Actinomycotic Osteomyelitis of Palate Masquerading Periapical Pathology: A Rare Case Report

Abstract
Osteomyelitis is an infection that is challenging to manage due to the poor vascularization of bone that favors the proliferation of microorganisms. We report a case of osteomyelitis occurring in endodontically treated teeth in the maxillary palatal region. Clinically and radiographically, it was initially diagnosed as osteomyelitis and was treated accordingly with antibiotics for 1 year with no reported healing. Later, biopsy was done and the findings were consistent with that of chronic osteomyelitis in association with infection by Actinomyces organisms. Thus, the case highlights the rare occurrence of actinomycotic osteomyelitis in maxilla and the importance of biopsy and histopathology which will help in correct diagnosis and rapid resolution through appropriate antibiotic therapy.

Keywords: Actinomycotic osteomyelitis, histopathology

Introduction
Osteomyelitis is an acute or chronic inflammatory process in the medullary spaces or cortical surfaces of bone that extends away from the initial site of involvement. The vast majority of osteomyelitis cases are caused by bacterial infections and result in an expanding lytic destruction of the involved bone with suppuration and sequestra formation. 

Osteomyelitis is a polymicrobial infection which can be induced by commensals present in the oral cavity associated with certain predisposing factors. One such inhabitant of the normal flora which can cause osteomyelitis is Actinomyces.

Actinomycosis is a rare saprophytic infection that is characterized by granulomatous and suppurative lesions. This infection typically spreads contiguously, frequently ignoring tissue planes and surrounding tissues or organs, ultimately producing multiple sinus tracts. It primarily affects soft tissues; osseous involvement occurs rarely. Actinomycotic osteomyelitis of maxilla is relatively rare when compared to mandible, probably because of better circulation in maxilla which provides increased oxygen supply.

This unusual case shows chronic actinomycotic osteomyelitis of maxilla with suppurative and sclerosing features which presented as a periapical infection in endodontically treated teeth.

Case Report
An 18-year-old male of moderate build reported with pain and discharge from the upper left back region of the jaw for 1 year. Medical and family history was insignificant. The patient had a habit of chewing tobacco around 8–10 times a day for 5 years. Generalized gingivitis and pocket associated in relation between 26 and 27 was present. History of the present illness revealed that the lesion started 1 year back and was gradually increasing in size. The present lesion was solitary, nonhealing ulcer measuring around 2 cm × 3 cm seen on the left side of the palate associated with 26 and 27. On inspection, it was observed that edges of the ulcer were sloping and the floor appeared to be filled with slough. On palpation, the swelling around the ulcer was firm and tender (Figures 1 and 2).

The patient was advised for radiographic examination and routine blood examination. Radiographic examination revealed radiolucency associated with endodontically treated 26 and 27. Well-defined area of radiolucency with cortical lining of dense bone was noticed along with sparse opacities separated by sclerotic

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bone, suggestive of sequestrum. Trabecular pattern of the involved area appeared to have poor density with fuzzy appearance. Hematological investigations were within normal limits [Figure 3].

On the basis of clinical and radiographic examination, provisional diagnoses were made as osteomyelitis in the region of upper left molars.

An informed consent was taken from the patient, and surgical excision of the lesion was done. The surgical procedure was performed under local anesthesia. Excisional biopsy and curettage were done and the defect was primarily closed by buccal pad fat [Figures 4 and 5].

Excised specimen was sent for histopathological examination.

H and E stained section revealed a stratified squamous epithelium with large clumps of bacterial colonies exhibiting central basophilic area and peripheral eosinophilic rods giving sunray appearance suggestive of actinomycotic colonies [Figure 6]. Decalcified section of bone showed bony trabeculae without osteocytes in lacunae [Figure 7].

The presence of actinomycotic colonies was further confirmed by use of a special stain Grocott methenamine silver which stained the organisms as black to brown in color [Figures 8 and 9].

Based on hematoxylin and eosin and Grocott methenamine silver staining, a final diagnosis of actinomycotic osteomyelitis was given.

**Discussion**

Actinomycosis is a chronic granulomatous infection caused by *Actinomyces* species which may involve only soft tissue or bone or the two together. *Actinomyces* are filamentous bacteria which resemble fungi. They are slow growing Gram-positive, nonacid fast, anaerobic, or microaerophilic bacteria.[5]

Von Langenbeck noted the first case of Human Actinomycosis in 1845 and attributed it to a fungus. Bollinger described the organism *Actinomyces bovis* and its ability to cause “lumpy jaw” in cattle. The word *Actinomyces* means “ray fungus,” and reflects the general belief at the time that the organism was a fungus. The organism was first isolated from humans in 1891 when Wolff and Israel reported culturing it anaerobically and growing only at body temperature. Actinomycosis is a Greek word comprising of “Aktino” meaning radiating appearance of sulfur granules and “mykos” which labels the condition as mycotic disease.

Actinomycetes like fungi form a mycelia network of branching filaments, but like bacteria, they are thin, possess cell walls containing muramic acid and are susceptible to antibacterial antibiotics. Thus, in the 1960s, Waksman concluded that *Actinomyces* was actually a Gram-positive bacteria.[6]

The most frequent clinical form of actinomycosis is the cervicofacial, abdominal, and pulmonary.

Cervicofacial actinomycosis classically involves the soft tissues of the jaws or neck region and characteristically
presents as brawny swelling which if untreated discharges through multiple sinuses on to the skin surface. The skin overlying the abscess is purplish red, indurated and has the feel of wood. It is common for the sinus through which the abscess has drained to heal but because of the chronicity of the disease new abscess develop and perforate the skin surface. Thus, the patient over a period of time shows extensive scarring and disfigurement. The infection of the soft tissues may extend to involve the mandible or
less commonly the maxilla which results in actinomycotic Osteomyelitis.

Primary actinomycotic osteomyelitis is rare, corresponding to about 12% of cases. It affects the cervicofacial region, typically the body of the mandible, followed by the region of the chin and angle of the mandible, but rarely affecting the upper jaw or temporomandibular joint. Actinomycosis in the maxilla accounts for only 0.5%–9% of all head and neck cases.

It is assumed that the mandibular predominance of the disease stems from the relatively poor vascualrization of the condensed cortical bone with a similar mechanism that predisposes it to osteoradionecrosis. Actinomycotic osteomyelitis of maxillary origin is extremely rare compared to mandibular actinomycotic osteomyelitis, probably because of the good blood supply which provides more oxygen and better circulation.

Infection tends to spread along planes of least resistance from the supporting structures of the affected tooth to various potential spaces in the vicinity. Thus, infection perforates bone where it is thinnest and weakest. In the maxilla, bone is weakest on the buccal aspect throughout and relatively thicker on the palatal aspect. In our case, the ulcer was present on the palatal aspect. The most common source of infection must be from the involvement of palatal root of the endodontically treated maxillary molar tooth. The presence of infection on palatal aspect itself suggests the aggressive nature of the microorganisms to invade the resistant structure.

Hence, the present case may serve as a reminder to consider actinomycosis as a possible cause of osteomyelitis in the maxilla in persistent infections and also to consider it as one of the differential diagnosis for nonhealing ulcers which do not respond to appropriate treatment.

Actinomycotic osteomyelitis pathomechanism is unclear. Actinomyces bacteria are inhabitant of the oral cavity in regions such as the palatine tonsils, gingival fluid, mucosal surfaces, dentin cavities, and sites of postextraction. It is suggested that the infection manifests especially when the normal composition of oral microbiota is disrupted, and chronic inflammation leads to localized pathological changes in the bone. It is an endogenous infection. The prerequisite for the development of this endogenous disease is the transport of pathogens into tissue layers with an anaerobic environment. These organisms have low potential for virulence and invasion, but the companion bacteria act as copathogens and participate in the production of infection by elaborating a toxin or enzyme. This polymicrobial associate flora works in a synergistic fashion to form a specific ecosystem with low oxidoreduction potential favorable for anaerobic growth. It destroys highly vascularized aerobic system and replaces it with a poorly irrigated granulated tissue thereby permitting anaerobic milieu. Marx et al. described it as mutualism than synergism because sulfur granules are produced in tissues but not under laboratory conditions suggesting the importance and contribution of associate bacteria in colony formation, growth, and evasion of host defenses. Once infection is established, the host mounts an intense inflammatory response. Suppuration, granuloma formation, and fibrosis then follows. The infection then produces draining sinus tracts.

In our case, a chronic, persistent, purulent, localized infection associated with endodontically treated tooth and poor oral hygiene was present. This showed that the penetration site of these organisms into deeper tissues was from gingiva, periodontal disease, or chronic periapical abscess which facilitated pathogenicity of these organisms. The presence of accompanying bacteria particularly streptococci and staphylococci had a synergistic effect in the pathogenicity of cervicofacial actinomycosis. However, it was not clear in the present case whether the actinomycosis was the primary infection or a secondary infection to a preexistent nonspecific local osteomyelitis of the alveolar bones.

Diagnosis of actinomycotic osteomyelitis is often delayed because of varied presentations, and they are cultured in fewer than 50% of cases and are the first element of diagnosis in fewer than 10% of cases.

If the culture is poorly executed, delayed, or suffers interference by a concomitant or recent antibiotic therapy by the patient, diagnosis may remain obscure. For this reason, a histopathological examination is highly recommended. Herein, the diagnosis depends on the morphology and staining characteristics of the microorganism. Actinomyces species strongly stain positive for H and E, Periodic acid–Schiff, and Giemsa (GMS). In addition, GMS staining is highly specific for demonstrating the filaments.

In our case also, the histological appearance of the biopsied material was the key for diagnosis which we confirmed by doing the special stain GMS.

As actinomycosis is generally considered a polymicrobial infection for a diagnosis involving osteomyelitis of the jaws, molecular testing is considered a suitable method. In a study by Hansen et al., a polymerase chain reaction (PCR) was used to detect Actinomyces israelii in bone specimens, which were decalcified in trichloroacetic acid and a remarkable reduction in sensitivity was reported. In a subsequent study by the same group, it was confirmed that PCR analysis of A. israelii resulted in a higher sensitivity if milder decalcification, such as with ethylene diamine tetraacetic acid was applied. The current PCR research in the bacteriological laboratory focuses on applying this technique to detect pathogens directly in clinical samples. It is clear that this approach has several
advantages over culture techniques for slowly growing or noncultivable bacteria. However, comparing PCR to conventional identification procedures, PCR is more expensive and requires experienced research personnel, and as our case was reported at an institutional level, we were not been able to perform PCR.

After arriving at a sound diagnosis, it is recommended that treatment of actinomycosis infection should be vigorous by removing the foci of infection, including resection of the sequestrated bone and curettage of all granulation tissue until healthy tissue is exposed. This is required for removal of necrotic tissue and penetration of antibiotics into the colony of microorganisms which is inaccessible otherwise, either because of fibrous tissue or surrounding edematous tissue. Additional exposure time to antibiotics is necessary because lysis of *Actinomyces* species occurs at a slow rate compared to most other bacteria.[16]

**Conclusion**

Maxillary osteomyelitis is an infection that is very rare, and in our case, the causative agent was *Actinomyces*, which makes it even more unusual. Actinomycosis has been referred to the great masquerade of the head and neck. The diagnosis of actinomycotic osteomyelitis often is overlooked because of its ability to mimic other conditions. Thus, in various head and neck infections and nonhealing ulcers, it should be considered as one of the differential diagnoses and careful microscopic and histologic examinations is required for early diagnosis and adequate therapy so that there is minimal functional damage and disfigurement.

Thus, in this case, the persistent nonhealing lesion was diagnosed appropriately through histopathology and successfully resolved with specific treatment against the targeted organisms. Clinicians should be aware of the possibility of actinomycotic osteomyelitis and the importance of histopathology in arriving at a definitive and timely diagnosis.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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