

Caso
clinico

Case
report

An atypical deep neck infection in a two-year-old child

Un'atipica infezione profonda del collo in una bambina di due anni di età

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■ CASE DESCRIPTION

A previously healthy 2-year-old girl was admitted to our Hospital for evaluation of enlarged left retro auricular and submandibular lymph nodes of 4-5 weeks duration, preceded by few days of fever. She had not family history of tuberculosis, no contact with pet or birds and no consumption of unpasteurised milk.

The child's physical examination revealed an important lymphadenitis about 5 cm in diameter, non-tender, painless, with normal overlying skin and no systemic symptoms.

She had been previously treated with amoxicillin/clavulanate first (50 mg/kg/day in two divided doses for 5 days) and then with cefpodoxime proxetil (8 mg/kg/day in two divided doses for 5 days), without benefit.

C-reactive protein was slightly higher (15.2 mg/L; n.v. <10), white blood cells (11,170/ μ l) and antistreptolysin O titer were normal and anti-DNase Abs were negative. She had a positive tuberculin skin test (induration 11 mm x 12 mm) while QuantiFERON[®] TB Gold test was negative. The neck ultrasonography showed a partial intranodal liquefaction involving only left submandibular lymph nodes.

The chest X ray showed no pulmonary lesions. The neck contrast-enhanced axial computed to-

mography (CT) displayed a bulky latero-cervical lymphadenitis of 6 cm size, widely runny with multiple necrotic foci.

The swelling constricted the left jugular vein. The CT scan revealed also a left lateral-retropharyngeal abscess of 3.3 cm size, which determined a deviation of pharynx lateral profile (Figure 1).

The following day she underwent an incisional biopsy for diagnostic purposes: histopathological examination of the sample showed necrotizing granulomas, without malignancy.

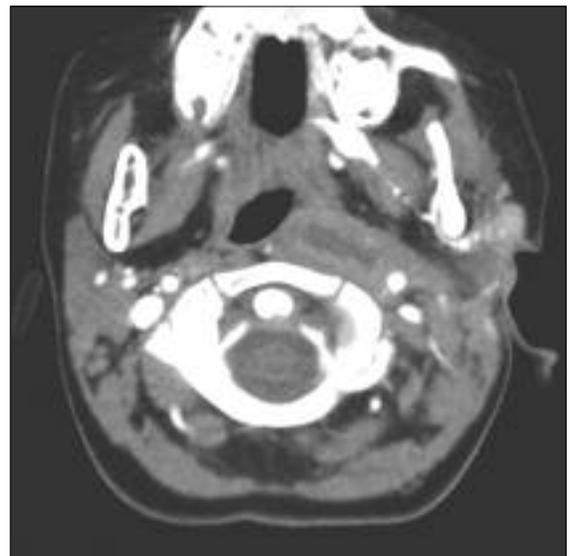


Figure 1 - Head and neck CT scan before pharmacological treatment. Axial contrast-enhanced tomogram shows a left lateral-retropharyngeal abscess.

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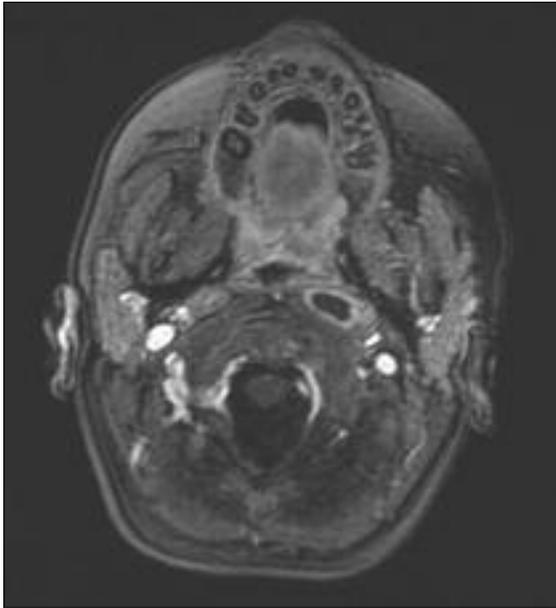


Figure 2 - Head and neck MRI after pharmacological treatment. Axial T2-weighted MRI shows the retropharyngeal abscess persistence, with size reduction.

The search for acid fast bacilli was positive and from the surgically removed material was isolated *Mycobacterium scrofulaceum*. On the basis of microbiological results, clarithromycin (125 mg twice a day) and ethambutol (250 mg once/daily) were started. Two weeks later the child underwent a complete left nodal excision. Few days after surgical intervention she presented a transient marginal mandibular nerve weakness. Antibiotic treatment was continued for six weeks. These drugs were well tolerated, without adverse events. To test visual acuity, periodic eye exams were performed during treatment with ethambutol.

Two months after drug interruption, magnetic resonance imaging (MRI) of the head and neck showed a persistence of the left lateral-retropharyngeal abscess, with size reduction to 1.86 cm (Figure 2).

The previously reported marginal mandibular nerve weakness was completely solved on follow-up. No relapse of disease or possible long-term complications were observed. The surgical wound healed completely, with normal overlying skin and good aesthetic result.

The MRI performed at the end of the follow-up, twelve months after the end of antimicrobial therapy, showed a complete spontaneous resolution of the lateral-retropharyngeal abscess.

DISCUSSION

Non-tuberculous mycobacteria (NTM) can cause a wide range of infections in children and adults.

The clinical manifestations caused by these organisms, in immunocompetent people, include lymphadenitis, pulmonary and skin/soft tissue infections. Lymphadenitis is more common in children, especially those healthy and aged 1-5 years. These are usually indolent, unilateral, involving cervical, submandibular and pre-auricular lymph nodes [1]. The diagnosis of a NTM subacute and chronic adenitis still represents a challenge for the pediatrician. The differential diagnosis includes tuberculous adenitis, infectious mononucleosis, cat-scratch-disease, brucellosis, actinomycosis, nocardiosis, toxoplasmosis, lymphoma and cystic hygroma [2]. A presumptive diagnosis could be made on the basis of clinical history, physical examination, epidemiological features (children coming from an area of low tuberculosis prevalence) and positive result of tuberculin skin testing. The latter remains the only practical and cheap skin test useful for the diagnosis of NTM adenitis, because the other skin tests with NTM-purified are no longer available on the market. Recommendations based on the literature of the 1980s considered an induration ≥ 15 mm to be more indicative of *Mycobacterium tuberculosis* infections. But recent studies demonstrate that a tuberculin skin test indurations ≥ 15 mm or ≥ 10 mm are common in children with NTM adenitis [1, 3]. This can be explained by the considerable cross-reactivity with other mycobacterial species of tuberculin test; thus sensitization to NTM can result in positive tuberculin test [4].

The newer assays based on the measurements of the release of interferon-gamma in whole blood or mononuclear cells after *in vitro* stimulation with specific *M. tuberculosis* antigens can enable us to differentiate between NTM and tubercular adenitis [1]. However the definitive diagnosis depends on the isolation and identification of the NTM causing adenitis. The material needed for recovering mycobacteria may be obtained by fine needle aspiration, incision and drainage or complete surgical excision [5]. Exact species identification can take up to eight weeks. Cultural growth of NTM pathogens from tissue specimens could be negative, even if the infection is present. Recently, positive cultures in 64-80% of cases have been obtained in some clinical laboratories, a rate much higher than previous-

ly reported [1, 2]. The use of Gen-probes and real time polymerase chain reaction has shown to be helpful in definitive diagnosis of NTM infections [6]. Sonographic findings can provide additional diagnostic clues for NTM lymphadenitis in childhood. A marked decrease of echogenicity in the early stages, with intranodal liquefaction in the advanced stages, are universal features, even though not entirely specific. CT and MRI are often used in the diagnostic pattern of NTM adenitis: they show a typical minimal inflammatory stranding of the subcutaneous fat and a lack of surrounding inflammation [1, 7]. The treatment of cervicofacial NTM lymphadenitis in children is still controversial. If surgical treatment is considered, radical exeresis rather than incision and drainage should be undertaken. The radical exeresis, in fact, allows to remove affected nodes and to obtain samples of tissue for diagnosis while incision and drainage may predispose the patient to a cutaneous fistulization [6, 8]. Pharmacological therapy containing a macrolide (azithromycin or clarithromycin) alone or in association with an antituberculous antibiotic (ethambutol or rifabutin) has been suggested [5]. Anyway, studies on wider pediatric populations, to better define the efficacy of chemotherapy versus placebo, are lacking. Lately Lindeboom et al. performed a randomized controlled trial (RCT) that showed a significantly higher success of surgical excision than antibiotic therapy with clarithromycin and rifabutin (cure rates of 96% and 66%, respectively) [9]. Another study led by Lindeboom et al. demonstrated that aesthetic outcomes of surgical intervention are more favourable than those of antibiotic therapy alone, in terms of extent of vascularization, thickness and surface of scar [10].

Our patient has also developed a retropharyngeal abscess, identified by CT images. NTM are rarely responsible of deep neck infections; to our knowledge, very few cases of NTM retropharyngeal abscess have been reported in literature so far [5, 11-13].

Concerning deep neck infections, acute constitutional symptoms, typically associated with polymicrobial abscess, are often absent in mycobacterial retropharyngeal infections [11]. Indeed, our patient has not shown any symptoms due to the presence of a deep neck infection.

In the present case the choice of treatment of the deep neck infection was towards antibiotics solely. The child was strictly monitored on clinical and radiological points of view for the onset of possible complications. This conservative choice was based on reports found in literature which support the medical treatment of deep neck infections, especially if they are not associated with septic shock, airway impairment or extension into other spaces, as in our case [14, 15]. This approach was associated with good clinical outcome and absence of complications. In conclusion, we have to consider NTM infection as a probable cause of subacute lymphadenitis in children. Tuberculin skin test and isolation of the pathogen are the better diagnostic means. The NTM adenitis therapy is still controversial. For this reason, more RCTs comparing surgical treatment versus antibiotic therapy alone, or observational-only strategy are needed. Finally, it is important to remind that it's necessary a careful monitoring of the patient with retropharyngeal abscess, considering the surgical approach in case of clinical worsening.

Keywords: Non-tuberculous mycobacteria, lymphadenitis, retropharyngeal abscess.

SUMMARY

Non-tuberculous mycobacteria are one of the major causes of lymphadenitis in children and seldom of deep neck infections. We reported the case of an immunocompetent two-year-old girl with adenitis and retropharyngeal abscess caused by an atypical mycobacterium. She had a positive tuberculin skin test, whereas the QuantiFERON® TB Gold test was negative. The child underwent a complete nodal excision. The search for acid fast bacilli was positive and *Mycobacterium scrofulaceum* was isolated from the surgically removed material. The

retropharyngeal abscess was treated only with antimicrobial therapy, which resulted in an appreciable size reduction of the abscess. After two months antimicrobial treatment was interrupted, and complete resolution was achieved after twelve months. No relapse of disease or possible long-term complications were observed. The surgical wound healed completely, with normal overlying skin and a good aesthetic result. The clinical management of atypical mycobacteria lymphadenitis and retropharyngeal abscess in children is discussed.

RIASSUNTO

I micobatteri non tubercolari sono tra i principali responsabili di linfadeniti nei bambini e, raramente, di infezioni profonde del collo. Riportiamo il caso di una bambina di 2 anni di età, immunocompetente, con adenite e ascesso retrofaringeo causati da un micobatterio atipico. Il test cutaneo alla tubercolina è risultato positivo, mentre il QuantiFERON® TB Gold test è risultato negativo. La bambina ha quindi subito un'exeresi linfonodale completa. La ricerca per bacilli alcool-acido resistenti, eseguita sul materiale rimosso durante l'intervento chirurgico, si è rivelata positiva ed è stato iso-

lato *Mycobacterium scrofulaceum*. L'ascesso retrofaringeo, sviluppato dalla nostra paziente, è stato trattato solo con terapia antibiotica, con notevole riduzione delle dimensioni dopo due mesi dalla sospensione della terapia, e con la completa risoluzione dopo dodici mesi. Non sono state osservate recidive né complicanze a lungo termine. La cicatrice chirurgica è guarita completamente, con cute sovrastante normale e buon risultato estetico. Questo articolo è utile alla discussione sulla gestione clinica, nei bambini, delle linfadeniti e degli accessi retrofaringei, provocati da micobatteri atipici.

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