# Maxillary Osteomyelitis Secondary To Mucormycosis In An Uncontrolled Diabetic - Case Report And Review

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**ABSTRACT:-** Mucormycosis or Zygomycosis is an infection (oportunistic infection) caused by saprophytic fungus mostly seen in immunocompromised patients .Infection generally starts via inhalational route and commonly affects the para nasal sinuses first and then invades vascular tissue to spread to different organs. When this infections spreads to adjacent jaw bones it can lead to thrombosis and subsequent necrosis of the involved bone . Many a times there is development of secondary bacterial infection .We are here presenting a case of 46 year uncontrolled diabetic patient who developed mucormycosis associated osteomyelyitis of left maxilla which required sequestrectomy and extensive medical management.

## I. INTRODUCTION

Mucormycosis is an opportunistic infection commonly affecting immunocompromised patients such as uncontrolled diabetes, AIDS affected individuals,Patients on chemotherapy and extensive malignancies<sup>1</sup>. Mucormycosis is a saprphytic fungi belonging to order mucorales class zygomycetes. The disease can have various presentation based upon the organ and site involved. After candidiasis and aspergillosis this is the third most common fungal infection seen<sup>2,3</sup>. We hereby present an interesting case in which there was extensive necrosis of left hemi maxilla of a 46 year old uncontrolled diabetic patient requiring sequestrectomy and buccal myo mucosal flap for closure and also discuss the available literature available.

### II. CASE REPORT

A 46 year old male patient reported to the OPD with complains of oral mal-odor and blood tinged discharge from oral cavity for past 15 to days . The patient was apparently normal 4 months back when he developed pain and heaviness involving left side of face which increased on bending forwards. The patient visited a local clinic which advised certain medications which the patient was not able to specify. He took them for 15 days and his symptoms resolved temporarily. He was then diagnosed to have a infected molar tooth on the left side and underwent an extraction of the same around 1 month back . Extraction didn't provide significant relief . Gradually patient noticed increasing mal odor and greenish white discharge from the oral cavity. Patient also developed mild fever and diffuse swelling of left side of the face.

Extra oral examination revealed a mild diffuse swelling involving left side of the mid face from corner of the mouth to the malar- infra orbital region. The overlying skin was normal with no evidence of pus discharge or sinus opening extra orally.

Intra-orally there was bony hard swelling involving the left maxilla almost reaching the midline with normal overlying mucosa . There was a sinus opening present distal upper left third molar and exposed bony surface. Routine blood work revealed elevated TLC - 14,000, ESR -43, RBS - 305 , Hba1c- 8.9.

Radiologically, cbct revealed extensive mixed radio-leucent and radio opaque destruction of the left maxilla with extension to the pterygomaxillary junction posteriorly and infra malar regionsuperiorly(Fig 1a,b,c). Anteriorly the lesion extended to the canine region and approached the midline of horizontal palate. There was evidence of sequestrum formation on the cbct.

Patient was stated on anti-hypoglycaemic medication including sliding scale insulin therapy for peri operative diabetic control. Patient was taken up for surgery under monitored anesthesia care. Intra oral vestibular incision was given from left canine to left third molar region with an anterior and distal release (Fig.2). Careful dissection was done to delineate the necrotic bone/ sequestrum which appeared greenish - grey(Fig. 3). The entire sequestrum was removed into-toto along with the involved teeth .Thorough debridement of the wound was done(Fig. 4,5,6). Haemostasis achieved. Buccal fat pad was released via distal exploration and was mobilized to cover the defect and subsequently secured . Two layer closure was then achieved via mobilisation of the buccal myo mucosal flap and tension free closure with the help of resorbable vicryl sutures. Post op recovery was uneventful and diabetic control was managed fastidiously (Fig 7).

Histopath analysis was done methodically which included gross macroscopic examination of both soft and hard tissues, both of which were processed. For soft tissues Hematoxylin and Eosin (H and E), periodic acid-Schiff (PAS), Gomori's methenamine silver (GMS) and Ziehl-Neelsen stain were done. Soft tissue examination showed highly necrotised tissue. GMS staining showed fungal organisms branching at right angles, with large non-septate PAS positive hyphae with round to ovoid spores.

The hard tissues was decalcified and processed, sectioned and stained with H and E stain. Hard tissue assessment showed bony trabeculae with fibrosis and inflamed marrow. Many resting and reversal lines were seen along with empty osteolytic lacunae.

The combined histopath features were suggestive of chronic osteomyelitis os maxilla with mucormycosis.

The patient was subsequently started on anti-fungal therapy with amphotericin B. The patient has been kept on regular follow up after the completion of therapy and has been disease free for past 9 months with good diabetic control.

#### III. DISCUSION

Mucormycosis is the third most common fungal infection caused by saprophytic fungus in an immunocompromised individual with a potential for extensive destruction<sup>4</sup>.Infection commonly arises due to inhalation and less commonly by direct inoculation or ingestion.Mucormycosis has a tendency to erode and invade small blood vessels causing thrombosis and subsequent vascular compromise leading to tissue necrosis<sup>5</sup>.Para nasal sinuses are generally first involved from there can be a spread to adjacent maxillary and cranial base stuctures<sup>6</sup>.Extensive cranial involvement has been associated with high morbidity and mortality<sup>7</sup>.

Oral lesions of mucormycosis must be differentiated from necrotisingsialometplasia, burn injury and carcinomas especially when there is oral mucosal involvement by the fungus. Definitive diagnosis is via histopathological assessment<sup>8</sup>.

Management of mucormycosis involves surgical debridement, restoration of immunity and medical therapy targeted at the fungus. Aphotericin B is the drug of choice for the medical management dose adjusted to the patients condition and weight.

Large defects resulting from surgery require precise and predictable reconstruction with local or distant soft tissue / hard tissue flaps and grafts. For maxillary defects, reconstruction with buccal fat pad allows for two layers closure and is easy simple , predictable without any significant co- morbidities. Bony and prosthetic rehabilitation is always challenging but is always secondary to control of infection.

### REFERENCES

- [1]. Fogarty C, Regennitter F, Viozzi CF. Invasive fungal infection of the maxilla following dental extractions in a patient with chronic obstructive pulmonary disease. J Can Dent Assoc 2006;72:149-52
- [2]. Leitner C, Hoffmann J, Zerfowski M, Reinert S. Mucormycosis: Necrotizing soft tissue lesion of the face. J Oral Maxillofac Surg. 2003;61:1354–8
- [3]. Lee DG, Choi JH, Choi SM, Yoo JH, Kim YJ, Min CK, et al. Two cases of disseminated mucormycosis in patients following allogeneic bone marrow transplantation. J Korean Med Sci. 2002;17:403–6
- [4]. Perusquía-Ortiz AM, Vázquez-González D, Bonifaz A. Opportunistic filamentous mycoses: Aspergillosis, mucormycosis, phaeohyphomycosis and hyalohyphomycosis. J Dtsch Dermatol Ges 2012;10:611-21
- [5]. Jayachandran S, Krithika C. Mucormycosis presenting as palatal perforation. Indian J Dent Res 2006;17:139-42.
- [6]. Garg R, Gupta VV, Ashok L. Rhinomaxillary mucormycosis: A palatal ulcer. Contemp Clin Dent 2011;2:119-23
- [7]. Tobón AM, Arango M, Fernández D, Restrepo A. Mucormycosis (zygomycosis) in a heart-kidney transplant recipient: Recovery after posaconazole therapy. Clin Infect Dis 2003;36:1488-91
- [8]. Jung SH, Kim SW, Park CS, Song CE, Cho JH, Lee JH, *et al*.Rhinocerebralmucormycosis: Consideration of prognostic factors and treatment modality. Auris Nasus Larynx 2009;36:274-9
- [9]. A. Maxillary necrosis by mucormycosis. A case report and literature review. Med Oral Patol Oral Cir BucalAuluck 2007;12:E360-4.
- [10]. Yeh CJ. Application of the buccal fat pad to the surgical treatment of oral submucous fibrosis. Int J Oral Maxillofac Surg 25:130, 1996
- [11]. Shah RJ, Katyayan MK, Katyayan PA, Chauhan V. Prosthetic rehabilitation of acquired maxillary defects secondary to mucormycosis: Clinical cases. J Contemp Dent Pract 2014;15:242-9.

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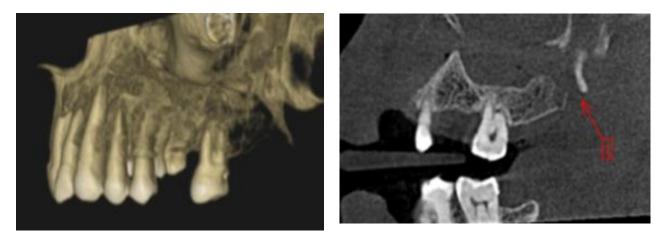


Figure 1a- 3d reconstruction view of left maxilla showing altered gross bony archietecture- osteomyelitis left maxilla

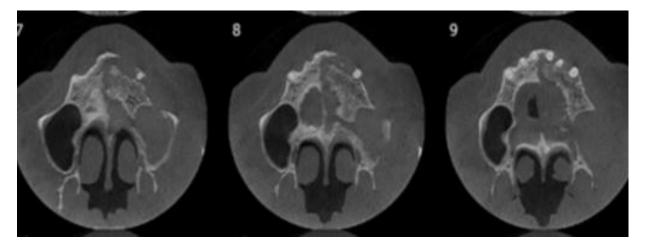


Figure 1b,c- saggital and axial images on cbct showing osteomyliticappearence of the Bone



Figure 2: Intra oral image of swelling involving left maxilla



Figure 3: - Intra oral image showing dirty greenish grey appearance of bone left maxilla



Figure 4 - After sequestrum removal and debridement



Figure 5 : Two layered closure with Buccal fat pad and Buccal myo-mucosal flap



Figure 6- Removed necrotic bone specimen



Figure 7: 1 month Post operative image

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