

An unusual case of maxillary osteomyelitis

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Abstract

Osteomyelitis is defined as inflammation of the medullary cavities, have raised system which if not treated will eventually involve the cortical bone. Involvement of maxilla is more infrequent compared to mandible. In addition to usual mixed bacterial infection, fungi are also implicated in certain cases especially in immune compromised host. A high index of suspicion is needed in this regard if the infection fails to respond adequately to antibiotics only. We hereby report a case of 50 year old male patient who presented with complaint suggestive of Oro antral fistula . On complete evaluation it turned out to be osteomyelitis of mixed bacterial and fungal etiology. Patient’s clinical management is discussed along with pertinent review of literature.

Keywords: Osteomyelitis, Fistula, Immune, Metrogyl

Introduction

Osteomyelitis is an infection which is difficult to treat, characterized by the progressive inflammatory destruction and new bone growth.[1,2]. Normal bone is highly resistant, needing a large bacterial inoculum, trauma, or the presence of a foreign body in order to initiate infection. [3,4]Although invasive candidal infections are being encountered with increasing frequency in compromised hosts, such as neonates and patients with intravascular access devices, osteomyelitis caused by *Candida* sp is a relatively rare infection.[5] However, with the increasing occurrence of factors predisposing to candidemia and invasive candidiasis, candidal osteomyelitis is being diagnosed more frequently. [6, 7, 8]What follows is a report of Oro antral fistula which turned out to be maxillary osteomyelitis of mixed bacterial and fungal etiology.

Case Report

A 50-year-old male patient reported to our OPD with a chief complaint of pain, pus discharge, and diffuse swelling of the right side of face following the extraction of teeth from the right maxillary quadrant. The extraction was performed 2 months before this visit. He was a known case of type II diabetes mellitus, and gave history of extraction. He was on intravenous administration of injection Amikacin and Metrogyl for further management of non healing extraction wound. Due to raised eosinophil count he was prescribed tab levocet for 21 days. His past records revealed that he had high fasting blood sugar (FBS) and postprandial blood sugar (PPBS (i.e., 139 mg/dl and 193 mg/dl, respectively) at the time of extraction due to which he was administered tab Glimiperide. He was also a known hypertensive and was on medication for same (tab Amlodipine) by his general physician. The wound failed to heal uneventfully, hence culture swab was sent. Culture sensitivity report revealed there was fungal etiology. There was no history of bisphosphonate use or substance abuse. Patient was tested negative for HIV. He had no history of chemotherapy or radiation therapy. He developed regurgitation of water and other oral fluids through the right nostril along with nasal congestion which was also later associated with pain and discomfort in the same region for which he reported to our OPD.

General Systemic examination was unremarkable and vital parameters were stable. Local examination revealed a diffuse swelling of the facial skin overlying the maxillary antrum which was tender on palpation. There was no neurosensory deficit in areas supplied by right maxillary and mandibular branches of trigeminal nerve. Intraoral examination revealed unhealed empty sockets with respect to 13, 14. Bone appeared denuded with no mucosal cover, pale yellow in color suggestive of necrosis. On application

of pressure purulent fetid discharge was seen from the unhealed sockets; suggestive of probable communication with the maxillary antrum. This was further confirmed, patient was given a glass of water to drink and was asked to tilt his head, on doing this, water flowed through his nostrils. an exposed necrotic alveolar bone in the area from 13 to 16 tooth sockets. The surrounding labial and palatal mucosa were inflamed, edematous, with irregular borders. Foul smelling pus oozed from the extracted teeth sockets.

Pre-Operative Clinical Images



Figure 1



Figure 2

The Orthopantomograph revealed a moth-eaten appearance, Waters view revealed haziness of the Right maxillary alveolar bone and haziness of the maxillary

antrum of the same side. Based on clinical and radiographic evidence a diagnosis of Osteonecrosis of maxilla was made. A pus sample was appropriately taken and sent to the microbiology department for routine culture and sensitivity test. His RBS was found to be raised. Culture report was suggestive of Klebsiella (bacterial origin) and biopsy was suggestive of fungal involvement. Hence a final working diagnosis of osteomyelitis of right maxilla with mixed bacterial and fungal origin was made which was probably triggered by extraction of teeth in the presence of uncontrolled Diabetes mellitus.

Intra-Operative Images

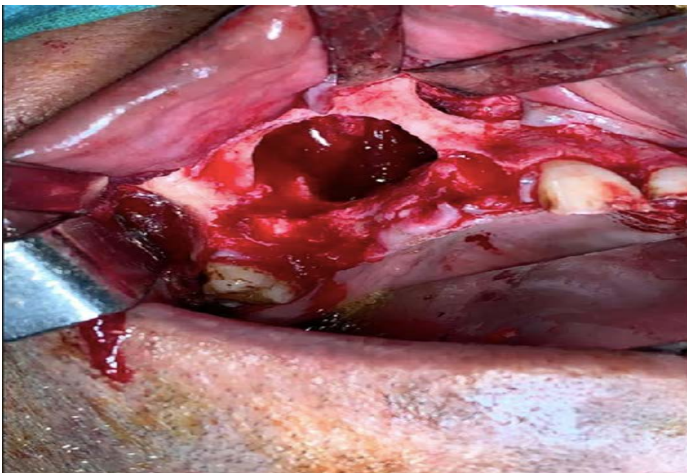


Figure 3



Figure 4

Post-operative Images (1 year)



Figure 5



Figure 6

Under general anesthesia and total aseptic condition, sequestrectomy and debridement of necrotic bone were done through the intraoral approach and Gentamycin pack was placed and removed after 48 hours. The excised hard and soft-tissue specimen was put in 10% formalin and sent for histopathological and microbiological examination, respectively. The microbiological examination involved screening for fungal hyphae using the Gomori methenamine silver (GMS) staining technique. The postoperative period was uneventful. The

antibiotic regime was continued for 5 days with regular twice daily dressings. The discharge sent for culture before surgery was sterile. The tissue was homogenized under aseptic condition and subjected to bacterial (aerobic and anaerobic), tubercular, fungal culture, Gram-stain, Ziehl–Neelsen (ZN) stain, and potassium hydroxide (KOH) mount examination. The antibiotic regime was continued for 5 days with regular twice daily dressings. Patient is being followed up, The postoperative period (1 year) was uneventful.

Discussion

Candida albicans is a saprophytic microorganism that may be isolated from skin and various mucous membranes in healthy individuals. Alterations of human flora, disruption of mucocutaneous membranes, leukopenia, and reduced cell-mediated immunity may predispose to fungal infection.⁶ *Candida* osteomyelitis is one of the less frequent manifestations of invasive candidiasis. This condition however carries significant morbidity, particularly when its diagnosis is delayed by lack of recognition of *Candida* spp as a potential bone pathogen. Osteomyelitis with *Candida* and other fungal species may occur in patients who have severely compromised host-defense mechanisms or who are receiving longstanding intravenous therapy or central parenteral nutrition.¹ Osteomyelitis due to *Candida* species can occur following either hematogenous dissemination or direct traumatic inoculation. Limited data are available regarding the incidence of candidal osteomyelitis. The pathogenesis in all instances is similar. A breach in the normal mucocutaneous barrier in a patient on antibiotics or immunosuppression becomes a portal of entry for *Candida* and, following an episode of fungemia, *Candida* can localize in any deep tissue.

Candida infection develops as a manifestation of systemic candidemia in most cases. Factors that predispose to

systemic infection with this agent include immunosuppression during the course of anti-cancer therapy, organ transplantation, parenteral hyperalimentation, indwelling arterial/venous catheters, intravenous drug addiction, diabetes, broad-spectrum antibiotic therapy, HIV infection, corticosteroid therapy, and myeloperoxidase deficiency. Direct implantation of *Candida* is a very rare cause. The usual species involved is *Candida albicans*, although examples of infection with *C. tropicalis*, *C. paratropicalis*, and *C. guilliermondii* have been reported.⁹ There are no distinctive clinical or radiologic characteristics indicative of the causative organisms, only careful microscopical examination and appropriate culture of biopsy material will permit a correct diagnosis.¹ Even when a bone culture yields a pathogen, sometimes it is not considered as the cause of the infection. The patient in this study had previous reports of *Candida*, but a diagnosis of candidal osteomyelitis had been considered unlikely. Some investigators recommend an amphotericin B dose of 1–1.5 g. In patients who are unable to start or complete an adequate course of amphotericin B, fluconazole seems to be a reasonable alternative and has been successful in treating this infection.^{10,11.} ⁷ According to some authors, patients should be treated for two to four weeks after resolution of clinical signs and symptoms of infection and when there is microbiological evidence of eradication of the infection. Others recommend that therapy should continue for at least six to twelve months beyond the resolution of clinical symptoms and signs of infection.^{6., 7.} The diagnosis of bone infection caused by *Candida* spp is not easy to establish. Blood cultures are usually negative. Biopsy of the involved bone is required to make a definitive diagnosis, and if adequate tissue samples are sent to the laboratory, cultures and special stains are usually positive. The diagnostic workup is of paramount

importance to differentiate between bacterial and fungal osteomyelitis. The biopsied bony tissues on decalcified sections show irregular bony trabeculae with empty osteolytic lacunae. The presence of fungal hyphae within the bone would essentially highlight the fungal nature. Culture sensitivity should be followed in all cases irrespective of its nature.

Conclusion

Osteomyelitis of facial bones needs to be investigated thoroughly as there is no difference in clinical presentation between bacterial and fungal osteomyelitis unless accompanied by maxillary sinusitis. The diagnostic workup with biopsy and culture sensitivity helps to identify the pathogen at the earliest. Fungal osteomyelitis is very rare and appropriate treatment with anti-fungal regime and timely surgical intervention, i.e., debridement, curettage, sequestrectomy will lead to successful resolution of the disease process.

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