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Case Report

Osteomyelitis of maxilla - a rare presentation: case report and review of literature

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ABSTRACT

Osteomyelitis involving the maxillofacial skeleton is a rare entity today. In maxillofacial region mandible is more commonly involved as compared to maxilla. It continues to remain one of the most difficult to treat infections with considerable morbidity and costs to the healthcare system. Hallmark of osteomyelitis are progressive bony destruction and formation of sequestrum. When present, the possibility of underlying malignancy or granulomatous diseases should be kept in mind and ruled out. We present a rare case of osteomyelitis involving the maxilla in a 64 year old male diabetic. The patient was managed with sequestrectomy and debridement by infrastructure maxillectomy via a midfacial degloving approach, appropriate parentral antibiotic therapy and glycemic control. The patient had an uneventful recovery.

Keywords: Maxillary osteomyelitis, Midfacial degloving, Infrastructure maxillectomy, Diabetes mellitus, Micromotor drill

INTRODUCTION

Osteomyelitis, once a dreaded condition, has lost its ability to instil fear predominantly due to advances in current resources for accurate diagnosis, surgical treatment, and potent antibiotic therapy resulting in better treatment outcomes. However, due to its varied symptomatology, mode of presentation and unpredictable course, the disease still continues to hold interest of clinicians. The condition is showing resurgence generally in patients with poor immunity like immunodeficiency syndromes, malnourishment, poorly controlled diabetes, patients on chemotherapy/radiotherapy.[1]

Osteomyelitis can be defined as an inflammation of the bone tissue caused by an infectious agent. It begins with the infection of the medullary cavity and spreads via the haversian system to involve the cortical bone and the periosteum.[2] It commonly involves the long bones and the vertebrae, cranial bones are involved infrequently.[3]

Osteomyelitis of maxillofacial region is uncommon and in maxillofacial region, mandible is more commonly involved than the maxilla. This has been attributed to cancellous bone and significant collateral blood flow in the maxilla.[4,5]

Management of osteomyelitis involves multi prong approach with excision of dead bone and removal of proliferating pathogenic microorganism by combining surgery, antibiotics and supportive care. Complicated maxillary osteomyelitis can spread and involve cranial bones and brain. Therefore, it is imperative that prompt diagnosis is made and aggressive treatment be initiated to avoid subsequent dreaded consequences.

We report a case of right chronic osteomyelitis in a 64 year old type II diabetic patient with poor glycemic control secondary to odontogenic infection and its managemen.

METHODS

A 64 year old male patient, a farmer by occupation, known case of diabetes mellitus with poor glycemic control and no habits, presented with history of pain right upper molars and premolars and swelling over right cheek for past 04 months. The patient had been under management by a dental surgeon. He had undergone dental extraction and drainage of pus and treated with oral antibiotics. He responded well but continued to have swelling over right side of maxilla along with tenderness.

On examination, the patient had a firm swelling over the right maxilla. Its surface was smooth and the swelling was mildly tender. Overlying skin was normal. He was partially edentulous, with the right upper incisor to 1st premolars having been extracted. There was a small oro–antral fistula extending from upper alveolus to maxillary antrum, through which foul smelling inspissated food material were extruding. Nasal endoscopy did not reveal any growth or mass or any purulent discharge (Figure 1 and 2).

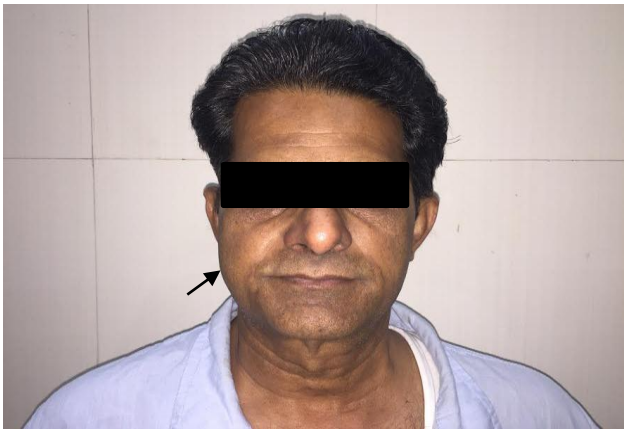

Figure 1: Notice slight bulge over right cheek area.

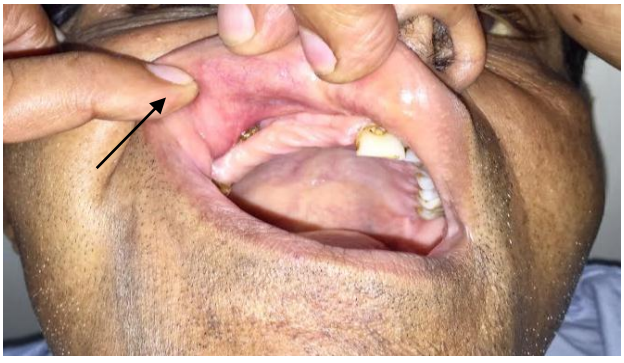

Figure 2: Intra-oral picture showing absent teeth on right and oro antral fistula with inspissated food particle (arrow).

Biopsies were taken from the maxillary antrum via the nasal cavity and via the pre-existing fistula above right upper alveolus. Histopathology report showed the presence of granulation and fibrosis. No evidence of granulomatous disease/malignancy was seen.

CECT PNS showed presence of extensive areas of bony destruction and lysis of right maxilla involving alveolar process, medial, anterior, posterolateral wall, along with erosion of right zygomatic arch and floor of right orbit. Minimally enhancing soft tissue was seen in the maxillarysinus (Figure 3).

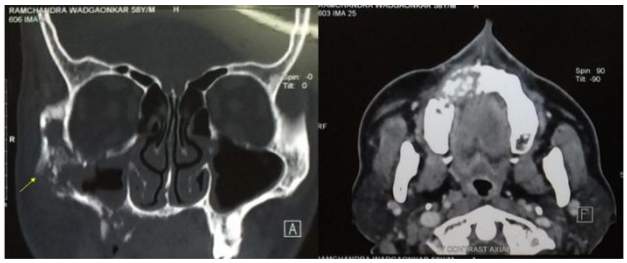

Figure 3: NCCT and CECT PNS showing extensive areas of necrosis and fibrosis involving the right maxillary sinus upto zygoma (arrow).

A contrast enhanced MRI was also done, which showed heterogenous areas of T2 hyperintensity and T1 iso intensity involving the posterior portion of right upper alveolar arch, anterior, medial and lateral walls of right maxillary sinus , floor of right orbit and proximal part of zygomatic process of right maxilla with overlying swelling of soft tissue. Post contrast, heterogenous contrast enhancement was seen. An impression of infective pathology, likely osteomyelitis was given (Figure 4).

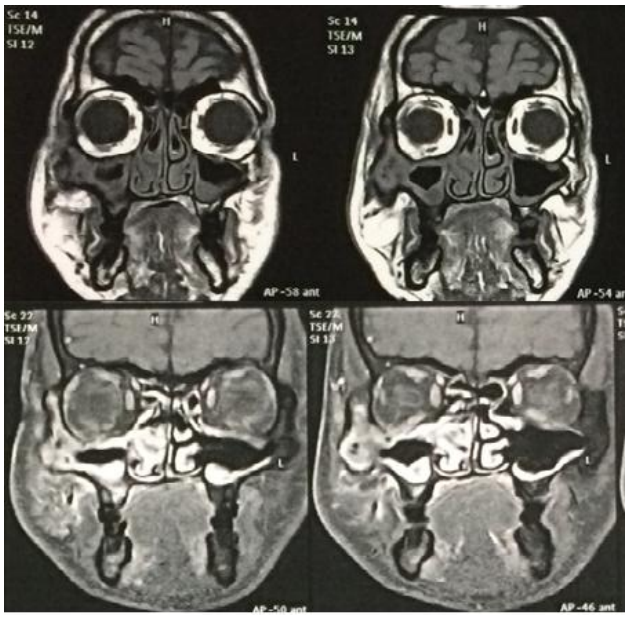

Figure 4: Pre and post contrast MRI T1W showing lesion involving right maxillary sinus.

After extensive evaluation and deliberation on above mentioned clinicoradiological findings and biopsy report, it was evident that we were dealing with an infective pathology and a provisional diagnosis chronic osteo-myelitis was made. However, in view of patient's age, and suspicion of a low grade malignancy, patient was still discussed in multidisciplinary tumour board and tentative plan of management was charted out. As the disease was extending supero-laterally upto zygomatic arch, we felt that reaching upto that farther location would be difficult by any other approach and also the patient wanted to avoid an external scar, so a midfacial degloving approach was taken (Figure 5). Standard incision was taken for midfacial degloving approach, periosteum over anterior aspect of maxilla was raised laterally upto zygomatic process. Palatal flap was raised using periosteal elevator in anticipation of performing a primary closure of operated cavity as soft tissue was not involved by disease. Anterior bony wall of maxilla was found to be brittle and necrosed which broke easily on pressure. A sufficient specimen of this bone, soft tissue from maxillary antrum was collected for HPR. Routine infrastructure bony cuts were taken using micromotor drill with fissure burr. Bony cuts over anterior wall and medial wall of maxilla were taken and maxillary antrum was examined. No obvious growth noticed inside the antrum except for hyperplastic mucosa. Thereafter rest of the infrastructure maxillectomy was performed using 6 mm round body cutting and diamond burr. All bony walls were excised (Figure 6). The purpose of using round burrs was to excise entire diseased bone in a controlled manner until underlying bone started bleeding, a sign of healthy bony wall could be appreciated. This prevented unnecessary excessive removal of bone. Medial aspect of zygomatic process was drilled out using diamond burr, leaving outer healthy cortex of bone. A thorough wash using diluted betadiene was given. After confirming that entire diseased bone is excised, primary closure was performed by suturing the palatal flap with mucosal surface of lip and buccal mucosa (Figure 7). The benefit of it, was that, roof of maxilla and zygomatic prominence could be preserved thereby maintaining the facial contour and symmetry, and any residual oro-antral fistula was avoided. Nasal packing was done. Post-operative period was uneventful. The patient was kept on nasogastric tube feeds for 7 days and thereafter started orally. Antibiotic cover was given with Inj. Teicoplanin, Amikacin and Metronidazole. The patient showed uneventful recovery. Histopathology of the operated specimen was consistent with osteomyelitis of maxilla. *Streptococcus pyogenes* was cultured from the excised specimen. Patient was prophylactically put on oral tab linezolid 600 mg q12 h for 4 weeks, based on culture and sensitivity report.

On follow up, 18 FDG PETCT was performed three months post op to assess for residual disease. The scan showed post-operative status devoid of significant FDG uptake (SUV max -1.94). Prosthetic rehabilitation is planned with removable maxillary denture. Presently patient is under regular follow up with no evidence of recurrence of disease at six months post op (Figure 8).

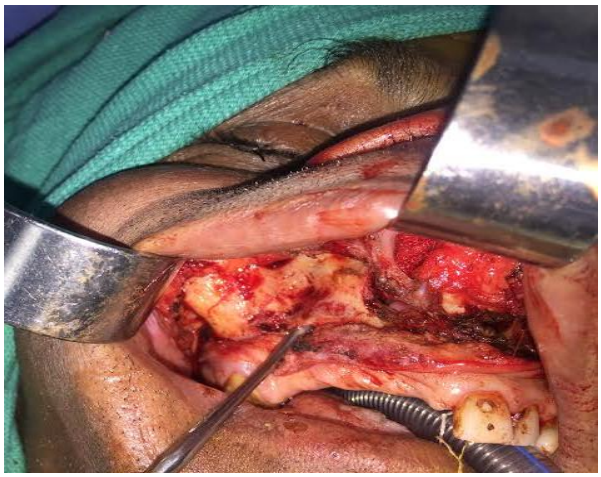

Figure 5: Intraoperative picture showing midfacial degloving approach and necrosed anterior wall of right maxilla (shown by a pointer).

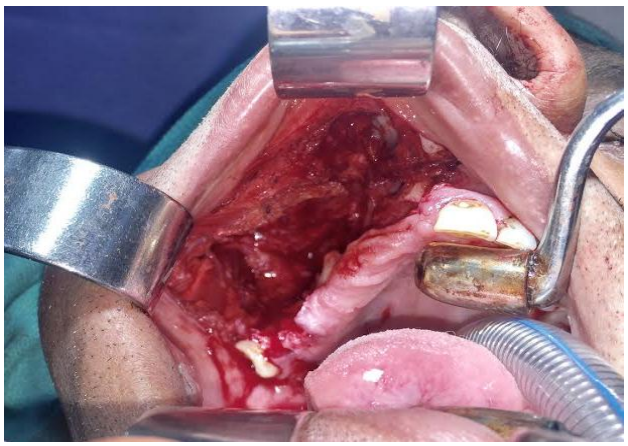

Figure 6: Intraoperative picture showing excised anterior, medial and posterolateral wall of maxilla.

Figure 7: Sutured palatal flap with mucosal surface of lip and buccal mucosa.

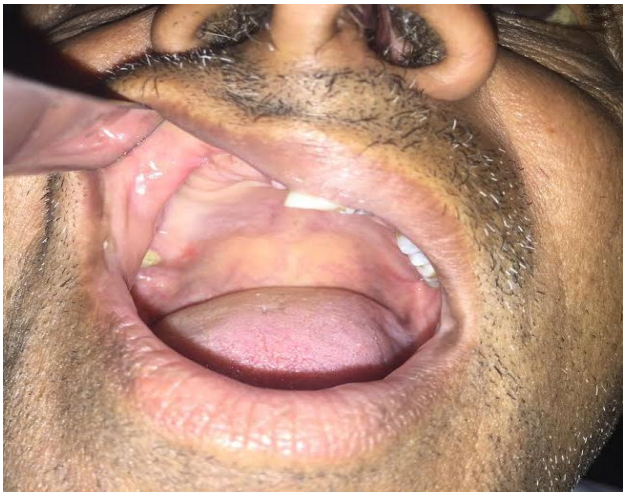

Figure 8: 3 months postoperative picture showing well healed surgical site.

DISCUSSION

In todays' era, osteomyelitis has become an uncommon entity and involvement of the maxillofacial skeleton is even rarer. The term osteomyelitis represents a wide spectrum of clinical conditions with a common pathology of inflammation and infection of the bone. Osteomyelitis of maxillofacial skeleton is seen more commonly involving the mandible.[6] It may result from hematogenous spread, contiguous spread from infectious focus, or direct bacterial inoculation into intact bone due to trauma.[7]

Diminished host defenses and immune compromised status like immunodeficiency syndrome, diabetes, autoimmune conditions, malignancies, malnutrition are major predisposing factors.[8] Hematogenous osteomyelitis is generally noticed in pediatric age group (85% patients aged below 17 years), whereas in adults up to 50% cases are post traumatic.[9]

Maxillary osteomyelitis can be classified based on following causes: [10]

a) Traumatic - following accidental or iatrogenic trauma. The primary site of infection may be antrum, teeth, or lacrimal sac
b) Rhinogenic - spontaneous spread of infection from the antrum and postoperative rhinogenic cases
c) Odontogenic - dental-root sepsis may progress to osteomyelitis.

Local periodontal infection is implicated as most common source of infection in cases of maxilla facial osteomyelitis. In one study, 51% patients of osteomyelitis had pre-existing periodontal disease.[11] In another study, odontogenic component as the source of infection could be established in 74%, followed by maxillary sinusitis and trauma as 16% and 6.4% respectively.[1]

Osteomyelitis of maxilla was originally described by 3 Rees in 1847.[12] By virtue of extensive blood supply, thin cortical plates and a relative paucity of medullary tissues in the maxilla, osteomyelitis is less frequent as compared to mandible.[1,5]

Once bacterial inoculation occurs there is alterations in pH and capillary permeability that contribute to regional edema, cytokine release, tissue breakdown, leukocyte recruitment, decreased oxygen tension, increased local pressure, small-vessel thrombosis, and bone deterioration. As the infection spreads into the medullary cavity, increased pressure causes its extension into the cortex by Haversian and Volkmann canals with subsequent spread into the subperiosteal space causing periosteal stripping, leading to loss of periosteal blood supply and consequent bone resorption and necrosis.[13]

Diabetes mellitus is a known suppressor of host immune response. It causes arteritis of small vessels compromising vascularity leading to reduced tissue perfusion, poor healing and poor host response against infection, thereby aggravating osteomyelitic process.[14] In one study, incidence of maxillary osteomyelitis among poorly controlled diabetics in rural Indian population was 45.1%.[1] Similar condition was noticed in our patient who had a poor glycemic control and local odontological infection.

Osteomyelitis of 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, Peptococcus, Pepto streptococci, Hemolytic streptococci, Pnemococci, Escherichia coli* and *Bacteriodes* are seen.[5] It is also been reported after maxillary necrosis secondary to invasive fungal infection like mucormycosis.[6]

Diagnosis of osteomyelitis is based on clinical and radiological criteria along with hematological and histological inputs. Acute infections are associated with leukocytosis and neutrophilia. Inflammatory markers such as ESR and CRP, are often elevated, however, these are nonspecific tests and are more important in the control of treatment.[15] Biopsy is essential and sample of soft tissue and bone sequestra should be sent for histological analysis, essentially to rule out any neoplastic process that may mimic osteomyelitis.[16]

A plain radiograph of PNS have poor sensitivity and specificity and is best avoided. Cross sectional imaging modalities such as computed tomography (CT) scanning and magnetic resonance imaging (MRI) are now considered standard for diagnosing osteomyelitis. These modalities provide excellent anatomic delineation of the infected area and surrounding soft tissue, assisting surgeon to plan optimal surgical management, avoiding morbidity and complications to adjacent critical structures.

CT is superior to MRI in detecting sequestra, cloacas, involucra. However, MRI is highly specific and sensitive for detecting osteomyelitis along with pathologic changes of bone marrow and soft tissue. It can detect inflammation as early as 3 to 5 days due to its ability to demonstrate changes in the water content of bone marrow. It also differentiates between retained secretion and soft tissue. MRI is referred during post treatment follow ups. Therefore both imaging modalities are complementary to each other.[17]

Nuclear medicine techniques, although highly sensitive, are sometimes nonspecific. These functional imaging are preferred in post treatment status where radiographic changes may still be present, despite adequate treatment. Fluorine-18 fluorodeoxyglucose (FDG) positron emission tomography (PET)- CT has highest diagnostic accuracy for confirming or excluding the diagnosis of chronic osteomyelitis in comparison with bone scintigraphy, MRI or CT individually.[17] It provides better spatial resolution, sensitivity, and specificity when compared to conventional scintigraphy (96% and 91%, respectively). Other advantages are - results available within 30 to 60 minutes of tracer administration, imaging is not affected by metallic implant artifacts, and is now available relatively easily.[17]

Technetium-99 metastable isotope (99 mTc) radionuclide bone scans can detect bony involvement even before a high resolution CT can demonstrate bone destruction. Isotope is absorbed by osteoclasts and osteoblasts. It has reasonable sensitivity (70–89%), but low specificity (16–36%) as bony remodeling continue to occur for a long duration, even if disease is not present.[17]

Gallium scintigraphy uses gallium-67 citrate which binds to transferrin and is absorbed by leukocytes showing increased isotope uptake in infection, sterile inflammatory conditions, and malignancy. It should be used in conjunction with bone scintigraphy, to distinguish bone and soft-tissue inflammation and show bone detail. The combined sensitivity and specificity are around 60% and 80% respectively.[17]

Indium-111 marked leukocyte scintigraphy is one of the best nuclear medicine scintigraphy for assessing patients with osteomyelitis, because it is independent of bone remodeling. The cost of procedure, complex to implement and limited availability are its major drawbacks. It has good sensitivity (84%) and specificity (80%).[18]

Cranial bones are infrequently involved, but infection spread to surrounding structures can lead to cerebral abscess, encephalitis, or meningitis.[3] Prompt diagnosis and aggressive management are crucial for best results. The goals of successful treatment are prompt diagnosis; correction of underlying disease process; improvement of the host's defenses; correct anatomical localization of bone involvement; adequate antimicrobial therapy; surgical debridement of all diseased tissue; and appropriate reconstruction and rehabilitation.[19] Appropriate culture-directed antibiotic therapy and an oncology type adequate debridement are the mainstay treatment. Broad spectrum high end antibiotics which concentrate in bone are preferred. In our case, we used inj. Teicoplanin, which has good bone penetration along with amikacin and metronidazole to cover the spectrum.[20] Patient's glycemic control was initially done by using plain insulin on sliding scale and thereafter patient was put on combination of oral hypoglycemic on advise of Endocrinologist. On discharge patient was put on prophylactic tab linezolid for 04 weeks, depending upon culture sensitivity test. Studies has shown that, Linezolid, a bacterioststic, protein synthesis inhibitor of oxazolidnone class has 100% bioavailability and good bone penetration, with bone levels in healthy adults undergoing hip replacement surgery of 50% of serum levels . Linezolid has demonstrated success rates comparable to vancomycin in clinical trials.[21,22]

Hyperbaric oxygen therapy (HBOT) has been employed to accelerate wound healing as adjuvant treatment in complicated or refractory cases. It involves respiration of 100% oxygen under pressures artificially elevated above the atmospheric pressure at sea level, with the patient being placed inside a pressure-resistant hyperbaric chamber. HBOT promotes fibroblasts and collagen production, angiogenesis, and healing in an ischemic or infected wound.[23] Limited availability, complex infrastructure, costs are its limiting factors.

HBOT along with antibiotic containing acrylic beads, micro vascular grafts has shown promising results in the management of patients with refractory osteomyelitis.[24,25]

Our basis of diagnosing this patient as osteomyelitis of maxilla following odontogenic infection was – a) patient was a diabetic with poor glycemic control. b) history of purulent discharge and swelling following dental extraction. c) bacteriology showed presence of *Streptococcus pyogenes*. d) biopsy from the involved bone- ruled out malignancy. e) radiological imaging favoured osteomyelitis.

CONCLUSION

Maxillary osteomyelitis is one of the most difficult to treat infectious disease, and is a challenge for both clinician and patient, despite many advances in diagnosis and treatment. Clinical suspicion is critical to initiate prompt and appropriate hematological, histological and radiological investigation. Aggressive medical management with adequate surgical intervention is the key to successful management.

In our case midfacial degloving approach along with use of micromotor drill round burrs was successfully employed for complete disease clearance, still remaining conservative enough to prevent facial disfigurement and

post-surgery oro antral fistula. The aim of this article is to focus on current knowledge of the disease, and progress made in understanding its pathogenesis, diagnosis and treatment.

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