

Oculoglandular tularemia: A case report

Oküloglandüler tularemi: Olgu sunumu

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ABSTRACT

Tularemia is an infection disease caused by *Francisella tularensis*. It is primarily a zoonosis, affecting mainly the rodents, which can serve as a reservoir for the microorganism. The transmission to human usually occurs through several ways such as a bite of the vectors, by handling an infected carcass or by taking in a contaminated food orally. There are seven clinical forms of the disease that makes up of depending on the body entrance. The ulceroglandular form is the most frequently encountered manifestation but those at least seen is the oculoglandular form. Tularemia may cause outbreaks in many part of the world. Small epidemics were reported from the Marmara and the Black Sea regions of Turkey. The adolesan case was presented with upper cervical lymphadenopathy and orbital swelling. It was diagnosed as tularemia by the positivity of the tularemia microagglutination test, and treated by streptomycin. We aimed to share in such as a rare case to refresh our knowledge and consideration at all.

Key Words: *Francisella tularensis*, tularemia, oculoglandular.

INTRODUCTION

Tularemia is an infection disease caused by *Francisella tularensis*, which is a small, pleomorphic, and gram-negative coccobacillus. It is primarily a zoonosis, affecting mainly the rodents that may serve as a reservoir for the microorganism. The transmission to human mostly occurs through a bite of some vectors such as ticks, flies or mosquitoes. Humans may also be infected by handling infected

ÖZET

Tularemi *Francisella tularensis*'in sebep olduğu bir enfeksiyon hastalığıdır. Başlıca, mikroorganizmalar için bir rezervuar olan kemirgenleri etkileyen zoonozdur. İnsana bulaşması vektörlerin ısırmasıyla, enfekte hayvan leşleri ile temas veya kontamine yiyeceklerin oral alınması gibi muhtelif yollarla olur. Vücuda giriş yerine bağlı olarak yedi klinik formu vardır. Ülseroglandüler form en sık, oküloglandüler form ise en az görülen şeklidir. Tularemi dünyanın pek çok yerinde salgınlara yol açabilir. Türkiye'de Marmara ve Karadeniz bölgesinde küçük salgınlara bildirilmiştir. Üst servikal lenfadenopati ve orbital şişlik ile başvuran adolesan hastaya tularemi mikroagglutinasyon test pozitifliği ile tularemi tanısı kondu ve streptomisinle tedavi edildi. Biz bu oldukça nadir görülen olguyu paylaşarak bilgi ve değerlendirmelerimizi tazelemeyi amaçladık.

Anahtar kelimeler: *Francisella tularensis*, tularemi, oküloglandüler.

animals or their carcasses, by consumption of contaminated water or foods or even rarely through inhalation.¹

Tularemia may cause outbreaks in many part of the world. Small epidemics were reported from the Marmara and the Black Sea regions of Turkey.^{2,3} The case is accepted a sporadic case as there had no family and journey history in the Yozgat, a city in the Middle Anatolia. We aimed to share in such as

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a rare case to refresh our knowledge and consideration at all.

CASE REPORT

A boy-14 years old, previously healthy, was presented with a right preauricular and cervical a hump and a swollen right upper lid (fig 1-2). He was complaining of fever, chills, sweating and severe headaches in addition to tenderness and rigidity of the lesion. On family inquiry, it was revealed that there had been eye redness and swelling just 3-4 days before regional lymph node enlargement in the same side. The symptoms had been persisting in spite of the triple antibiotics treatment concurrently (trimethoprim-sulfisoxazole, metronidazole, penicillin G), and the cervical mass had become progressively larger during last ten days. When taken closely the history he was bitten by a number of the flies perched upon a lemming carcass whilst shepherding in a rural area. On a physical examination, the certain easily palpable masses were seen on both preauricular (1x1 cm) and cervical region (5x5 cm) with a warm-reddish skin (Figures 1-2).

Laboratory results were gained as hemoglobin 11.3 g/dl, white blood count 12.400/mm³; platelet count 524.000/mm³, sedimentation rate 72mm/h, C-reactive protein 10mg/L and liver transaminases were within normal limits. The Mantoux tuberculin skin test was negative. Chest X-ray also was normal. Ultrasonography analyzing of the lesion confirmed multiple lymphadenopathy as predicted initially on physical examination. A large hypo echoic cavity under the skin (5x4x4cm³) was interpreted as a central necrosis, then, the sufficient material, gained by a needle aspiration, was send for the microbiological examination to determine the etiologic agent but none was able to be seen on the gram stain excepted many polymorphonuclear leukocytes. Multiple culture of the purulent material also failed to find out any microorganism. Tularemia microagglutination test performed in the patient's serum and was found positive at 1/1280 titer in Reference Laboratory for Turkey (Refik Saydam National Public Health Agency, Department of Communicable Diseases Research, National Tularemia Reference Laboratory, Ankara). PCR test and culture for tularemia was not studied. Streptomycin was given in a dose 40mg/kg daily and he quickly improved after a ten days therapy. On follow-up, he was evaluated as

having completely recovered a couple of months afterwards.



Figure 1. Note preauricular small and cervical large lymphadenopathy



Figure 2. Large cervical mass with little redness can be seen.

DISCUSSION

Tularemia is caused by a small, gram-negative, pleomorphic coccobacillus called *Francisella tularensis*. It is primarily an infection of wild animals that is transmitted to humans mainly through infected animal or insect bites especially in hot seasons. The symptoms appear after the incubation period of 3-7 days. When tularemia is acquired via the skin, a primary ulcer is often detected around the bite, and regional lymph nodes become prominently enlarged in general.¹

Tularemia has an acute onset with the symptoms associated such as fever, chills, lymphade-

nopathy, weakness, myalgia, arthralgia, vomiting and diarrhea.⁴ The diagnosis of tularemia is most commonly established through the use of a standard and highly reliable serum agglutination test. Therefore, the positive agglutination test (1/160 titer above) in a patient with a compatible history and physical finding is nearly only way to diagnose due to some troubles in culturing and isolating the bacteria. The prognosis is well enough if diagnosed rapidly and treated with appropriate antibiotic (aminoglycosides). First option in the treatment is streptomycin for a couple of weeks. Otherwise, serious consequences may develop especially in systemic involvements.¹ There are seven distinguishing clinical forms of tularemia, depending on the body entrance of the bacteria. The ulceroglandular form is the commonest manifestation. The others are called as the glandular, pulmonary, oropharyngeal, intestinal, typhoidal, and the oculoglandular form which is very rare. When the oculoglandular tularemia occurs, the conjunctiva should be the gate of entry and is probably contacted with contaminated fingers directly or bitten by a vector.

Conjunctiva inoculation may result in the orbital infection, which is a significant component of this form together with preauricular lymphadenopathy. The conjunctiva is painful and inflamed with regional lymphadenopathy which is referred to as Parinaud's oculoglandular syndrome.¹ Parinaud's oculoglandular syndrome, very rare entity, is known as a unilateral granulomatous follicular conjunctivitis associated with mucopurulent discharge as well as painful preauricular and submandibular lymphadenopathy. Little corneal ulceration or perforation may occur and might easily be seen by a fastidious examination. The patient usually shows a number of systemic signs like fever, malaise, headache, fatigue, and myalgia.

A history of exposure to a contaminated animal should be interrogated.^{5,6} Because *Francisella tularensis* can not easily be isolated on chemically supplemented chocolate agar to confirm the diagnosis. Owing to both miscellaneous causatives of the simi-

lar clinical manifestations and to difficulty in isolation of etiologic agent, the diagnosis is quite hard. The entities including tuberculosis, mononucleosis, toxoplasmosis, cat-scratch disease and malignant tumors such as lymphomas should be considered at differential diagnosis. In this case, the patient's confession to have touched a lemming carcass or bitten by the flies suggested that tularemia could be thought of the etiologic agent. There was no history of contact in the water sources. The clinical picture was also quite clear in terms of a well history of contacting with lemming carcass or insect bites, and the most likely source of infection was presumed to be one or both of them. The primary ulcer can generally be seen in conjunctiva, but sometimes it may be to have already healed in time of examination, as supposed to be the case, or not be formed yet by the time of the examination. Diagnostic problems and delays are not small enough to ignore even at this century.

In conclusion, tularemia should be suspected, if a patient is seen with appropriate clinical findings and sufficient investigations should be completed to set up a proper diagnosis and treatment.

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