



## MAXILLARY MUCORMYCOSIS – REPORT OF TWO CASES.

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**ABSTRACT**

Mucormycosis is an uncommon saprophytic opportunistic fungus causing localized cutaneous infection associated with high morbidity and, on dissemination, high mortality. Mucormycosis typically develops in patients with compromised immune system as a consequence of uncontrolled diabetes mellitus, renal failure, organ transplantation, chemotherapy, severe burns, and malnutrition. Rhinocerebral mucormycosis is the most common type while occurrence on palate is rare & late. Here we present two confirmed and one suspected case of mucormycosis presenting as maxillary osteomyelitis.

**KEYWORDS :** Mucormycosis, Zygomycetes, diabetes mellitus, hyphae.**Introduction:**

Mucormycosis is the name ascribed to infections caused by usually nonseptate fungi belonging to the class Zygomycetes (Phycomycetes) of the order Mucorales (genera Rhizopus, Mucor and Absidia). Organisms of class Zygomycetes are ubiquitous saprophytic filamentous fungi having low intrinsic pathogenicity<sup>1</sup>.

It is one of the most rapidly fatal fungal infections known to man. This fungus invades the arteries, forms thrombi within the blood vessels that reduce blood supply and cause necrosis of hard and soft tissues. Once entered into the arteries, the fungus can spread to orbital and intracranial structures<sup>2</sup>. Rhinocerebral Mucormycosis is the most common type and its extension to the orbit and brain is quite usual<sup>3</sup>. It is opportunistic infections that have been recognized, in association with diabetes, hematologic malignant disease, immunosuppressive therapy, thermal burns and surgery<sup>4</sup>.

Fifty to seventy five percent patients have poorly controlled diabetes & Ketoacidosis<sup>5</sup> causing localized cutaneous infection associated with high morbidity and on dissemination, high mortality<sup>6</sup>. Mucormycosis rarely affects healthy people.

In this paper we have presented two cases of mucormycosis.

**CASE REPORT****CASE 1**

A 55 years old male patient reported for evaluation of pain and swelling in upper left jaw from past 3 months. Pain was moderate in nature and aggravated on chewing of food. Swelling was firm and extended from 23 to 26 regions. Chronic sinusitis and oral –antral communication was present since 4 years. Patient also gave history of purulent discharge and foul odor. On clinical examination discolored, yellowish necrotic bone of 1cm was found mesial to 26. Medical history revealed that patient was a known patient of chronic sinusitis and diabetes from past 4 years.

The patient was on treatment with oral hypoglycemics for 6 months but due to financial constraints the patient did not take the medication as per the prescribed dose after 6 months and avoided laboratory investigations. On radiographical examination paranasal sinus view (PNS) view showed hazy left maxillary sinus (Figure 1) and computer tomography (CT scan) revealed destruction of upper alveolus, left hard palate and walls of left maxillary antrum (Figure 2). Biopsy was advised and gross findings revealed 8 small bits of soft & hard tissues, which were creamish - grey & grey black in color. Histopathological findings revealed pseudostratified lining of maxillary sinus (Figure 4) exhibiting hyperplasia and squamous metaplasia. Dense chronic inflammatory infiltrate was also found (Figure 5) along with necrotic bone (Figure 6) and fungal hyphae at right angle (Figure 7).

**CASE 2:**

A 45 years old male patient reported with chief complain of opening

in the left palate, denudation of maxillary alveolar bone, exfoliation of maxillary teeth and Oro-nasal communication. Patient complained of discomfort in chewing of food and gave history of purulent discharge and foul odor. Clinical features revealed Oro-nasal communication and chalky denuded bone in the left maxillary anterior region extending from 11 to 16 regions.

Radiographical findings in CT scan showed thickening of sinus lining and destruction of palatal bone (Figure 3). Biopsy was advised and gross findings revealed 5 small bits of hard tissues which were grey brown in color. Histological findings revealed pseudostratified lining of maxillary sinus exhibiting hyperplasia, squamous metaplasia along with dense chronic inflammatory infiltrate. Necrotic bone and fungal hyphae at right angles (Figure 8) were also present.

**DISCUSSION:**

The first case of mucormycosis was reported by Paultauf in 1885<sup>7</sup>. Exact frequency is not known but is higher in immunocompromised patients and diabetics. Despite advances in diagnosis and treatment, a high mortality still exists. Mortality rates of 30-70% are quoted in the literature. There is no specific predisposition for sex or race. Disease is seen in all age groups<sup>5</sup>. Six different manifestations of mucormycosis based on clinical presentation and involvement of a particular body site, are: (1) rhinocerebral, (2) pulmonary, (3) cutaneous, (4) gastrointestinal, (5) central nervous system, and (6) disseminated/miscellaneous<sup>8</sup>. Few of the predisposing factors are enumerated in table 1.

**Predisposing factors<sup>9</sup> (Table 1)**

S. No.	Predisposing Factors	Common site	% cases
1	Poorly controlled insulin dependent diabetes mellitus (IDDM)	Any	60-80 %
2	Malignancies, steroid therapy, chemotherapy, neutropenic state	Any	9.7%
3	Iron or aluminum over load specially with desferroxamine therapy	Any	6.2 %
4	IV drug abuse	Central nervous system & cardiovascular system (CNS & CVS)	?
5	Protein energy malnutrition	Gastro intestina	0.5%
6	Burns & sustained skin trauma	Cutaneous	1%
7	Diarrhea & acidosis in small children	Any	7%
8	Chronic renal failure on hemodialysis	Any	7%

### Mechanism of spread -

The fungus is present in air, dust, plants and decaying matter. It adheres to the dust particles and is inhaled and deposited in the nose and paranasal sinus mucosa. The warm moist environment with the decreased immunity of the host enhances the growth of fungus. It then invades the blood vessels and causes plugging by the fungal mycelia. This leads to thrombosis and ischemic necrosis. It also acts by inducing IgE hypersensitivity, which is enhanced in a hypoxic environment<sup>10</sup>. The ability to scavenge free iron from the host is essential for the pathogenesis. Interactions between iron and fungal spores appear to be important in the rate of replication and survival of fungi in the human host<sup>8</sup>.

The patients with this condition usually present with orbital & facial pain, headache, fever, nasal discharge, visual changes & sinusitis. On examination there may be periorbital and facial swelling with sign of orbital cellulitis like proptosis and ophthalmoplegia. On nasal examination black necrotic tissue may be visible on nasal turbinates & septum. In later stages the patient becomes confused and then slips into coma. The patients are usually immunocompromised due to use of steroids or cytotoxic drugs. They have gross metabolic derangements like liver failure, renal failure, uncontrolled diabetes and ketoacidosis<sup>5</sup>.

For the diagnosis of this condition biopsy of involved necrotic tissue is indicated this shows broad nonseptate hyphae. Fungal culture and C.T. scan can be done to evaluate the extent of the disease. In our both cases 1 and 2, biopsies were taken from the left maxillary antrum. On hemotoxylin and eosin (H&E) examination they showed aseptate fungal hyphae branching at right angled. C.T scan showed obliteration of the left maxilla.

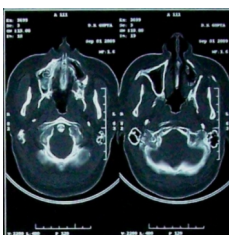
This disease is managed by treating the underlying medical disease. Correction of hypoxia, acidosis hyperglycemia & electrolyte abnormalities should be done. Any steroid or immunosuppressant medication is discontinued if possible. Renal functions should be monitored closely<sup>3</sup>. Standard therapy: conventional and liposomal amphotericin B is effective against it. The liposomal form offers less infusion site side effects and milder nephrotoxicity, however, it generally costs more. The duration of therapy varies from weeks to months depending on the site and severity of the infection. Experimental therapy: newer antifungal medications are being currently developed. The orally administered posaconazole, from the family of azoles, recently showed promising results against the mucorales species. Iron chelation is a novel adjunctive therapy that has potential role in the treatment of mucormycosis<sup>11</sup>. Rehabilitation (closure of the oronasal and/ or oroantral fistulae) can be done surgically or by construction of a prosthetic appliance<sup>3</sup>.

### RADIOGRAPHIC FINDINGS

#### Case 1- Fig 1 – PNS View

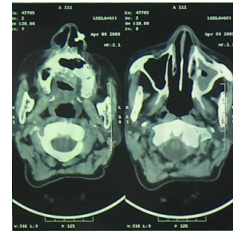


On PNS view hazy left maxillary sinus seen  
Fig 2- CT SCAN



Destruction of upper alveolus seen in left hard palate, on left side of scan and in right, walls of left maxillary antrum seen.

#### Case 2 – Figure 3 – CT SCAN



Shows thickening of sinus lining on left side of scan and destruction of palatal bone on right side of scan.

### HISTOLOGICAL PICTURES

#### CASE 1

Fig 4 (10X) - Pseudostratified lining of maxillary sinus exhibiting hyperplasia and squamous metaplasia

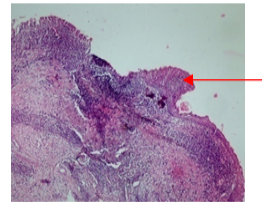


Fig 5 (40X) – Chronic inflammatory infiltrate

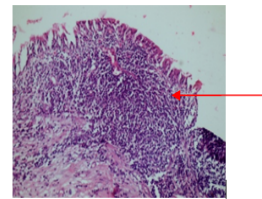


Fig 6 (40X) - Necrotic bone

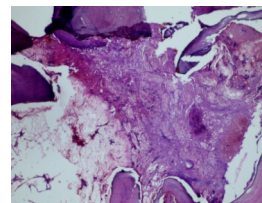
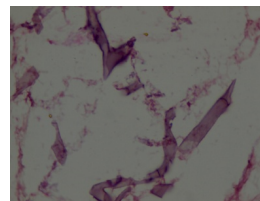


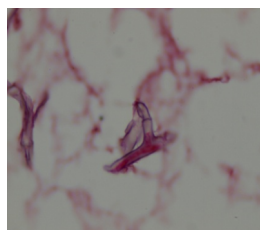
Fig 7 (40X)



H&E picture showing Fungal hyphae at right angle.

#### CASE 2

Fig 8 (40X)



H&E picture Fungal hyphae at right angle.

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