

## Case Report

# Brucellar terminal ileitis and epididymo-orchitis in an adolescent; case report and review of the literature

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### Abstract

Although brucellosis is a multi-systemic illness, terminal ileitis or colitis due to brucellosis is reported anecdotal in literature. Genitourinary manifestations of Brucella, namely epididymo-orchitis is very rare in childhood brucellosis. Herein, we present a case of brucellosis in a child with a rare combination of terminal ileitis and epididymo-orchitis not reported previously in the literature.

**Key words:** acute abdomen; Brucella; children; brucellar gastrointestinal manifestations; brucellar genitourinary manifestations.

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### Introduction

Brucellosis is an important endemic zoonotic disease, which may cause a wide spectrum of clinical symptoms in humans, from a mild flu-like illness to systemic manifestations [1]. Systemic symptoms such as arthralgia, myalgia and weight loss are seen frequently in brucellosis. Gastrointestinal manifestations are diverse and often characterized by loss of appetite, vomiting and abdominal pain. Colitis and ileitis have been reported as rare sporadic cases in the literature [2]. Brucella epididymo-orchitis may be seen with a rate of 2-20% in human brucellosis. In addition, brucellar epididymo-orchitis may lead to serious complications such as necrotizing orchitis [3]. We report herein a rare case of a 17-year-old male patient who presented with acute abdomen clinic and diagnosed with brucellar terminal ileitis and subsequently with brucellar epididymo-orchitis.

### Case Presentation

A 17-year-old male was admitted to the emergency department with complaints of abdominal pain and fever for 3 days. There was no one in his family who had similar symptoms. The family was dealing with livestock. The patient had a history of consuming cheese from unpasteurized milk. The patient had no symptoms of nausea, vomiting and diarrhea. He had no gaita excretion for 3 days. In the physical examination; he appeared toxic and his body temperature was found

as 38,5 C° pulse as 90/minutes and blood pressure as 110/75. In addition, there was strong tenderness and rebound on the lower right quadrant in abdomen examination. Other system examinations were normal. Laboratory test results for hemoglobin was 12.6 g /dL, for white blood cell count 8670/mm<sup>3</sup>, for platelet count 141000/mm<sup>3</sup>, for the sedimentation rate 28 mm/hr. and for CRP 50 mg/L (0-4). Since he was appeared toxic and had strong tenderness and rebound on the lower right quadrant in abdomen examination, an abdominal ultrasonography was performed in order to investigate the cause of the abdominal pain. On the abdominal ultrasonography, terminal ileum was monitored as edematous. Multiple enlarged mesenteric lymph nodes and free peritoneal fluid were detected by abdominal ultrasonography. The spleen was enlarged with a length of 146 mm. Our preliminary diagnoses were infections, Familial Mediterranean Fever (FMF), inflammatory bowel diseases (IBD) and malignancy. To investigate infectious etiologies two blood cultures were obtained. The serology for Epstein-Barr virus, mumps, parvovirus, cytomegalovirus was obtained and found negative. To exclude enteric infections, stool microscopy was obtained and found negative for parasites, erythrocytes and leucocytes. Because he had the history of consuming cheese from unpasteurized milk, Brucella agglutination test and Brucella serology were performed. He had no history of recurrent fever and abdomen pain and there were no one in his family

with FMF. To exclude hematologic malignancies peripheral blood smear was examined and no atypical leucocytes were seen. Colonoscopy was planned to exclude IBD. On the second day of his hospitalization; pain, swelling and redness was observed in his right testicle. Scrotal Doppler ultrasonography showed that, the dimensions of the right epididymis were increased and had a heterogeneous appearance. Appendix-epididymal torsion was seen at the head of the right epididymis with nodular hypoechoic area. Complete urinalysis revealed no significant feature and there was no growth in the urine culture. The patient was evaluated as epididymo-orchitis. Scrotal elevation and nonsteroidal anti-inflammatory drug therapy were initiated. On the third day of his hospitalization Brucella agglutination test resulted as positive in 1/640 titer. Blood cultures were negative. Brucella enzyme-linked immunosorbent assay (ELISA) immunoglobulins (Ig) M and G in serum were positive and 16.3 and 30.1 respectively. The patient was started on oral doxycycline 100 mg, rifampicin 300 mg twice daily for 6 weeks. His complaints were significantly regressed on the 3rd day of treatment, while symptoms of scrotal swelling and abdominal pain disappeared on the 10th day. Priory planned colonoscopy was not performed because of the positive Brucella serology and improved clinic of the patient with the treatment.

## Discussion

Brucellosis is a multi-systemic infectious disease, which can involve many organs and tissues. Middle East countries including Syria, Turkey, Iran, Iraq, and Saudi Arabia have highest incidence rate for human brucellosis [4]. Its incidence has been reported as 1-26.7 % in Turkey [5-8]. The disease is transmitted from animals to humans mainly by consumption of unpasteurized milk and dairy products, consumption of undercooked meat, or skin penetration of those in contact with livestock. Other rare transmission forms are inhalation of contaminated aerosols, conjunctival inoculation, blood transfusions and transplacentally from mother to fetus [9]. The family of our patient was also dealing with livestock and abortion was reported in one of their cattle.

The patient in this case had positive Brucella agglutination test and positive IgM and IgG serology for Brucella. The brucellosis is diagnosed with potential exposure, clinical features, and serological tests with and without positive culture. The positive culture is the gold standard of the diagnosis. When no positive culture was obtained, the serological tests are the main tools of the diagnosis of brucellosis. [10] Our patient

had a history of consumption of unpasteurized cheese and atypical clinical features which could be explained by Brucellosis. In spite of his negative blood cultures, Brucellosis was diagnosed due to clinical suspicion, a positive history and serology in this case.

Gastrointestinal findings of Brucella infection are relatively rare. The most common gastrointestinal findings include nausea, vomiting, diarrhea and constipation. Acute abdominal pain and terminal ileitis clinic have rarely been reported in the literature [3,11]. A case of acute Brucella ileitis was described for the first time by Petrella and Young in 1988 [12]. Our patient is important for being the first reported case of terminal ileitis in childhood age group. Brucella can present with gastroenteritis and/or constipation. The patient also complained of constipation with abdominal pain at admission. In this case, terminal ileitis was developed due to Brucella infection and the patient presented with an acute abdomen clinic. Acute abdomen clinic secondary to cholecystitis, pancreatitis, peritonitis and mesenteric lymphadenitis has been reported in patients with brucellosis in the literature [13,14]

Brucella-related genitourinary complications are more common than gastrointestinal findings and reported as 2-20% in various studied in the literature [15]. In a retrospective 7-year study by Yurdakul *et al.* from our country, 17% of patients with Brucella had epididymo-orchitis [16]. Epididymo-orchitis which is known as inflammation of epididymis and testis, may develop secondary to sexual activity, heavy physical exercise or direct trauma (cycling) in postpubertal men. Whereas bacterial epididymo-orchitis is more common among prepubertal boys and is associated with structural urinary anomalies. Chlamydia, *N. gonorrhoea*, *E. coli* and viruses are common in sexually active adolescents, while mycoplasma, enterovirus and adenovirus are the most common causative agents in those without sexual activity. Brucella should be definitely kept in mind as a causative factor of epididymo-orchitis in countries where Brucella is endemic, however. Epididymo-orchitis is diagnosed clinically and by Doppler ultrasonography. Urine analysis and culture are recommended to be performed in all patients with epididymo-orchitis, although studies have shown that it is found to be positive in 7 to 28% of patients. Urethral exudate or intraurethral swab culture and gram staining are recommended, as well as investigation for syphilis and HIV with the first-pass urine especially in sexually active individuals [17].

It is recommended to initiate empiric antibiotic therapy in patients with the diagnosis of epididymo-

orchitis and to revise the diagnosis and treatment if there is no improvement after 3 days [18]. In our patient, hematuria and pyuria were not observed in urine examination and there was no growth in the culture. And there was not any improvement in his clinic while he was getting non-steroidal anti-inflammatory treatment. Brucella epididymo-orchitis is distinguished from other acute nonspecific type orchitis by a gradual onset, long duration, and animal contact or a history of cheese consumption from unpasteurized milk [15]. However, in our patient scrotal pain showed an acute and rapidly progressing clinical course. Cases of Brucella epididymo-orchitis progressing to orchiectomy due to delayed diagnosis have been reported in the literature. For this reason, early diagnosis is essential [19].

## Conclusion

Brucellar terminal ileitis and epididymo-orchitis are unusual manifestations of brucellosis, especially in children. The history of consumption of unpasteurized milk and dairy products and contact with livestock should be examined carefully in patients with acute abdomen and scrotal disease in Brucella endemic countries. Recognition of unusual manifestations of Brucella is important in order to diagnose and treat brucellosis early. This will lead to complete recovery without complications of this illness. We presented this successfully treated case, because the combination of these findings had not been previously reported in the literature.

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