CASE REPORT

A cluster of atypical brucellosis in the same family

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ABSTRACT

Brucellosis is a systemic and chronic zoonotic disease passed by *Brucella* bacteria, which is endemic in our country. Patients presenting with different clinical manifestations are often accompanied by nonspecific symptoms like fever, night sweats, loss of appetite, weight loss and polyarthralgia. Although Brucellosis can affect all systems. Examples of musculoskeletal and hematological involvement will be discussed in the following two cases. Peripheral arthritis, sacroiliitis, spondylitis, tenosynovitis and osteomyelitis are seen in musculoskeletal involvement, while leukopenia, anemia, and rarely pancytopenia are seen in hematological involvement. Psoas abscess is a rare infection that is difficult and late to diagnose and categorized as primary and secondary. The most common cause of primary psoas abscess is *Staphylococcus aureus* and it usually occurs by hematogenous spread from a focus of infection in another part of the body. We aimed to present two cases from the same family, one of whom had primary psoas abscess and spondylodiscitis secondary to brucellosis; the other of whom had pancytopenia due to brucellosis.

Keywords: psoas abscess, Brucella, pancytopenia, spondylodiscitis

ÖZET

Aynı aileden eş zamanlı yatırılan iki atipik bruselloz olgusu

Bruselloz, *Brucella* cinsi bakterilerin yol açtığı, ülkemizde endemik seyreden, sistemik ve kronik seyirli zoonotik bir hastalıktır. Farklı klinik tablolarla başvuran hastalara sıklıkla ateş, iştahsızlık, gece terlemeleri, halsizlik, kilo kaybı ve poliartralji gibi nonspesifik semptomlar eşlik eder. Bruselloz tüm sistemleri tutabilmekle beraber aşağıdaki iki olguda kas iskelet sistemi ve hematolojik tutulum örneklerinden bahsedilecektir. Kas iskelet sistemi tutulumunda periferik artrit, sakroileit, spondilit, tenosinovit ve osteomyelit görülürken, hematolojik tutulumda sıklıkla lökopeni, anemi, nadiren de pansitopeni görülür. Psoas apsesi nadir görülen; tanısı genellikle zor ve geç konulan, primer ve sekonder olarak sınıflandırılan bir enfeksiyon hastalığıdır. Primer psoas apsesinin en sık sebebi *Staphylococcus aureus* olup sıklıkla vücudun başka bir bölgesindeki enfeksiyon odağından hematojen yayılım sonucu oluşur. Bu çalışmada biri bruselloza sekonder gelişen primer psoas apsesi ve spondilodiskiti olan; diğeri pansitopenisi olan aynı aileden iki olgunun sunulması amaçlanmıştır.

Anahtar kelimeler: psoas absesi, Brusella, pansitopeni, spondilodiskit

INTRODUCTION

Brucellosis is a zoonotic infection represents reticuloendothelial granulomatous reaction caused by gram-negative coccobacillus called Brucella spp. [1]. The main risk factors are consumption of the infected animal's raw milk and dairy products and dealing with animal husbandry. Since the disease involves many organs and systems, it can present with different clinical pictures [2]. Symptoms of vomiting, abdominal pain, diarrhea and constipation may occur due to gastrointestinal involvement [3]. It most commonly affects the musculoskeletal system, causing sacroiliitis and spondylitis [4].

Psoas abscess is a rare infection that is often difficult and late to diagnose, categorized as primary and secondary. The most common cause of primary psoas abscess is *Staphylococcus aureus* and it usually occurs by hematogenous spread from a focus of infection in another part of the body. Patients usually present with complaints of fever, fatigue, sweating, weight loss, abdominal pain, limping, lower back and leg pain [5]. Although hematological changes such as bone m0arrow and spleen involvement are frequently encountered in brucellosis, pancytopenia is an unusual complication of brucellosis [6]. Hypersplenism, disseminated intravascular coagulation, hemophagocytosis, bone marrow suppression and destruction of platelets are thought to have a role in the pathogenesis [7]. The pancytopenia

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in brucellosis ends with the treatment of the underlying brucellosis [8].

CASE REPORT

Case 1

A 72-year-old, immunocompetent female patient, who had no comorbidities except known hypertension and cardiac arrhythmia, applied the emergency department with complaints of the low back, hip and knee pain that had continued for about a month, and with complaints of loss of appetite, nausea, vomiting, chills, and sweating for the past few days.

She lives in the countryside of Çanakkale, was engaged in animal husbandry, attended to animal births but not consumed of fresh milk and cheese.

On physical examination, body temperature was 36.5ºC, pulse was 76 beats/min and blood pressure was 130/80 mmHg. The other PE findings were within normal range. The patient, who had no signs of peripheral arthritis, had sensitivity in the corse of percussion on sacral and lumbar regions. Lumbar vertebral column movements were painful and limited in all directions, accompanied by bilateral paravertebral muscle spasm but there was no pedro pons sign observed in lumbosacral and sacroiliac joint radiographs, no sacroiliitis was detected.

Laboratory findings admission were as follows: white blood cell (WBC): 8800/uL, C-Reactive Protein (CRP): 14 mg/dL, erythrocyte sedimentation rate (ESH): 65 mm/s, Alanine aminotransferase (ALT): 31 U/L, Aspartate aminotransferase (AST):44 U/L, Alkaline phosphatase (ALP): 668 U/L, Gamma glutamyl transferase (GGT): 526 U/L, direct bilirubin: 1.1 mg/dL, total bilirubin: 1.3 mg/dL. The patient's Rose Bengal Lam Agglutination test performed before admission was positive. The abdomen computed tomography (CT) showed no abnormal findings.

The patient was consulted for gastroenterology because of elevated ALP and GGT. With the preliminary diagnosis of brucella cholangitis, elective magnetic resonance cholangiopancreatography (MRCP) planned but couldn't performed. Ursodeoxycholic acid treatment was given to the patient.

Oral treatments of doxycycline 2x100 mg, rifampicin 1x600 mg and intramuscular streptomycin 1x1 g treatment were started. Contrast-enhanced lumbar spine magnetic resonance imaging (MRI) revealed spondylodiscitis at the L2-L3 level and a 2.5 cm abscess in the right psoas muscle. Abscess drainage was performed by interventional radiology under the guidance of CT, and 2 abscess aspirate cultures, aerobic and anaerobic, were taken. In both cultures, Brucella spp. reproduced. Complaints of the patient regressed after drainage of the abscess. She was discharged on the 9th day of the treatment after oral intake improved.

The patient was diagnosed as acute complicated brucellosis clinically and serologically and started on a 3-month treatment consists of oral rifampicin and doxycycline and two weeks of streptomycin.

On the first outpatient clinic examinations of 23rd day of the treatment after discharge, ALT: 16 U/L, AST: 15 U/L, ALP: 124 U/L, GGT: 34 U/L, direct bilirubin: 0.3 mg/dL, total bilirubin: 1.8 mg/dL, CRP: 1.8 mg/dL and it was seen to regress.

Case 2

A 67-year-old female patient with a known diagnosis of hypertension who is the sister of case 1 applied to our outpatient clinic simultaneously with her sister with complaints of joint pain, nausea, vomiting, chills, tremor, oral food intake disorder and weight loss for 1.5 months. It was learned that; she lost about 10 kg in the last 2 months and had night sweats at nights and also intramuscular (IM) vitamin B12 treatment was started for vitamin B12 deficiency anemia due to pancytopenia in the examinations performed in an external center 1 month ago, and despite 10 times of IM injections, there was no regression in her complaints.

Her physical system examination was normal. Laboratory findings were as follows: hemoglobin (Hgb): 8.4 g/dL, platelet (Plt) count: 116000/uL, WBC: 1500/uL, (neutrophil: 580/uL, lymphocyte: 840/uL), CRP: 3.2 mg /dL, creatinine: 0.80 mg/dL, AST: 56 U/L, ALT: 14 U/L. Rose Bengal Lam Agglutination test was positive and Brucella Coombs agglutination test was determined as 1/1280 and above.

The patient was admitted to our ward because of systemic symptoms and pancytopenia. It was observed that there was an iron deficiency in the anemia parameters, and vitamin B12 levels were above the normal limit after vitamin B12 replacement. Patient's pancytopenia in the blood table is associated with brucellosis. The patient was diagnosed as acute non-complicated brucellosis clinically and serologically and started on streptomycin 1x1 gr IM, doxycycline 2x100 mg oral and rifampicin 1x600 mg oral treatment.

Nausea was terminated on the 6th day of follow-up of total parenteral nutrition (TPN) treatment, which was started due to oral intake disorder. On the 9th day of the follow-up, the patient's appetite and oral intake improved, and her complaints completely regressed and discharged from service.

It was planned to complete the existing streptomycin treatment in 10 days, and the doxycycline and rifampicin treatments in 6 weeks.

First control examination of the patient's on 20th day of outpatient treatment, laboratory findings were found that WBC: 6000/uL, neutrophil: 3200/uL, lymphocyte: 2400/uL, Plt: 276000/uL, Hgb: 10.5 g/dL. It was observed that pancytopenia was resolved with the treatment.

DISCUSSION

Human brucellosis is caused by Brucella bacteria and widespread zoonotic diseases worldwide with more than half a million new cases diagnosed each year. It has been eradicated in many countries around the world; however, it is still responsible for significant morbidity and mortality in Turkiye, the Middle East,

Sub-Saharan Africa, Central and South America and India. It is transmitted from animals to people through touch with infected animals and consumption of infected milk-milk products and it can affect almost all systems such as the musculoskeletal, gastrointestinal, genitourinary and central nervous system and may present with different clinical pictures [9,10].

Although fatigue, joint pain, and fever were frequently observed in the patients at admission, the first case we followed up had low back and hip pain that limited walking, although there was no fever. In the etiology of psoas abscess which is seen as a rare complication, Brucella spp. may be a rare cause [5,11]. While psoas abscess is mostly seen in children and young people, but the patient in our case is 72 years old [5]. Most psoas abscesses usually develop secondary to the spread of infection from adjacent tissues. When it rarely develops by hematogenous route, the most common cause is Staphylococcus aureus [12]. Antibiotherapy and abscess drainage are administered together in the treatment of psoas abscess. Because of it is minimally invasive, percutaneous abscess drainage with USG or CT, and accompanying antibiotic therapy is the most preferred treatment method in psoas abscesses [13]. In our first case, 2.5 cm abscess was seen in the right psoas on MRI and it was drained by interventional radiology. Brucella spp. was detected in the culture of the abscess. The patient's complaints regressed with the given antibiotic therapy.

Osteoarticular complications are most common in brucellosis. In addition, gastrointestinal involvement may occur together with hematological disorders. Symptoms such as loss of appetite, abdominal pain, dyspepsia, vomiting, diarrhea, constipation, hepatomegaly, splenomegaly, and tenderness on the abdominal examination have been reported in 70% of patients [3]. Both of our cases had complaints of nausea, vomiting and loss of appetite, which caused a serious oral intake impairment. Abdominal examinations were normal. Rose Bengal Lam agglutination test was positive (+) in both of our cases, and Brucella Coombs agglutination test was positive (+) at 1/1280 and over 1/1280 titers, respectively. In the USG of our first case, no pathology was found except splenomegaly, and MRCP was planned for the preliminary diagnosis of cholangitis because of the high ALP and GGT levels. In both of our cases, it was observed that oral intake improved with treatment; and ALP and GGT levels which were initially high in our first case regressed.

In brucellosis, the blood tests change in many patients as the bone marrow and spleen are affected; however, pancytopenia is a rare manifestation of brucellosis [6]. Hemophagocytosis syndrome, hypersplenism, destructions in platelets and disseminated intravascular coagulation are responsible for the pathogenesis here [14]. Pancytopenia seen in brucellosis resolves spontaneously with the treatment of brucellosis without the need for additional treatment [7,8].

In our second case, there was a deepening pancytopenia (Hgb: 8.4 g/dL, Plt: 116000/uL, white blood cell: 1500/uL, neutrophil: 580/uL, lymphocyte: 840/uL) and improved with antibiotherapy. It was reported that pancytopenia seen during the course of brucellosis regresses with treatment [15]. In our patient, control blood tests were within normal limits, except for hemoglobin (white blood cell: 6000/uL, neutrophil: 3200/uL, lymphocyte: 2400/uL, Plt: 276000/uL, Hgb:10.5 g/dL). In addition, it was reported that pancytopenia develops in 8% of cases with brucellosis and granulomatous lesions are seen in bone marrow examination in 67% of them [15]. For our second patient who presented with pancytopenia, advanced hematological examination could not be performed because of the unavailability hematology department in our hospital.

In two meta-analyses, assessments about antibiotic therapy in brucellosis, the combination of streptomycin and doxycycline was reported to be superior to the combination of doxycycline and rifampicin [16,17]. In both of our cases, a combination of doxycycline, rifampicin and streptomycin was given and clinical and laboratory improvement was observed in a short time.

The diagnosis may be delayed in brucellosis, especially if there are unusual findings like pancytopenia, and the clinical presentation may mimic histiocytosis or other clinical conditions that lead to bone marrow infiltration. Therefore, brucellosis should be well-considered in various diagnosis of all these terms that cause pancytopenia. In addition, it should be taken into attention that brucellosis, which is unusual but endemic in our country, may cause psoas abscess in some cases.

In conclusion, with appropriate treatment in brucellosis, complications show a good course and recovery is seen in a short time.

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