

## Cervical Lymphadenitis by *Mycobacterium triplex* in an Immunocompetent Child: Case Report and Review

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**Abstract** *Mycobacterium triplex* was first described in 1996. This nontuberculous *Mycobacterium* causes a severe pulmonary disease in immunocompromised patients but it can involve also healthy patients. A literature search was made on the PubMed database and it produced only few cases of children with cervical lymphadenitis due to this *Mycobacterium Triplex*. We are describing a case of *M. triplex* cervical lymphadenitis in an immunocompetent child.

**Keywords** *Mycobacterium triplex* · Cervical lymphadenitis · Immunocompetent child

### Case report

A 4 years old boy was admitted to the Pediatric Surgery of University of Siena with cervical lymphadenopathy since about one month. Background showed a single episode of fever spontaneously regressed in about 48 h five days before the onset of enlargement of the cervical nodes. He

underwent to antibiotic therapy with amoxicillin clavulanic acid and rifampicin for 15 days, without clinical remission. For this reason was referred to our Clinic. A physical examination revealed a right swollen submandibular lymphnode (3 × 2 cm), feel rubbery, movable and painful. Overlying skin was healthy. A neck and abdomen ultrasound were performed. It was showed the presence of bilateral cervical lymphadenopathy with inflammatory aspect and no significant alterations in the abdomen. Blood exams were normal. There were no fever or others signs and symptoms as night sweats, unexplained weight loss, sore throat or difficulty in swallowing or breathing. Based on the clinical situation and according to parents a surgical excision of cervical lymph node was performed. The histopathology revealed necrotizing granulomatous lymphadenitis (Fig. 1). The cultural examination identified the presence of *Mycobacterium*'s specie. The growth and biochemical characteristics were most closely compatible with *Mycobacterium triplex* (*M. triplex*). The susceptibility to isoniazid and ethambutol was decreased by in vitro testing. This suspect was confirmed with molecular identification by PCR amplification and gene sequencing study of the 16S rRNA. Indeed, it showed that our isolate exhibited 100 % homology with the reference of *M. triplex*. The patient was discharged one day after surgery without any antimicrobial therapy. The clinical complete improvement was within 7 days. To date the patient is well without any health problems.

The incidence of infections caused by non tuberculous *Mycobacterium* (NTM) has increased in recent years [1]. They are ubiquitous organisms, commonly isolated from environmental and animal sources, whose pathogenicity may vary according to the host's immune status. Although exposure to NTM frequently causes no symptoms [2]. *M. triplex* represents a unique species of *Mycobacterium* firstly

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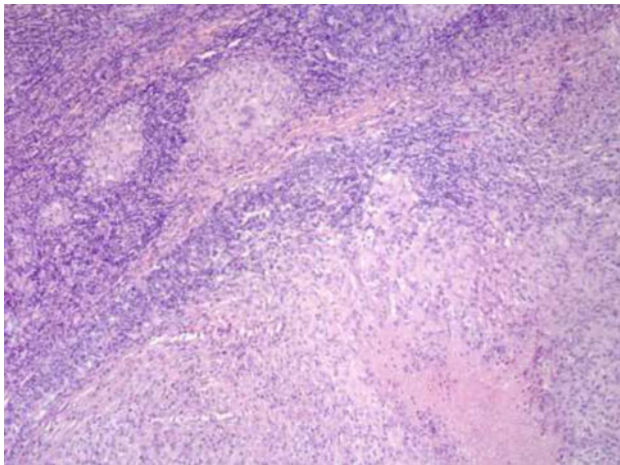
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**Fig. 1** Histopathologic features

described in 1996 by Floyd et al. [3]. The evidence is based on sequencing the 16S rRNA hypervariable region [4]. Currently the clinical relevance has not been systematically studied for *M. triplex*. A literature search was made on the PubMed database and it showed that it is more commonly associated with infections in immunocompromised adults patients. *M. triplex* has been rarely isolated from immunocompetent host and in children. Table 1 summarized the previous published cases of infection by *M. Triplex* with the main clinical features [5–12]. Our review of the Literature confirmed that the descriptions regarding lymphadenitis caused by this type of *Mycobacterium* in healthy children is very poor. To our knowledge, our patient is the second documented report of cervical lymphadenitis in a healthy child and the first in Italy. Also reviewing our

**Table 1** Published cases of human infection with *M. triplex* and related site involved

Site	Patient (age and sex) and references	Patient's immunocompetency	Clinical/instrumental manifestations	Diagnostic methods	Treatment	Outcome
Lung	54 year-old female (10)	Yes	Cough, fatigue CT: lung nodule, cavitations and bronchiectases	Culture of bronchoalveolar lavage and bronchial aspirate genetic diagnosis	Chemotherapy RMP-EMB-INH; EMB-CLA; EMB-CLA-LVX;	A&W
	54 year-old female (9)	Yes	Hemoptysis, cough, fever, fatigue CT: lung infiltrate and nodule (0.3 cm)	Culture of bronchoalveolar lavage genetic-diagnosis	Chemotherapy RMP-CLA-INH	A&W
	67 year-old male (8)	Yes	Hemoptysis CT: bronchiectases, alveolar opacities, and micronodules.	Culture of bronchial aspirate and sputum genetic-diagnosis	Chemotherapy RMP-CLA-CIP-EMB	A&W
Neck	4 year-old female (11)	Yes	Preauricular mass, submandibular adenopathy	Culture of lymph-node biopsy specimen genetic-diagnosis	Chemotherapy RMP-EMB-CLA; RFB-EMB-CLA; Surgical incision.	A&W
	4 year-old male (our case)	Yes	Submandibular lymph-node (3 × 2 cm) feel rubbery, movable and painful. no alterations of overlying skin. afebrile.	Culture of two pem lymph-node genetic-diagnosis	Surgical excision	A&W
Brain	41 year-old male (7)	Hiv-infection	Fever, cachexia, several edema of the lower limbs, diarrhea, ascite, cough.	Culture of sputum, ascitic fluid genetic diagnosis	Chemotherapy RMP-EMB-INH-CLA-PZA + Antiretroviral therapy (RITONAVIR, INDINAVIR ZIDOVUDINE AND LAMIVUDINE)	Death

**Table 1** continued

Site	Patient (age and sex) and references	Patient's immunocompetency	Clinical/instrumental manifestations	Diagnostic methods	Treatment	Outcome
Other						
Disseminated disease	40 year-old male (5)	Hiv-infection	Fever, night sweats, chills, weight loss and articular pain. MRI/CT: colliquative abscess of the left knee; spleen abscess, multiple lymphadenopathy.	Culture of joint fluid, bone and blood genetic diagnosis	Chemotherapy CLA-ETIO Antiretroviral therapy was modified to include quadruple combination of 2 new nucleoside analogues (didanosine and stavudine) and 2 protease inhibitors (ritonavir and saquinavir hard-gel capsules). Surgical drainage	Condition is slowly worsening
Pericardial and peritoneal fluid	13 year-old female (6)	Drug-induced immunodepression	Ascitis, pericarditis	Culture of pericardial and peritoneal fluid <b>GENETIC DIAGNOSIS</b>	Drainage procedure (paracentesis and pericardiocentesis)	A&W
Lymph-nodes	47 year-old male (12)	Hiv-infection	Axillary adenopathy	Culture of lymph-node biopsy specimen	Chemotherapy INH-RFP-ETB (3mo) and INH-RFP (2mo)	Death

RMP rifampicin, CLA clarithromycin, CIP ciprofloxacin, EMB ethambutol, INH isoniazid, LVX levofloxacin, ETIO ethionamide, PZA pyrazinamide, RFB rifabutin

series [13, 14], on 261 cases of cervical adenopathies we found no cases by *M. triplex*. The clinical picture of our child strongly suggested the diagnosis of nontuberculosis mycobacterial lymphadenitis and the clinical features were similar to another reported case [11]. According to the Literature we believe that the surgical excision represents the gold standard therapy in nontuberculous Mycobacterial lymphadenitis. Our documented case may suggest to clinicians and medical microbiologists to keep in mind the *M. triplex* in the etiological differential diagnosis of cervical lymphadenitis in otherwise healthy children. In any case, the differential diagnosis should be always considered thought this diagnostic improvement has only a taxonomic and epidemiological value: the clinical features and the treatment are the same as all other adenopathies caused by nontuberculous Mycobacteria. We also believe that important questions regarding epidemiology and pathogenesis of the disease caused by these organism remain today unanswered. It is possible that the few reported cases can be due to a failure diagnosis for difficult isolation and specificity in cultural examination.

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