Osteomyelitis of the Maxilla : A Case report of three cases

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Abstract
Though now-a-days the prevalence Osteomyelitis is decreasing, it still remains a challenging clinical entity. In recent times there is an increasing incidence of systemic diseases that compromise the host immunity such as diabetes mellitus, HIV infection etc. In the present work, three cases of maxillary osteomyelitis secondary to odontogenic infection among immune-compromised individual are reported.

Key words: Osteomyelitis, Maxilla Trauma, Periodontitis

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Introduction

Osteomyelitis of the maxilla is a rare condition, the gravity of which was appreciated by Hippocrates as long ago as the 5th century BC [1]. Since its first description by Rees, isolated cases have been reported from time to time [2]. Kelly called the condition as "empyema of the antrum of Highmore"[2]. After the introduction of antibiotics, acute phases of osteomyelitis are often concealed by antimicrobial drugs without fully eliminating the infection. Subacute or chronic forms of osteomyelitis have therefore become more prominent, lacking an actual acute phase.

Osteomyelitis of the mandible is very common compared to maxilla because of its bone architecture and vascularity. Maxilla is an irregular bone, containing a large air sinus whose walls are mainly composed of thin compact bone, below which lies a mass of spongy medullary alveolar bone where infection is more likely to gain a footing and spread in the alveolar bone mass, than in the relatively compact bone of the antral walls. Compared to the frontal bone, arrangement of the arteries and its lack of diploic veins make maxilla vulnerable to infection [3,4]. Infection may be derived from the antrum, lacrimal apparatus, the dental germ or may be blood-borne. Maxilla derives its arterial supply from the internal maxillary artery, whose branches form anastomosing loops or arcades. Hence sequestra may be localized, but when the internal maxillary artery is itself thrombosed, the whole maxilla sequestrates [1,3]. Most cases of the maxillary OM reported so far have been in infants where infection is considered to arise from the nursing mother or attendant, the organisms entering through abrasions of the gum [1]. Herein, we report 3 cases of chronic suppurative maxillary OM in adults, two odontogenic and one traumatic in origin with diabetes and malnutrition been the predisposing factors.

Case reports

Case-1: A 45 year old female patient was referred to the Dental department from Ear, Nose and Throat (ENT) section for evaluation of antro-cutaneous draining sinus in relation to left middle half of the face. Patient complained of blurring vision since 6 months which has slowly progressed to bilateral complete blindness, intermittent fever and progressive weight loss since a year. She also complained of a painful pustule on the left cheek a year back which ruptured 6 months later and was followed by formation of an extra oral sinus which discharged blood tinged pus. Patient was a Type II diabetic and was on insulin therapy since 4 years. Family history was insignificant. Patient was poorly built and nourished and weighed 32 kgs. All the vital signs were within the normal range. An extra-oral draining sinus with yellow-colored, muco-purulent, blood tinged, foul smelling discharge was seen on the left cheek. The sinus was communicating with the left maxillary antrum, the necrosed walls of which were visible through the sinus. The maxillary antrum in turn was communicating with the nasal cavity through which nasal septum was viewed (Figure-1). Root apices of maxillary left 2nd and 3rd molar were visible through perforation of floor of the maxillary sinus. Intraoral examination revealed poor oral hygiene, chronic
generalized periodontitis and partial edentulism (missing teeth with respect to 14, 15, 24, 25). Upper left molars showed grade III mobility. Patient was subjected to various biochemical investigations and culture test. Culturing of the pus revealed α-hemolytic streptococci, staphylococci, oral anaerobes such as Fusobacterium and Prevotella. Patient was anemic (Hb-9gm%). Radiological examination showed ill-defined moth eaten radiolucency with ragged borders extending from the distal aspect of left upper canine to the maxillary tuberosity. There was generalized alveolar bone loss indicating chronic generalized periodontitis.

Histopathological examination of the specimen obtained by incisional biopsy revealed inflammatory infiltrate composed predominantly of mononuclear cells, necrotic bone with empty lacunae (sequestrum) and marrow spaces with numerous chronic inflammatory cells. Based on the clinical, radiographic and histopathologic picture a diagnosis of odontogenic circumscribed osteomyelitis of the maxilla with draining extra-oral sinus was confirmed. Based on culture and sensitivity testing, antibiotic therapy with intravenous injections of ampicillin (1gm) and metronidazole (500mg) 6 hourly for 2 days followed by amoxicillin and clavulanate (875/125mg) bid and metronidazole tid for 3 weeks was given. Patient was referred for consultation and treatment to the endocrinologist and general physician. Sequestrectomy and curettage was performed followed by full-mouth extraction and fistulectomy. There was uneventful healing and patient is under follow-up. Patient was referred for fabrication of complete denture.

**Case-2:** A 41 year old male patient reported with a complaint of pain and pus discharge from upper anterior teeth since 10 days. Patient gives a history of tooth extraction following an abscess formation in relation to upper anterior and right posterior teeth (16 to 22), five months back. Intraoral examination revealed missing maxillary left central and lateral incisor (21, 22) and on the right quadrant from central incisor to first molar (11-16) all the teeth were missing. The underlying necrosed alveolar bone was visible in relation to 11-16, extending superiorly up to maxillary buccal vestibule and inferiorly 3 to 4 mm above free gingival margin (Figure-2). Surrounding mucosa was erythematous, edematous and pus exudation was seen along with mobility of supporting alveolar bone segment. Patient was type 2 diabetic under oral glycemic drugs since 2 years. Patient was subjected to various biochemical investigations. Radiological examination revealed...
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Surgical excision of the alveolar segment between 11 and 16 was done and sent for histopathological examination which showed bony trabeculae with ragged borders and absence of osteoblastic lining and osteocytes within the lacunae. Marrow spaces showed chronic inflammatory cells and endothelial lined blood vessels with red blood cells. Microbial pus culturing showed α-hemolytic streptococci, gram–ve bacilli, normal flora of the oral cavity. Patient Haemoglobin level was 12 gm%.

Based on the clinical, radiographic and histopathological examination, a final diagnosis of chronic suppurative maxillary osteomyelitis was given. Based on culture and sensitivity testing management entailed a course of antibiotics (Clindamycin) in combination with surgical debridement (sequestrectomy). Following surgical excision, uneventful healing was noticed. All missing teeth were replaced by removable partial denture. Patient is now under follow up.

**Figure-2: Intraorally necrosed alveolar bone in relation to 11, 12, 13, 14, 15 and 16.**

**Case-3:** A 22-year-old male patient reported with a complaint of painful swelling on left lower half of the face since 5 days. Onset of the swelling was insidious, initially small and gradually increased to the present size of 3x3cm. The swelling was febrile, tender, soft in consistency and associated pain was continuous and radiating to head and neck region. There was a history of uneventful sports trauma to the upper anterior region, 8 years back. Two years later patient developed recurrent episodes of pain and swelling in the upper left central incisor region which was taken care by over the counter medication. Two years back endodontic treatment was done with respect to maxillary left central incisor which failed to control the infection following which it was extracted. Family history was insignificant.

Intraoral examination revealed missing maxillary left central incisor and dental caries with respect to 25, 26. The underlying necrosed alveolar bone was visible in relation to 23, 24 and 25 and in between 26 and 27, extending superiorly up to maxillary buccal vestibule and inferiorly 3 to 4 mm above free gingival margin (Figure-3). Surrounding mucosa was erythematous, edematous and pus exudation was seen in relation to 23, 24 and 25 along with mobility of supporting alveolar bone segment. Patient was subjected to various biochemical investigations. Patient was anemic. Radiological examination revealed alveolar bone loss till the apical third of 23, 24, 25 and 26 and periapically was associated with moth-eaten radiolucency. Surgical excision of the alveolar segment between 23 and 26 was done and sent for histopathological examination which showed bony trabeculae with ragged borders and absence of osteoblastic lining and osteocytes.
within the lacunae. Marrow spaces showed chronic inflammatory cells and endothelial lined blood vessels with red blood cells (Figure 4). Culturing of pus revealed α-hemolytic streptococci, gram negative bacilli, normal flora of the oral cavity. Based on the clinical, radiographic and histopathological examination, a final diagnosis of chronic suppurative maxillary osteomyelitis was given. Based on culture and sensitivity testing Clindamycin 600 mgs, 6 hourly intravenously for 3 days followed by 300mg tid for 21 days was advised. Uneventful healing was noticed and patient is under follow up. Missing teeth were replaced by removable partial denture.

**Figure-3: Intraorally necrosed alveolar bone in relation to 23, 24, 25 and 26.**

**Discussion**

Osteomyelitis (OM) of maxilla is a rare entity with the advent of antibiotics, improved nutrition, dental care, early diagnosis and intervention based on new imaging modalities [3,5]. OM is a challenging disease for clinicians and patients, despite many advances in diagnosis and treatment planning. In the past, OM was encountered frequently and dreaded because of its prolonged course, uncertainty of outcome and occasional disfigurement resulting from loss of teeth and bone, and the accompanying facial scarring but with improved social conditions it is disappearing except in rare cases of malnutrition, immunosuppression, or conditions associated with reduced vascularity of the bone [5].

MacBeth classified maxillary osteomyelitis as traumatic (following injury or surgery, the primary site of infection may be antrum, teeth, or lacrimal sac), rhinogenic (spontaneous spread of infection from the antrum and postoperative rhinogenic cases) and odontogenic (dental-root sepsis may progress to osteomyelitis) [1]. In our first case the origin of infection was not clear, whether it was odontogenic or rhinogenic in origin. The patient had generalized periodontitis with attachment loss and an extraoral draining sinus on
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the left cheek communicating with the maxillary antrum and nasal cavity as well. The intimate relationship between the blood vessels supplying the teeth and their periodontal membrane with the antral mucosa make spontaneous spread of infection from the antrum to the maxilla possible. The floor of the antrum which is formed by the alveolar bone has a very thin layer of compact bone, a little thicker than the wall of the cancellous spaces of the alveolar bone itself [2,3]. Periodontal disease, which leads to breakdown of the periodontal barrier membrane, facilitating deep invasion pathogens, seems to be an important condition leading to osteomyelitis. Significant periodontal disease was found in 51% of the OM patients in a retrospective study [6]. OM has been associated with a variety of systemic diseases and pathological conditions. Every systemic disease with concomitant alterations in the host defences may influence profoundly the onset and course of acute and chronic OM. This patient was also immunocompromised, an uncontrolled diabetic, anemic and malnourished contributing to the severity of the disease. Our second patient was also anemic and diabetic under medication. Role of Diabetes Mellitus as a suppressor of the host immune response is a well-established fact. It also may play a significant role in osteomyelitis of the jaw bones by compromising the vascularity due to arthritis of the smaller vessels [7]. Among diabetics mechanism facilitating bone infection include diminished leukocyte chemotaxis, phagocytosis, and lifespan; diminished vascularity of tissue due to vasculopathy, thus reducing perfusion and the ability for an effective inflammatory response; slower healing rate due to reduced tissue perfusion and defective glucose utilization [8]. Incidence of maxillary osteomyelitis among poorly controlled diabetics in rural Indian population was 45.1% in contrast to 0% in other studies [9].

Our third patient had a history of trauma which could have led to compromised vascularity, predisposing the bone to OM. Patient was a chronic alcoholic and tobacco user. In a retrospective study of 244 cases of acute and secondary chronic osteomyelitis of the jaws, alcohol and tobacco consumption were observed in 33.2 and 47.5% of the cases respectively. In addition, all our patients were from poor socioeconomic status which could have attributed to malnutrition contributing to the development of osteomyelitis. All the three patients were negative for Mantoux test and HIV test. In a review of 141 cases of jaw osteomyelitis in Nigeria, Adekeye and Cornah, it was found that odontogenic infections was the cause of 38% of mandibular, and 25% of maxillary involvement [5]. Similarly, Balm et al found odontogenic sources to be the most common cause of mandibular osteomyelitis. Koorbusch et al, found an even distribution of OM caused by trauma and odontogenic infections with 36% each [10]. Unlike osteomyelitis in other regions of the body, which is predominantly due to Staphylococcus aureus, osteomyelitis of the maxilla is typically a polymicrobial infection that is caused by many types of odontogenic microbial flora. Both
gram-positive and gram-negative microorganisms, including Staphylococcus aureus, epidermidis, streptococci and Bacteriodes are seen [11]. Deep boring facial pain, pyrexia, swelling, trismus, facial anesthesia, and nasal discharge are common symptoms of maxillary OM. Ocular symptoms include epiphora, proptosis, impaired mobility and blindness. Swelling may be found additionally over the ascending process of the maxilla, in the canine fossa, over the hard palate, and the lateral nasal wall [3].

Imaging of the suspected OM in the jaws is accomplished by conventional radiography, supplemented by computed tomography, magnetic resonance imaging and radionuclide bone scanning aids in determining the extent and degree of disease, location of sequestra, and in treatment planning. The radiographic changes in OM usually demonstrate a “moth-eaten” appearance due to enlargement of medullary spaces and widening of Volkmann’s canals resulting from destruction by lysis and replacement with granulation tissue, or bone destruction of varying extent in which there are “islands” of sequestra and at times a sheath of new bone (involucrum) separated from the sequestra by a zone of radiolucency, or stippled or granular densification of bone caused by subperiosteal deposition of new bone obscuring the intrinsic bone structure [4]. Histopathologic appearance of OM shows necrotic bone with loss of osteocytes from their lacunae, peripheral resorption, and bacterial colonization. The periphery of the bone and the haversian canals contain necrotic debris and an acute inflammatory infiltrate consisting of polymorphonuclear leukocytes [11-13] usually requires medical and surgical treatment, although occasionally antibiotic therapy alone is successful. The principles of treatment are evaluation and correction, to the extent possible, of compromised host defense along with empirical administration of culture-guided antibiotics, removal of loose teeth and sequestra, debridement, decortication, resection and reconstruction. Adjunctive hyperbaric oxygen therapy (HBO) for nonradiation OM may be considered in refractory infections and among medically compromised with no HBO contraindications[11-14].

Conclusion
To conclude, on one hand occurrence of osteomyelitis seems to have become rarer with the advent of newer antibiotics, imaging techniques and better social conditions, but on the other hand with the increasing prevalence of immunocompromised conditions like diabetes mellitus, HIV infection, etc which act as predisposing factors osteomyelitis seems to be on the rise. The cause of this disease is multifactorial and its presentation varies. Infection of the maxilla can cause serious complications for the patient such as infection of cranial cavity and brain. Thus, it is essential that any maxillary osteomyelitis be treated aggressively to avoid subsequent dreaded consequences.
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Reference