

Complicated Ulceroglandular Tularemia

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Abstract

A 52-year-old woman presented with a tender swelling in the right axilla, fever, a headache, nausea, and general weakness. On examination, she was found to have lymphangitis on the right arm and red papules on the 1st and 2nd fingers of the right hand. She had had prepared wild rabbit stew 5 days before disease onset. Serology and an ultrasound of the right axilla confirmed the diagnosis of ulceroglandular tularemia. The lymphadenitis did not resolve after streptomycin treatment so an incision was made and 30 cc of purulent fluid drained. Over the course of the next 3 months, the fluid continued to drain. A radiographic fistulography was performed and it revealed a short main channel with a few long channels of varying caliber branching out from it, all terminating in a conglomerate of necrotic axillary lymph nodes. The lesions healed spontaneously and completely over the following 12 months without additional antibiotic therapy. Radiographic fistulography can help plot the course of the fistula/fistulas and demonstrate the anatomic features of the lesion in resource poor settings.

Keywords: Case report, fistula, fistulography, lymphadenitis, tularemia

INTRODUCTION

Tularemia is a zoonotic and occasionally vector-borne bacterial disease that is almost exclusively found on the northern hemisphere.^[1-3] Several clinical phenotypes of the disease exist: Ulceroglandular tularemia, glandular tularemia, oculoglandular tularemia, oropharyngeal tularemia, typhoidal tularemia, and pneumonic tularemia.^[3] Tularemia can rarely be complicated by external or internal spontaneous fistulization. Ulceroglandular and glandular tularemia are usually managed conservatively utilizing antibiotic monotherapy, but the need for surgery occasionally arises.^[1,3] Surgery is usually indicated to remove necrotic tissue or to drain an abscess.^[3] Even though sophisticated diagnostic techniques are now available to aid the clinician in evaluating the damage caused by this destructive infection, much less costly, yet safe and time-honored procedures such as radiographic fistulography are also useful in this context. Here, we present a complicated case of ulceroglandular tularemia with chronic drainage form an external skin fistula diagnosed with radiographic fistulography.

The patient's permission was obtained and the study was conducted in accordance to the Declaration of Helsinki.

CASE REPORT

A 52-year-old woman presented to the University Clinic for Tropical and Infectious Diseases in Belgrade, Serbia, with a tender right axilla and acute onset fever, chills, headache, nausea, and malaise. On examination, she was found to have right axillary lymphadenitis, lymphangitis on the right arm, and red papules on the fingers of the right hand. The patient's medical and family history was inconspicuous. She had had prepared wild rabbit stew 5 days before symptom onset.

The diagnosis of ulceroglandular tularemia was made by serology (tube agglutination with a titer of 1/320) and an ultrasound of the right axilla. The patient was started on intravenous streptomycin. After 10 days of treatment, all the symptoms abated save the swelling in the axilla that did not recede and fluctuation was noted during follow-up. An incision was made and 30 cc of sanguinopurulent material was evacuated. Bacterial cultures of the material remained sterile. However, a draining fistula was formed by the

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procedure and it did not heal over the course of the next 3 months.

In order to visualize the cavity, a radiographic fistulography was performed since magnetic resonance imaging was not readily available at the time. Biligriffin was used as a contrast medium. The radiograph revealed a short and narrow main channel branching out into a few long channels of varying caliber all terminating in a conglomerate of necrotic axillary lymph nodes [Figure 1]. Since no internal fistulization was observed, we opted for a watchful waiting approach instead of additional surgery and/or antibiotics. The lesions healed completely and uneventfully over the following 12 months.

DISCUSSION

In patients with glandular and ulceroglandular forms of tularemia suppuration and fistulization are sometimes observed and may occur despite specific antibiotic therapy.^[1-3] Ultrasound and contrast-enhanced computed tomography of the lymph nodes usually reveal a hypoechoic/hypodense pattern, necrosis, and an absence of a discernible hilum.^[4] Surgical

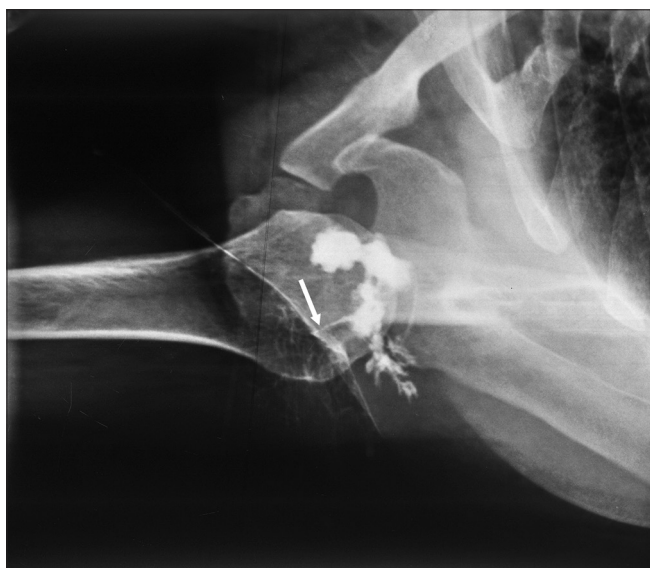


Figure 1: Fistulography of the right axilla (the white arrow points to the main external canal of the fistula)

drainage is indicated if antibiotic failure occurs.^[3] However, in our patient, surgical drainage leads to the formation of a chronic draining fistula. Radiographic fistulography helped visualize the extent of the problem and revealed that a successful surgical intervention was nearly impossible due to the impressive branching of the primary fistula [Figure 1]. More importantly, it reassured us that the fistula had no internal portal making it much less worrisome. In conclusion, radiographic fistulography can help plot the course of the fistula/fistulas and demonstrate the anatomic features of the lesion in resource-poor settings. By extension, it may also be helpful in patients with pulmonary tularemia and osteomyelitis associated with tularemia.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

Research quality and ethics statement

The authors followed applicable EQUATOR Network (<https://www.equator-network.org/>) guidelines, notably the CARE guideline, during the conduct of this report.

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Conflicts of interest

There are no conflicts of interest.

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