



## Similar presentation of cervical lymphadenitis of different etiology in two siblings

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Here, we report chronic suppurating submandibular swelling occurring in two siblings, with one case caused by nontuberculous mycobacteria and the other caused by *Bartonella henselae*. These two infections share a similar clinical presentation, but the treatment modalities differed. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;122:e51-e54)

Nontuberculous mycobacteria (NTM) and *Bartonella* species often cause chronic cervicofacial lymphadenitis, especially in children between 1 and 5 years of age.<sup>1,2</sup> Such infections are usually unilateral and occur in the submandibular or preauricular area. The nodes eventually suppurate, forming a chronic sinus tract. Here, we describe the cases of two siblings with chronic suppurating submandibular lymphadenitis.

### CASE REPORT

The first patient was a 4-year-old girl who presented to our department with a 2-month history of a swelling of the right submandibular lymph node. She had been healthy and did not exhibit fever, fatigue, anorexia, respiratory or gastrointestinal symptoms, or weight changes. She had no history of a cat scratch, and the family had no pets at home. Hematologic findings were normal, and serologic tests were negative for cytomegalovirus, toxoplasmosis, and *Bartonella henselae*.

Evaluation of the submandibular swelling revealed skin discoloration and a fluctuant lymph node that was 2 cm in diameter (Figure 1). Ultrasonography showed an enlarged lymph node 2 cm in diameter with central necrosis located in the right submandibular angle. Mantoux skin testing was negative. Fine-needle aspiration biopsy (FNAB) of the enlarged lymph node was performed, and material was submitted to the microbiology and pathology departments. Real-time polymerase chain reaction (PCR) analysis revealed *Mycobacterium avium*. All affected submandibular lymph nodes were excised with the patient under general anaesthesia. The patient experienced good recovery and was discharged from the hospital. *M. avium* was eventually identified in the laboratory culture. No recurrence was observed during follow-up.

Four weeks after the patient's discharge, her 2-year-old brother arrived at the outpatient clinic with right submandibular swelling. The swelling had been present for

4 weeks, and the parents were concerned that he had contracted the disease from his sister. He was healthy and had no symptoms other than the submandibular swelling. Anamnesis included no recollection of a cat scratch. Physical examination revealed lymph node swelling in the right submandibular region and red, discolored skin (Figure 2). Normal results were obtained from investigations of full blood count, electrolytes, liver function tests, and cross-reactive protein. The erythrocyte sedimentation rate was slightly elevated (20). Serologic tests for cytomegalovirus, Epstein-Barr virus, herpes simplex virus, toxoplasmosis, and *B. henselae* showed no evidence of ongoing infection.

Ultrasonography revealed an enlarged hypoechoic lymph node of a 2-cm diameter. The Mantoux skin test was negative. FNAB of the enlarged lymph node was performed, and material was submitted to the microbiology and pathology departments. PCR analysis revealed *B. henselae*, and the patient was diagnosed with cat scratch disease (CSD). No antibiotics were prescribed. The patient was monitored biweekly at the outpatient clinic until resolution of the enlarged lymph node occurred after 4 months. Immunologic evaluation included complete blood count and differential, detection of interferon-gamma receptor 1, quantitative immunoglobulins, and cytokine analysis to rule out immunologic abnormalities in the two children. These analyses revealed no immunologic abnormalities.

### DISCUSSION

The report describes two cases of chronic head and neck infection occurring in two siblings. The older sister presented with a cold abscess caused by *M. avium*. Four weeks later, her younger brother presented a similar clinical picture, which made us consider that he might also have NTM lymphadenitis. However, diagnostic testing led to the diagnosis of *B. henselae* infection in the younger brother.<sup>3</sup> To our knowledge, this case is the first report of siblings with a cervical lymphadenitis caused by either an NTM species or a *Bartonella* species. Both infections are well known causes of chronic cervicofacial lymphadenitis, and as such, the infections in a brother and a sister in the presented cases were considered a coincidence. Children with abnormalities of the interleukin-12–interferon-gamma pathway or genetic disorders, such as Mendelian susceptibility to mycobacterial disease are more prone to infections.<sup>4</sup>

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Fig. 1. A 4-year-old girl with swelling of the right submandibular lymph node. Red discoloration of skin is clearly visible.



Fig. 2. The 2-year old brother with an identical clinical picture.

Immunologic evaluation, however, showed normal results for both patients described here.

Bacterial cervicofacial lymphadenitis is often caused by the *Staphylococcus* or *Streptococcus* species, and infected children commonly present with a short history of an enlarged lymph node accompanied by fever and malaise. Chronic lymphadenitis is often caused by infectious diseases, but malignancies, autoimmune diseases, and chronic inflammatory processes may also be the reason. Clinicians differentiate between malignancy and benign causes on the basis of clinical and laboratory findings.<sup>5</sup> Bacterial causes include NTM, *S. agalactiae*,

*M. tuberculosis*, *B. henselae*, *Legionella*, and *Francisella tularensis* infections.<sup>5,6</sup> Infections caused by anaerobic bacteria are associated with dental caries and periodontal disease.

In a study by Choi et al.<sup>6</sup> persistent cervical lymphadenitis in children was mainly caused by infections with NTM, accounting for 73% of cases, followed by *Bartonella* and *Legionella*, each accounting for 12% of cases.

Malignancies that present with chronic lymphadenitis may be primary lesions or metastasis. Malignant lymph nodes may enlarge in a rash or with a slow course.<sup>5</sup> Hodgkin lymphoma shows slow progress in lymph node size, but non-Hodgkin lymphoma is abrupt. Clinical manifestations that may indicate malignancy are lymph nodes that do not decrease in size, in combination with organomegaly, leukocytosis, leukopenia, high erythrocyte sedimentation rate, lactate dehydrogenase and uric acid levels.<sup>6,7</sup>

In a child with a history of an enlarged cervicofacial lymphadenitis for a period longer than 3 weeks and with negative serology for other infectious causes of chronic lymphadenitis (cytomegalovirus, Epstein-Barr virus, *Bartonella*, and *Toxoplasmosis*), purified protein derivative (PPD) skin testing and head and neck ultrasonography are recommended.<sup>8</sup> A skin reaction larger than 5 mm without a history of tuberculosis exposure or Bacillus Calmette-Guérin vaccination is strongly suggestive of NTM infection<sup>8</sup> and indicates the need for lymph node excision as the most optimal treatment for NTM lymphadenitis. FNAB is recommended to obtain material for microbiologic investigation (PCR testing for mycobacterial species, *Bartonella*, acid-fast staining, cultures) and cytology, if the PPD reaction is negative. Surgical excision is indicated if PPD testing and PCR results are negative and no alternative diagnosis can be established. Histopathology and immunohistochemistry of biopsy material can help exclude malignant causes of lymphadenopathy, and rare diseases, such as Rosai-Dorfman disease or Kikuchi disease, might be found during the analysis.

In an otherwise healthy child showing necrotizing cervicofacial lymphadenitis without systemic symptoms, NTM and *B. henselae* lymphadenitis should be considered in the differential diagnosis.<sup>1,2</sup> However, both CSD and NTM lymphadenitis remain unrecognized in many cases.<sup>1,2</sup> NTM and *B. henselae* lymphadenitis show similar clinical presentations: usually an inflammatory lymphadenopathy—typically in the submandibular or preauricular region—and a protracted course involving suppuration of lymph nodes with abscess and fistula formation. As such, the clinical features do not reliably differentiate NTM from *B. henselae* cervicofacial lymphadenitis, and the

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