

SPINAL AND TESTICULAR SOFT TISSUE INVOLVEMENT CAUSED BY BRUCELOSIS: A CASE REPORT

BRUSELLA KÖKENLİ OMURGA VE TESTİS YUMUŞAK DOKU TUTULUMU: OLGU SUNUMU

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SUMMARY:

Brucellosis is a systemic infection, caused by facultative intra-cellular bacteria of the genus Brucella, that can involve many organs and tissues. We present a 42-year old man with spinal and testicular soft tissue involvement caused by brucellosis. Complete recovery was achieved after 12 weeks triple antibiotic treatment.

Key words: *Brucellosis, spinal brucellosis, testicular infection, treatment*

Level of Evidence: *Case report, Level IV.*

ÖZET:

Brusellozis, brucella bakterisinin doku içi fakültatif yerleşimi sonucu gelişen, bir çok doku ve organı tutabilen sistemik bir enfeksiyonudur. Bu çalışmada brusella nedeniyle omurga ve testiküler yumuşak doku tutulumu olan 42 yaşında bir erkek hasta sunulmuştur. 12 haftalık antibiyoterapi ile tam bir iyileşme sağlanmıştır.

Anahtar Kelimeler: *Brusellozis, omurga brusellozisi, tetiküler enfeksiyon, tedavi*

Kanıt Düzeyi: *Olgu sunumu, Düzey IV.*

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INTRODUCTION:

Brucellosis is a systemic infection, caused by facultative intra-cellular gram-negative bacteria of the genus *Brucella*, that can involve many organs and tissues. For this reason, brucellosis is frequently included in the differential diagnosis of a variety of clinical pictures that involve many medical specialists⁽⁶⁾. The spine is the most common site of musculoskeletal involvement, followed by the sacroiliac joint⁽⁹⁾. Four species are responsible for brucellosis in humans: *Brucella abortus*, *Brucella suis*, *Brucella canis* and *Brucella melitensis*^(1,17). Brucellosis occurs worldwide, with high endemicity in areas such as the Mediterranean countries, the Arabian Peninsula, Latin America, and Asia⁽¹²⁾. The brucellian infection mainly affects organs rich in mononuclear phagocytes, such as the liver, the spleen, the lymph nodes and the bone marrow⁽¹¹⁾. The aim of imaging in spinal brucellosis is to allow an early diagnosis and a precise assessment of the extent of the disease, so that possible neurological complications may be detected. The early diagnosis of brucellosis is very important because of the different organs and tissue involvement on late-onset.

CASE REPORT:

A 42-year-old man presented with a history of lumbal pain and a fever of 37.5-38°C that occurred sometimes. He had been admitted to another hospital with the complaints of fever, cough without sputum and lumbal pain and antibiotic therapy was given for a suspected atypical pneumonia. He had a history of consuming cheese made from raw cow milk. Neurological examination was normal. Physical examination revealed bilateral testicular odemea, erytemea and calor. Ultrasonogram

of the testicles disclosed enlargement of the testes and epididymes and nonhomogenous hypoechoic focal echo patterns. Laboratory test results were normal except for the C-reactive protein (CRP) (60 mg/dl), and erythrocyte sedimentation rate (ESR) (80 mm/hr). The Rose Bengal test was positive, and the *Brucella* seroagglutination test (Wright agglutination test) was positive at a titer of 1/1280. The BACTEC 9240 blood culture system (BD, USA) for initial blood cultures with subculturing onto chocolate blood agar at 37°C in CO₂ was used. At least 2 blood cultures were carried out before treatment. Diagnosis of brucellosis was established by isolation of *Brucella melitensis* in blood. The MRI imaging revealed soft tissue involvement at the level of L3-L4 (not abscess). Osteoarticular complication was defined as pain and two or more inflammatory signs in any peripheral joint (heat, redness, swelling, definite restriction of movement) or inflammatory pain (pain unrelieved with rest) together with radiologic alteration and/or radionuclide uptake in a deep location evaluated by both the clinician and the radiologist independently⁽⁶⁾. All clinical findings (including testicular soft tissue involvement) lead us to think that these complications were because of brucellosis. Triple antibiotic treatment was administered (doxycycline 2x100 mg per oral, streptomycin 1x1 gram intramuscular, and ciprofloxacin 2x500 mg per oral for 12 weeks). Complete recovery was found at the follow-up examination, laboratory tests and MR imaging.

DISCUSSION:

Brucella species can infect a wide variety of domestic and wild animals⁽¹²⁾. *B. melitensis* is

the most common agent in Turkey among animals, especially sheep and goats ^(1,13). It is primarily found in animals and is transmitted to human by direct contact with infected animals or by consuming contaminated unpasteurized milk ^(5,13,16). The clinical manifestation of brucellosis is very great, ranging from asymptomatic infection to serious debilitating disease ⁽¹²⁾. Brucellosis infection spreads hematogenously to tissues and almost every organ can be affected ^(1,2). Symptoms are nonspecific and may include fever, chills, weight loss, sweats, headache, myalgia, fatigue, and depression ⁽⁷⁾.

According to Colmenero et al. ⁽⁶⁾ 31% of patients had focal forms, rates similar to those reported by others ^(4,10). Among focal forms, spinal involvement is among the most important complications of human brucellosis, and it may have widely variable manifestations. The disease can affect the musculoskeletal system at virtually any site, but in adults the axial skeleton is the most frequently involved. Sacroiliitis and spondylitis account for more than 80 % of patients with osteoarticular involvement. This tendency for axial involvement in adults may be related to closure of the metaphysis, change in bone vascularization, and redistribution of the bone marrow from the long to the axial bones, which occurs from the second decade of life ⁽⁶⁾.

Spinal involvement has increased rapidly around the world in recent years, especially in the endemic countries ⁽³⁾. In particular, more widespread use of MRI has allowed more accurate and frequent detection of the spinal form of this infectious disease ⁽¹⁴⁾. Clinical symptoms of spinal brucellosis may include moderate fever and spinal pain of variable intensity and of mixed type. Physical examination usually shows a "spinal

syndrome", with a segmental spinal rigidity and paravertebral muscle contracture. Pressure applied to the spinous process of the involved vertebrae elicits pain. In spondylitis, the lumbar segment is the most frequently involved, followed by the thoracic, and finally the cervical segments ⁽⁶⁾. It is uncommon for brucellar spondylitis to present with spinal cord or nerve root compression. The long latent stage between the onset of symptoms and the appearance of the radiologic changes (from 2 weeks to 8 weeks) may prevent early diagnosis ⁽¹⁾.

MRI is the imaging method of choice for the diagnosis and follow-up of brucellar spondylitis. MRI has high sensitivity for detecting the disease in the early stages and provides excellent definition of paravertebral and epidural extension ^(1,13,15,17).

Treatment of brucellosis must control the illness effectively and prevent complications, including relapse. The regimen of choice and duration of antimicrobial therapy should be based on the presence of focal disease and underlying conditions that contraindicate certain specific antibiotics. Currently, the most commonly used antibiotics in the treatment of brucellosis are tetracycline, rifampicin, aminoglycosides, trimethoprim-sulphamethoxazole, and quinolones. Combined drug therapy with a prolonged course is recommended ⁽¹⁴⁾. Only rapid and effective management may prevent irreversible neurological and bony complications ^(3,14).

Second in frequency to osteoarticular are genitourinary complications, with an incidence of between 6 % and 17.5 % ⁽⁶⁾. The marked affinity of different *Brucella* spp in reproductive systems of cows, goats, and sheep has been known for a long time. The tropism for the genital organs of ruminants may be related to

the high concentrations in these locations of erythritol, a carbohydrate that allows *Brucella* to grow. Although in humans the concentration of erythritol in seminal fluid and prostatic secretions is far lower than that found in ruminants, there are high concentrations of other carbohydrates, which might explain the fact that genitourinary complications are also frequent in humans ⁽⁶⁾.

According to Savaş et al. osteoarticular involvement, and orchitis were observed in 64.9 %, 38.0 % (of male patients), of the patients, respectively ⁽¹²⁾, and according to Colmenero et al. ⁽⁶⁾ osteoarticular complications were the most frequent focal forms, being present in 21.3 % of all patients and 66 % of the focal forms; and 3.4 % (5.1 % when only the male population was considered) had genitourinary complications. Orchitis occurs in up to 20 % of males with brucellosis and is an important focal form of human brucellosis which may cause serious complications ⁽¹²⁾. The genitourinary complications of brucellosis usually respond favorably to treatment, but testicular atrophy and abscesses requiring surgical treatment have been described ⁽⁹⁾.

The indiscriminate use of antibiotics in patients with fever of unknown nature frequently masks *Brucella* infection, because this bacterium is sensitive to a wide range of antimicrobial agents that, however, do not

eradicate the infection. The infection usually manifests again after several weeks, frequently as a focal complication ⁽⁶⁾ (as seen in our case).

Other than blood cultures and SAT, hematologic testing, such as white blood cell count, erythrocyte sedimentation rate, and biochemical testing, such as aspartate aminotransferase, alanine aminotransferase, lactate dehydrogenase, CRP, blood urea nitrogen, and creatinine, have been of little value ⁽¹⁶⁾.

In conclusion, brucellosis will continue to be a public health problem in countries where consumption of unpasteurized dairy products and stockbreeding are widespread ⁽¹²⁾. However, when therapeutic failure, relapses, and mortality were considered together, the risk of an unfavorable evolution was significantly greater in patients with focal forms. Given the worse prognosis, knowledge and early diagnosis of the focal forms of *B. melitensis* infection is especially important ⁽⁶⁾. Prevention of human brucellosis depends on the elimination of the disease from domestic livestock by vaccinations of the susceptible animals, skin test for sheep, serologic test on milk, blood samples for cattle, and pasteurization of milk. Moreover, educating the people working with these susceptible animals is important in the prevention of brucellosis ⁽¹²⁾.

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