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## Oronasal mucormycosis in two dental patients- diagnostic dilemma and review of literature

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### Abstract

Mucormycosis (previously known as Zygomycetes) is one of the emerging mold infections across the globe, considered as an emergency in any medical setup. Being an angioinvasive disease, it tends to grow within vascular lumen and invade deeper into the tissues. Diabetes mellitus and diabetic ketoacidosis are risk factors associated with these infections like any other opportunistic fungal infections. We are presenting case series of two pediatric patients who took dental treatment few weeks back and now presented in Dental OPD with complaints of Dental pain, swelling on face. On examination patient had black discoloration and eschar formation on oral and nasal mucosa. Mycological Examination revealed as case of Mucormycosis. Patients were hospitalized, given antifungal drugs but prognosis was very poor.

**Keywords:** Oronasal, mucormycosis, dental, patients-diagnostic, literature

### Introduction

Zygomycosis is an opportunistic fungal infection caused by class Zygomycetes such as the Genera Rhizopus, Rhizomucor, Lichthemia, Mucor etc. and may present clinically as cutaneous, subcutaneous, systemic and rhino cerebral or rhino orbital infections [3]. Zygomycetes thrive in a highly acid condition that has rich carbohydrate; therefore, a diabetic ketoacidosis person has a more risk of quick invasion [4]. Cases are commonly observed whenever there is any breach in aseptic precautions [5].

### Case Report 1

10 years old, female child was brought in Dental OPD with complaints of toothache and swelling on face since last 2-3 months. Patient gave history that she took dental treatment few weeks back in a private clinic. On oral examination there was blackening in oral mucosa and child was found to be diabetic. Patient was referred for Pediatric and ENT checkup and was referred with complaints of right sided suborbital swelling which was gradually increasing in size along with sinusitis and pyrexia. Her routine investigation, urine, blood and nasal scraping were taken for microbiological evaluation where on nasal scraping's broad aseptate fungal mycelium were seen. Sample was sent for KOH mount and fungus culture. In KOH mount non-septate, thick fungal mycelium were seen. On SDA Agar (A selective medium for fungus culture) after 4 days of incubation in BOD chamber, thick, cottony, wooly fungus was grown. By LPCB was identified as Mucor spp. As patient was diabetic, she was hospitalized in PICU and systemic Liposomal Amphotericin B was started along with other antibiotics and treatment. Patient was responding to treatment but after fifth day patient left hospital against medical advice (LAMA) and follow up could not be done.

### Case Report 2

7 year old, diabetic female child was brought in Dental OPD with complaints of toothache, epistaxis and pyrexia and there was swelling on face, which was increasing in size gradually. Patient gave history that she took dental treatment few weeks back in a private clinic and her tooth extraction was done. On oral and nasal examination there was blackening in oral and nasal mucosa.

Patient was given nasal packing and admitted in PICU, despite of continuous high group of antibiotics patient general condition deteriorated. Her blood, urine and nasal scrapings were received for evaluation, in urine large number of budding yeast cells were seen on culture *C. krusei* was isolated, in nasal scrapings large number of fungal mycelium were seen and on culture *Rhizopus* spp. was grown. Patient was hospitalized in PICU and systemic Liposomal Amphotericin B was started. Despite of Liposomal Amphotericin B, patient deteriorated and left hospital against medical advice (LAMA) as attendants wanted to shift patient in a higher centre, but patient passed away when she was going to higher centre in a Ambulance.

### Case Number 1



Fig 1, 2, 3, 4: Culture growth on SDA

### Case Number 2



Fig 5, 6: Fungus Culture growth on SDA

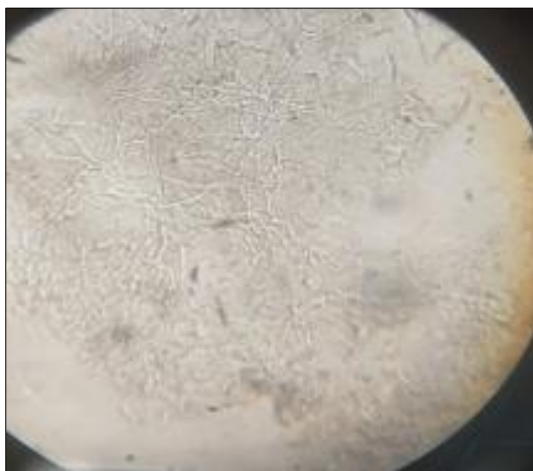


Fig 7: Fungal Hyphae seen in KOH MOUNT

### Discussion

Mucormycosis (previously known as Zygomycetes) is one of the emerging mold infections across the globe, acquired by spore inhalation, particularly prevalent in Asia [3, 5]. This is usually considered as an emergency in any medical setup. Being an angioinvasive disease, it tends to grow with in vascular lumen and invade deeper into the tissues. The risk factors associated with these infections are like any other opportunistic fungal infections, but diabetes mellitus is the most prominent one, particularly the state of diabetic ketoacidosis. If a person with diabetes should develop any of these symptoms, medical attention should be sought as quickly as possible. This fungal infection carries a high mortality rate (25-80%) dependent upon which area of the body is being attacked.

There are six clinical types of mucormycosis: Rhinocerebral, Pulmonary, Gastrointestinal, Cutaneous, Renal and Disseminated mucormycosis. Out of these Rhinocerebral is the commonest one and if not managed immediately, proves to be fatal. The incidence of pulmonary and gastrointestinal disease has enormously increased over the year. The disease among combat-trauma cases has been reported particularly by *Saksenea erythrospora*, *Apophysomyces variabilis* and *Actinomyces elegans var. kuwaitiensis* [6].

- Rhinocerebral mucormycosis affects the sinuses and the brain, and is the most common of the Mucormycosis infections [7]. What may begin like a normal sinus infection progresses to infect the eye socket, and also inflames the cranial nerves? Blood clots may develop in the vessels leading to the brain [8, 9 10]. Symptoms include fever, swelling of the eye area, pain, pus discharge from the nose, and nasal scabbing.
- Pulmonary mucormycosis involves the lungs and resembles pneumonia. If left untreated, this swiftly advancing infection could spread throughout the chest, heart and brain. The patient may have difficulty breathing, a fever, coughing, and may begin to cough up blood.
- G.I.T. mucormycosis present as pain abdomen and hematemesis.
- Renal mucormycosis is unique clinical entity, where in young individuals present with flank pain, fever and oliguria without any underlying risk factors.
- Cutaneous mucormycosis is presenting as necrotizing fasciitis, among immunocompetent individuals, particularly after the intramuscular injections [11]. The primary cutaneous disease should be suspected when ulcerative, rapidly progressive necrotic lesions develop within a few days of traumatic injury specially when there is no response to routine antibacterial therapy in an otherwise healthy individual.

### Diagnosis of Mucormycosis

Earlier the diagnosis of mucormycosis used to be established during postmortems but now antemortem diagnosis is quite possible due to substantial awareness among the clinicians. The index of suspicion of this disease should be kept very high by clinicians because most of such cases are invariably confused with the bacterial causes, thereby specimens are not sent for mycological investigations. The final diagnosis is established by conventional direct microscopy using KOH and Calcofluor white wet mounts; histopathology by H&E, PAS and GMS,

which reveals sparsely septate, broad, ribbon-shaped hyphae with right-angled branching in the tissue section with thrombosed vessels [12]. The inflammatory response is quite variable but most histopathological descriptions show acute inflammation with necrosis. The tissue morphology is similar in all mucormycosis hence exact etiological diagnosis can only be made when it is grown on culture media. Moreover, differentiating from similarly looking disease like basidiobolomycosis (*Basidiobolus ranarum*) and pythiosis (*Pythium insidiosum* & *P. aphanidermatum*) is also pertinent [6]. Wherever, culture is not feasible, molecular techniques like PCR, roll circle amplification (RCA), are useful in screening the mucormycetes [13].

### Management of Mucormycosis

The role of dentists is critical because mucormycosis primarily occurs around rhinomaxillary or rhinocerebral areas involving facial tissues, palate, alveolar bone, and mandibular bone. Therefore, dental professionals should be mindful of symptoms of mucormycosis. In addition to palatal lesions, atypical symptoms such as sinus pain, facial pain, unanticipated odontalgia of otherwise sound teeth, or patient deterioration after dental therapeutic interventions should alert clinicians to seek confirmation of the diagnosis and promptly start optimal treatment [14]. The illness usually progresses rapidly in debilitated or immunocompromised patients. Recent rise in the incidence of rhinomaxillary mucormycosis has been noticed and has become an emergency presume. it is necessary for a dental.

Practitioner to be alert to initial signs and symptoms of this disease, specifically when evaluating the high-risk patients. Patient management depends upon tackling the underlying risk factors i.e. diabetes mellitus, extensive surgical debridement of the necrosed tissue combined with antifungal therapy. Liposomal amphotericin B is the ideal one and is available as intravenous as well as topical ointment [15]. Besides this, oral posaconazole is the only azole derivative indicated for the treatment of this disease. A timely diagnosis entailing proper treatment has very good prognosis.

### Conclusion

Mucormycosis is an aggressive, frequently fatal invasive fungal infection that can develop in patients with a number of predisposing conditions. In the susceptible patient, it can be triggered by minor interventions. Expedient diagnosis, systemic amphotericin B therapy, aggressive surgical debridement and optimal medical management are critical for patient survival.

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