

**CASE REPORT**

## **An unusual cause of orchiectomy: Brucella epididymo-orchitis that does not respond to the treatment**

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### **ABSTRACT**

Brucellosis is an endemic disease in Turkey, and epididymo-orchitis is a rare complication of brucellosis. A 21-year-old married male presented to infectious diseases clinic with 4 weeks history of swelling, increased temperature on right testicle and acute scrotal pain for 2 days. His physical examination pointed an epididymo-orchitis within his radiological findings; patient was started on standard epididymo orchitis treatment. Upon increase in the patient's complaint, control radiological evolution was carried out after the completion of antibiotic therapy and showed no resolution of lesions, so patient underwent surgery. Herein, a case of a brucellar epididymo-orchitis that does not respond to the treatment which was resulted in orchiectomy is presented. *J Microbiol Infect Dis* 2015;5(2): 89-92

**Key words:** Brucella, epididymo-orchitis, radiodiagnostic imaging, orchiectomy

### **Orşiektominin nadir bir sebebi: Tedaviye cevap vermeyen brusella epididimiorşiti**

#### **ÖZET**

Bruseloz Türkiye'de endemiktir ve brusella epididimiorşiti nadir görülen fakat kolaylıkla atlanılan bir komplikasyonudur. 21 yaşında evli erkek hasta son 4 haftadır devam eden sağ skrotal kızarıklık, 2 gündür artan akut skrotal ağrı şikayeti ile enfeksiyon hastalıkları kliniğine başvurdu. Hastanın fizik muayene ve radyolojik bulguları epididimiorşiti işaret etmesi üzerine standart tedavi başlandı. Hastanın şikayetlerini artması üzerine yapılan kontrol radyolojik değerlendirmede lezyonda herhangi bir gerileme saptanmaması üzerine hasta cerrahiye devredildi. Burada tedaviye cevap vermeyen ve orşiektomi ile sonuçlanan epididimiorşit vakası takdim edilmektedir.

**Anahtar kelimeler:** Brucella, epididimiorşit, radyolojik görüntüleme, orşiektomi

#### **INTRODUCTION**

Brucellosis is a systemic disease that affects many tissues and organs.<sup>1</sup> Genitourinary system involvement is the second most common complication after musculoskeletal system involvement, epididymo-orchitis is the most frequent type of genitourinary complication.<sup>2-5</sup> One of the treatment options is doxycycline + rifampicin combination for six weeks and patient's recovery ends uneventful in most cases.<sup>6</sup> Herein, we aim to present a rare involvement of brucellosis that deals with unexpected outcome which is scarce in literature and to point the clinicians that brucellosis can end with orchiectomy. In this connection, we suggest similar case reports to be published to focus this issue.

#### **CASE**

A 21-year-old married male presented to infectious diseases clinic with 4 weeks history of swelling, increased temperature on right testicle and acute scrotal pain for 2 days that developed abruptly. In his physical examination, he had a swollen right testis and was found enlarged about twice the normal size. The right testis was tender on palpation. The skin over the swelling was red with local rise of temperature. There was no significant finding except palpation of liver enlarged by 2 cm compatible with hepatomegaly in abdominal exploration. His temperature was 38.6°C with a pulse of 88/min heart rate and he had a history of undulant fever. In deepening his history, it was learned that he was a

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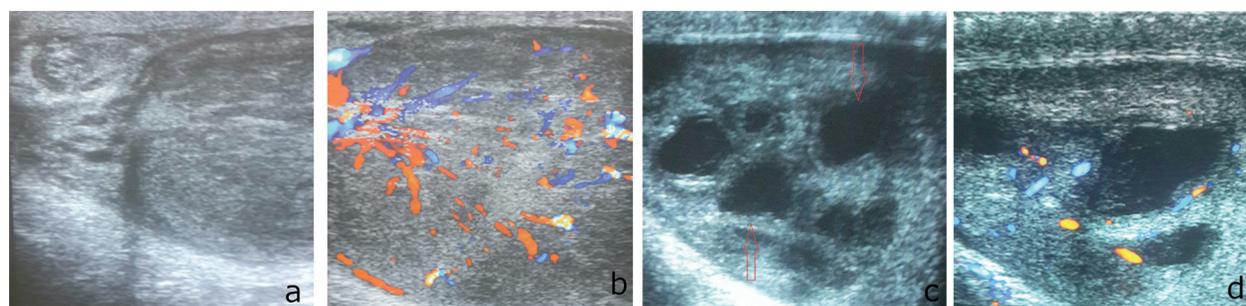
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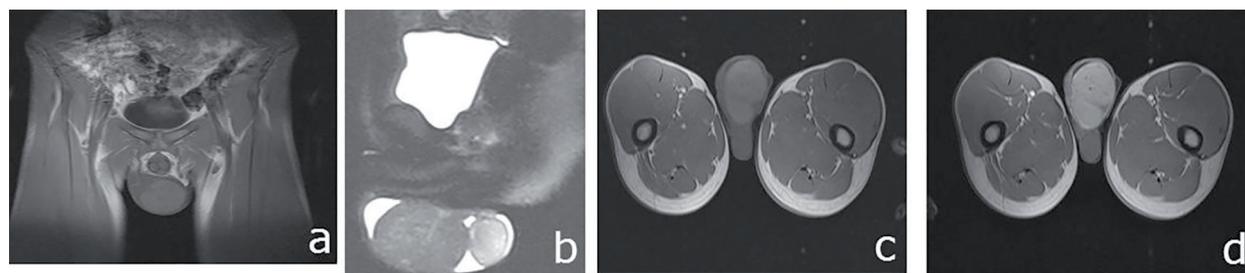
farmer and had habitual history of consuming unpasteurized milk and milk products. Bloods results on admission revealed a hemoglobin level of 12.5 mg/dL,  $5.2 \times 10^9/L$  leukocyte count with lymphomonocytic cell dominated, C-Reactive Protein levels were also significantly higher (70 mg/l) and a rate of 67 mm/h erythrocyte sedimentation. Alanine amino transferase (ALT) levels were accelerated with a level of 110 IU/L and there was no significant finding in other serum chemistry and urine analysis. His initial laboratory results were negative for Widal test, Mantoux test, Venereal Disease Research Laboratory test (VDRL); his urine and blood cultures were negative also. For differential diagnosis, tumor markers of testicular carcinoma were found to be within the normal limits (AFP: 1.26 U/L, hCG: <1 U/L) and there was no evidence of HIV serologically. Rose Bengal test was positive and Coombs test was significantly positive at a value of 1/5120.

The brucella culture couldn't perform on testicular aspirate as patient refused. Ultrasonographic evaluation revealed heterogeneous echogenicity within the right epididymis extending to right testicle based on swelling in the tail of the epididymis and there was increased doppler blood flow also (figure 1). Despite his fever and local symptoms decreased in the following 72 hours; scrotal swelling appeared to be larger on inspection. A pelvic-scrotal MRI was performed to rule out any pelvic pathology and testicular abscess. MRI scan demonstrated overall decreased signal intensity on T1W images and T2W images on the right epididymis and testicle compared with the normal side; and a slight scrotal ef-

fusion also seen. There was a diffuse enlargement of the affected side with mild contrast enhancement (figure 2). There was no evidence of possible testicular abscess or masses radiologically. Control ultrasonography (US) was carried out after the completion of antibiotic therapy. There were multiple large hypoechoic lesions filling the affected side with non-doppler activity signing the necrotic areas (figure 1) and slide increase doppler blood flow in the rest of the testicular parenchyma due to inflammation; indicating response to the therapy. With these clinical findings the patient was started on standard epididymo orchitis treatment with combination of doxycycline (200 mg/day) plus rifampicin (600mg/day), non-steroid anti-inflammatory medication, complete bed rest with scrotal elevation and local hypothermia application. Within the presence of urologic consultation, as the efficiency of each combination seems to be similar in literature, treatment was performed for 42 days without modification of antimicrobial combination. Upon increase in the patient's complaint, control radiological evolution was carried out after the completion of antibiotic therapy and showed no resolution of lesions, so patient underwent surgery. This case was unresponsive to the standard therapy and resulted in orchietomy. After surgery, patient's recovery was uneventful. As the most important complication of the patient was infertility, semen analysis was performed after the operation and reported as a sufficient number and motility of sperm content for fertilization; gratifying the patients fertile status.



**Figure 1.** First evaluation US images. a) US reveal the heterogeneous echogenicity in epididymis and testicle. b) There is significantly increased doppler activity due to acute infection. Follow-up US images. c) Multiple large hypoechoic lesions signing the necrosis (red arrows)filling the affected side a (after treatment). d) The necrotic parenchyma shows no signal activity. There is slide increase Doppler blood flow in the rest of the testicular parenchyma.



**Figure 2.** On coronal MRI images the testicular parenchyma shows hypointensity on T1W images (a) and, the scrotal effusion can also see on T2W images with hypointense testicular parenchyma (b). T1W axial non-contrast (c) and post contrast (d) MRI images of the lesion. There is a mild contrast enhancement.

## DISCUSSION

Brucellosis is an endemic zoonotic disease that can involve many organs and tissues. The incidence of brucellosis in developed countries is low, but it occurs sporadically in occupationally exposed groups, including farmers, veterinarians, and laboratory and slaughterhouse workers.<sup>3,4</sup> Brucellar epididymo-orchitis (BEO) which is a focal complication of human brucellosis, and by the men one-sided granulomatous EO are the most common complication. The main treatment option for BEO is besides medical treatment.<sup>4</sup> The most effective antibiotic combination for therapy is tetracycline (particularly doxycycline) and aminoglycoside. But, we used doxycycline+rifampicin combination. If it is unable to respond to this treatment, then surgical or orchidectomy as draining of testicular abscess treatments are activated. Moreover, the above mentioned characteristics are not always present in patients with BEO and therefore differential diagnosis between BEO and non specific EO is often difficult.<sup>7</sup> The diagnosis of brucellosis requires microbiological confirmation by isolation from blood culture or the detection of specific antibodies by serological tests. Although the isolation of *Brucella* spp. from blood, bone marrow, or other tissue cultures is considered the gold standard<sup>7</sup> the reported bacterial isolation rates in the literature range from 15% to 90% depending on the methodology used. Previous antibiotic use significantly decreases the likelihood of bacterial isolation in blood cultures in chronic cases.<sup>7,8</sup> Accordance with the literature of our case had similar results; urine and blood cultures were negative also. As the patient had used antibiotics effective on brucella infection before he was admitted to our hospital, *Brucella* had not been isolated from blood cultures.

In the diagnosis of BEO, brucellar agglutination tests and scrotal US are crucial diagnostic meth-

ods for the differential diagnosis of EO in endemic regions of brucellosis.<sup>7</sup> US is the best imaging technique for diagnosis of epididymitis and/or EO. Enlargement of affected side, swelling in the tail of the epididymis, heterogeneous echogenicity within the parenchyma of testis-epididymis and increased doppler blood flow are the main findings in acute period. If lesions don't resolve in 6 weeks, circular hypoechoic areas without any doppler activity signs necrotic areas due to infarct as a complication.<sup>9</sup> In selected patients magnetic resonance imaging (MRI) is an alternative imaging tool. Diffuse or focal enlargement of parenchyma with decreased signal intensity and diffuse contrast enhancement on T1W images are typical for acute EO. On T2W images the signal intensity varies hypointense to hyperintense approaching the signal of normal testis varying according to acute or chronic stage.<sup>10</sup> Antimicrobial therapy is generally enough in brucellosis with uncomplicated EO. There are several articles about the treatment efficiency of the combination therapies in the literature and the efficiency of each combination seems to be similar. The treatment period should be at least 6 weeks and the duration of antibiotic therapy for BEO varies considerably among studies.<sup>6,11-15</sup> Human brucellosis continues to pose a therapeutic problem because of the intracellular localization of the brucella within the host's reticulo endothelial cells, a site relatively inaccessible to antibiotics. Inappropriate choice, dosage and length of antimicrobial therapy, failure of patients to take prescribed drugs and, very rarely, antibiotic-resistant brucella strains are associated with relapses and therapeutic failure. The percentage of therapeutic failure or relapse ranges from 0% to 40%.<sup>6,13-15</sup> BEO usually have a favorable outcome with standard treatment of brucellosis, except for some rare cases that present necrotizing orchitis and require surgical treatment.<sup>5,15</sup> In patients with focal hypoechoic le-

sions in the testis seen on US, orchiectomy is usually performed.

In conclusion, testicular involvement of brucellosis is a rare but can lead to orchiectomy. Therefore, in the presence of EO and fever, brucellosis should be considered particularly in the endemic areas.

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