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# Cervical tularaemia in a non-endemic area

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## Abstract
Tularemia is a zoonotic disease caused by Francisella tularensis. The microorganism is transmitted to humans by contact with, or ingestion of, infected animal tissues, by insect bites, consumption of contaminated food or water, or from inhalation of aerolized bacteria. In this report we describe a case of tularemia presenting with multiple cervical lymphadenitis in Asturias (Spain). Final diagnosis was established based on a serological test. The patient was successfully managed with surgery and streptomycin for 2 weeks. The ulceroglandular form of tularemia should be considered in the differential diagnosis of cervical lymphadenitis, particularly in those not responding to penicillin treatment. To our knowledge, this is the first case described in Asturias, a north coast county of Spain.

***Key words:*** *Tularaemia, Francisella tularensis, zoonosis, lymphadenopathy.*

## Introduction
Tularemia is a zoonotic infection caused by *Francisella tularensis* (*F. tularensis*), a non-capsulated, Gram-negative coccobacillus. Two main types of *F. tularensis* have been described, type A and type B, which have differences in their epidemiology and virulence (1). *F. tularensis* biovar type A has the highest mortality rate in humans and is predominantly seen in North America; type B, a less virulent biovar, is widely distributed throughout the Northern Hemisphere. In Spain, this is a rare illness with sporadic reports. The first tularaemia outbreak in northwestern Spain (Castilla-León) occurred in 1998 with 585 cases (2), and there was another in 2004 with 13 cases (3). Between 2000 and 2003, seven cases were reported in that region. The last outbreak was reported in 2007 with 362 cases confirmed in the same zone of the country (3).
We report one case of ulceroglandular tularemia in Asturias, north coaster county of Spain. The disease had not been previously reported in this region.

## Case Reports
A 77-year-old woman arrived at the department of oral and maxillofacial surgery of the University Central Hospital (Asturias, Spain) in August 2007 with a 2-months

history of fever and left cervical mass. The patient did not refer arthralgia or joint swelling. There was no history of recent travel or exposure to ill people. She had received non-specific treatment (oral amoxicillin/clavulanate) for 2 weeks without improvement.

In the physical examination she had fixed left cervical mass, of 8 cm in diameter. It was more painful on palpation and exhibited increased local surface temperature and redness. Other physical examination signs were normal. No cutaneous or intraoral lesions were observed.

A detailed examination of the oral cavity did not identify a cause. Orthopantomography did not reveal radiolucencies or dental pathology.

In the laboratory investigations, the leukocyte count was 9800 (63% neutrophills, 34% lymphocytes, 3% monocytes), haemoglobin 12.3 g/dl, platelet count 352,000/mm3 and the erythrocyte sedimentation rate was 53 mm/h. Serum electrolytes and liver and kidney function tests were normal.

A fine-needle aspiration biopsy of the cervical mass revealed lymphadenitis with a focal necrotic area. Cytologic evaluation showed no evidence of cancer. Computed tomography revealed multiple left cervical and supraclavicular lymphadenopathies with necrotic areas (Fig.1).

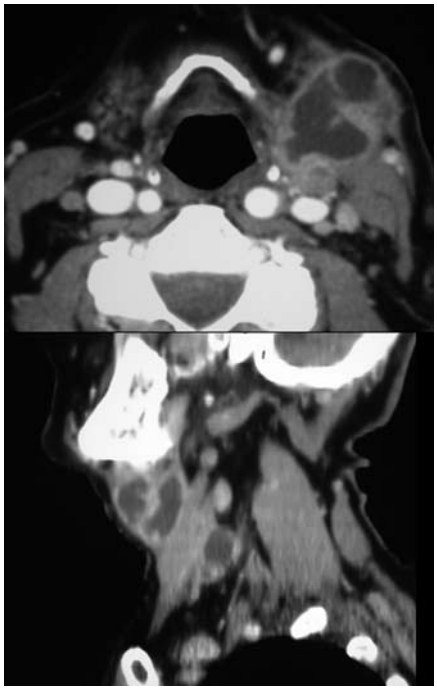

**Fig. 1.** Computed tomography image showing left cervical and supraclavicular lymphadenopathies with necrotic areas.

The patient was taken to the operating room in October 2007 and the abscessed cervical and supraclavicular mass was drained and removed. Intraoperative biopsy was consistent with granulomatous lymphadenitis. Stained cultures were obtained to look for tuberculosis and aerobic and anaerobic organisms.

The pathologists entertained the possibility of tuberculosis and recommended additional test to search for a bacteriological source. Confirmatory diagnosis of ulceroglandular tularaemia was made by haemaglutinin titers. Serologic tests later showed that the patient had a *Francisella tularensis* infection, and antibody levels were 4096 mg/UI. Antimicrobial therapy (streptomycin 1 g every 12 hours IM) was given for 14 days and the patient recovered completely. During the patient´s recovery, she related that feeds rabbits in her country house. These animals were the most likely vector for the infection.

## Discussion

The first report of *F. tularensis* infection was reported in 1911 by McCoy in Tulare County, California (4). *F. tularensis* is a short rod-shaped or coccoid, faintly staining, strictly aerobic Gram-negative bacterium which comprises two predominant subspecies, *F. tularensis* spp. tularensis (Jellison type A) and *F. tularensis* spp. holarctica (Jellison type B). *F. tularensis* type A is isolated in North America while *F. tularensis* type B is spread more widely over the Northern hemisphere and is the sole subspecies isolated in European countries (5).

A variety of small mammals (as rodents and lagomorphs) are natural reservoirs of F. tularensis, which they acquire through bites by ticks, flies and mosquitoes or by contact with contaminated soil or water. The microorganism is transmitted to humans by direct contact with infected animals, by insect bites, by inhalation or by contact with contaminated meat or water (6).

The incubation period averages 3 to 5 days, but ranges from fewer than 1 to 21 days (1,7). After this period, symptoms consist of fever, chills, headache, cough, and myalgias. Ulceroglandular tularaemia is most frequently caused by vector-borne transmission, in the USA and Central European regions by ticks and in Northern Europe by mosquitoes. Patients with ulceroglandular tularemia have a local cutaneous papule which develops at the time of onset of general symptoms (1). Within a few days, the papule may become pustular and ulcerate and after this, regional lymph node enlargement is noticed by the patient. Enlarged lymph nodes, especially in the cervical and periauricular area, are seen in approximately 85% of patients and may be the only clinical sign in nearly 50% of infected persons (8).

Respiratory tularaemia is contracted by inhalation of aerosolised F. tularensis. Compared with the ulceroglandular form, outbreaks of respiratory tularaemia occur infrequently, especially in Europe (1).

As the culture and isolation of the bacterium is difficult, the microbiological diagnosis of tularaemia relies mainly on serology (9). Tube agglutination and microagglutination are usually preferred for diagnosis, although during the last decades, have been replaced by enzyme-linked immunosorbent assay (ELISA). Results become positive 10-14 days after the onset of the disease (6). In the pathological examination of the tularaemia lymph nodules, lesions are characterized by the presence of histiocytes, macrophages, lymphocytes and giant cells in addition to necrosis areas. Results in routine laboratory tests are non-specific (10).

The drug of first choice for the treatment of all forms of tularemia is streptomycin, although gentamicin is an acceptable substitute. Betalactams, macrolides, lincosamides, and trimoxazole are not reliable for treatment of tularaemia. Ciprofloxaxin has also been demonstrated to be effective in vitro and in infected animals. The therapy must be given for 7-14 days (1,10).

In comparison with the Nordic countries, the incidence is generally lower in central and southern Europe. In Spain, tularaemia in humans was first reported in 1998, when 585 cases occurred in Castilla-León, a big inner county on north-western (2). In 2004 were detected 13 cases. Recently, in 2007 have been reported 362 cases in the same region (3). In the 1998 outbreak, the mechanism of transmission was by contact, while in 2007 inhalation has been more frequent. To our knowledge, there has not been reported any case of tularaemia previously in Asturias, a small coaster county on north Spain.

## Conclusion

This case of ulceroglandular tularemia demonstrate that while rare in a nonendemic area, the disease must be considered in patients with lymphadenitis with necrotic areas who fail to improve with antibiotic therapy. Contact with infected animals could guide the diagnosis that must be confirmed with serologic test or cultures.

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