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A Case Report of Delayed Diagnosed Oropharyngeal Tularemia Complicated with Retropharyngeal Abscess

Retrofarengeal Apse Komplikasyonu Gelişmiş Gecikmiş Tanılı Bir Orofarengeal Tularemi Olgusu

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Abstract

Tularemia is a zoonotic bacterial infection caused by *Francisella tularensis* with diverse clinical presentations. We present an oropharyngeal tularemia case complicated with retropharyngeal abscess which is a rare reported complication of tularemia. A twenty-one-year old male farmer was admitted to our hospital with a history of fever, headache, generalized aches and tender lump on the left side of neck. Physical examination revealed tender lymphadenopathy on the left posterior cervical region and retropharyngeal abscess. Surgical intervention was performed under general anesthesia. The diagnosis was made by serological tests. After antimicrobial therapy the patient recovered completely without sequelae.

Keywords: Francisella tularensis, Tularemia, Retropharyngeal abscess, Early diagnosis.

Özet

Tularemi *Francisella tularensis*'in etken olduğu, çeşitli klinik tablolar ile prezante olan bir zoonotik bakteriyel enfeksiyondur. Bu yazıda tulareminin ender olarak raporlanmış bir komplikasyonu olan retrofarengeal apseli bir orofarengeal tularemi olgusu sunulmuştur. Yirmi bir yaşında erkek hasta ateş, baş ağrısı, yaygın vücut ağrısı ve boyun sol yanında boyunda şişlik şikayetleri ile hastanemize başvurdu. Fizik muayenesinde posterior servikal bölgede ağrılı lenfadenopati ve retrofarengeal apse varlığı saptandı. Hastaya genel anestezi altında cerrahi müdahale yapıldı. Tularemi tanısı serolojik testlerle konuldu. Hasta antimikrobiyal tedavi sonrası sekelsiz olarak taburcu edildi.

Anahtar kelimeler: Francisella tularensis, Tularemi, Retrofarengeal apse, Erken tanı.

This study presented as a poster presentation at the XVI. KLİMİK Congress was held in Antalya on 13-17 March 2013. (Poster No: P13-05, Congress Book page: 291).

Bu çalışma 13-17 Mart 2013 tarihlerinde Antalya'da düzenlenen XVI. KLİMİK Kongresi'nde poster bildirisi olarak sunulmuştur (Poster No: P13-05, Kongre Kitabı sayfa: 291).

Introduction

Tularemia is a zoonotic bacterial infection caused by *Francisella tularensis* with a worldwide distribution and with diverse clinical presentations. The disease occurs predominantly in the northern hemisphere, but has rarely been found in the southern hemisphere. F. tularensis can infect a wide range of animals (more than 250 animal species). The family *Francisellaceae* includes two species in the genus Francisella and four *F. tularensis* subspecies (tularensis, holarctica, novicida ve mediasiatica). Although all of those have been associated with human disease, only the *tularensis* and *holarctica subspecies* of *F. tularensis* are relatively common. *F. tularensis* is transmitted to humans by direct contact with infectious animals, arthropod bites, aerosols, or ingestion of contaminated food or water [1,2].

Case Report

In this report, a delayed diagnosis of oropharyngeal tularemia case complicated by multiple suppurative cervical lesions and retropharyngeal abscess has been presented. Our case is distinctive for reporting retropharyngeal abscess, a rare reported complication of tularemia. A twenty-one-year old male farmer was admitted to otorhinolaryngology department of our institution in Erzurum with a history of fever, headache, generalized aches and tender lump on left side of his neck. His medical history revealed that his complaints had begun four months ago resembling an upper respiratory tract infection and he had been empirically treated with antipyretics and beta-lactam/beta-lactamase inhibitor combinations as the initial diagnosis was common cold and acute tonsillopharyngitis. He had also been hospitalized in the department of otolaryngology in a tertiary teaching hospital because of the persistence of the symptoms and the occurrence of the swelling. Physical examination revealed tender lymphadenopathy on the left posterior cervical region with a diameter of five centimeters and retropharyngeal abscess. In the laboratory investigations, leukocyte count was 8720/mm3 (65% neutrophils, 32% lymphocytes, 3% monocytes), haemoglobin level was 14.2 g/dl, platelet count was 275.000/mm3, C-reactive protein level was 18.3 mg/L and the erythrocyte sedimentation rate was 28 mm/h. Serum electrolytes with liver and kidney function tests were normal. Reactive lymphoid tissue located around the submandibular gland and bilateral cervical cysts presenting abscess formation were observed in the ultrasound scan of the neck. Computed tomography showed bilateral cervical lymphadenopathies with necrotic areas and retropharyngeal abscess (Figure 1). Open surgical drainage and excision biopsy were performed under general anesthesia. Microscopical examination of the Gram and Ehrlich-Ziehl-Neelsen stained smears revealed PMNL. Routine cultures of the material were negative for bacteria and fungi. Cytologic evaluation showed no evidence of cancer and pathologic examination was reported as subacute inflammatory process. Throat swab, lymph node aspirate and serum samples were sent to the reference laboratory for investigation for tularemia.

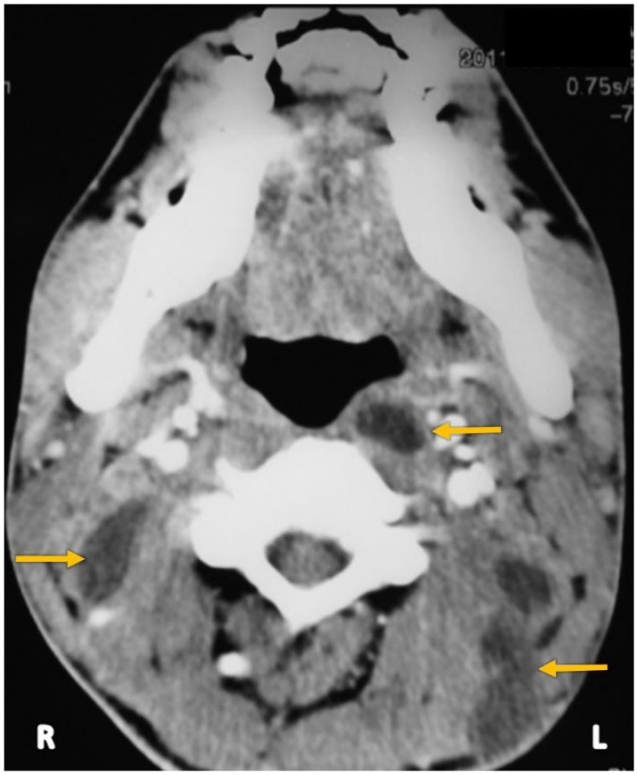

Figure 1. Head and neck computed tomography scan revealed bilateral cervical lymphadenopathies with necrotic areas and retropharyngeal abscess (white arrows). R and L signify right and left side respectively.

Due to the suspicion of tularemia, the patient was treated with doxycycline (2x100 mg/day) and ciprofloxacin (2x500 mg/day) initially. Tularemia diagnosis was made serologically. Microagglutination test yielded positive with an antibody titer of 1/2560. Antimicrobial therapy discontinued after three weeks and the patient recovered completely without sequelae.

Discussion

The clinical consequences of tularemia depend on the virulence of the particular organism, the portal of entry, the extent of systemic involvement, and the immune status of the host. Especially farmers, hunters, walkers, and laboratory personnel are most at risk of contracting tularemia. Patients who seek medical attention usually present with at least one of six classic forms of tularemia: Ulceroglandular, glandular, oculoglandular, pharyngeal, typhoidal, and pneumonic. Ulceroglandular tularemia is most common and frequently caused by vector-borne transmission in the USA and Central European regions by ticks and in Northern Europe by mosquitoes [3-5]. Although the ulceroglandular form is the most common throughout the endemic areas of the world, oropharyngeal tularemia with oral ulcers, tonsillopharyngitis and enlargement of cervical lymph nodes are the most commonly occurring syndromes in Turkey and this form is mostly acquired by drinking contaminated water or ingesting contaminated food [5,6]. Affected persons may develop stomatitis, pharyngitis or tonsillitis with pustules and ulcers. A retropharyngeal abscess or suppuration of regional lymph nodes may occur. The lymph nodes are usually enlarged and tender [5,7]. We reported a typical oropharyngeal tularemia case with an atypical complication, retropharyngeal abscess. His occupation is farmer and he stated that he had drunk well-water. Only a few cases of tularemia with retropharyngeal abcess complication have been reported in literature [7-9].

The diagnosis of tularemia ultimately rests on clinical suspicion. Due to the relatively unspecific general symptoms of tularemia and the variety of the primary disease patterns, clinical diagnosis is not easy. Early diagnosis depends on a precise medical history, physical examination and epidemiological data regarding animal contacts, occupation, and insect bites and allows medical therapy with effective antibiotics and prevents the complications. Then the diagnosis should be confirmed through molecular methods like PCR and specific serological tests [1,2,5].

The drug of first choice for the treatment of tularemia is streptomycin, whereas gentamicin is acceptable substitute. Doxycycline and fluoroquinolones have also been demonstrated to be effective. Surgical intervention is mostly necessary unless diagnosis and therapy are delayed [2,4,10]. Failure of therapy with common antibiotics such as beta-lactam/beta-lactamase inhibitor combinations was the key feature leading to final diagnosis in our case. In conclusion, clinicians should consider tularemia in the differential diagnosis of patients with painful lumps in the neck who did not recover with empirical antibiotic therapy directed against pharyngitis or tonsillitis particularly in endemic areas for early diagnosis and preventing complications.

Declaration of interest: The authors declare no conflict of interest and alone are responsible for the content and writing of the paper.

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