Salivary Gland Nocardiosis in an Immunocompetent **Patient**

Prashanth V. Shetty, Odaiyappan Kannappan and Yashdeep Sinha Sarma, Department of General Surgery, Kasturba Medical College, Manipal, Karnataka, India.

We report a case of nocardiosis in an immunocompetent patient who presented with pain and multiple swellings in the face. Nocardia asteroides was isolated from the parotid and submandibular salivary glands. The patient was successfully treated by surgical drainage and oral administration of trimethoprim and sulfamethoxazole. To the best of our knowledge, this is the first reported case from India on N. asteroides affecting the salivary gland. [Asian J Surg 2011;34(2):99-101]

Key Words: Nocardia asteroides, salivary gland abscess

Introduction

The Nocardia species are aerobic, branching filamentous, gram-positive, weakly acid-fast soil saprophytes belonging to the "aerobic nocardioform actinomycetes" subgroup. They cause pulmonary, central nervous system, cutaneous, and subcutaneous infections. They are common in immunocompromised hosts.² Cutaneous nocardiosis can be subdivided into four clinical types: mycetoma, lymphocutaneous infection, superficial skin infection (abscess or cellulitis), and disseminated disease with cutaneous involvement. Primary skin and subcutaneous lesions are rare manifestations of this infection. We herein report a case of multiple salivary gland abscesses caused by Nocardia asteroides.

Case report

A 33-year-old female was admitted to our hospital with complaints of pain followed by swelling below the right ear lobe and on either side of the lower jaw. She had a fever and difficulty opening the mouth. She also had a seizure disorder but stopped taking phenobarbitone 5 years prior because there was no seizure recurrence. She was not a diabetic and was not on any immunosuppressive drugs. She used broomsticks, which are used for cleaning the floor, as toothpicks after meals.

Physical examination showed swelling in the right parotid and bilateral submandibular regions with classic signs of inflammation, fluctuant with pus pointing (Figure 1). Laboratory investigations showed neutrophilic leucocytosis. Serology results for HIV-1 and -2 were negative. Chest X-ray and brain computed tomography results were normal. Based on the results of clinical and laboratory investigations, a provisional diagnosis of multiple abscesses was made. The abscesses were incised and drained. Endovenous therapy with amoxicillin/clavulanic acid at 6.6 g per day was started. A gram-stained smear of purulent discharge showed variable filamentous organisms. A modified Ziehl-Neelsen stain showed acid-fast organisms that resembled Nocardia. Culture on nutrient agar and biochemical test results identified the isolate as *N. asteroides*. A biopsy confirmed the diagnosis of abscess. Antibiotic sensitivity testing of the isolate revealed sensitivity to amoxicillin/clavulanic acid, co-trimoxazole, doxycycline, etc. The patient was started on co-trimoxazole in spite of sensitivity to amoxicillin/clavulanic acid for a better response and to prevent relapse. At the 6-month follow-up, the wound had healed well with no facial nerve deficit (Figure 2).

Address correspondence and reprint requests to Dr Prashanth V. Shetty, Department of General Surgery, Kasturba Medical College, Manipal, Karnataka, India.

E-mail: drprashanthvshetty@yahoo.co.in • Received: Jul 5, 2010 • Revised: Dec 24, 2010 • Accepted: Apr 14, 2011

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Figure 1. Abscess at the parotid and submandibular region preoperatively.



Figure 2. Areas showing healed abscess cavity after 6 months of treatment with co-trimoxazole.

Discussion

Actinomyces are rare bacterial pathogens associated with suppurative salivary glands.³ Most reported cases involve sporotrichoid forms on the limbs of otherwise healthy patients. N. asteroides, a less pathogenic and true opportunistic agent, is an exceptional causative agent in immunocompetent patients. Nocardia species are ubiquitous environmental saprophytes. Human infections arise from direct inoculation of the skin or soft tissues or by inhalation. Immune responses to Nocardia species are primarily T cell-mediated. *Nocardia* is more problematic in patients with impaired cell-mediated immunity. 4 N. asteroides is relatively resistant to neutrophil-mediated killing. It inhibits phagosome-lysosome fusion, giving rise to L-forms. Cell wall-deficient forms (L-forms) of Nocardia species have been isolated from serious human infections. This may explain the late relapse of *Nocardia* infections.⁵ Retrograde contamination of the salivary ducts and parenchymal tissues by bacteria that reside in the oral cavity may account for the mechanism of infection.³ Ultrasound scans should be performed in all patients with an acute swelling of the parotid region irrespective of age, number of leucocytes, or typical fluctuation.⁶ Needle aspiration of the purulent gland may yield the causative organism. If no pus is aspirated, introduction of sterile saline and subsequent aspiration may yield organisms. Specimens should not be taken from Stensen's duct because of the potential for oropharyngeal contamination.3 Nocardia will grow on most non-selective media used routinely for culture. The yield is increased by the use of selective media, such as Thayer-Martin agar, paraffin agar, or buffered charcoal-yeast extract medium.⁷ Polymerase chain reaction, restriction fragment length polymorphism analyses, and DNA sequencing have enabled rapid and accurate species identification of *Nocardia*. Patients benefit from aggressive surgical debridement (incision and drainage).⁸ Surgical drainage of a parotid abscess should be performed under myoelectric monitoring. In previous case reports, oral minocycline¹ or intravenous co-trimoxazole (sulphamethoxazole/trimethoprim, 400/80 mg) twice a day followed by doxycycline 100 mg twice a day² were used. We used co-trimoxazole twice a day for 6 months. In adults with normal renal function and localised disease, the recommended dose of trimethoprim-sulfamethoxazole is 5–10 mg/kg trimethoprim and 25–50 mg/kg sulfamethoxazole in 2–4 divided doses.⁹

Non-immunosuppressed patients with pulmonary or systemic nocardiosis should be treated for at least 6 months, and those with central nervous system involvement for 12 months. ¹⁰ Amikacin, imipenem, minocycline, and linezolid can also be used.

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