

## ACTINOMYCOTIC OSTEOMYELITIS OF LOWER JAWS: REPORT OF TWO CASES AND A REVIEW OF LITERATURE

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### ABSTRACT

*Actinomycotic osteomyelitis effecting the facial bones is rarely seen. The most causative bacterial pathogen is Actinomyces israelii. The route of entry is trauma, surgical extraction, endodontic treatment and periodontal infections. Mandibular actinomyces osteomyelitis is seen with higher incidence compared to the maxillary due to poor vascularization. This report presents two cases of mandibular actinomycotic osteomyelitis, with the diagnosis particularly based on histological procedures.*

**KEYWORDS:** Actinomycosis, Osteomyelitis, Hyaluronidases

### INTRODUCTION

Actinomycosis is a chronic granulomatous suppurative and fibrosing disease caused by anaerobic or microaerophilic, gram-positive non-acid fast, branched filamentous bacteria. The most commonly isolated organism is Actinomyces israelii, although A. naeslundii, A. viscosus, A. odontolyticus, and A. propionica have been shown also to cause the human disease [1]. A. israelii accounts for 52% of the infections, whereas Actinomyces viscosus (40%), Actinomyces odontolyticus (5%), Arachnia propionica (2%) and Actinomyces meyeri (2%) respectively [2]. Osteomyelitis is inflammation of the bone and its marrow contents. Changes in the calcified tissue are secondary to inflammation of the soft tissue component of the bone [1]. Osteomyelitis due to actinomyces has been reported infrequently.

### CASE REPORTS

#### Case 1

A 60 years old male patient was referred to the Department of Oral Surgery Faculty Clinic because of pain in relation to lower left back region of the jaw since two months. On intra oral examination, there was a necrotic bone exposed in relation to left lower posterior edentulous region. The size of the lesion is 2 cms and white in colour (Fig-1) with palpable left lower submandibular lymphnodes. Radiological features showed radiolucent and radio opaque areas in relation to left posterior edentulous region (Fig-2) and given provisional diagnosis as Osteomyelitis. Incisional biopsy was done and necrotic bone is removed under local anesthesia and sent to the Department of Oral Pathology and Microbiology of G.Pulla Reddy Dental College and Hospital for final diagnosis. The given grossing specimen was whitish in colour, irregular in shape, and hard in consistency. On histopathological examination the Hematoxylin and eosin stained sections showed, fragments of bone and granulation tissue. Hard tissue specimens included trabeculae of woven bone enclosing marrow tissue and a number of partly resorbed bony sequestra with extensive involvement of numerous actinomycotic

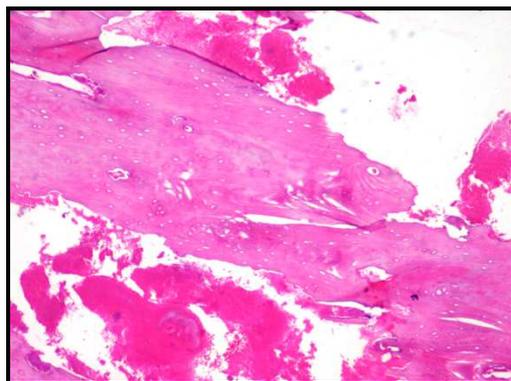
colonies consisting of club-shaped filaments with basophilic central core and eosinophilic peripheral portion were seen with some neutrophils (Fig-3). The bony trabeculae showed empty lacunae without Osteocytes (Fig-4). The granulation tissue showed chronic inflammatory cells with thin walled blood capillaries. The histological appearance were consistent with those of osteomyelitis due Actinomyces.



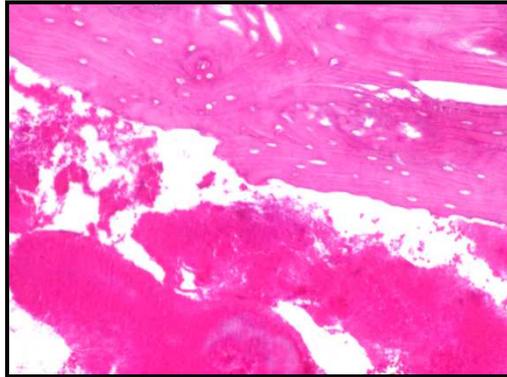
**Figure 1: Intra-Oral**



**Figure 2: Orthopantamograph**



**Figure 3: H & E - Stained Section Showing Bacterial Colonies (4x)**



**Figure 4: H & E – Stained Section Showing Bony Trabeculae with Empty Lacunae (10 x)**

### Case 2

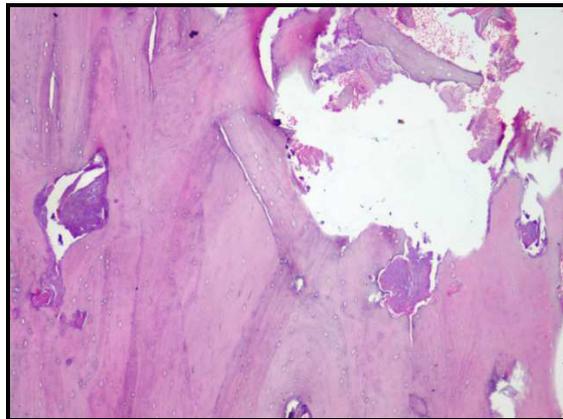
A 58 years old male patient came to the Department of Oral and Maxillofacial Surgery of G.Pulla Reddy Dental College and Hospital, Kunool. Patient gives a chief, complaint of pain in the right lower posterior region of the jaw. On intra oral examination necrotic bone was seen in relation to lower right mandibular premolar region. Patient underwent extraction of tooth in mandibular right posterior region after radiotherapy for throat cancer. The lesion was 1.5 -2 cm in size dull brown in color (Fig-5). Radiographic features showed sclerotic bone in the right mandibular posterior region (Fig-6). With this information, provisional diagnosis was given as Necrotic bone. Biopsy was done and sent to the Department of Oral Pathology and Microbiology for final diagnosis. The given grossing specimen was hard in consistency, with irregular borders and brownish in colour. The histopathological examination of the corresponding tissue section showed bony trabeculae with empty lacunae without osteocytes (Fig-7). At focal areas bacterial colonies was present which are centrally basophilic and eosinophilic peripherally with club shaped filaments suggesting of Actinomycosis (Fig-8). Surrounding the bony trabaculae granulation tissue was present with chronic inflammatory cells and few thin walled tiny blood vessels concluding as Actinomycotic osteomyelitis.



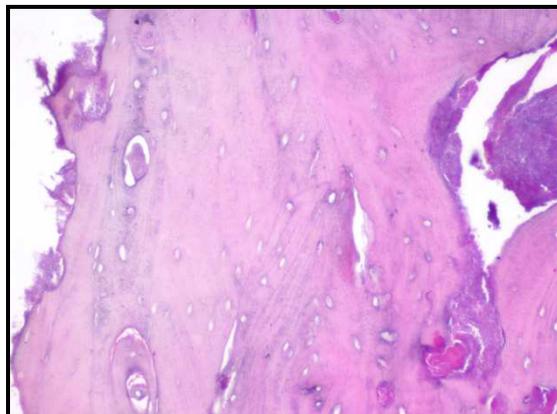
**Figure 5: Intra Oral**



**Figure 6: Orthopantamograph**



**Figure 7: H & E Stained Section Showing Bony Trabeculae with Bacterial Colonies (4x)**



**Figure 8: H & E Stained Section Showing Bacterial Colonies with Empty Lacunae (10x)**

## DISCUSSIONS

In 1877, pathologist Otto Bollinger described the presence of *A. bovis* in cattle, and shortly afterwards, James Israel discovered *A. israelii* in humans. In 1890, Eugen Bostroem isolated the causative organism from a culture of grain, grasses, and soil. After Bostroem's discovery, a general misconception existed that actinomycosis was a mycosis that affected individuals who chewed grass or straw [3]. Actinomyces resembles both bacteria and fungi, thus they were often considered to be transitional between the two groups. However, most of the fundamental characteristics of Actinomyces indicate that they are, in fact, bacteria[4].

Cope in 1938 suggested that the infection may be classified anatomically as cervicofacial (i.e. lumpy jaw), thoracic or abdominal. Cervicofacial is the most common among all and accounts for more than half of the reported cases[5]. Although rarely seen, due to its aggressive and locally destructive nature, actinomycosis of the oral cavity is a highly significant condition[6]. In cervicofacial actinomycosis which is the most frequent the mandible is more commonly involved than maxilla (4:1). It requires a break in the integrity of the mucous membranes and the presence of devitalized tissue to invade deeper body structures and to cause disease which occurs through oro-maxillofacial trauma, dental extractions, dental caries or most probably through any dental intervention as the causative organism[5].

Osteomyelitis due to Actinomyces has been rarely seen. The spread of Actinomyces by hematogenous route with intraosseous granuloma formation and minimal subperiosteal bone reaction has been reported by Bala et al[7]. Mandibular actinomycotic osteomyelitis is usually underappreciated by many clinicians in their assessment of head and neck infections. Most of the cases are traced to an odontogenic source with periapical tooth abscess and posttraumatic or surgical complication as the key antecedent events[8]. From the past 48 years period, a systematic search of MEDLINE data base was performed and reported 30 cases of Actinomycosis Osteomyelitis affecting the jaws [4]. Actinomyces bacteria enters through the Jaw fracture, oral surgery, an infected tooth socket, deep periodontal pockets, and a root canal and leads to the consequent development of Actinomycosis[4].

Therefore, the prerequisite for the development of endogenous disease is the transport of pathogens into tissue layers with an anaerobic environment [9]

These organisms lack tissue decomposing enzymes (hyaluronidases), therefore, they require the aid of other accompanying bacterial flora to achieve pathogenicity. The presence of accompanying bacteria, particularly streptococci and staphylococci, having a synergistic effect in the pathogenicity of cervicofacial actinomycosis[10, 11]. Although the pathogenesis of actinomycosis osteomyelitis is unclear, it is suggested that inflammation begins when the normal composition of the microbial flora is disturbed, and chronic inflammation leads to localized pathological changes in the bone. It is assumed that the mandibular predominance of the disease stems from the relatively poor vascularization of the condensed cortical bone in the mandible with a similar mechanism that predisposes it to osteoradionecrosis[12].

Clinical diagnosis of actinomycosis may be difficult because the condition might not provoke pain at any stage of the disease and the cause is frequently not recognized on presentation[13]. Unidentified mass, facial swelling, or persistent infection particularly after endodontic therapy or tooth extraction, regardless of its nontraumatic history is suggestive of actinomycosis[14, 15]. In our cases also the patient gives an history of pain and history of tooth extraction in second case.

Radiologic features depend on the type and stage of the disease. It varies from the diffuse lytic changes with fuzzy and indistinct bony trabeculae to diffuse patchy, sclerosis of bone often described as 'cotton-wool' appearance [1]. In our cases also, the first case showed radiolucent and radioopaque areas, whereas second case well sclerotic bone is seen..

Histologic examination reveals only a dense mass of bony trabeculae with little interstitial marrow tissue. The osteocytic lacunae appear empty. The bony trabeculae exhibit many reversal and resting lines giving pagetoid appearance. If the interstitial soft tissue is present, it is generally fibrotic and infiltrated by small number of lymphocytes [1]. In our cases also both the tissues showed dense bony trabeculae with empty lacunae without osteocytes. The granulation tissue showed numerous chronic inflammatory cells predominantly lymphocytes. In between bony trabeculae the granulation tissue showed numerous bacterial colonies which are club-shaped filamentous with basophilic central core and eosinophilic

peripheral portion suggesting all the features of osteomyelitis due to actinomycosis.

A biopsy should be performed on any persistent periapical lesion with osteomyelitis even though a chronic draining sinus or cervicofacial abscess does not exist. The most important clinical relevance is to send the discharge for microbial examination rather than only biopsy. Even though the proper surgical incision and drainage and the administration of antibiotics caused the lesion to regress, sometimes, it can prove potentially fatal. Hence, this disease needs to be considered frequently in the diagnosis of head, neck and intraoral infections. In the background of debilitating bacterial infections, a vigorous antimicrobial regimen should be followed before undertaking any surgical procedure. Actinomycotic osteomyelitis is a chronic specific suppurative osteomyelitis which is not refractory to treatment. In our case, the patient developed actinomycotic osteomyelitis after tooth extraction as a postprocedural complication. As actinomycotic osteomyelitis develops in patients with poor host immune response, the morbidity and mortality rate should be reduced with proper management [16]

The initial treatment for Actinomycotic osteomyelitis consists of high doses of penicillin that, depending on the severity of the case, can be administered by intravenous infusion in doses ranging from 3-12 million units daily or oral administration of 2-4 gm per day for a period ranging from 3-12 months, depending on the response of the host to the infection. Other antibiotics used in actinomycotic osteomyelitis is clindamycin, erythromycin, chloramphenicol, cephaloridine, minocycline, imipenem. Metronidazole and Aminoglycosides are ineffective against *A. israelii*. Sometimes it is necessary to curette the region, remove bone sequestration, and refer the patient to maxillofacial reconstruction in cases of substantial loss of bone and soft tissue [17].

## CONCLUSIONS

Mandibular osteomyelitis is an infection that is challenging to manage due to the poor vascularization of bone that favors the proliferation of microorganisms. In this case, the causative agent was *Actinomyces*, which makes it even more unusual. The origin of the microorganism has not been clearly established; however, the diagnosis allows long-term treatment with antibiotics.

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