CASE REPORT

Maxillary Mucormycosis

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ABSTRACT

Mucormycosis, also known as zygomycosis; is an uncommon saprophytic opportunistic fungus caused by fungi of order Mucorales. These fungal spores are found in the soil and in decaying vegetation. Most individuals are exposed to these fungi on a daily basis, but people with weakened immune systems are more susceptible to infection. Mucorrmycosis typically develops in patients with compromised immune system as a consequence of uncontrolled diabetes mellitus, renal failure, organ transplantation, chemotherapy, severe burns, and malnutrition It causes localized cutaneous infection associated with high morbidity and, on dissemination, high mortality.. Rhinocerebral mucormycosis is the most common type while occurrence on palate is rare & late. Hereby we present a case of mucormycosis presenting as maxillary osteomyelitis.

KEY WORDS: Mucormycosis, Zygomycetes, diabetes mellitus, hyphae.

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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[1].

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[2].

Rhinocerebral Mucormycosis is the most common type and its extension to the orbit and brain is quite usual[3]. It is opportunistic infections that have been recognized, in association with diabetes, hematologic malignant disease, immunosuppressive therapy, thermal burns and surgery[4]. Fifty to seventy five percent patients have poorly controlled diabetes & Ketoacidosis[5] causing localized cutaneous infection associated with high morbidity and on dissemination, high mortality[6]. Mucormycosis rarely affects healthy people. Hereby we describe a caase of mucormycosis involving the maxilla, after obtaining the patient's consent.

CASE REPORT

A 55 years old female patient visited the out-patient department in July 2010, with the chief complaint of pain and swelling in upper jaw from past 3 months. Pain was gradual in onset, continuous with moderate intensity, throbbing type and aggravated on having of food. Patient also gave history of purulent discharge and foul odor. Patient's attendant revealed a past dental history of chronic sinusitis and oral –antral communication since 4 years. Patient was known a diabetic and was under medication since past 4 years. On intraoral examination, edentulous arches and discolored yellowish necrotic bone of was found extending from 16 to 26 regions, which was covered with slough (Fig 1 and 2). Considering the history and clinical findings a provisional diagnosis of osteomyelitis of maxilla and a differential diagnosis of midline lethal granuloma and noma /cancrum oris was thought of. Midline lethal granuloma is also a rare condition involving progressive destruction of the midface region which includes the nose, sinuses, palate and even the eyes, with history of blood discharge and stuffy nose. Noma or cancrum oris is a gangrenous disease leading to tissue destruction of the face, especially of the mouth and cheek.

The patient was subjected to the following investigations- complete hemogram, random and fasting glucose levels and computed tomography. Hemogram report revealed Hb- 10gm%, microcytic hypochromic anemia and hyperglyceamia.

Computed tomography (CT scan) revealed destruction of upper alveolus, hard palate and walls of left maxillary antrum and thickening of sinus lining on left side of scan and destruction of palatal bone on right side (Figure 3). Further before proceeding with the incisional biopsy, the patient was referred to a physician for complete evaluation and consent.

Incisional biopsy was advised and gross findings revealed 8 small bits of soft & hard tissues, which were creamish - grey & grey black in color (Fig 4). Histopathological findings revealed pseudo- stratified lining of maxillary sinus exhibiting hyperplasia and squamous metaplasia. Dense chronic inflammatory infiltrate was also found along with necrotic bone and fugal hyphae at right angle (Figure 6) suggestive of mucorrmycosis.

Patient was under a medical supervision and was prescribed with hypoglycemics and antifungal therapy of amphotheracin-B: 80mg/day BD, fluconazole 80mg/day BD. The patient was treated with bone denudation and regular dressings (Fig 5). The patient was followed up for the next 6 months and a complete denture was fabricated for both the arches.

DISCUSSION

The first case of mucorrmycosis was reported by Paultauf in 1885[7]. Exact frequency is not known but is higher in immuno-compromised 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[5]. Six different manifestations of mucorrmycosis 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[8]. Few of the predisposing factors are enumerated in table 1.

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[10]. 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[8].

Predisposing factors [9] (Table I)

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%

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 opthalmoplegia. 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[5]. 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

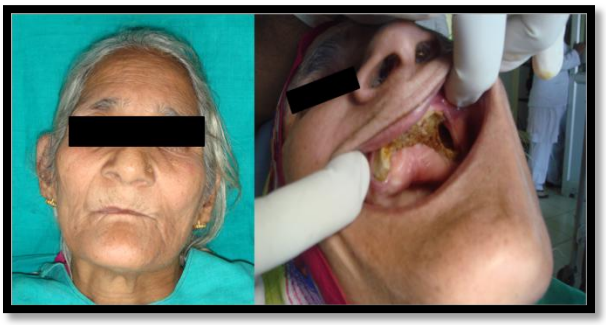

Fig 1 *– Extraoral and intraoral presentation of the patient*

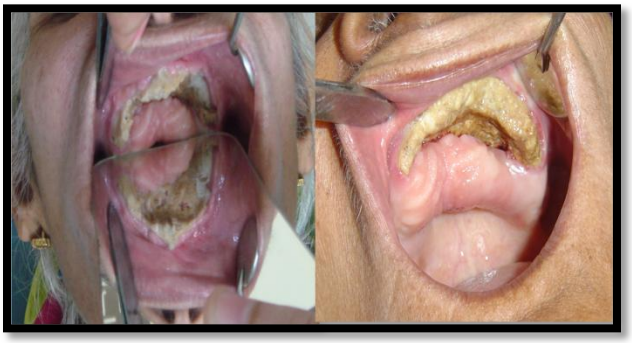

Fig 2*- Intraoral examination reveals a edentulous arches and discolored yellowish necrotic bone of was covered with slough found extending from 16 to 26 region*

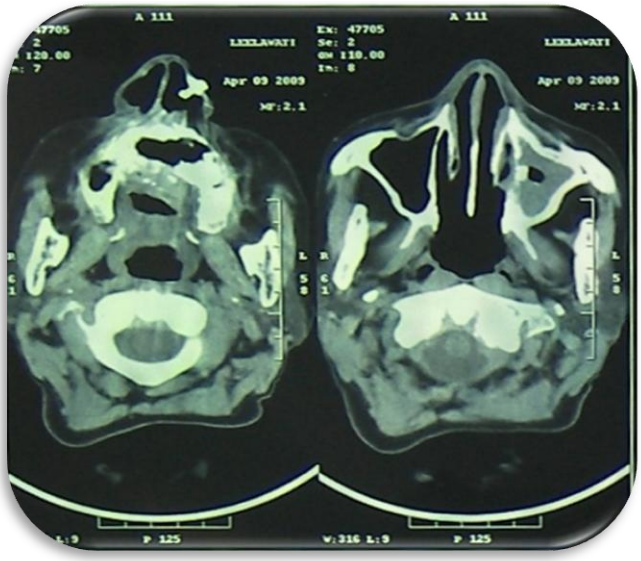

Fig 3*- CT scan reveals destruction of upper alveolus, hard palate and walls of left maxillary antrum and thickening of sinus lining on left side of scan and destruction of palatal bone on right side.*

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[3].

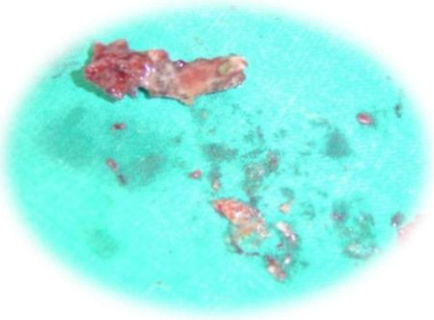

Fig 4 *- Gross findings revealed 8 small bits of soft & hard tissues, which were creamish - grey & grey black in color.*

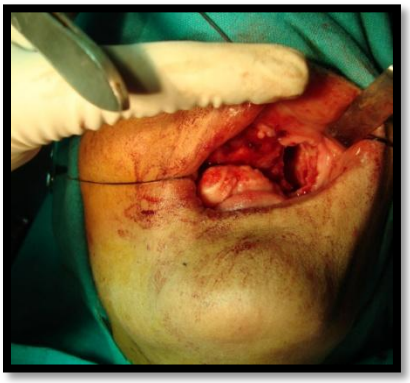

Fig 5*: Bone denudation of the maxilla was done.*

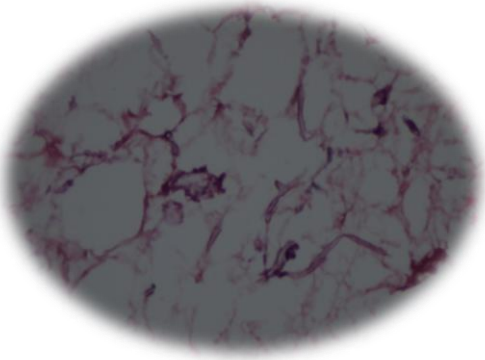

Fig 6*: (H& E stain 40x) showing Fungal hyphae at right angle.*

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[11]. Rehabilitation (closure of the oronasal and/ or oroantral fistulae) can be done surgically or by construction of a prosthetic appliance[3].

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