Actinomycotic Osteomyelitis of the Mandible - A Rare Case Report

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Abstract

Actinomycetes are a relatively sporadic cause of infection of the head-and-neck region and their appearance is usually uncharacteristic, and hence pose a challenge for the diagnosis. The present article intends to exhibit this rarity afflicting mandible and highlight its management. The present report describes a case of a 55-year-old countryside female who presented with pain and swelling affecting the left side of the mandible. Orthopantomograph and cone-beam computed tomography imaging showed multiple ill-defined radiolucencies and perforations of the buccal and lingual cortical plates. Fine-needle aspiration microbiology was used to ascertain the microbial organism and the patient was treated with amoxicillin + clavulanic acid with curettage of the infected site. The patient responded well to prompt systemic antibiotics and local surgical measures with complete resolution of the infection and spontaneous bone regeneration. Although rare actinomycosis of the mandible is curable and should be included in the differential diagnosis of osteomyelitis of the jaw. Early and accurate diagnosis and prompt intervention confirm better outcomes.

Keywords: Actinomycosis, filamentous, jaw infection, osteomyelitis, ray fungus, suppuration

Introduction

Actinomyces are commensals of the oral cavity, oropharyngeal, and gastrointestinal regions. Actinomycotic infection has a typical subacute to chronic clinical course primarily afflicting soft tissues and rarely the bones. *Actinomyces israelli* is found to be primarily associated with the cervicofacial actinomycosis.[1] It is a slow-progressive, gram-positive, anaerobic to microaerophilic, branched, and filamentous bacteria. Actinomycotic osteomyelitis of the mandible is an unusual sequel of odontogenic infections. Its diagnosis is particularly challenging, as its presentation often overlaps with other infectious and noninfectious disease both clinicoradiologically.

Characteristically, a breach in the integrity of mucous membranes by preceding infection, trauma, or the surgery may help bacteria gain entry to deeper body structures and lead to disease progression. Predominantly, the spread of infection is by direct invasion and rarely by the hematogenous or metastatic spread. The aim of the current article is to exhibit our experience with this rarity and document its successful treatment.

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Case Report

A systemically healthy, 55-year-old countryside female reported to the emergency department complaining about pain and swelling for 5 months with a history of dull aching, poorly localized, progressively intensifying pain of the lower left back region of the jaw radiating to the left preauricular, and temporal region. At the time of presentation, a single, large, tender, and diffuse swelling measuring 4 cm × 2 cm was noted along the left half of the mandible with intact overlying skin. Intra-oral examination revealed moderate gingival inflammation with a periodontal pocket of 4 mm in the left mandibular second molar without any history of pus discharge. An expansion of the buccal cortical plate over the molar region was noted [Figure 1]. The patient reported

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no former history of trauma, surgery, or infection at the same local site. Orthopantomograph and cone-beam computed tomography demonstrated multiple ill-defined radiolucencies from the symphyseal region extending posteriorly involving the condyle. The lower border of the mandible appeared intact with no root resorption or displacement of the tooth [Figure 2a and b]. The differential diagnosis included malignancy, osteosarcoma, chronic relapsing multifocal osteomyelitis, and granulomatous diseases.

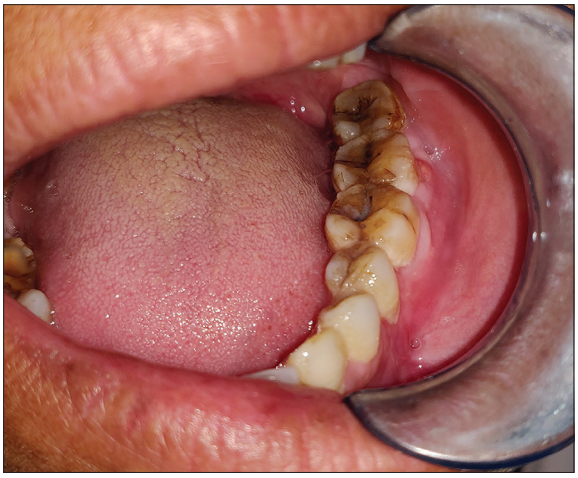

Figure 1: Intraoral view

Fine-needle aspiration cytology and microbiology were demanded to ascertain the nature of the lesion that revealed necrotic granular material with extensive inflammatory infiltrate surrounding the bony spicules with capillary proliferation and fibrosis. Furthermore seen in smears were the cell debris, areas of bone necrosis, and actively phagocytizing macrophages. Abundant colonies of filamentous bacteria were suggestive of Actinomyces. The 40X view of microscopy identified the species as *A. israelli* [Figure 3a].

Empirically, amoxicillin + clavulanic acid 1.2 g intravenous 12 hourly was administered and the patient was scheduled for open debridement, curettage, and saucerization. Significant granulation tissue with lytic loculated lesions was seen underneath the periosteum from which frank purulent discharge was expressed. Curettage and saucerization were performed, preserving the inferior alveolar nerve. Several areas of significant decortication and rough osteomyelitic bone were debrided [Figure 2c]. No postoperative complications occurred. The patient was discharged on the 5th postoperative day and further reviewed monthly for clinical and radiographic assessment for 6 months. Antibiotics continued for 4 weeks.

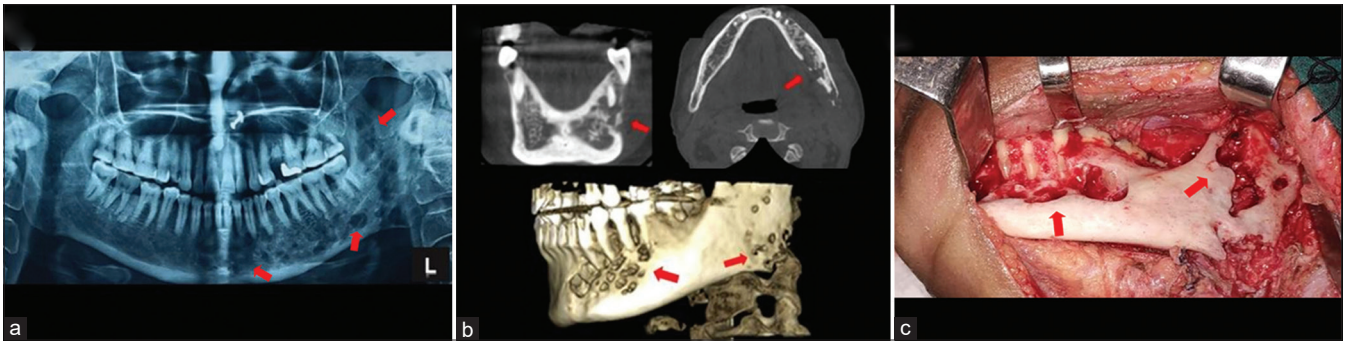

Figure 2: (a) Preoperative orthopantomograph presenting an extensive ill-defined radiolucent lesion over the left side of the mandible (arrows). (b) Cone beam computed tomography presenting with mandibular osseous lesion and cortical erosion over the left side of the mandible (arrows). (c) Intraoperative clinical view depicting decortication and debridement of the left side of the mandible (arrows)

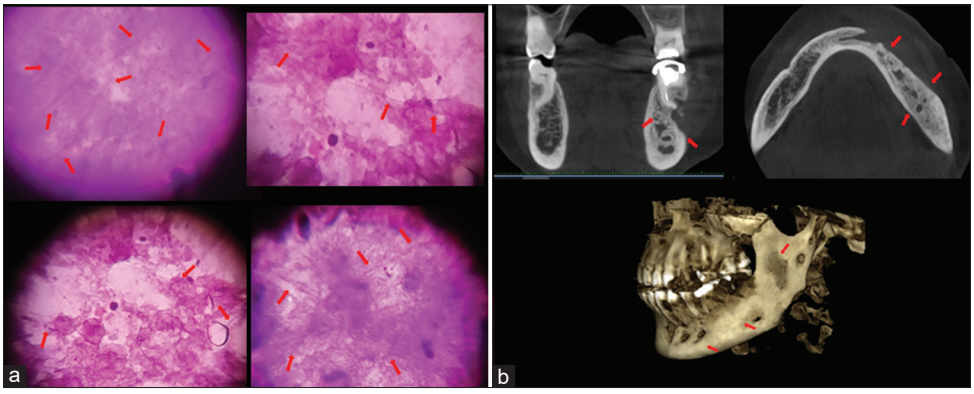

Figure 3: (a) Histopathology demonstrating filamentous actinomyces israelii colonies surrounded by inflammatory cells and necrotic material suggestive of acute suppurative osteomyelitis with actinomyces (Leishman staining, 40X) (arrows). (b) Postoperative 6 months follow-up radiograph showing resolution of infection and neo-bone formation (arrows)

The surgical site showed significant healing and remained well without any signs of infectious relapse [Figure 3b].

Discussion

Actinomyces are unique enough to be discussed and categorized as bacteria that are prokaryotic organisms. Cope (1938) on the basis of involvement of the internal organs, classified it as cervicofacial (i.e., most common), abdominal, or thoracic. The incidence of cervicofacial actinomyces is usually once per year at any major medical center and referred to as the chameleon of the head–and-neck region. The mandible is more commonly involved than maxilla (4:1).[2,3] Its diagnosis is easily missed out due to its ability to mimic granulomatous diseases and neoplasms clinicoradiologically. Therefore, it has been given the title "Great masquerader of head and neck disease."[4]

Actinomycosis of the oral cavity is highly unpredictable, due to its locally and aggressively destructive nature in bone, which was comparable to our case as it caused erosion of the left half of mandible. It is usually a slowly expanding mass that might not aggravate pain with an unrecognized cause. The characteristic board-like or "woody" appearance eventually appears, disseminating liberally by the fascial planes. It is generally described by its contiguous spread, suppurative and granulomatous inflammation along with the formation of fistulae or sinus tracts that may discharge with a thin, watery characteristic "sulfur granules."[5]

"Lumpy jaw" is caused by actinomycetes, due to direct transport of infection into adjacent tissue by reducing local oxygen tension. Similarly, our patient revealed a periodontal pocket that was the portal for access into deeper tissue. Unlike other infections, actinomycosis is not virulent and does not follow the natural anatomical planes but relatively warrens through them and turn into a lobular "pseudotumor".[6] Inflammation initiates when the standard composition of the bacterial flora is distorted; these organisms lack hyaluronidases (tissue decomposing enzymes); therefore, they require the presence of other additional bacterial flora, mainly streptococci, and staphylococci to attain its pathogenicity.[7] Owing to the condensed cortical bone of the mandible and poor vascularization to medullary tissue, intraosseous granulation tissue formation, and subperiosteal bone reaction with characteristic lesions of the dead bone (sequestrum) separated from surrounding healthy tissue by the reactive sheath of new periosteal tissue (involucrum) forms.[8] Radiological features vary from the diffuse lytic changes with hazy and fuzzy bony trabeculae to diffuse irregular, sclerosis of the bone often described as "cotton-wool" appearance.[9] Our case showed irregular and patchy radiolucent areas over the entire left half of the mandible.

The typical clinical picture should be confirmed by biopsy (bacteriological and/or histopathological examination) for the final diagnosis of invasive actinomycosis. The outcomes of both examinations are not always constant. Bacteriological examinations should undergo proper collection, transport, and culture of specimens under anaerobic conditions, which are essential for the growth of *A. israelii* whereas histopathological tests should be undertaken before any antibiotic therapy to prevent any false-negative results.

Therapeutically, higher concentrations of antibiotics are mandatory to infiltrate remote areas of microorganism colonies, due to fibrosis and surrounding edematous tissue over weeks or months. The lysis of Actinomyces species occurs at a dawdling rate compared to most other bacteria; therefore, surplus exposure time to antibiotics is obligatory.[10] Early diagnosis, vigorous antibiotic treatment with surgical debridement is the basic form of therapy. Actinomyces are exquisitely vulnerable to beta-lactam drugs and parenteral administration of penicillin G remains gold standard for its management. Administrative dose ranging from 2 to 20 million units/day parenterally or 2–4 g/day orally, divided every six hourly for at least 4–6 weeks extending up to 3–12 months depending on pathological and clinical response to avert any deterioration. An alternative to allergic patients would be macrolides, clindamycin, tetracycline, erythromycin, carbapenem, imipenem, and cephalosporin. Commonly least effective antimicrobials against Actinomyces species include aminoglycosides, cephalexin, and penicillinase-resistant penicillins (e.g., nafcillin and oxacillin).

Stand-alone medicinal management is not always curative. Although wide surgical debridement is recommended to achieve two paramount goals: drainage of abscesses along with the removal of necrotic tissue or bone and penetration of therapeutic dose of antibiotics into dense fibrous and edematous tissue. Therefore, the surgical intervention for radical eradication of infectious foci becomes inevitable along with profound empirical therapy. A persistent and dedicated follow-up is essential to preclude any further recurrence.

Conclusion

Actinomycotic osteomyelitis involving the mandible is underreported disease and often overlooked. Microorganisms generally gain access into the deeper tissues through the oral mucous membrane. As a clinician, prompt diagnosis and management ensure better predictable prognosis. Close follow-up is crucial in determining the trajectory of the treatment.

Declaration of patient consent

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

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Conflicts of interest

There are no conflicts of interest.

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