

# Oropharyngeal Tularemia: A Case of Ulcerative Pharyngitis and Necrotizing Pyogranulomatous Lymphadenitis

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## Keywords

tularemia, rabbit fever, cervical adenopathy, necrotizing granuloma, necrotic tonsil

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**T**ularemia is a rare zoonotic disease that often goes undiagnosed for several weeks, leading to delayed treatment. A 19-year-old woman presented with necrotic oropharyngeal lesions, fever, and odynophagia. Due to low clinical suspicion and nonspecific symptoms, her diagnosis was not made for 2 months. This case highlights the importance of maintaining tularemia in the differential diagnosis, especially when the patient has a history of travel to a region with previous outbreaks. St. Elizabeth Youngstown Institutional Review Board granted an exemption for this report.

## Case

An otherwise healthy 19-year-old woman presented with 10 days of severe sore throat, odynophagia, and fever to a local health facility in Martha's Vineyard, Massachusetts. Despite treatments with oral steroids, clindamycin, and penicillin on 2 separate occasions, her symptomatology persisted. Otolaryngology service was consulted as she returned home to Ohio after 2 weeks. Upon further questioning, she did admit to interacting with a rabbit. Physical examination revealed bilateral 2-cm tender, nonfluctuant cervical adenopathy without skin changes, 2+ mild necrotic tonsils (**Figure 1**), and diffuse aphthous ulcerations throughout the posterior pharyngeal wall, tongue base, and palate. There was no involvement of the hypopharynx or larynx on flexible laryngoscopy. A respiratory panel with the most common types of viral and bacterial pathogens was

negative. Infectious disease service placed the patient on empiric intravenous antiviral, antifungal, antibacterial, and steroid medications. Her symptoms subsided, and she was subsequently discharged. However, she had multiple readmissions due to relapsing fevers and cervical adenopathy. A contrast-enhanced computed tomography (CT) demonstrated bilateral cervical adenopathy with focal necrosis (**Figure 2**). Excisional biopsy revealed friable lymphoid tissue with copious amount of purulent exudate bilaterally. Pathology showed lymphoid material with pyogranulomatous and necrotizing granulomatous inflammation. Histological stains were negative for mycobacteria and fungal organisms. Viral cytopathic effect was not identified. Ultimately, universal polymerase chain reaction was positive for *Francisella tularensis*. The patient was then treated with intravenous gentamicin and ciprofloxacin and responded appropriately.

## Discussion

*F tularensis* is a highly virulent pathogen that could infect a host with as few as 10 organisms.<sup>1</sup> Thus, this gram-negative, coccobacillus, facultative intracellular bacteria was touted as a possible biological form of terrorism.<sup>1</sup> Despite its long history, the diagnosis still often eludes physicians to this day.

Tularemia comprises 6 unique types. Ulceroglandular and glandular tularemia make up the majority of infections seen in the United States, with oculoglandular, typhoid, and pneumonic being far less common. Pharyngeal tularemia is the rarest form of the disease, as seen in this case. It has

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**Figure 1.** Clinical photograph showing a necrotic lesion on the right palatine tonsil.



**Figure 2.** Coronal view of a contrast-enhanced computed tomography demonstrating bilateral enhancing masses with loculations and necrosis.

been reported more frequently in Europe and Asia, and it is most commonly seen during the summer months in children younger than 14 years.<sup>2</sup>

These infections have a strong geographic distribution, with many cases being reported in Arkansas, Oklahoma, and Missouri. Historically, rabbits are the most common disease reservoir, but the organism has been found in contaminated water, carcasses, domestic and wild animals, and other rodents. Since the cottontail rabbit was introduced onto Martha's Vineyard, this isolated island has experienced 2 outbreaks of pneumonic tularemia. Reviews of these 2 outbreaks performed by Feldman et al<sup>3</sup> did not report any cases of pharyngeal tularemia. Two risk factors that were seen in 1 case-control study were lawnmowing and brush cutting. This patient denied any such exposures.

Multiple factors often lead to a delay in diagnosis and proper treatment of tularemia, one of which is the difficulty in obtaining a definitive pathologic diagnosis. *F tularensis* requires cysteine rich culture and up to 2 weeks of incubation prior to growth. Diagnosis may be reached by microagglutination test using antibody titers that appear after 2 weeks of infection or universal polymerase chain reaction.<sup>4</sup> In places with infrequent cases, the laboratories may lack the necessary equipment, and initial seronegative cultures will add to the delay in diagnosis. A case series performed by Çağlı et al<sup>5</sup> found the mean number of days to diagnosis was 51 days, compared to 48 days in this case. Other factors leading to long diagnostic delay include the rarity of disease, misdiagnosis and subsequent treatment, and patient inattention. The uncommon presentation of a rare disease, multiple unsuccessful treatments, and geographic location all contributed to the final diagnosis of tularemia in this patient. This case demonstrates the ever-important history of present illness, varied clinical manifestations of rare diseases, and maintaining a high clinical suspicion.

#### Author Contributions

**Kyle P. Quillin**, drafting, critical revision, final approval, accountability for the work, substantial contributions to the conception and design of the work; **Brandon E. Fornwalt**, drafting, critical revision, final approval, accountability for the work, substantial contributions to the conception and design of the work; **Eugene L. Potesta Jr**, drafting, critical revision, final approval, accountability for the work, substantial contributions to the acquisition, analysis, and interpretation of data for the work; **Cang T. Nguyen**, drafting, critical revision, final approval, accountability for the work, substantial contributions to the acquisition, analysis, and interpretation of data for the work.

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