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Case Report

Actinomycosis of the Salivary Gland

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Abstract

Actinomycetes are saprophytic bacteria of the oral cavity. They can cause a rare, chronic and suppurative reaction of the teeth and mandible, and then involve the cervical region. We present a case of salivary gland actinomycosis mimicking a neoplasm. The patient underwent surgical excision and pathology confirmed the diagnosis of actinomycosis. He took an oral form of amoxicillin for 2 months after the operation. At the 1-year follow-up, there was no evidence of recurrence. Primary actinomycosis of the salivary gland is very rare and can mimic a neoplasm both clinically and radiologically. Successful treatment relies on adequate surgical drainage or excision and prolonged antibiotic therapy. (*Tzu Chi Med J* 2008;20(3):218–220)

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1. Introduction

Cervicofacial actinomycosis is a rare disease that is caused by an inhabitant of the normal oral flora (1). It can be found as a saprophyte in the oral cavity and has not been isolated *in vitro* (2). Actinomycosis was first reported in humans by Von Lanfenbeck in 1845 and was thought to be a fungus. Waksman showed that *Actinomyces* was an anaerobic Gram-positive bacterium in the 1960s (3). Five species of *Actinomyces* have been identified, and the most common in humans is *Actinomyces israelii* (1,3).

Most major medical centers report an average of one actinomycosis case per year (2). The incidence of actinomycosis in all submitted specimens ranges from 0.1% (4) to 0.37% (5). There is a male predominance (ranging from 1.5:1 to 3:1) reported in many series and the fifth decade of life is the most affected (2,3). The clinical symptoms depend on the location of the disease. The common locations of actinomycosis are: cervicofacial (40–60%), abdominopelvic (20%), thoracic (15%) and central nervous system (2%) (1). The facial skeleton is often affected (1).

The main causes of actinomycosis include dental infection, manipulations and oromaxillofacial trauma (1–3). Disease occurs almost exclusively by direct invasion, and rarely by metastatic or hematogenous spread (2). An immunosuppressed state, diabetes and malignancy are predisposing factors (1–3,6). The clinical manifestations include two forms: one is chronic, progressive and indolent with indurated infiltration, and multiple abscess and fistulas; the other

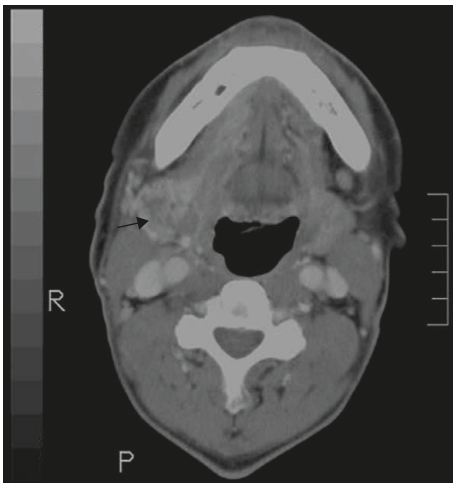

Fig. 1 — Neck computed tomography with contrast medium enhancement shows a heterogeneous mass (arrow) over the right submandibular region.

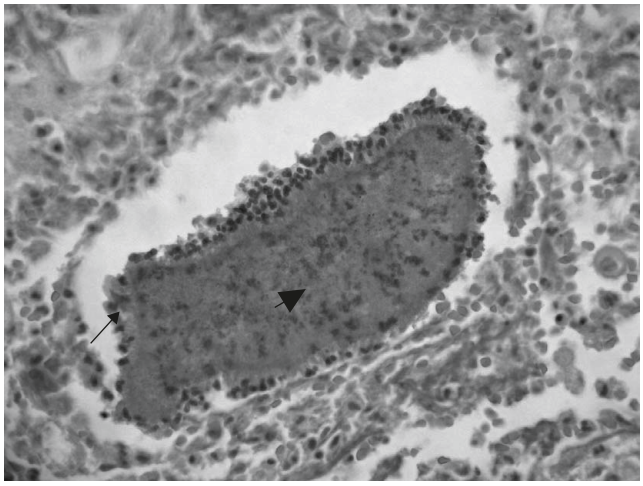

Fig. 2 — *Actinomyces* (arrowhead) and sulfur granules (arrow) (hematoxylin & eosin, 400×). A large ball-like cluster of *Actinomyces* with filaments or sulfur granules (located at the edges) and aggregates (central location) in a background of acute inflammation.

is more acute and rapidly progressive, with fever and a sore, fluctuating swelling that cause it to resemble a typical pyogenic infection [1].

Imaging studies, such as computed tomography (CT) or magnetic resonance imaging (MRI), help to define the extent of the disease. Definite diagnosis relies on positive cultures and biopsy showing the bacteria. The propensity of this disease to mimic carcinoma or tuberculosis is well known [1–3,6–9]. Chiang et al [7] reported a nasopharyngeal actinomycosis case imitating nasopharyngeal carcinoma. It must be considered as the great masquerader of head and neck disease [2].

2. Case report

A 62-year-old male patient visited our Ear, Nose and Throat (ENT) Outpatient Department with a history of right submandibular swelling for about 10 years. The mass had enlarged rapidly in recent days. Examination of the head and neck showed a firm, fixed and tender mass over the right submandibular area. The size of the mass was 3×3 cm, and did not fluctuate in size after eating. The patient also had Addison's disease that had been treated with cortisone for 3 years, and his oral hygiene was poor.

Imaging study (CT scan with contrast medium enhancement) was performed and showed a 3×3 cm heterogeneous mass over the right submandibular area (Fig. 1). Laboratory studies, including complete blood count and biochemistry profile, were all within normal limits. The patient underwent surgical excision and pathology revealed *Actinomyces* flora with sulfur granules on hematoxylin and eosin staining (Fig. 2). After the operation, the patient was prescribed an oral form of amoxicillin 500 mg to be taken 3 times daily for 2 months. At the 1-year follow-up, the patient was healthy and there was no sign of recurrence.

3. Discussion

Actinomycosis is a chronic and suppurative infection that can occur in the head and neck region. Initial presentation is usually a palpable mass and it is sometimes accompanied by surrounding skin erythematous changes. The clinical diagnosis is difficult to establish because the onset and duration of the disease are not specific, and the differential diagnosis should cover a wide range of diseases from tumor to chronic infections such as tuberculosis [3].

The imaging modalities cited in the literature are CT, sialography, radionuclide imaging, ultrasonography (US) and MRI [1]. CT is more effective at defining the extent of the abscess and the surrounding anatomical structures [1]. Usually, actinomycosis appears as an ill-defined soft tissue mass with rim enhancement on CT, and the radiological differential diagnoses include branchial cleft cyst, lymph node metastasis, inflammatory lymphadenopathy and vascular tumor [3]. MRI is sometimes helpful in defining the relation between the inflammatory process and cervical soft tissues [3].

Actinomyces is noted for forming characteristic sulfur granules in infected tissue but not *in vitro* [2,3,8,9]. The term *sulfur granule* has been a misnomer for a long time; the name actually reflects the yellow color of the granules in pus as the granules

contain no sulfur at all [2]. They are composed of an internal tangle of mycelial fragments and a rosette of peripheral clubs. In our case, we saw the typical findings of *Actinomyces* and sulfur granules with hematoxylin and eosin staining (Fig. 2).

Definite diagnosis of actinomycosis relies on positive cultures and histopathology [2,3]. Therefore, fine needle aspiration cytology or open biopsy for pathology should be performed to make the diagnosis.

Since the advent of antibiotic therapy, the principles for management of cervicofacial actinomycosis have emphasized a prolonged treatment schedule coupled with surgical management if necessary. High-dose penicillin remains the treatment of choice [2]. A 2-month course of oral penicillin V, 2–4 g/day in four divided doses, or oral amoxicillin, 500 mg three times daily, are equally efficacious [2]. Surgical intervention is recommended for excision of necrotic tissue and curettage of affected bony tissue [10]. The duration of therapy should be related to the severity of the disease and ongoing assessment for clinical or pathological remission. Intensive, prolonged penicillin therapy (12–18 months) and surgery were described by Harvey et al in 1957 [11]. Alternatives to penicillin include doxycycline, ceftriaxone, clindamycin, erythromycin and chloramphenicol [12].

There are no specific methods to prevent cervicofacial actinomycosis. Maintenance of good oral hygiene and appropriate plaque removal can limit the tendency of *Actinomyces* to establish dense colonization and subclinical infection.

Actinomycosis should be considered when patients present with cervicofacial swelling.

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