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SHORT REPORT

The First Tularemia Case in Thrace Region of Turkey in the Last 60 Years*

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Tularemia is caused by *Francisella tularensis* which is a zoonosis frequently fatal in animals such as rats, guinea pigs, rabbits etc., but rarely fatal in humans (1). This disease may occur as an epidemic or can be seen sporadically. The biggest epidemic defined in Turkey started in the vicinity of Bursa in 1988, then other cases were reported from the Black Sea Region, Ankara, and more recently from Kocaeli and Konya (2). The first outbreak of tularemia occurred in the European part of Turkey (3, 4). This outbreak area included 34 villages of Kırklareli and Tekirdağ Provinces in 1936 as well as Lüleburgaz, a district of Kırklareli. A second outbreak reemerged also in Lüleburgaz in 1945. A total of 150 and 18 patients were reported in the first and second outbreaks, respectively (3-5). After that time, tularemia has not been reported in this region. We present here the first case of tularemia diagnosed in 60 years.

Case Report

The patient was a 30 year old woman with a complaint of a neck mass which appeared one month prior to admission to hospital. She had suffered from sore throat and fever two weeks before the development of the mass. Although she was treated with antibiotics

such as telithromycin, penicillin and cefazolin, there was no regression. The patient was admitted to the Otorhinolaryngology Clinic of Trakya University Hospital on 28th of February 2005. She was hospitalized with probable diagnoses of tuberculosis lymphadenitis or infected branchial cyst. She had a fixed, rigid, painful, partly hyperemic lymphadenopathy (LAP) with dimensions of 2x4 cm at the upper jugular region of the neck. Her haemogram was normal but erythrocyte sedimentation rate was high (55 mm/h). Chest x-ray did not show any abnormality. Conglomerated submandibular LAP with necrotic character, composed of a degenerating area, was detected on ultrasonography. The size of the thyroid was normal, and its activity homogenously increased as determined by scintigraphy. Two LAPs were excised by explorative cervicotomy on 2nd of March. Caseified granulomatous lymphadenitis was defined by using a pathological assay. Aerobic, anaerobic cultures, and acid fast staining of samples aspirated from LAP were negative. Tularemia antibody titer was positive in 1/10.240 dilution by tularemia microagglutination test. The patient was evaluated as a case of oropharyngeal tularemia. She was cured by daily administration of doxycycline 200 mg plus streptomycin 1 g for 10 days.

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The clinical course of tularemia may vary according to the site of entry and the virulence of the microorganism. Infected individuals may remain asymptomatic or the disease may progress to clinical forms, such as ulceroglandular, ocular, oropharyngeal, glandular, typhoid, pleuropulmonary, and gastrointestinal forms (6). The ulcero-glandular form is the most commonly reported clinical presentation in the literature (7), however recent reports from Turkey defined the oropharyngeal type as the predominant form (7, 8). In the oropharyngeal form, which was the clinical manifestation of our case, the causative organism enters the human body by consumption of contaminated water or food (9). Patients with oropharyngeal form of tularemia have fever, sore throat and cervical LAP. This form of the disease may mimic various diseases such as lymphoma, infected branchial cyst, LAP related to tuberculosis (10).

Tuberculosis is endemic and an important problem in Turkey, therefore in patients who have fever, sore throat plus prolonged and suppurated cervical LAP, it is the first considered disease. The pathological evaluation of the cervical LAP in tuberculosis usually reveals caseified granulomatous lymphadenitis, which is also a finding in tularemia. Microbiological procedures may be insufficient to confirm or reject the diagnosis of tuberculosis. Therefore, patients receive empirical anti-tuberculous therapies. Patients with tularemia recover if there is streptomycin in the combination of anti-tuberculous therapy. Nevertheless, patients with tularemia may unnecessarily be exposed to side effects related to prolonged and toxic therapies (10). On the other hand, since tularemia had not been reported for 60 years

misdiagnosis of tularemia in this region is likely. We presented the first case diagnosed in the last 60 years in Thrace Region of Turkey. Therefore, when tularemia is reported in a region, cases may appear in the following years, even latter decades.

Granulomatous LAP is a common pathological sign of some other bacterial (yersiniosis, brucellosis, salmonellosis, cat-scratch disease etc.), parasitic (toxoplasmosis, leishmaniasis etc.) and fungal infections (sporotrichosis, candidiasis etc.). Therefore, tularemia should always be distinguished from these infections by using serological assays and/or microbiological culture. The definitive diagnosis of tularemia requires isolation of the causative agent, which is rather difficult. Thus, tularemia is serologically distinguished from other infections by demonstration of antibody against *F.tularensis*, but the sera should also be examined by using Rose-Bengal test for brucellosis to eliminate cross-reactions (6). Confirmation of this presented case was done by observation of high antibody titers for tularemia but not for brucellosis.

In conclusion, tularemia should be considered in the differential diagnosis of patients with complaints of fever, sore throat and neck mass, especially when there is no response to non-specific antibiotic therapies, even in non-endemic regions.

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