**Open Journal of Surgery**

**ISSN: 2639-3611**

**Volume 1, Issue 2, 2018, PP: 15-19**

# A Case of Glandular Tularemia Applied to Surgical Drainage Due to Late Diagnosis

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**Abstract**

*Tularemia is a zoonotic infectious disease caused by Francisella tularensis, a gram-negative bacterium. The microorganism is transmitted to humans by direct contact with infected animals, through vectors such as flies and ticks, by inhalation of contaminated waters and infected aerosols. Depending on the route of transmission, infection may occur with different clinical pictures. In this study, we present a 13-year-old girl who presented to our clinic with a complaint of neck mass and then diagnosed as glandular tularemia. It is emphasized that the cases diagnosed late should not respond to medical treatment and surgical treatment is required. Diagnosis and treatment methods have been reviewed.*

**Keywords:** *Pediatric Patient, Surgical Drainage, Tularemia*

## Introduction

Tularemia is a zoonotic infectious disease caused by *Francisella tularensis*, a gram-negative bacterium (1). The microorganism is transmitted to humans by direct contact with infected animals, through vectors such as flies and ticks, by inhalation of contaminated waters and infected aerosols (2). Butchers, farmers in endemic areas, animal breeders, hunters, veterinarians and laboratory workers are risk groups (3-4). Incubation time is between 3-5 days, disease occurs at the earliest 2. day at the latest 14. days after infection (3). Depending on the virulence of the agent, the source of inoculation and the immunity of the host, the clinic of the disease appears (5). Depending on the route of transmission, infection may occur with different clinical pictures. Ulceroglandular form is seen most commonly in the World, but oropharyngeal form is seen most commonly in Turkey. (6).

In this study, we present a 13-year-old girl who presented to our clinic with a complaint of neck mass and then diagnosed as glandular tularemia. It is emphasized that the cases diagnosed lately should not respond to medical treatment and surgical treatment is required. In addition diagnosis and treatment methods have been reviewed.

## Case Presentation

A thirteen-year-old girl reffered by the external center because of a growing mass in her neck for a month. In the neck ultrasonography (USG), the 31x14 mms on the right side and 28x14 mms size on the left side were observed with several lymph nodes at the cervical vascular chain. The patient was directed to us for further examination and treatment. We were informed from the patient's history that he lived in the village, drank tap water, and his family were engaged in farming. His medical history and family history were absent. On physical examination, two 3x3 cms lymphadenopathy (LAP) were palpated and other system examinations were normal (Picture 1). Hemoglobin 12,2 g/dL, Leukocyte 11400/uL, Thrombocyte 377000/uL, C-Reactive Protein 0,26 mg/dL and erythrocyte sedimentation rate (ESR) 42 mm/h was detected in the laboratory results. Tularemia agglutination test was sent.

The patient was diagnosed with glandular tularemia and was started on gentamicin treatment 5 mg/kg IM dose. On the tenth day of the treatment a 3x3 cms on the right side, 7x7 cms lymphadenopathy on the left side

**Fig1.** *When the patient comes first*

was palpated and fluctuations were observed (Picture 2). The result of microagglutination test (MAT) was reported to be positive at 1/1280 titer. Gentamicin was discontinued and ciprofloxacin was started. Two days after the antibiotic change, when the patient was checked, there was no regression and drainage was performed by the pediatric surgery. Many leukocytes were seen in the culture, and epithelial and dominant microorganisms were not observed. Ciprofloxacin treatment was given for 21 days.

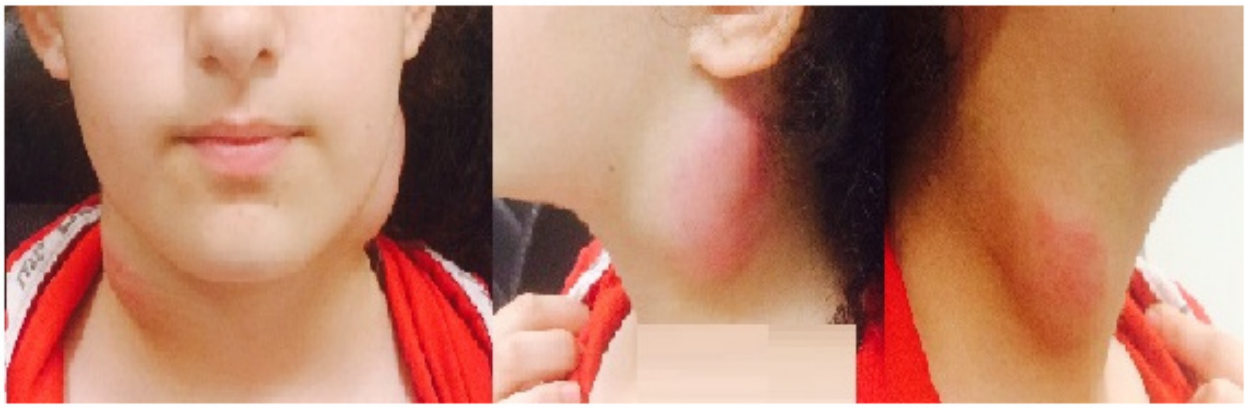

**Fig2.** *The was not improvement after antibiotherapy, fluctuations evolved.*

At the post-treatment polyclinic control ultrasound, showed that the biggest one 12x19 mm size in the right cervical vascular chain and the biggest one 15x23 mm size in the left cervical vascular chain several lymph nodes were observed. Suggestions were made for the patient whose complaints were regressed and the tests improved (ESR 20 mm/h) (Picture 3). The patient was called for check-up three months later. Consent was obtained from patient parents for case report.

**Fig3.** *After surgical drainage*

## Discussion

Tularemia is a zoonotic disease, which is usually limited to the northern hemisphere countries (North America, Japan, Finland, Sweden, Austria, Germany, Spain, Hungary and Bulgaria), sometimes seen endemic, sometimes in the form of outbreaks (7). The first study describing the etiological factor of tularemin in the literature was published in 1912 after the isolation of the agent from the dead ground squirrels in Tulare, California by McCoy and Chapin(8). Tularemia was first detected in 1936 in Turkey. In the Thrace region, it was due to the epidemic seen with 150 patients (9).

*F. tularensis*, *F. tularensis subsp. tularensis* (type A*), F. tularensis subsp. holarctica* (type B), *F. tularensis subsp. Mediasiatica* and *F. tularensis subsp. novicidi* to four subtypes have been defined. *F. tularensis subsp. Tularensis* (type A) is the most virulent type and is common in North America. *F. tularensis subsp. holarctica* by (type B) are subtypes that cause outbreaks in Turkey and northern sphere, and is less virulence (10).

The most common form in the world is ulceroglandular form and is mostly observed after tick bite or contact with infected animal tissues (10). Zargan et al. between the years 1936-2011 reported that they screened emerging cases of tularemia in Turkey. They determined that the majority of tularemia cases were oropharyngeal tularemia and the cause of the infection was 90% determined that they are waterborne (11,12). In studies conducted in Turkey is the most commonly reported form of oropharyngeal. Our patient was evaluated as glandular form.

The microorganism can easily pass through the skin and mucous membranes in tularemia.Consequently, the infection is frequently caused by contact with the skin or mucosa, infected animal tissue or body fluid. Also infected mosquito, tick etc. transmitted after being bitten by the arthropod. Contaminated dust may be transmitted by inhalation of food and water. However, this organism is not transmitted from human to human. In publication conducted in Turkey it was by seen water borne epidemics (12). Leblebicioğlu et al. in their case-control study, It was reported that ingestion of food collected from the environment and drinking of tap water was a risk factor for tularemia (9). In our case, he said he drank from fountain water. We thought that it was a water-borne infected.

Fever, chills, tonsillopharyngitis, sore throat, headache, weakness, anorexia, oral mucosa ulcers and usually unilateral cervical lymphadenopathies are seen. The most important complaint of patients with oropharyngeal type is sore throat and fever, and glandular tularemia has a mass in the neck. The most common complication of the disease is lymph node suppuration and fluctuations. Late initiation of treatment increases the likelihood of suppuration (13). There was a mass in the neck in our case. In our patient, she had developed fluctuations due to late diagnosis.

In the diagnosis phase, especially the patient living in an endemic area or visiting to that region should remind tularemia. Resumes and physical examination are important, should be done carefully. Animal and tick exposure with fountain water drinking is important for diagnosis in Tularemia. Routine cultures have low sensitivity in the diagnosis of tularemia(12). The exact diagnosis is made by the production of the bacteria. The diagnosis is made by serology tests in practice, because it is difficult to produce the bacteria. Since tularemia agent is a single antigenic type, it is possible to diagnose with agglutination, MAT and ELISA. The response in the agglutination test occurs between the 1st and 2nd week of the disease.The ≥1:160 in agglutination test and ≥1:128 in MAT is considered significant. Also, it can also be diagnosed by PCR (9,13). In our patient, the result of MAT was positive at 1/1280 titer and tularemia was diagnosed.

In differential diagnosis, bacterial adenitis caused by staphylococcus and group A streptococci, cat-scratch disease, mycobacterium tuberculosis related lymphadenopathy, toxoplasmosis, infectious mononucleosis, anthrax, plague and Kawasaki disease should be considered (12). In our patient, a preliminary diagnosis of tularemia was made by a result of physical examination and resume, MAT were requested.

*F. tularensis* is sensitive to aminoglycosides; Streptomycin and gentamicin are the first options for the treatment of tularemia in children. Although oral alternative drugs are ciprofloxacin and doxycycline, there are not enough clinical data about their efficacy. The treatment duration is usually 14-21 days. The use of tetracycline and chloramphenicol in the pediatric age group due to high probability of recurrence is limited. Treatment is continued for 7-10 days, especially antibiotic treatment started in the first three weeks has been shown to be effective in improving infection (10,12,14). *F. tularensis* is resistant to beta-lactam antibiotics. In patients initially treated with this antibiotic, the diagnosis is delayed and the likelihood of chronicity increases (3). Çağlı et al. reported that delayed treatment resulted in complications requiring surgical intervention (15). Surgical drainage of the lymph node is recommended if abscesses develop in patients, but excision of the mass is not recommended (13). In our case, gentamicin treatment was given for 10 days and ciprofloxacin treatment was given because of the lack of response. Despite the treatments given in our patient, the existing LAP sizes increased and fluctuation was observed. Therefore, surgical intervention was performed.Ciprofloxacin treatment was completed to 21 days due to the possibility of recurrence after surgical drainage.

In conclusion, tularemia is a rare infectious disease that causes a mass in the head and neck.Tularemia should be considered among the differential diagnoses in cases presenting with lymphadenitis in childhood. The presence of similar disease in where they live, animal contact, use of tap water and travel should be questioned.In the early period, patients should be suspected and MAT is required and starting the appropriate treatment will be the main factor in the treatment of the disease. As in our patient, there is a permanent scar attached to surgical drainage with time and cost loss in late diagnosed cases.

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**Citation:** **Muhammet Mesut Nezir Engin, Önder Kılıçaslan, Ramazan Cahit Temizkan, Kenan Kocabay.** *A Case of Glandular Tularemia Applied to Surgical Drainage Due to Late Diagnosis. Open Journal of Surgery. 2018; 1(2): 15-19.*