

Tularaemia presenting as parapharyngeal abscess: case presentation

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Abstract

Objective: We report an extremely rare case of the oropharyngeal form of tularaemia, causing a parapharyngeal abscess.

Case report: A 48-year-old woman presented with fever, sore throat, breathing difficulty and a right-sided neck swelling. This mass had previously been treated with penicillin without response, and had already been surgically drained once in another hospital. On physical examination, the tonsils were exudative and hypertrophic and the pharynx was hyperaemic. A fluctuant, 4 × 4 cm mass was seen on endoscopic examination, originating from the left parapharyngeal area and protruding towards the pyriform sinus, and partly obstructing the airway. Microagglutination test antibody titres for *Francisella tularensis* were positive (1/1280). The patient healed completely after definitive drainage of the abscess and antimicrobial therapy for 14 days (streptomycin, 2 × 1 g intramuscularly).

Conclusion: Tularaemia should be considered in the differential diagnosis of patients presenting with tonsillopharyngitis, cervical lymphadenitis and parapharyngeal abscess who do not respond to treatment with penicillin, even if they do not live in an endemic region.

Key words: Tularaemia; Pharyngeal Disease; Abscess; Diagnosis; Oropharynx; Francisella Tularensis

Introduction

Tularaemia is a zoonotic illness caused by *Francisella tularensis*, a non-encapsulated, Gram-negative coccobacillus.

There are two types of *F tularensis* subspecies: *tularensis* (known as type A) and *holarctica* (type B), distinguished by bacterial virulence and clinical characteristics.¹ *Francisella tularensis* subspecies *tularensis* (type A) is restricted almost completely to North America, whereas *F tularensis* subspecies *holarctica* (type B) is predominantly seen in the Northern Hemisphere.² Widespread outbreaks of tularaemia have been reported in European and Asian countries such as Sweden, Turkey, Bulgaria, Finland, Russia, Spain and India.^{3–9}

Type A tularaemia generally spreads by inhalation and has a more severe clinical course than type B, with more frequent serious complications such as septic shock and rhabdomyolysis.¹ The illness spreads to humans by direct contact with infected animals and arthropods, by inhalation, or from contaminated food and water. The incubation period is usually 3 to 6 days, but has been reported to range from 1 to 21 days.^{9–11}

Clinical forms may differ according to the bacterial virulence and route of spread and the patient's immunological status.

Here, we present a case of the oropharyngeal form of tularaemia, which caused a parapharyngeal abscess.

Case report

A 48-year-old woman was admitted to hospital with fever, sore throat, difficulty breathing and a neck swelling. Her

present complaints had started a week before and had progressively worsened. She reported a painful, approximately 3 × 2 cm swelling in the right posterior triangle of her neck. This swelling had not responded to treatment with penicillin, and had already been surgically drained once in another hospital. One month previously, another painful swelling had occurred in level three of the patient's neck on the left side, but had spontaneously discharged.

The physical examination findings were as follows. The patient had a fever of 38.6°C. Lymphadenopathy was found on the left side of the patient's neck, with a tender, non-fluctuant, 3 × 3 cm mass. The tonsillae were exudative and hypertrophic, and the pharynx was hyperaemic in appearance. A fluctuating, 4 × 4 cm mass was seen on endoscopic examination, originating from the left parapharyngeal area and protruding towards the pyriform sinus, and partly obstructing the airway. Other physical examination findings were normal.

Biopsy specimens from the lymphadenopathy showed granulomatous inflammation on histopathological examination.

Computed tomography (CT) scanning of the patient's neck showed an extensive, 35 × 25 mm abscess extending from the fossa of Rosenmüller inferiorly to the level of the epiglottis and narrowing the nasopharyngeal and oropharyngeal lumen (Figure 1).

Laboratory tests gave the following results: leucocytes, 6000/ml (72 per cent neutrophils, 16.7 per cent lymphocytes, 6.2 per cent monocytes); haemoglobin concentration,

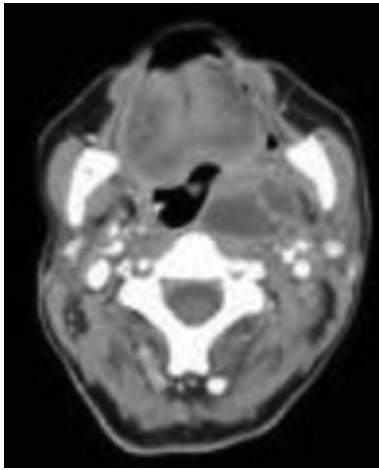


FIG. 1

Axial computed tomography scan of the patient's neck.

11.8 gr/dl; platelet count, $218 \times 10^3/\mu\text{l}$; erythrocyte sedimentation rate, 27 mm/h; C-reactive protein (CRP), 12.3 mg/l; and negative results for purified protein derivative (PPD) test and anti-streptolysin O (ASO). Results for serum electrolytes and liver and kidney function tests were normal.

Two cultures were prepared from specimens taken from the parapharyngeal abscess, but no growth was observed in either.

Antibody titres of the microagglutination test for *F tularensis* were positive (1/1280).

No pathological change was observed in the chest X-ray.

After drainage of the abscess and antimicrobial therapy for 14 days (streptomycin 2×1 g intramuscularly), the patient healed completely.

Discussion

Infection with *F tularensis* was first reported by McCoy in 1911 in California, USA.²

There are six forms of the disease: ulceroglandular, glandular, oropharyngeal, typhoid, pneumonic and oculoglandular.

The ulceroglandular form is seen most frequently. This results from skin trauma from an infected animal or insect (e.g. cats or mosquitoes) and causes fever, an ulcer or pustule at the inoculation site, and lymphadenitis with suppuration.¹²

The glandular form comprises lymphadenopathy without a skin lesion.

The oculoglandular form occurs as a result of bacterial inoculation of the conjunctiva. Cervical or pre-auricular lymphadenopathy is seen along with conjunctivitis.¹³

Airways infection is seen in the pneumonic form, which results from inhalation of the micro-organism.

Systemic findings are observed in the typhoid form.^{14,15}

The oropharyngeal form is caused by contact with contaminated water or food. This form has been reported to constitute between 1 and 4 per cent of all tularaemia cases. Fever, cervical lymphadenopathy, ulcerative pharyngitis and tonsillitis are seen, with the most frequent symptoms being fever and sore throat.⁴ This is the form of *F tularensis* infection most frequently encountered by the ENT specialist.

Tularaemia has also been reported to have other clinical presentations, including acute otitis media and retropharyngeal abscesses.¹⁵ The occurrence of a parapharyngeal

abscess caused by the oropharyngeal form of the disease, as seen in the present case, has rarely been reported.

The fever, sore throat and lymphadenopathy caused by oropharyngeal tularaemia may be seen in other diseases such as acute bacterial tonsillitis, infectious mononucleosis and tuberculosis. As tularaemia is an infrequent illness with no specific findings, its diagnosis is generally delayed. Routine biochemical and haematological tests may show non-specific inflammatory findings, such as a raised CRP and leukocyte count. The T-cell count may also be raised, as is the case with other intracellular pathogens.¹⁶ In the present case, the leukocyte count, lymphocyte count and CRP level were all increased.

Tularaemia may be diagnosed using a direct enzyme-linked immunosorbent assay against *F tularensis* lipopolysaccharide. Quicker diagnosis is also possible by applying the polymerase chain reaction technique to specimens of exudate, blood and secretions.¹⁷ The micro-organism requires cystine (or culture media containing cystine) in order to grow, and growth takes 10 or more days; these two factors frequently cause false negative culture results. Third-degree laboratory safety precautions are required in order to protect laboratory personnel from secondary infection.¹⁰ In the present case, our two attempts at culturing fluid drained from the parapharyngeal abscess both gave negative results.

The most frequently used diagnostic method for tularaemia is serological examination.¹¹ Cross-reaction may occur with brucella, legionella, salmonella and yersinia antibodies. Enzyme-linked immunosorbent assay or agglutination methods are used. Patients without *F tularensis* infection have titres of 1/10–1/80. A titre of over 1/160 in tube agglutination, over 1/128 in microagglutination or a four-fold rise in two to four weeks are diagnostic. As the test result may be negative in the acute phase, serological tests should be done at least 10 days after the beginning of the illness.⁷ In the present case, the tube agglutination test result on the 15th day was 1/1280.

Histopathologically, tularaemia has chronic granulomatous inflammation characteristics. Epithelioid histiocytes, macrophages, lymphocytes and giant cells may be seen in the lesions.¹⁵ In the present case, a granulomatous inflammation was observed upon histopathological examination of the cervical lymphadenopathy specimen. Lymph nodes with central necrosis may develop into an abscess over time and spontaneously discharge.¹⁸ Our patient's diagnosis had been delayed and her cervical lymphadenopathy had developed into abscesses; one had already discharged spontaneously and another had been drained elsewhere but had not resolved.

- **Tularaemia is a zoonotic illness caused by *Francisella tularensis***
- **Its diagnosis requires a high index of clinical suspicion**
- **Oropharyngeal tularaemia should be considered in cases with tonsillopharyngitis, cervical lymphadenitis and parapharyngeal abscess**

Imaging findings are non-specific for tularaemia. Enlarged lymph nodes and abscess formation associated with oropharyngeal tularaemia may be seen by contrast-enhanced CT or

magnetic resonance imaging.^{19,20} In our patient, CT imaging revealed a parapharyngeal abscess.

The diagnosis of tularaemia requires a high index of clinical suspicion. The diagnosis is based on combined evaluation of the clinical features, histopathological findings, serological tests and culture results.¹⁸ The differential diagnosis of otolaryngological tularaemia includes streptococcal and staphylococcal tonsillitis, Epstein–Barr virus infection, diphtheria, tuberculosis, cat-scratch disease, Lyme disease, fungal infections, toxoplasmosis, and malignant conditions.^{17,21,22}

Francisella tularensis is resistant to penicillins. Thus, aminoglycosides, macrolides and fluoroquinolones play an important role in the treatment of tularaemia. Streptomycin is the first choice for the treatment of all forms of tularaemia except that involving meningitis. Medical treatment should continue for 7 to 14 days.^{23,24} Our patient healed completely following drainage of her abscess and 14 days of antimicrobial therapy (streptomycin 2 × 1 g intramuscularly).

Tularaemia is not easy to diagnose due to its non-specific symptoms and relatively rare occurrence. Clinicians who work outside endemic areas may not be aware of this disease. However, early diagnosis of tularaemia is important. Tularaemia should be considered in the differential diagnosis of patients presenting with tonsillopharyngitis, cervical lymphadenitis and parapharyngeal abscess who do not respond to treatment with penicillin, even if they do not live in an endemic region.

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Dr S Koc takes responsibility for the integrity of the content of the paper

Competing interests: None declared
