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Case Report

Bilateral parotitis caused by *Mycobacterium chelonae* in an immunocompetent child

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This report is of a healthy 3-year-old boy with bilateral parotitis caused by *Mycobacterium chelonae*. He was treated with antibiotics, but the symptoms did not improve. The biopsy pathology report revealed chronic caseating granulomatous inflammation. After 2 weeks, *Mycobacterium chelonae* was identified from the biopsy specimen culture. The antibiotics were changed to amikacin and clarithromycin, according to the susceptibility test. Two weeks later, he underwent debridement surgery. Only partial excision of the infected tissue was performed because of the possibility of facial nerve injury. After another 2 weeks of treatment with amikacin and clarithromycin, parotidectomy was performed. The patient then received a 6-month course of oral clarithromycin. At the 1-year follow up, he was well and without residual mass. His immunologic examinations were all within normal limits. This is the first report of bilateral parotitis caused by *Mycobacterium chelonae* in an immunocompetent boy in the English-language literature.

Key Words: Amikacin; Clarithromycin; Debridement; Mycobacterium; *Mycobacterium chelonae*; Parotitis

Introduction

The most common pathogens associated with acute suppurative parotitis are *Staphylococcus aureus*, anaerobic bacteria, and Gram-negative organisms [1]. Non-tuberculous mycobacteria (NTM) are rare causes of parotitis [1,2]. *Mycobacterium chelonae*, a non-tuberculous mycobacteria, is classified as Runyon group IV, a rapidly growing mycobacteria. Clinical diseases caused by *M. chelonae* include lung disease, disseminated cutaneous infection, localized cellulitis, abscess, joint infections, osteomyelitis, ocular disease, surgical site infections, and catheter infections [3]. Unlike other NTM, *M. chelonae* seldom causes cervical lymphadenitis [3,4]. To date, there have been no reports of bilateral parotitis caused by *M. chelonae* in the English-language literature. This patient is the first with bilateral parotitis caused by *M. chelonae*.

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Case Report

A 3-year-old previously healthy boy presented with a fever of 40°C and left parotid gland swelling for 1 week in November 2006. He had received antibiotic treatment (cephalexin) from a local pediatrician but the fever and parotid mass were persistent. Three days later, the right parotid gland became enlarged and tender.

The patient was admitted to the Mackay Memorial Hospital, Hsinchu, Taiwan, and received intravenous (IV) oxacillin and gentamicin. At admission, the bilateral parotid glands were tender and a reddish-violet color (Fig. 1). The consistency of the bilateral parotid glands was initially hard, but the center of the masses became soft and fluctuated in size. Greenish-yellow pus was aspirated from the parotid masses. The pus culture was negative for bacterial growth.

Computed tomography (CT) scan of the head and neck region demonstrated diffuse enlargement of both parotid glands and multiple small abscesses inside the parotid glands (Fig. 2). These small abscesses had

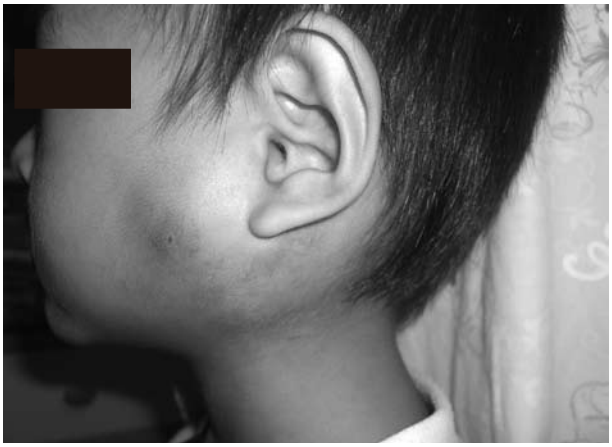

Fig. 1. Bilateral parotid masses with reddish-violet overlying skin on the tenth day of the clinical presentation.

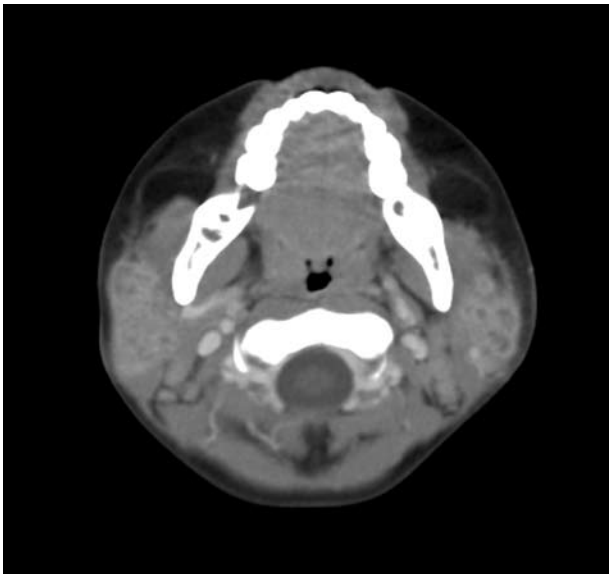

Fig. 2. Computed tomography scan of both parotid glands with enlargement and multiple small abscesses.

hypodense foci, measuring up to 2.5 cm in diameter, with rim enhancement. A biopsy was performed as the symptoms did not improve after 2 weeks of antibiotic treatment. The pathology report of the specimen indicated chronic caseating granulomatous inflammation. A few acid-fast bacilli were identified. The antibiotics were changed to isoniazid, ethambutol, and rifampin, due to the suspicion of *Mycobacterium tuberculosis* infection. After 2 weeks, *M. chelonae* was isolated from the biopsy specimen culture. The pathogen was identified as *M. chelonae* based on the rate of growth on conventional solid media of less than 1 week; the colony morphology type was rough; the colony pigmentation was non-photochromogen; and the results of biochemical tests of nitrate reduction were negative

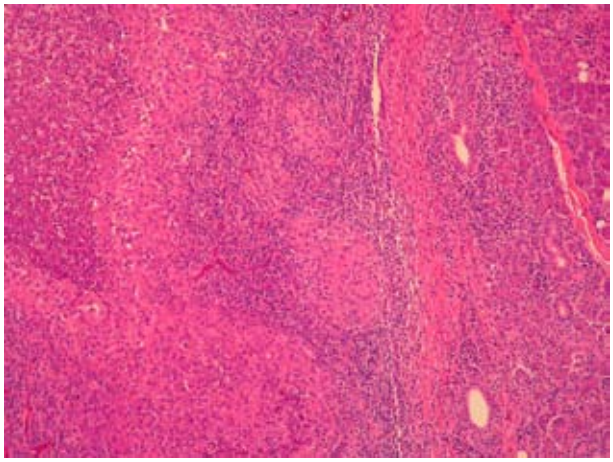

Fig. 3. Histology of the parotid gland revealing chronic caseating granulomatous inflammation (hematoxylin and eosin stain; original magnification, × 60).

and the arysulfatase 3-day test was positive. Susceptibility tests showed that the isolate was susceptible to amikacin, clarithromycin, tobramycin, imipenem, and cefoxitin, but was resistant to doxycycline, trimethoprim-sulfamethoxazole, and ciprofloxacin. IV amikacin and oral clarithromycin were prescribed, according to the susceptibility test.

The patient's symptoms of fever and parotid masses with pus drainage from the biopsy wound were persistent despite amikacin and clarithromycin antibiotic therapy for 2 weeks. Debridement surgery was performed. The proximity of the facial nerve to the parotid glands meant that only partial parotidectomy and debridement surgery could be performed. The biopsy specimen showed chronic caseating granulomatous inflammation involving the parotid gland and adjacent soft tissue (Fig. 3). Amikacin and clarithromycin were continued after surgery. After 2 weeks, when the inflammation of the parotid masses and surgical wound had decreased, a second parotidectomy was performed. As much of the infected tissue as possible was removed. After the second parotidectomy, clarithromycin was administrated for 6 months. At the 1-year follow-up after discontinuation of clarithromycin, the boy was well and without residual sequelae or facial nerve injury.

Tuberculin skin test was negative. The results of immunological examinations, including lymphocyte markers, immunoglobulin levels, total hemolytic complement (CH50) test, and polymorphonuclear leukocyte functions of bacterial killing, phagocytosis, and chemotaxis, were all within normal limits. His serum human immunodeficiency titer was negative.

According to these immunological examinations and the boy's past medical and follow-up clinical history, the patient was considered to be immunocompetent.

Discussion

There are only a few reports describing parotitis caused by NTM infections [5-13]. This patient had bilateral *M. chelonae* parotitis that was treated successfully. *M. chelonae* was first identified in 1903, when Friedman isolated an acid-fast bacillus from the sea turtle Chelonae corsicata [14]. *M. chelonae* is a ubiquitous organism, and can be found in soil and water worldwide [15]. Young children have a tendency to put objects contaminated with soil, dust, or standing water into their mouths. Pathogens may invade the parotid gland via the salivary duct, or enter the body through oral mucosal breaks caused by teething or pharyngeal trauma [4,5]. This patient was a 3-year-old boy and it is likely that *M. chelonae* entered his parotid glands via one of these routes.

M. chelonae can cause various clinical syndromes, including disseminated disease. Disseminated disease occurs almost exclusively in immunodeficient patients, especially those with acquired immunodeficiency syndrome [3]. Primary and secondary immunodeficiencies that result in impaired T-cell function or macrophage activation result in an increased risk for mycobacterial infection [16,17]. This patient was immunocompetent with bilateral *M. chelonae* parotitis, because he did not have a history of severe infectious disease and he became well after discontinuation of clarithromycin for more than 1 year. His immunological study results were all within normal limits.

The diagnosis of NTM infection relies on histopathologic findings or culture of the organism. Microabscesses, ill-defined granuloma, non-caseating granuloma, and scarce giant cells are likely to be present in lymph nodes infected with NTM. Caseous necrosis may be present in advanced lesions. In this patient, the pathologic finding was chronic caseating granulomatous inflammation. However, the definitive pathogen was found by culture of the specimen. Tuberculin skin testing with *M. tuberculosis* purified protein derivative may elicit small reactions with some NTM [18]. In this patient, the tuberculin skin test was negative.

Surgery is the principle therapy for NTM parotitis. Coulter et al reported 98 patients with NTM adenitis during a 14-year period, 28 (29.5%) of whom required multiple operations [19]. Disease of the preauricular nodes and the parotid glands cause particular problems due to the involvement of the facial nerve, and total parotidectomy cannot always be achieved. Early research suggested the use of preoperative antibiotic treatment for NTM lymphadenitis, including parotitis [20,21]. After primary surgery, if there is continued discharge with active inflammation, postoperative administration of chemotherapy should be considered before further surgery is undertaken [19]. *M. chelonae* is one of the most antibiotic-resistant species of the rapidly-growing mycobacteria. Clarithromycin is thought to be the most effective bactericidal drug, and many reports have confirmed that clarithromycin has been effective in *M. chelonae* infections [18]. In Taiwan, according to a report by Yang et al in 2003, 48% of *M. chelonae* isolates were susceptible to clarithromycin, and 82% were susceptible to linezolid [22]. Activities of cefoxitin, doxycycline, trimethoprim-sulfamethoxazole, and ciprofloxacin against *M. chelonae* isolates are poor. All rapidly-growing mycobacteria species are usually susceptible to amikacin [18,21-22]. Multidrug regimens containing amikacin are usually recommended for therapy of infections caused by all rapidly-growing mycobacteria species including *M. chelonae* [22]. In this patient, amikacin was selected with clarithromycin on the basis of the susceptibility test of the organism. As bilateral parotitis caused by *M. chelonae* is an uncommon presentation, clarithromycin was prescribed to this patient for 6 months after surgery. The results of the treatment were good and there was no recurrence after 1 year of follow-up.

This is the first report of bilateral *M. chelonae* parotitis. Surgical excision of the lesion is the best treatment. However, if complete excision is not possible, amikacin with clarithromycin chemotherapy for inflammation control before complete excision should be considered. A 6-month course of clarithromycin chemotherapy can prevent recurrence if there are residual organisms after surgery.

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