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CASE REPORT

Actinomycosis osteomyelitis of the jaws: Report of four cases and a review of the literature

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Abstract Actinomycosis osteomyelitis of the jaw bones, particularly in the maxilla, is an extremely rare disease. This report presents two cases of maxillary and two cases of mandibular actinomycosis osteomyelitis, with the diagnosis particularly based on histological procedures. The highly diversified pathogenicity of the phenomenon and the absence of solid diagnostic criteria are discussed. Laboratory challenges are emphasized, and a comprehensive overview of the entity including treatment alternatives is given along with a review of the relevant literature.

Introduction

Actinomycosis of the jaws is a relatively uncommon infection that produces abscesses and open draining sinuses. The principle cause of cervicofacial actinomycosis is *Actinomyces israelii*. However, *Actinomyces naeslundii*, *Actinomyces viscosus*, and *Actinomyces odontolyticus* are occasionally identified. *Actinomyces* produces chronic, slowly developing infections, particularly when normal mucosal barriers are disrupted by trauma, surgery, or a preceding infection.[1] A break in the integrity of the mucous membranes and the presence of devitalized tissue can result in invasion of the deeper body structures and cause illness.[2]

Actinomyces strains resemble both bacteria and fungi, thus, they were often considered to be transitional

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between the two groups of microorganisms. However, most of the fundamental characteristics of *Actinomyces* indicate that they are, in fact, bacteria. They are anaerobic or facultative in contrast to pathogenic fungi, which are uniformly aerobic. In addition, *Actinomyces* does not contain sterols in its cell walls, and is sensitive to antibacterial chemotherapeutic agents.[3]

Actinomycosis is generally a polymicrobial infection requiring the presence of companion bacteria, most frequently anaerobic streptococci, fusiform or Gram-negative bacilli, and *Haemophilus* species. The associated flora form a kind of symbiosis with *Actinomyces* species and may cause an anaerobic environment which furthers the growth of this species.[4] Hence, these associated bacterial species act as copathogens and participate in the production of infection by elaborating a toxin or enzyme or by inhibiting host defenses. Furthermore, these accompanying species enhance the relatively low invasive power of *Actinomyces* by eliciting early manifestations of the infection and by treatment failure.[4–6]

Involvement of bone is rare, but osteomyelitis sporadically occurs, secondary to the primary infection at primary sites. The infection progresses by direct extension into adjacent tissues. Unlike other infections, actinomycosis does not follow the usual anatomical planes but rather burrows through them and becomes a lobular "pseudotumor".[7]

The purpose of this report was to present four cases of *Actinomyces* osteomyelitis and review the possible pathogenesis of the disease along with the outcomes after proper treatment modalities. A review of the literature on clinical sites, diagnostic methods, and treatment procedures is also included.

Case reports

Case 1

A 37-year-old woman was admitted to the Department of Oral Surgery Clinic in May 2002. The patient's medical history was noncontributory. Root-canal treatment had been completed in her left upper first premolar 2 years previously. This was followed by progressively increasing swelling in the oral vestibular region adjacent to the tooth. The swelling also mildly involved the left buccal area. She also described a continuous pain in her tooth. The patient was empirically treated by her dentist with oral administration of ampicillin/sulbactam (50 mg/kg), but after 1 week of gradual and partial recovery, the swelling returned. Because her pain was still present, she asked for her tooth to be extracted and the extraction was performed 2 months prior to her admission to the Department of Oral Surgery Clinic. The patient reported that she had been prescribed oral spiramycine for 20 days after the extraction, but the treatment failed to resolve her pain and swelling. She described a sense of "itching" on her cheek, and an ongoing sensation of pressure and intermittent discomfort around the tooth. She also described pain in the neighboring molar tooth with a gross amalgam restoration (Fig. 1B). Clinical examination revealed an unhealed tooth socket. The color of the adjacent gingiva showed slight erythema resembling desquamative gingivitis (Fig. 1B). Additionally, exposed sequestra were present. On an X-ray examination, the maxillary bone adjacent to the tooth socket showed destruction of the alveolar bone (Fig. 1A). As a result of the clinical and radiological findings, the patient underwent surgical intervention with a preliminary clinical diagnosis of actinomycosis infection.

Local infiltration anesthesia was induced, a full mucoperiosteal flap was elevated, and the defect was curetted. The right maxillary first molar adjacent to the area of the lesion was extracted due to the extensiveness of the lesion and complaints of the patient. Curetted tissue from the surgical site was submitted for histopathological and microbiological examination [Fig. 1A(i)]. In hematoxylin-and-eosin (H&E)-stained sections, fragments of bone and granulation tissue were observed. Hard-tissue specimens included trabeculae of woven bone enclosing marrow tissue and a number of partly resorbed bony sequestra with extensive involvement of microorganisms. For the histological differential diagnosis, sections were also stained with tissue Gram, Giemsa, periodic acid Schiff (PAS), Gomori methenamine silver (GMS), and Ziehl Neelsen stains. The histological appearances were consistent with those of osteomyelitis in association with infection by *Actinomyces* organisms (Fig. 2A and B).

Culture of the involved tissue did not demonstrate the presence of *Actinomyces*. Similarly, no *Candida* colonies were observed. However, cultures were positive for Gram-positive microorganisms that are common inhabitants of the oral cavity.

Case 2

A 24-year-old otherwise healthy man was referred to the Department of Oral Surgery Faculty Clinic because of an unhealed extraction socket in the region of the lower left first molar that had been extracted 2 years prior to referral. The tooth had been asymptomatic, and the patient could not recall ever experiencing pain. However, on clinical examination, the socket seemed like a freshly extracted one [Fig. 1B(i)]. A panoramic radiograph of the area revealed ill-defined bony changes with osteolytic and osteosclerotic areas (Fig. 1B). There was no soft-tissue involvement.

Actinomycosis was suspected in accordance with clinical and radiological findings; therefore, the patient underwent surgical intervention with a preliminary clinical diagnosis of actinomycosis. Necrotic tissue curetted during the surgical intervention [Fig. 1B(ii)] was submitted for histopathological evaluation, and sections were primarily stained with H&E, and then similar staining procedures were performed with each of the stains used in Case 1. On histopathological examination, trabeculae of necrotic woven bone enclosing *Actinomyces* granules with bone marrow and a number of partially resorbed bony sequestra, nonspecific inflammatory cell infiltrates, vascular proliferations, and granulation tissue were seen. Within the granulation tissue were granules surrounded by polymorphonuclear leukocytes (Fig. 2B). The periphery of the *Actinomyces* granules showed radiating, basophilic filaments and eosinophilic, club-shaped ends (Fig. 2B and C). However, culture of the tissue was