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# A Rare Presentation in Tularemia: Inguinally Located Glandular Tularemia

## Tularemide Nadir Bir Prezantasyon: İnguinal Yerleşimli Glandüler Tularemi

Ayşe SAĞMAK TARTAR, Ayhan AKBULUT

Firat University Faculty of Medicine, Department of Infectious Diseases and Clinical Microbiology, Elazığ, Turkey

**Keywords:** Tularemia, glandular tularemia, inguinal lymphadenopathy, ciprofloxacin, treatment**Anahtar Kelimeler:** Tularemi, glandüler tularemi, ingüinal lenfadenopati, siprofloksasin, tedavi

Dear Editor,

Tularemia is a zoonotic disease caused by the Gram-negative bacteria *Francisella tularensis*. It is seen in the northern hemisphere<sup>[1]</sup>. *F. tularensis* is widespread in nature and has been isolated in more than 250 animal species. Natural reservoirs of the bacteria include rodents such as wild rabbits, squirrels, shrews, voles, and mice as well as beaver and deer. Humans and domestic pets are incidental hosts of *F. tularensis*<sup>[2]</sup>. The disease is transmitted to humans through the tissues of infected animals or by direct contact of their bodily fluids to the skin or mucosal surfaces; arthropod bites; consumption of contaminated water and food; and inhalation of infected aerosols<sup>[2,3]</sup>. The incubation period is about 2–5 days, and the disease develops 1–21 days after inoculation<sup>[4]</sup>. Human to human infection has not been documented. Depending on the route of infection, the disease may manifest as ulceroglandular, glandular, oculoglandular, oropharyngeal, typhoidal, or pneumonic tularemia<sup>[4]</sup>.

The aim of this paper was to present an adult who presented with inguinal lymphadenopathy (LAP) and was diagnosed with glandular tularemia, which is a relatively rare site of involvement in Turkey.

A 26-year-old male patient presented with fever, fatigue, and inguinal swelling which started 20 days ago. The patient resided in Bingöl and had previously been treated with amoxicillin/clavulanate by a physician at another center. After seven days of

antibiotic therapy, there was no fever but the inguinal swelling persisted. In his epidemiological history, he reported working with livestock and he had no history of tick bites. His family did not have similar symptoms. He had not consumed any game animals and reported drinking from the municipal water supply. On physical examination, a 4x8 cm palpable LAP with fluctuation was evident in the left inguinal zone and multiple palpable LAPs with millimetric diameter were detected in the bilateral inguinal zone (Figure 1). Other physical examination findings were normal. Results of laboratory tests were white blood cells count: 16.890/mm<sup>3</sup> (neutrophil 70.4%), hemoglobin: 13.9 gr/dL, hematocrit: 44.6%, platelet count: 235.000/mm<sup>3</sup>, aspartate transaminase: 18 U/L, alanine transaminase: 9 U/L, urea: 20 mg/dL, creatinine: 0.7 mg/dL, erythrocyte sedimentation rate: 29 mm/hour, C-reactive protein: 57 mg/L. *Brucella* tube agglutination, herpes simplex virus, toxoplasma, cytomegalovirus, Epstein-Barr virus, and syphilis serological tests were negative. Oral ciprofloxacin 500 mg q12h and metronidazole 500 mg q6h were initiated as empirical therapy. Interventional radiology was consulted and 15 mL of yellow, purulent fluid was aspirated from the LAP in the left inguinal zone. The fluid was analyzed by Gram, Giemsa, and acid-resistant bacilli (ARB) staining, culture, and cytology examination. No growth was observed in the culture; Gram and ARB staining were unremarkable. Cytological examination and Giemsa staining revealed predominant leukocytes with polymorphic nuclei. Quantiferon test for tuberculosis was negative. A serum sample submitted to the National Tularemia Reference Laboratory of the



Address for Correspondence/Yazışma Adresi: Ayşe Sağmak Tartar MD, Firat University Faculty of Medicine, Department of Infectious Diseases and Clinical Microbiology, Elazığ, Turkey

Phone: +90 543 408 84 68 E-mail: [dr.ayse01@mail.com](mailto:dr.ayse01@mail.com) ORCID: [orcid.org/0000-0002-9052-7986](http://orcid.org/0000-0002-9052-7986)

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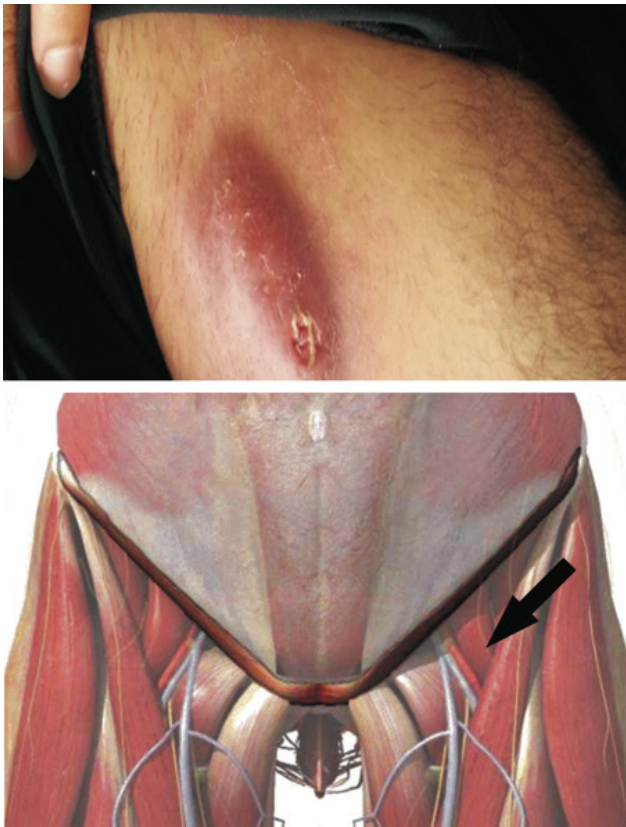


Figure 1. Glandular tularemia with inguinal focus

Public Health Agency of Turkey returned a microagglutination test result of 1/160, and a sample submitted 14 days later returned a result of 1/640. Metronidazole was discontinued after 14 days of antibiotherapy, while ciprofloxacin was administered for 21 days. The patient was discharged with full recovery.

Tularemia is a zoonotic disease. Except for the typhoidal and pneumonic forms, the mortality rate is less than 1%. Globally, the most common mode of transmission for the disease is via contact with infected animals and ticks; however, in Turkey it is most often transmitted through consumption of contaminated water and food. Therefore, oropharyngeal tularemia is the most prevalent clinical form in Turkey<sup>[5]</sup>. If diagnosis of oropharyngeal tularemia is delayed, glandular forms characterized by cervical lymphadenitis may develop. An analysis of the recent cases revealed that the glandular form is the second most common manifestation of tularemia<sup>[5]</sup>. The most common form worldwide is the ulceroglandular form, which develops after contacting infected animals and ticks<sup>[3]</sup>. Glandular tularemia accounts for 15–44% of all cases of tularemia worldwide<sup>[6]</sup>. These patients exhibit painful LAP primarily involving the submandibular, cervical, axillary and inguinal lymph nodes. If fever is absent, diagnosis may be delayed. Later, suppurative lymph nodes may develop fluctuation. Glandular tularemia is more common in children than adults<sup>[6]</sup>. Nevertheless, our adult patient was diagnosed

with glandular tularemia. On presentation, he exhibited a lymph node with fluctuation due to delayed diagnosis. The differential diagnosis of ulceroglandular and glandular tularemia includes diseases such as cat scratch disease, malignancy, mycobacterial infections, syphilis, lymphogranuloma venereum, chancroid, streptococcal or staphylococcal lymphadenitis, fungal infection, toxoplasmosis, rat bite disease, herpes simplex infection, and anthrax<sup>[7]</sup>.

According to the guidelines on tularemia from the World Health Organization, tularemia can be confirmed when at least one microagglutination titration is  $\geq 1:128$  or when titration quadruples between serum samples obtained at an interval of 14 days<sup>[2]</sup>. In our case, microagglutination test results were 1/160 in the initial sample and 1/640 in the sample obtained 14 days later. The patient showed full response to treatment and was discharged with complete recovery.

There have been cases of glandular tularemia with inguinal LAP reported in Turkey, but they were in pediatric patients<sup>[8]</sup>. Glandular tularemia cases in adults are generally in the form of cervical and axillary LAP. There is a report of an adult patient from Ankara with bilateral inguinal LAP and intraabdominal abscess<sup>[9]</sup>. Although tularemia-related inguinal LAP is rarely seen in Turkey, it accounts for 30% of the tularemia cases in North America<sup>[6]</sup>.

To date, there have been three cases of tickborne tularemia reported in our country, and all were ulceroglandular tularemia<sup>[10,11]</sup>. No data are available in Turkey with regard to the presence of *F. tularensis* in ticks.

Early treatment of tularemia helps reduce complications, relapse, and mortality. Delayed diagnosis leads to more complications, suppuration, and the need for surgical intervention. However, drainage may be required in spite of appropriate and early antibiotic therapy<sup>[1]</sup>. Our case presented late, so the abscess was drained. Tularemia is diagnosed sporadically in our region, and typically present nationwide as oropharyngeal and glandular tularemia.

In conclusion, although oropharyngeal tularemia is the most common clinical form in Turkey, it may exhibit atypical involvement in endemic regions, as in our case.

#### Ethics

**Informed Consent:** Written informed consent was obtained from patient who participated in this paper. Authors declared that the research was conducted according to the principles of the 'World Medical Association' Declaration of Helsinki "Ethical Principles for Medical Research Involving Human Subjects", (amended in October 2013).

**Peer-review:** Externally and internally peer-reviewed.

## Authorship Contributions

Surgical and Medical Practices: A.A., Concept: A.S.T., Design: A.S.T., Data Collection or Processing: A.S.T., Analysis or Interpretation: A.S.T., Literature Search: A.A., Writing: A.S.T.

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