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A Tularemia Case with Bilateral Inguinal Lymphadenopathy and Abdominal Abscess

Bilateral Inguinal Lenfadenopati ve Abdominal Apse ile Seyreden Bir Tularemi Olgusu

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SUMMARY

Tularemia is a zoonotic disease caused by Francisella tularensis. Humans can acquire the infection through bites from arthropods, contact with animal tissues, consumption of the infected water and inhalation of aerosolized bacteria. The oropharyngeal form is the most common presentation in Turkey, reflecting the consumption of contaminated water. While the most common complication of the disease is the suppuration of the lymph nodes, abscess formation can be a very rare complication. In this study, we report a glandular tularemia case with bilateral inguinal lymphadenopathy and intra-abdominal abscess. A 51-year-old woman came to the infectious diseases clinic with fever and swelling in the groin ongoing for two months. She had been started on a treatment of amoxicillin-clavulanic acid at another hospital for two weeks and had a history of watering her garden bare feet with natural pool water. She had fever and bilateral inguinal lymph nodes in her physical examination. One of her lymph nodes was extracted for diagnosis. The pathological examination result was granulomatous lymphadenitis. The sample's culture and PCR results were negative for tuberculosis and tularemia but the F. tularensis microagglutination test (MAT) was positive. She had received treatment for Tularemia but her clinical and laboratory findings had not improved. Pelvic lymphadenopathies and an intra-abdominal abscess were observed in an MRI. Percutaneous drainage was done and the patient's condition improved. This tularemia case was presented to emphasize that in regions, like our country, where tularemia is epidemic, if specific treatment is late, rare complications can be seen.

Key Words: *Tularemia; Intraabdominal abscess; Lymphadenopathy*

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ÖZET

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Tularemi, Francisella tularensis'in sebep olduğu zoonotik bir hastalıktır. İnsana eklembacaklıların ısırığı, hayvan dokularıyla temas, infekte su içimi ve damlacık oluşturmuş bakterinin solunmasıyla bulaşır. Kontamine su alımını yansıtan orofarengael formu ülkemizde en sık karşılaşılan formudur. En sık karşılaşılan komplikasyonu lenf nodlarının süpürasyonu iken apse formasyonu oldukça nadir görülebilen bir komplikasyonu olabilmektedir. Biz bilateral inguinal lenfadenopati ve intraabdominal apse ile seyreden bir glandüler tularemi olgusu bildirdik. Elli bir yaşında kadın hasta infeksiyon hastalıkları kliniğine iki aydır devam eden ateş ve kasıkta şişlik şikayetiyle başvurdu. Başka bir hastanede iki hafta amoksisilin-klavulanik asit kullanımı ve doğal su havuzundan çıplak ayakla bahçe sulama öyküsü mevcuttu. Fizik muayenesinde ateş ve bilateral inguinal lenf nodları mevcuttu. Teşhis için lenf nodlarından bir tanesi çıkarıldı. Patolojik inceleme sonucu granülomatöz lenfadenitti. Örneğin kültür ve polimeraz zincir reaksiyonu (PCR) sonucu tüberküloz ve tularemi açısından negatifti fakat F. tularensis mikroaglütinasyon testi (MAT) pozitifti. Tularemi tedavisi aldı ancak klinik ve laboratuvar bulguları iyileşmedi. Manyetik rezonans görüntülemede pelvik lenfadenopatiler ve intraabdominal apse gözlendi. Perkütanöz drenaj yapıldı ve hastanın durumu düzeldi. Bu olgu bizim gibi tularemininin epidemik olduğu bölgelerde spesifik tedavi gecikirse nadir komplikasyonların görülebileceğini vurgulamak amaçlı sunulmuştur.

Anahtar Kelimeler: *Tularemi; İntraabdominal apse; Lenfadenopati*

INTRODUCTION

Tularemia is a zoonotic disease caused by *Franciella tularensis* gram-negative aerobic coccobacilli, infecting vertebrates and invertebrates in the northern hemisphere[1]. Some mammals and rodents might act as reservoirs while some ectoparasites and blood sucking flies, mites, fleas might act as vectors[2-4] The disease is transmitted to humans by contact with the infected animal's tissue and body fluid to the mucous membrane or injured skin or arthropod (tick, fly) bite or consumption of the contaminated water and food or breathing the contaminated aerosols[2,4,5]. Depending on the way of contact with the microorganism, Tularemia has six different clinical forms: ulceroglandular, glandular, oculoglandular, oropharyngeal, typhoidal and respiratory[1,2]. While in countries like the USA, Scandinavia, and Japan the disease is endemic and the most common contact is direct contact with animal -mostly arthropod- and the most common clinical form is the ulceroglandular form, in counties like Russia, the Balkans and Turkey, the disease is epidemic and the most common contact is consuming contaminated water and the most common clinical form is the oropharyngeal form [2,4,6]. The most common complication is the suppuration of the infected lymph nodes even after treatment. Meningitis, pericarditis, peritonitis, hepatitis, hepatic abscess, and brain abscess are rare complications[1,2,5].

In this report, we presented a Tularemia case with bilateral inguinal lymph nodes and intra-abdominal abscess originating from contact with the contaminated water in order to attract attention to the rare complication of the disease after delayed diagnosis and treatment.

CASE REPORT

A 51-year-old female patient living in the Beypazarı region of the province Ankara sought treatment in another hospital's emergency room for fever and swelling in the groin in November, 2011. With a diagnosis of lymphadenitis, the

patient was given amoxicillin/clavulanic acid (2 x 1000 mg PO) treatment. After a two-week therapy, she sought treatment in the Infectious Diseases Clinic for continued fever, increased swelling in the groin, dizziness, headache and myalgia and was admitted to hospital. In her history, she had watered plants in her garden from a natural water pool with bare feet. There was no problem in her family members and family history. At the time of her admittance, she was conscious and oriented. She had 38.5°C fever. Her blood pressure was 110/70 mmHg. Her heart rate was 80. In her physical examination, she had bilateral painful lymph nodes in both groins, without fluctuation, sensitive with palpation. Other systemic examinations were normal. In her blood count, leucocyte was 10.4 K/µL (67.6% PNL and 26.1% lymphocyte) (10.4 µl), thrombocyte was 471 x103/µL (N: 150-300 x103/µL), Hb 11.7 g/dL and erythrocyte sedimentation rate (ESR) was 94 mm/hour. C-reactive protein (CRP) was 77.2 mg/L (N: 0-8 mg/L). Liver and kidney function tests were normal. There wasn't any problem with the PA lung X ray. Grubel Widal test, HBsAg, anti-HCV, anti-HIV tests were negative.

With an USG investigation, there was one (15 x 7 mm) heterogeneous echogenicity, lobule contoured, solid lesion in the right inguinal region and two (25 x 20, 21 x 15 mm) lymph nodes with hypo echoic structure in the left inguinal region. One of the lymph nodes was extracted and sent to the pathology laboratory. In the histopathological examination, there was a non-necrotized epithelioid granuloma with a few small caseous granulomas and in some places suppurative granuloma. With PAS EZN, Giemsa and Grocott stains, there wasn't any specific microorganism. These findings were compatible with granulomatous lymphadenitis. The samples and serum taken from the patient were sent to the Public Health Institution Reference Laboratory to be tested for tuberculosis. EZN and PCR tests were negative for tuberculosis. In view of the patient's history and physical exam and the fact that she lived in a region with tularemia, she was tested for tularemia. Microagglutination test (MAT) was 1:1280 and *F. tularensis* specific culture and PCR were negative. In order to make sure there was no cross reaction, the patient was tested for *Brucella.* Tube agglutination test was negative.

After positive MAT test, the patient was diagnosed with glandular tularemia and antibiotic treatment was started with a combination of doxycycline (200 mg/d) and ciprofloxacin (1000 mg/d). At the 5th day of treatment, the fever, headache and myalgia disappeared. After a few days without fever, the fever recurred. There was an increase in the CRP level and the lymph node size was unchanged. The patient's fever etiology was investigated again. A new MAT test titer had increased (1/2560). In abdominal USG, there were two LAPs at the right and left upper region. In response to this result, the patient had an Abdominal Magnetic Resonance Imaging (MRI) test. There was pelvic lymphadenopathy one next to the bladder at the right (32 x 22 mm) and one at the left (27 x 12 mm) (Figure 1), and there was liquid lobulated with septum (abscess 15 x 9 cm) at the right region of the pelvic bone (Figures 2,3). The abscess was drained by percutaneous drainage and moxifloxacin (400 mg/d) treatment was started. The patient's fever decreased and ESR and CRP values returned to the normal level. The investigation of the abscess material by culture, microscope and PCR was

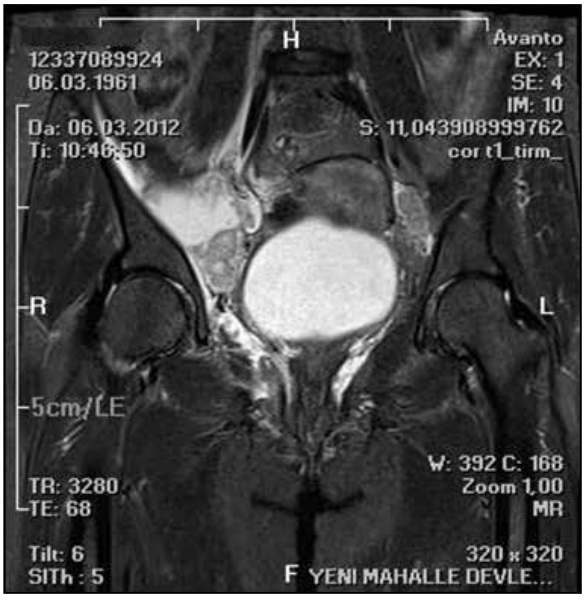

Figure 1. Coronal MR image shows bilateral lymphadenopathy located superolaterally to the bladder.

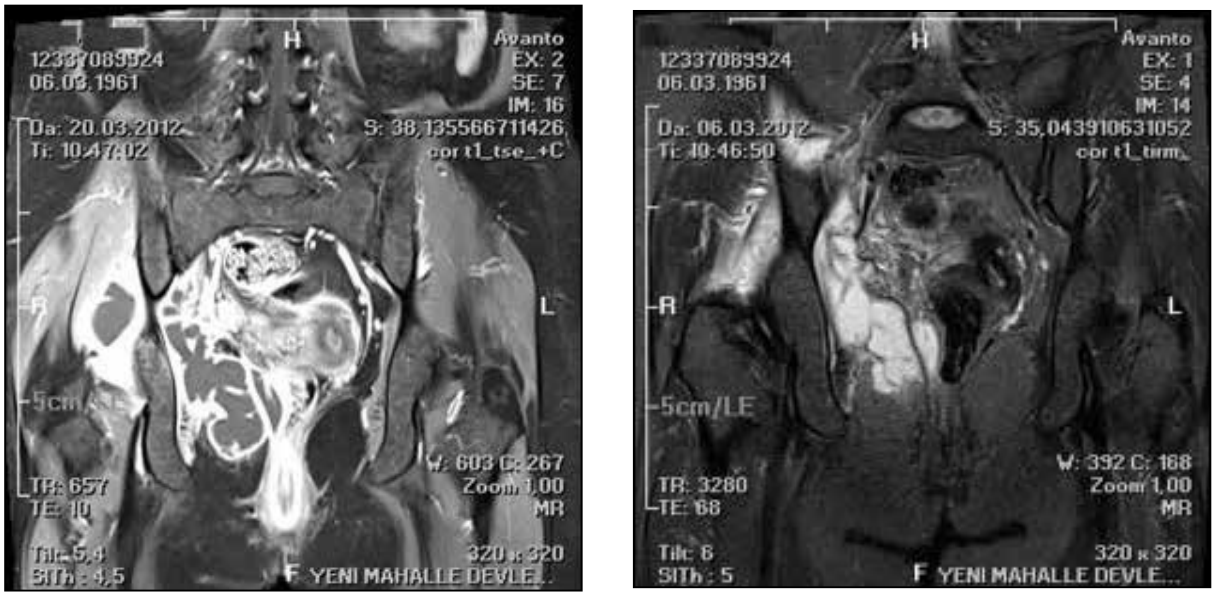

Figure (2,3). Coronal T1 with contrast medium and T2-weighted images show the cyst with septa, consistent with abscess, surrounding the right pelvic bone.

negative for tuberculosis and *F. tularensis.* The patient had clinical and laboratory improvement after abscess drainage and was discharged. One month after the drainage, the *F. tularensis* agglutination titer was 1:1280 and the leucocyte, ESR and CRP tests were normal. In her follow up, there was not any problem remaining in abdominal USG test performed two months later. *F. tularensis* agglutination test was 1:320 after one year and there was no laboratory or clinical problem.

DISCUSSION

Tularemia came into prominence after the new outbreak in Anatolia in Turkey after 2005. The oropharyngeal clinical form is the most common clinical form in Turkey due to contaminated water contact. In the world, on the other hand, the most common form is the ulceroglandular form [6,7]. The ulceroglandular clinical form occurs after an arthropod bite or direct contact with a contaminated animal or carcass. In some cases, healing of the ulceration before lymphadenitis or atypical or minimal ulceration will cause the clinical form characterized by lymphadenitis (glandular tularemia)[4-6].

In general, inguinal lymphadenopathy in adults and occipital lymphadenopathy in children have been the most common involvement regions in the tularemia cases related to tick bites[4-6]. In northern America, up to 30% of the inguinal lymphadenopathy reported has resulted from tularemia[4,5]. In Turkey, only a few tularemia cases with inguinal lymphadenopathy have been reported[8,9]. Our case had bilateral inguinal lymphadenopathy, rarely found from tularemia in Turkey. A granulomatous lymphadenitis histopathological result most commonly suggests tuberculosis in our country[9,10]. The most common clinical problems with granulomatous lymphadenitis histopathological result are tuberculosis and tularemia. These two clinical forms have some clinical and histopathological similarities which can cause clinical confusion. Cases with granulomatous lymphadenitis have increased in our country lately and can be related to either of these two diagnoses[11]. For this reason, our case was tested for both diseases. The culture and PCR tests were negative for both diseases but the MAT test was positive at the high titer. Diagnosis for this case was possible with only serological test.

In this case, during the investigation of the fever etiology, bilateral inguinal lymphadenopathy and intra-abdominal abscess were determined. Abscess complication for tularemia is very rare as with a few brain abscesses, hepatic abscess and pharyngeal abscess[4,5,12,13]. In our case,

the intra-abdominal abscess complication was not reported before. It was cured only after drainage.

Prior to specific treatment, the patient had spent two months with the symptoms. Failure in treatment and abscess complication was related to the delay in diagnosis. Under treatment, there is a suppuration possibility in lymph nodes for Tularemia. This is especially true with cases where the diagnosis is late (> 2 weeks). Despite specific treatment, a 30% lymph node suppuration was reported[1]. The abscess in our case was possibly related to a suppurated lymph node. The negative result for the *F. tularensis* culture and PCR might have been related to the sterile suppuration result of the recurrent specific treatment[14,15]. Although microorganism was not shown in the abscess material, the patient did not recover clinically until the abscess had been drained. This, in combination with the specific treatment, confirmed that *F. tularensis* was the origin of the abscess. After abscess drainage, there was a remarkable improvement in the ESR, CRP and the agglutination titer.

Glandular tularemia with bilateral inguinal lymphadenitis is a rare clinical situation in Turkey. Intra-abdominal abscess has not been reported before as a Tularemia complication. In regions where tularemia is epidemic, some rare complications might be seen. Tularemia is a possible disease causing granulomatous lymphadenitis other than tuberculosis.

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