Case Report

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Diagnostic Dilemma in Maxillary Osteomyelitis: A Case Report

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ABSTRACT

Background: Maxillary osteomyelitis is rare compared to mandibular osteomyelitis owing to its extensive blood supply and unique structure. We present case report of maxillary osteomyelitis, where clinical, radiographical, histopathological features were perplexing, hence leading to dilemma in diagnosis. A 62-year-old male patient came with chief complain of painful swelling near his left maxillary molars along with pus discharge. His main concern was cessation of pus discharge and removal of the pathology without long-term morbidity. On examination, multiple draining sinuses were present with segmental mobility of left maxillary arch with unusual paper-thin bone texture. Incisional biopsy suggested tuberculous osteomyelitis. After initial biopsy Anti Koch therapy was initiated. Later surgical excision and debridement was performed which led to final diagnosis of fungal osteomyelitis. To establish definitive treatment plan, it is necessary to assess credibility of current methods of investigations.

Keywords: Maxilla; osteomyelitis; tuberculosis; case report.

INTRODUCTION

Osteomyelitis" (OM) is inflammation of bone. In maxillofacial skeleton, it occurs when both medullary and cortical bone undergoes inflammatory process[1]. Thus, osteomyelitis is inflammation of basal and alveolar bone[2]. Mandible is more commonly invaded by infection[3]. However, OM of maxilla is a rare entity due to its rich vascularity[4].

CASE REPORT

Patient concerns

A 62-year-old male with complaint of swelling near upper left posterior teeth since one month came to our centre. The patient's history revealed pus discharge since eight days from the same region, forming multiple draining sinuses. Patient was known case of Type II Diabetes, on medication since two years [Tab Glynase MF (USV private limited) 1 BD]. Patient had fluctuating glycaemic control despite medication. Patient had no deleterious habit.

On clinical examination a tender swelling was present over his palate in relation to left maxillary molars approximately 1.5 x 1.5 cm in size. Multiple draining sinuses were present with active pus discharge in relation to 24, 25, 26 region (figure 1). On digital palpation, segmental mobility of the left posterior maxillary arch was noted which was unusual, the texture of bone was paper-thin bone.

Diagnostic aids

Basic investigations revealed, Haemoglobin (Hb) 10.5gm/dl, high neutrophil count. Erythrocyte sedimentation rate (ESR) value was very high, 58

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mm of the first hour indicating deep-seated infection. Random Blood Sugar (RBS) was 192mg/dl and HbA1c was 10.26 and mean plasma glucose was 248, which suggested uncontrolled diabetic condition. Empirical antibiotic therapy was started until the culture sensitivity report was out. The culture sensitivity report suggested no specific organism.

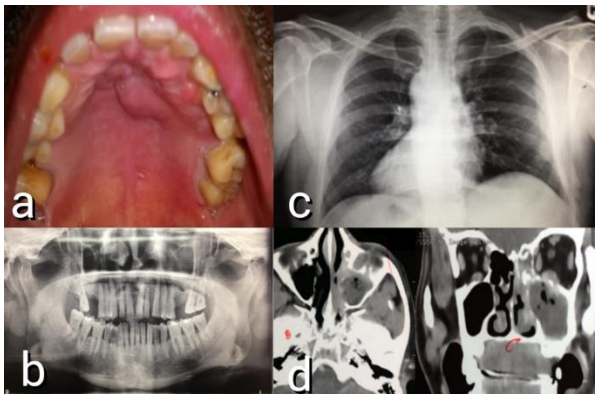

Fig 1: a. Intraoral image showing multiple draining sinus b. orthopantomogram c. PA chest d. CT images.

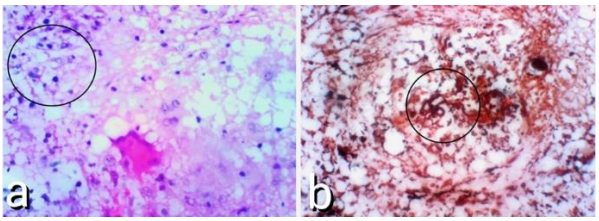

Fig 2: a. Preoperative biopsy with plenty of Langhans Types of Giant cells (encircled) b. Postoperative biopsy shows non septal fungal hyphae(encircled).

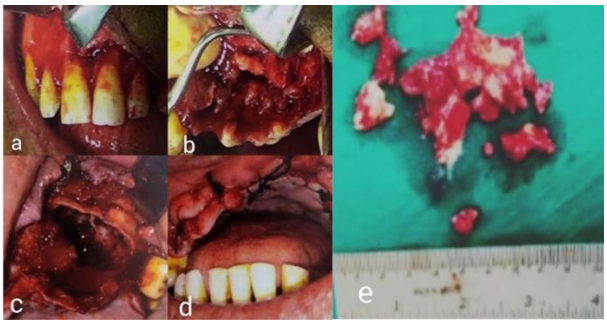

Fig 3: a-d. Intraoperative images. e. Specimen.

Orthopantomogram (OPG)showed diffuse bone loss of maxilla, involving maxillary teeth with diffuse maxillary sinus lining on left side. On Computed tomography (CT) scan a small lytic area was noted in maxilla at 28 region of 9x9mm in cross-section with dehiscence of maxillary sinus at the same level. Soft tissue opacity was noted in maxillary, frontal, ethmoid sinuses with bony remodelling of anterior, medial, and posterolateral walls of left maxillary sinus. Chest X-ray was normal. (figure 1)

Treatment

An incisional biopsy (figure 2-a) suggested polypoidal inflammatory mass showing heavy pyogenic exudates with plenty of granulomas showing epithelioid cells and langhan's type giant cells. Histopathology revealed tuberculous granulomatous inflammation with secondary pyogenic abscess formation. Hence Anti Koch's Therapy was started.

Patient's oral antidiabetic medications were altered and shifted to Human Actrapid insulin therapy with 3 dose regimens, 10 to 15 units depending on RBS levels measured. After glycaemic control, surgical intervention was done under General Anesthesia. Lesion was completely excised (figure 3) and enucleated, offending teeth were extracted and closure was done. Specimen was sent for histopathological examination (figure 2-b).

An endoscopic biopsy was taken from the ethmoid region and sent for Gene X-pert test. Gene X-pert report and polymerase chain reaction (PCR) report were negative.

Operative biopsy suggested GMS stain showing hyphae with distended large spores surrounded by granulomatous inflammation showed fungal organisms, leading to the diagnosis of fungal osteomyelitis.

Patient was also prescribed Tablet Itraconazole (200mg), OD for ten days. Healing was uneventful and healthy mucosa confirmed diagnosis correctly.

Patient had no history of previous Tuberculosis and chest X-rays as well as Gene expert reports were negative. This led to a dilemma for diagnosis of this peculiar case.

Outcome

Biopsy report was a turning point for the treatment plan. Presence of numerous granulomas and giant cells put diagnosis into confusion. There was no

evidence of pulmonary TB and further investigations like Polymerase chain reaction (PCR) and Gene X-pert reports, which were negative. Decision of re-evaluating histopathological reports and sticking to evidence-based treatment plan made us conclude diagnosis of fungal osteomyelitis.

Follow up

Follow up was done for 3 years without any complaint.

DISCUSSION

Maxillary fungal osteomyelitis is rare condition due to rich vascular supply. Fungal infections are opportunistic and occur either in an immunocompromised patient or after a surgical procedure like tooth extraction. However, fungal infections occur seldom without typical clinical presentations, which can create a diagnostic dilemma[5]. Clinical presentation of fungal osteomyelitis is similar to bacterial osteomyelitis. First and foremost, being pain and swelling in region of the lesion. In both type of infections, it primarily attacks maxillary sinus region and thus patient complains of maxillary sinusitis with pus discharge and sinus tract formation. Both infections predominate in Diabetic patients[6].

Tuberculous osteomyelitis is rare, constituting only 2% of maxillofacial skeleton. Radiographic presentation of TB osteomyelitis cannot be distinguished from other pyogenic osteomyelitis and cannot help in clearing the dilemma. *Only a typical clinical presentation of* oral *TB osteomyelitis is moving of entire maxillary arch like paper-thin bone*[7]. Tubercular osteomyelitis can be initially detected through standard Monteux test, positive Acid-Fast Bacilli (AFB) test, and can be confirmed with newer technique of Polymerase Chain Reaction (PCR) and Gene Expert test[8].

PCR and Gene expert were negative thus need arose to get back to every report and re-evaluate the same. The histopathological reports were the salvage. Re-evaluation of histopathological reports ended and it confirmed to be fungal osteomyelitis.

Treatment plans for both lesions vary drastically. For Tuberculous osteomyelitis, Anti-tubercular drugs [Rifampicin (450mg), Ethambutol (800mg), Isoniazid (300mg) and Pyrazinamide (750mg)] play a major role in this lesion. Fungal osteomyelitis needs surgical intervention as a major treatment plan along with anti-fungal drugs like Itraconazole, Amphotericin-B, Flucytosine & the lesion needs to be resected completely[9].

Kumar et al noted that tuberculous osteomyelitis can be a primary case instead of secondary spread[10]. Hence in our case when routine tests did not detect tuberculosis, we still could not rule out its possibility.

CONCLUSION

Need for better and more specific diagnostic aids for detecting osteomyelitis is now a requisite and hence we should encourage more studies for the same. Some lesions are rare and mimic other types of lesions in every way. This rare case of maxillary infection which mimicked Tuberculous Osteomyelitis in every aspect finally came out to be fungal osteomyelitis. The diagnostic dilemma for the lesion was necessary to be cleared out for correct treatment plan as both the infections carry entirely different treatment plans.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent form. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initial will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

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