptomycin was started. Treatment for spondylodiscitis was applied to the patient for 12 weeks in total including streptomycin and doxycycline treatment for three weeks

and single doxycycline treatment for nine weeks. Laboratory parameters and clinical findings were recovered and partial recovery was monitored on radiological scans; therefore, total treatment was discontinued by completing to 12 weeks.

Since brucellosis may involve all organs and systems, a range of different clinical findings may be noted. In endemic regions, brucellosis should always be considered in the differential diagnosis of lesions which involve cervical vertebrae.

Conflict of Interest

Authors reported no conflicts of interest.

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