Case Report

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A Rare Case of Mucormycosis in Oral Cavity: A Case Report

Abstract:
Maxilla is one of the facial bones with rich vascular supply. Necrosis of maxillary bone is rare and may occur due to infection, trauma and rare metabolic disorders. Maxilla is essential bone which forms the roof of oral cavity. Mucormycosis is highly lethal opportunistic fungal infection, which affects especially in diabetic and immunocompromised patients.It is caused by filamentous fungi of the class zygomycetes. In this article we report a case of 64-year-old male patient with unknown diabetes and had undergone extraction of maxillary tooth 6 months back and had reported with unknown fungal infection.

Keywords: Necrosis of maxillary Bone; Mucormycosis; Fungal Infection.

Introduction :

Fungi are normally seen in soil, manure, fruits and vegetables which are the main sources of fungal infection. Spores of fungi can also be inhaled from dust or air from airconditioners.[5]

The nose and paranasal sinuses are commonly involved by infections caused by organisms entering through inhaled dust. Paltauf in 1885 was the first to describe mucormycosis. The disease is caused by a saprophytic fungus, mainly Mucor or Rhizopus, which is considered the most deadly and rapidly progressive form of fungal infection in humans.[5]

Mucormycosis is a rare opportunistic fungal infection caused by "bread mold fungi" of the genera Mucor, Absidia, Rhizopus and Cunninghamella, also collectively known as Phycomycetes, which represents the third most common angio-invasive fungal infection after candidiasis and aspergillosis and is considered as one of the most important medical complications in immunocompromised patients.[18,9]

The fungal class of Zygomycetes (Phycomycetes) can be divided into three orders: Mucorales, Entomophthorales, and Zoopagales. Organisms of the order Mucorales are responsible for the disease process "mucor." Four of the fourteen families within the order are pathogenic, with the family Mucoraceae being most significant. The most commonly isolated organisms in patients with mucor are of this family: Rhizopus, Rhizomucor (Mucor), and Absidia.[17]

Mucormycosis can manifest itself as six different syndromes: rhinocerebral, pulmonary, gastrointestinal, CNS, subcutaneous, and disseminated forms.[17]

Patients may also develop cellulitis, fever, headache, necrotic turbinates, and nasal discharge. If left untreated the diseases may spread into the brain resulting in death. Healthy individuals are rarely affected by mucormycosis and those affected have predisposing risk factors such as a history of tooth extraction, pneumonia, severe burns, and gastrointestinal or rhinocerebral infections. Mucormycosis is generally observed after the third decade of life.[13]

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Quick Response Code

The most common risk factor for mucormycosis is summarized in the table:9

Underlying disease	Therapy	Transplantation	Local condition	General condition
Leukemia	Antineoplastic agents	Solid organ	Burns	Malnutrition
Lymphoma	corticosteroids	Bone Marrow	Trauma	
Multiple Myeloma	Antibiotics	Peripheralbloodstem cells		
Neutropenia	Anti rejection agent			
Metabolic disorders	Intravenous drug abuse			
Cirrhosis				
Acute renal failure	Radiation			
	Deferoxamine			

Case Report:

A 64-year-old male patient reported to the Department of Oral and Maxillofacial surgery, MPCD&RC Gwalior with a chief complain of unhealed socket on left maxillary molar region since 6 months. Patient complains of swelling on left palatal region. History of present illness revels pain in upper left back tooth regionin the past 1-2 months pain was insidious in onset, dull gnawing, intermittent in nature and lasts for some hours and get relived on its own. There was no history of paresthesia and anesthesia in the area of interest. There was no relevant past medical history. Past dental history revealed that patient had visited a local dentist with chief complain of pain and loosening of upper back tooth region.The patient hadundergone extraction of the involved teeth 6 months back. Past history revealed that patient was smoker and use to smoke 2-3 bidi bundles daily since 40 years. General examination revealed that he had poor built and nourishment with presence of pallor and signs of anemia. The patient had lymphadenopathy without any incidence of swelling and tenderness. Intraoral examination revealed that there was missing left 1st and 2nd molars, generalized signs of periodontitis and exposed necrotic bone in the area of left maxillary alveolar region, which was by large palatal swelling present on left maxillary alveolar region which was accompanied by large palatal swelling. The swelling extends mediolaterally from maxillary molar alveolar region to midline of the hard palate. The swelling also extended anteroposteriorly from canine region to maxillary molar region on left side. The alveolar region of left maxillary molar region was covered by brownish yellow slough, deprived of soft tissue which covers whole alveolar region mediolaterally and anteroposteriorly from maxillary 1st molar region extending upto left maxillary tuberosity.

The lesion measured 6cm x 4cm in size and had denduded bone on with irregular margins and sloping edge. The brownish slough on the lesion was scrapable with evidence of bleeding and the surrounding mucosa was indurated. Soft tissue margin around the mucosa was normal with no signs of inflammation.

Based on this clinical finding and history, a provisional diagnosis of Fungal infection was made with differential diagnosis of chronic non healing ulcer, Mucormycosis, Squamous cell carcinoma, Aspergillosis, Midline lethal granuloma and Osteomyelitis.

The history of extraction and nonhealing wound with wide extensions, and osteomyelitic type of destruction of the left maxilla in the absence of systemic signs and symptoms of bacterial osteomyelitis prompted us to investigate further.
The patient was later subjected to the following biochemical and blood investigations. The hemoglobin, which was found to be 12.2 gm%, and fasting blood sugar, thus it was 160 mg/dl which prompted thus patient was asked to go for Hba1c the value was 8.1.After that patient was asked to consult a physician for blood sugar level maintainance.Prophylactic antibiotic was prescribed to the patient and recalled for biopsy.

Hard tissue Biopsy under local anesthesia was planned and specimen was obtained from the alveolar region: Patient was operated under local anesthesia with 1:80,000 adrenaline.

The biopsy report revealed:

1) Biopsy shows bony trabeculae with intertrabecular spaces showing necrotic tissue with 2types of PAS –D positive fungal hyphae, one slender and the other broad, aseptate.
2) Features are suggestive of fungal sinusitis.
   Based on biopsy report and fungal culture the final diagnosis of mucormycosis (zygomycosis or phycomycosis) was made.

Sequence of events

Frist weak:

Local surgical debridement was planned and done under local anesthesia.

Patient was draped with double layer of sterile sheet and was painted with betadine solution. Local anesthesia was given with 1:80000 adrenaline. The surgical site was exposed with No 15 BP blade. A crevicular incision was given on the left upper maxillary region from 23- 28 tooth region.

The infected bone was removed till maxillary sinus and then extraction of involved tooth was done. Then that area was closed in layer with 3-0 vicryl suture.

The patient was advised for mouth rinsewith 2% diluted hydrogen peroxide and Betadine® mouthwash (povidone-iodine USP 1.0% w/v) 3 times a day after meals for 15 days. Medical treatment included use of antifungal drugs such as posaconazole 300 mg, 2 times a day after meals for 15 days. Anti-inflammatory& Analgesic drug such as aceclofenac 100 mg,serratiopeptidase in combination withparacetamol 325 mg, 2 times a day (6–8 hourly), was also given for 5 days. The patient was recalled after 15 days. He was also referred to a general physician for evaluation of diabetes.

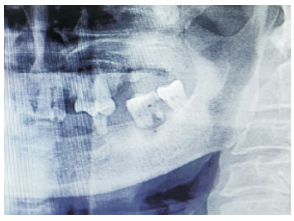


Figure 1: Orthopantomogram

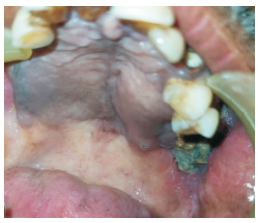


Figure 2: Intraoral Photograph Showing Necrotic Bone On Left Alveolar Ridge

SURGICAL PATHOLOGY REPORT @

SPECIMEN	:	Maxillary lesion biopsy.
CLINICAL HISTORY	:	Mucormycosis
GROSS	:	Received 1 bony tissue bit measuring 0.7 x 0.6 x 0.5 cm.
MICROSCOPY & IMPRESSION	:	**Maxillary lesion biopsy :** **1. Biopsy shows bony trabeculae with intertrabecular spaces showing necrotic tissue with 2 types of PAS-D positive fungal hyphae, one slender and the other broad, aseptate.** **2. Features are suggestive of fungal sinusitis.**
ADVISED	:	Fungal culture.
HISTOPATH NO	:	[546968 : Entire tissue -decal]

Figure 3: Biopsy Report

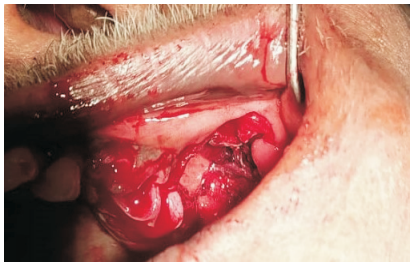


Figure 4 : Intra Operative Pic

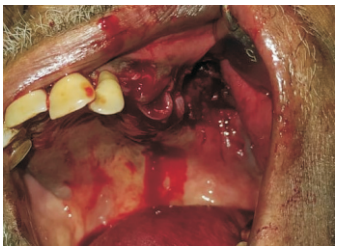


Figure 5 : Complete Excision Of Lesion

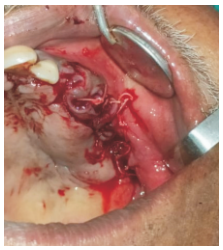


Figure 6: Closure Done With 3-0 Vicryl Suturing

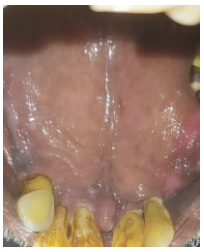


Figure 7 : Post Operative After 7 Days

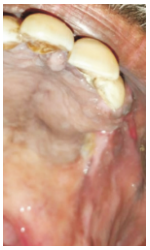


Figure 8 : Post Operative After 1 Month

Discussion:

Mucormycosis is found in fruits, soil, dust, and manure and can be cultured from the nasal mucosa of normal persons, where it may not cause clinical signs of infection. The

organisms are aerobic, but can live 2 to 5 days in vitro. Although infection usually occurs after inhalation through the nose or mouth, a skin laceration can also become an opening for mycotic entry. The terms phycomycosis and zygomycosis are occasionally used; however, mucormycosis is the most frequent term. For many years the term mucormycosis has been interchangeably used with zygomycosis in the medical literature. Currently, in line with a recent classification, we prefer to use the term mucormycosis rather than zygomycosis.[3]

According to the new taxonomic revision of the fungi, the phylum Zygomycota, subphylum Zygomycotina, and class Zygomycetes disappeared in their widely understood sense to be replaced by the phylum Glomeromycota (for arbuscularmycorrhizal fungi). Glomeromycota has a subphylum Mucormycotina, which includes the order Mucorales, which in turn are considered as etiologic factor of mucormycosis infection.[9]

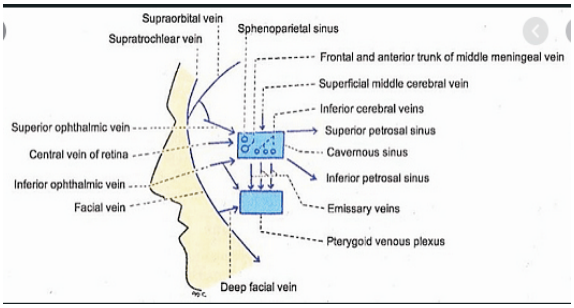
The term rhinocerebral mucormycosis should only be used if the orbit, paranasal sinuses, and brain are involved. While zygomycosis has been reported to occur in otherwise healthy individuals; a predisposing medical condition is virtually always present of these, type I diabetes mellitus is the most common and is associated with 40% of zygomycosis cases overall and 70% of patients with rhinocerebral (craniofacial) disease.[1]

Predisposing conditions for zygomycosis
Uncontrolled diabetes (particularly patients who are acidic)
Blood dyscrasias, leukaemia
Malignant conditions, lymphoma
Renal failure
Burns
Protein-calorie malnutrition
Cirrhosis
Corticosteroid
Immunosuppressive therapy
Organ transplant

Patients with mucormycosis in the early stage of disease often exhibit facial cellulitis and anesthesia, nasal discharge, necrotic turbinates, fever, headache and lethargy. If the infection extends to the nasal turbinates, the orbit can become involved. Infection can lead to proptosis, periorbital edema, chemosis, ophthalmoplegia, and loss of vision if the orbital apex becomes involved.[19]

Mucormycosis is aggressive and potentially fatal in diabetic patients because of impaired host defense mechanism and increased availability of micronutrients such as iron. Disseminated involvement of mucormycosis is observed in diabetic patients with ketoacidosis, which favors rapid proliferation of the fungus and its invasion into the orbit and cerebrum.[18, 19]

Infection of the CNS is usually attributed to direct extension from the nose or paranasal sinuses or through vascular channels, the supraorbital fissure, or the cribiform plate. If the disease invades the mouth, a black, necrotic eschar is often found in the palate, and ischemic, necrotic turbinates may be found in the nose. Because mucormycosis often invades blood vessels; infarction, necrosis, and thrombosis are the major characteristics seen. Therefore, it is possible to have CST following mucormycosis. There are several pathways through which an infection can reach the cavernous sinus. The valveless superior and inferior ophthalmic veins allow 2-way communication among the face, nasal cavity, pterygoid plexus, and the dural sinuses, including the cavernous sinus. The infraorbital and deep facial veins drain into the pterygoid plexus, which communicates with the cavernous sinus through the emissary plexus of the foramen ovale. The superficial middle cerebral vein and sphenoparietal sinus also drain into the cavernous sinus. Therefore, an infection from the maxillofacial region can potentially enter the cavernous sinus directly, indirectly, or by reverse flow from various veins.[3]



Therefore, a team of specialists including a dentist, an ENT surgeon, an ophthalmologist, a neurosurgeon, physicians and a maxillofacial surgeon is required for management of such patients.[8]

Differential diagnosis .[18]
Differential diagnosis of the lesion should include
1) Squamous cell carcinoma
2) Chronic granulomatous infection like tuberculosis,
3) Tertiary syphilis,
4) Midline lethal granuloma,
5) Wegener's granulomatosis and
6) Other deep fungal infection.[18]

In this case it was provisionally diagnosed as squamous cell carcinoma of maxillary alveolar bone because squamous cell carcinoma presents as chronic persistent ulcer with raised margins and exposure of the underlying bone, with other features like local pain, swelling, epistaxis, nasal discharge, epiphora, diplopia or numbness. However, if the lesion is associated with diabetes mellitus or immunosuppression, a diagnosis of deep fungal infection is favored, which was later confirmed after histopathologic examination in this case.

The initial medical approach to mucormycosis is to treat any underlying predisposing disorder.

To treat this following treatment protocol can be used:

Surgical:

Surgical management also should be initiated early in the course of treatment. This should involve debridement of all infected tissues. In some cases, radical resection may be required, which can include partial or total maxillectomy, mandibulectomy, and orbital exenteration.[3,16]

Medicine:

The use of amphotericin B in patients with mucormycosis has been a widely published and accepted treatment, with a survival rate of up to 72%. Although combined treatment of surgery and amphotericin B has a survival rate of 80%, 70% of those who do survive will encounter some type of functional deficit ie, blindness or cranial nerve palsy.

Rifampin has also been used to treat mucormycosis because of the reported synergistic effect of amphotericin B and rifampin against Rhizopus in vitro.

Posaconazole has variable in vitro activity against Mucorales, which is species-dependent. A study of 131 clinical isolates showed that the median MICs of posaconazole for various Mucorales species varied widely between 1.0 and 8.0 µg/mL . In laboratory animal studies, experimental infections produced by Mucor spp. were most responsive to posaconazole, while those caused by Rhizopus spp. were usually non-responsive.These data raise concerns on the clinical efficacy of posaconazole, at least in the current standard dose of 300 mg/day of extended release tablets, as Rhizopus is among the most common agents causing mucormycosis.

However, other studies have shown minimal, if any, increased activity in vivo when amphotericin B is combined with rifampin. Therefore, some authors do not recommend these therapies for general use in most patients diagnosed with mucormycosis.3, 1

Hyperberic Oxygen Therapy

Hyperbaric oxygen (HBO) has also been used to treat rhinocerebralmucormycosis. A few studies have shown that HBO has direct in vitro fungistatic activity and reduces tissue hypoxia, which may reverse the hypoxic acidosis that helps the fungus to proliferate. However, a retrospective study showed no significant difference between the effectiveness of therapy with and without HBO.[3, 1]

Conclusion:

Mucormycosis is lifethreatening fungal infection that frequently occurs in immunocompromised patients. Such as uncontrolled diabetes, renal failure, organ transplant, long term corticosteroid, immunosuppressive drugs,cirrhosis and AIDS. In diabetic patient it can get triggered by minor dental procedures such as tooth extraction. In such cases attempt should be made for early diagnosis of the disease. Any fungal infection should be treated as early as possible at least in oral cavity so that lesion does not get extended to cavernous sinus (which can further cause cavernous sinus thrombosis). Aggressive surgical excision of lesion with anti-fungal medicine(Posaconazole & Amphotericin B)is the only way for success of this kind of infection.

References:

1. Tugsel Z, Sezer B, Akalin T. Facial swelling and palatal ulceration in a diabetic patient. Oral surgery, oral medicine, oral pathology, oral radiology, and endodontics. 2004;98(6):630-6.
2. Pogrel MA, Miller CE. A case of maxillary necrosis. Journal of oral and maxillofacial surgery. 2003 Apr 1;61(4):489-93.
3. Kim J, Fortson JK, Cook HE. A fatal outcome from rhinocerebral mucormycosis after dental extractions: a case report. Journal of oral and maxillofacial surgery. 2001 Jun 1;59(6):693-7.
4. Hibbett DS, Binder M, Bischoff JF, Blackwell M, Cannon PF, Eriksson OE, Huhndorf S, James T, Kirk PM, Lücking R, Lumbsch HT. A higher-level phylogenetic classification of the Fungi. Mycological research. 2007 May 1;111(5):509-47.
5. Mucormycosis of the oral cavity ,4nne Cale Jones, DDS,' Teresa Youngblood Bentsen, DDS,h and Paul D. Freedman, DDS,' Gainesville, Fla., and Flushing, N.Y. LIN1VtRS1TY OF FLORIDA COLLEGE OF DENTISTRY AND BOOTH MEMORIAL MEDICAL CLNTtR.
6) Afroze SN, Korlepara R, Rao GV, Madala J. Mucormycosis in a diabetic patient: a case report with an insight into its pathophysiology. Contemporary clinical dentistry. 2017 Oct;8(4):662.
7. Mengji AK, Yaga US, Gollamudi N, Prakash B, Rajashekar E. Mucormycosis in a surgical defect masquerading as osteomyelitis: a case report and review of literature. Pan African Medical Journal. 2016 Mar 11;23(1).
8. Mignogna MD, Fortuna G, Leuci S, Adamo D, Ruoppo E, Siano M, Mariani U. Mucormycosis in immunocompetent patients: a case-series of patients with maxillary sinus involvement and a critical review of the literature. International Journal of Infectious Diseases. 2011 Aug 1;15(8):e533-40.
9. Mignogna MD, Fortuna G, Leuci S, Adamo D, Ruoppo E, Siano M, Mariani U. Mucormycosis in immunocompetent patients: a case-series of patients with maxillary sinus involvement and a critical review of the literature. International Journal of Infectious Diseases. 2011 Aug 1;15(8):e533-40.
10. Manjunatha BS, Das N, Sutariya RV, Ahmed T. Mucormycosis of the hard palate masquerading as carcinoma. Clinics and practice. 2012 Dec;2(1):66-8.
11. TAYLOR R, SHKLAR G, BUDSON R, HACKETT R. Mucormycosis of the oral mucosa. Archives of dermatology. 1964 Mar 1;89(3):419-25.
12. Gluskin M, Solomon MP, Gold B, Corrado ML, Berger J. Mucormycotic slough of nasal floor and palate in the anephric patient.
13) Kalaskar RR, Kalaskar AR, Ganvir S. Oral mucormycosis in an 18-month-old child: a rare case report with a literature review. Journal of the Korean Association of Oral and Maxillofacial Surgeons. 2016 Apr;42(2):105.
14. Rai S, Misra D, Misra A, Jain A, Jain P, Dhawan A. Palatal mucormycosis masquerading as bacterial and fungal osteomyelitis: A rare case report. Contemporary clinical dentistry. 2018 Apr;9(2):309.
15. Van der Westhuijzen AJ, Grotepass FW, Wyma G, Padayachee A. A rapidly fatal palatal ulcer: rhinocerebral mucormycosis. Oral surgery, oral medicine, oral pathology. 1989 Jul 1;68(1):32-6.
16. Elinav H, Zimhony O, Cohen MJ, Marcovich AL, Benenson S. Rhinocerebral mucormycosis in patients without predisposing medical conditions: a review of the literature. Clinical microbiology and infection. 2009 Jul 1;15(7):693-7.
17. Peterson KL, Wang M, Canalis RF, Abemayor E. Rhinocerebral mucormycosis: evolution of the disease and treatment options. The Laryngoscope. 1997 Jul;107(7):855-62.
18) Doni BR, Peerapur BV, Thotappa LH, Hippargi SB. Sequence of oral manifestations in rhino-maxillary mucormycosis. Indian Journal of Dental Research. 2011 Mar 1;22(2):331.
19. Kumar JA, Babu P, Prabu K, Kumar P. Mucormycosis in maxilla: Rehabilitation of facial defects using interim removable prostheses: A clinical case report. Journal of pharmacy & bioallied sciences. 2013 Jul;5(Suppl 2):S163.
20. Aggarwal P, Saxena S, Bansal V. Mucormycosis of maxillary sinus. Journal of Oral and Maxillofacial Pathology. 2007 Jul 1;11(2):66.