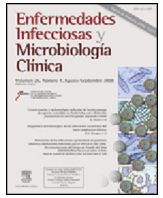




Enfermedades Infecciosas y Microbiología Clínica

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Diagnosis at first sight

Cervical tumor unusual etiology[☆]

Tumor cervical de etiología inusual

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A previously healthy 39-year-old woman visited the emergency department due to a tumor in her left cervical region which had appeared in the past 15 days and was gradually increasing in size, accompanied by pruritus and night sweats. She had no pain or dysthermia. She had received an anti-inflammatory drug and spiramycin plus metronidazole for a week with no improvement. Her prior history of note included migraine headaches and allergic rhinitis. She reported no contact with animals or consumption of raw dairy products. A physical examination revealed a fixed mass with no pain or inflammation occupying the superior 1/3 of the left half of the neck (Fig. 1). There was no lymphadenopathy on the right side of the neck or in the rest of the examination. Nasofibroscope revealed normal mucosa. It was decided to hospitalise the patient for study.

Laboratory testing revealed leukocytosis $12.2 \times 10^9 l^{-1}$, neutrophilia $9.64 \times 10^9 l^{-1}$, ESR 49 mm/h and CRP 3.39 mg/dl. Serology tests for HIV, HBV, HCV, EBV, CMV and syphilis were negative. A neck and chest CT scan showed a multinodular mass on the left side of the neck at levels IIA-IIB and III with central necrosis measuring 4 cm \times 2.8 cm (Fig. 2). A chest X-ray was normal. The patient was suspected of having disease of infectious etiology or lymphoproliferative disease. An FNAB was performed but was inconclusive due to a limited sample.

The patient was empirically treated with cefazolin 1 g/8 h plus methylprednisolone 40 mg/12 h. Following nine days of treatment with limited improvement, a biopsy was performed for study. A histopathology examination ruled out necrosis but not malignancy. A microbiological culture isolated a coccobacillus positive for Gram staining, catalase and urease and negative for pyrazinamidase and alkaline phosphatase.

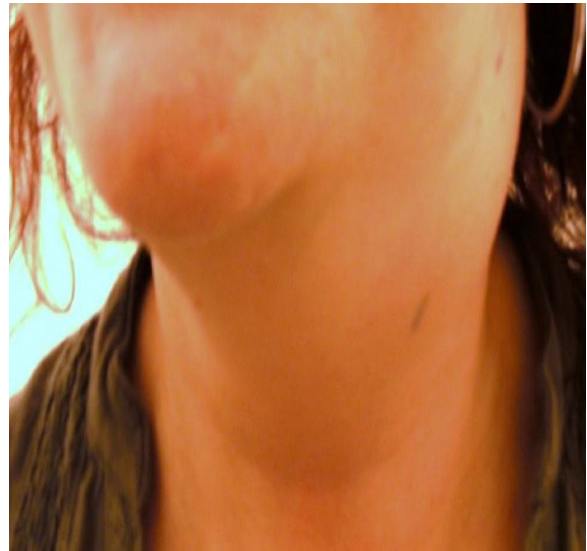


Fig. 1. Left lateral cervical tumor.

Clinical course

The bacterium was identified as *Corynebacterium pseudotuberculosis* (*C. pseudotuberculosis*) (Fig. 3) using API-Coryne (bioMérieux[®]) and confirmed using mass spectrometry (Bruker Biotyper MALDI-TOF), with a score of 2.339 at a referral hospital. An antibiogram was prepared according to EUCAST criteria. Susceptibility to penicillin, clindamycin, rifampicin, tetracycline, vancomycin, ciprofloxacin and linezolid and resistance to gentamicin were obtained.

The patient was treated with oral erythromycin. She had slow progression which required readmission after a week for parenteral treatment with clindamycin (600 mg/8 h) plus amoxicillin-clavulanic acid (1 g/8 h) for seven days. After she improved, she was discharged with oral erythromycin (500 mg/8 h) for two more weeks. Her signs and symptoms completely resolved.

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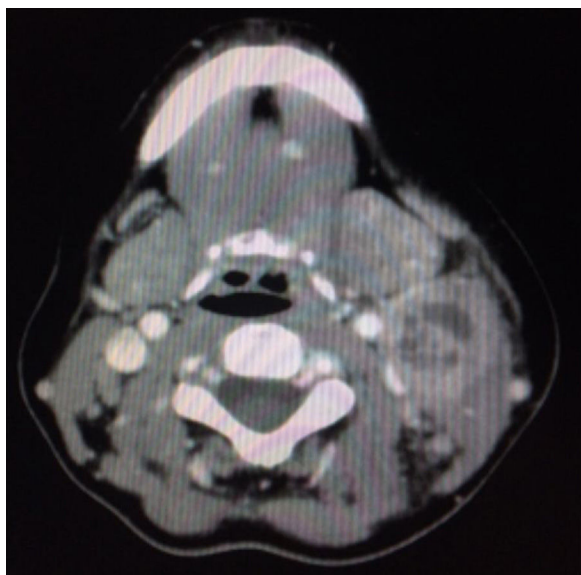


Fig. 2. CT scan. Lymphadenopathic mass, with central necrosis, in left cervical space.



Fig. 3. Isolation of *Corynebacterium pseudotuberculosis* in blood agar.

Final comments

The main causes of lymphadenitis are infectious and tumoral. Infectious causes may be acute (bacterial or viral) or chronic (mycobacteria or chlamydiae).

In isolating *C. pseudotuberculosis*, it is important to confirm species identification in order to distinguish it from corynebacteria which cause similar signs and symptoms (*Corynebacterium imitans*) and other diphtheria toxin-producing species such as *Corynebacterium diphtheriae* (*C. diphtheriae*) and *Corynebacterium ulcerans*.¹ A positive urease test presumptively distinguishes it from *C. diphtheriae*.² Unconventional techniques such as *rpoB* gene

sequencing, 16S rRNA sequencing and MALDI-TOF^{1,3} are needed for proper identification.

Lymphadenitis caused by *C. pseudotuberculosis* is an uncommon disease in humans. It mainly affects previously healthy male rural workers in contact with sheep and goat livestock.^{4–6} Transmission mainly occurs through skin lesions by means of direct contact (animals or animal products) or contact with contaminated environments.⁶ This infection has also been associated with consumption of raw dairy products from contaminated livestock, as well as inhalation of droplets containing the bacterium.^{5–7} The cervical location of the lymphadenopathy suggested oral transmission of the microorganism through raw milk from contaminated livestock.⁸ Although the patient did not report having ingested raw dairy products, we believe this to be the most likely route of acquisition as unpasteurised goat cheese is often consumed in our setting.

The most common manifestations of the disease are suppurative lymphadenopathy in different locations (axillary, inguinal and cervical), fever and/or myalgia with a chronic or subacute course.^{9,10}

The patient was treated with macrolides, as recommended by the therapeutic guidelines. Surgical lymph node drainage is sometimes needed to ensure a favourable clinical course.^{7,8} In our case, it was not needed.

We wish to emphasise the importance of identifying this uncommon etiological agent in similar clinical cases for proper treatment and resolution of signs and symptoms.

References

- Bernard K. The genus *Corynebacterium* and other medically relevant coryneform-like bacteria. *J Clin Microbiol.* 2012;50:8–3152.
- Public Health England. Identification of *Corynebacterium* species. UK Standards form Microbiology Investigations; 2014. Available from: https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/371490/ID_2i4_1.pdf
- Konrad R, Berger A, Huber I, Boschert V, Hormansdorfer S, Busch U, et al. Matrix-assisted laser desorption/ionisation time-of-flight. (MALDI-TOF) mass spectrometry as a tool for rapid diagnosis of potentially toxigenic *Corynebacterium* species in the laboratory management of diphtheria associated Bacteria. *Euro Surveill.* 2010;15, pii=19699. Available from: <http://www.eurosurveillance.org/ViewArticle.aspx?ArticleId=19699>
- Bregenzer T, Frei R, Ohnacker H, Zimmerli W. *Corynebacterium pseudotuberculosis* infection in a butcher. *Clin Microbiol Infect.* 1997;3:696–8.
- Heggelund L, Gaustad P, Håvelsrud OE, Blom J, Borgen L, Sundset A, et al. *Corynebacterium pseudotuberculosis* pneumonia in a veterinary student infected during laboratory work. *Open Forum Infect Dis.* 2015;2:1–6. Available from: <http://ofid.oxfordjournals.org/content/2/2/ofv053.full>
- Estevao Belchior SG, Gallardo AA, Abalos MA, Alvarez LA, Nuñez NC, Guevara D, et al. *Corynebacterium pseudotuberculosis*, potencial agente zoonótico. Revisión de casos (Zoonotic Potential of *Corynebacterium pseudotuberculosis*, Revision cases). *REDVET Rev Electrónica Vet [Online].* 2009;10. Available from: <http://www.veterinaria.org/revistas/redvet/n101009/100912.pdf> [accessed 11.06.16].
- Join-Lambert OF, Ouache M, Canioni D, Beretti JL, Blanche S, Berche P, et al. *Corynebacterium pseudotuberculosis* necrotizing lymphadenitis in a twelve year old patient. *Pediatr Infect Dis J.* 2006;25:484–851.
- Romero-Pérez JC, Suárez-Machado M, Batista-Díaz N. Linfadenitis por *C. pseudotuberculosis* en una paciente joven. *Rev Clin Esp.* 2004;204:338–90.
- Allen C, Goldberger MD, Benjamin A, Lipsky MD, James J, Plorde MD. Suppurative granulomatous lymphadenitis caused by *Corynebacterium ovis* (*pseudotuberculosis*). *Am J Clin Pathol.* 1981;76:486–90.
- Hémond V, Rosenstingl S, Auriault ML, Galanti MJ, Gatfosse M. Axillary lymphadenitis due to *Corynebacterium pseudotuberculosis* in a 63-year-old patient. *Med Mal Infect.* 2009;39:136–9.