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# Mucormycosis – A Rare Case Report

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***Abstract:*** *Mucormycois is a devastating opportunistic fungal infection that occurs mostly in diabetics and immunocompromised patients including those with hematologic malignancies. Neutropenia induced by bone marrow suppression in leukemic patients make them a significant risk for opportunistic fungal infection. We report a case of mucormycosis in a 17 yr old male patient, under chemotheraphy for Acute myeloid leukemia. Diagnosis was made by histopathologic examination and confirmed by special stain which revealed the characteristic appearance of the fungus.The present case highlights the importance of considering mucormycosis even in patients with mild immunosuppression.*

***Keywords:*** *Phycomycosis, Acute Myeloid Leukemia*

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## I. Introduction

Mucormycosis is an opportunistic deep fungal infection caused by mucorales including Mucor, Absidia, Rhizopus and Cunninghamella, primarily affecting patients with decreased immune survillience[1]. Zygomycosis is the 2nd most common filamentous mycosis in patients with hematologic neoplasm and 3rd most common opportunistic fungal infection after candidiasis and aspergillosis[2,3].Mucorales are ubiquitous saprophytic organism found in soil ,manure, vegetables and as bread mould. An increased frequency of opportunistic fungal infection have been noted over the past two decades which can be due an increase in number of immunocompromised patients especially those with hematologic malignancies, AIDS, neoplastic disease, advanced age, long standing diabetes mellitus and those receiving immunosuppressive therapy[4]. Among patients with malignancy, hematologic malignancies are much more frequently associated with mucormycosis which account for less than 1 percent of patients. Literature review showed 929 cases of mucormycosis reported, of which 31 cases presented in the oral cavity[5].

## II. Case Report

A 17yr old male patient , reported with complaints of pain and swelling in the upper front tooth region. He was under chemotheraphy for Acute myeloid leukemia(AML) for the past 1.5 years . The intra oral examination showed a necrotic slough which was creamy white with brownish black in colour extending from palatal mucosal region with respect to distal aspect of 17 to mesial aspect of 24 including anterior hard palate. Provisional diagnosis of leukoplakia was made (fig 1).Grade II mobility of 21 and 22 ,Grade I mobility of 23 was noted. Laboratory evaluation revealed the following : total white blood cell count was 40580/mm$^3$ ; Hemoglobin 8.8 g/dl ; MCH-30.18pg ; MCHC- 36g/dl ;HCT-23fl;Platelet count -27,000/mm$^3$. Intra oral periapical radiograph of 21, 22 region showed periodontal widening and periapical radiolucency . Incision biopsy was taken and microscopic section showed granulation tissue composed of fragments of eosinophillic rod like structures of varying length which resembled broad, non-septate hyphae of mucor species with branching at right angle , degeneratd muscle bundles ,diffuse inflammatory cell infiltrate ,thickened vascular channels filled with RBC's . Hyphal structures are seen invading the vessels and within the lumen and a histopathologic diagnosis of fungal infection was made (fig 2). Special stain with Grocott-Gomori's Methenamine silver showed numerous ribbon like non-septate hyphae with branching at right angles (fig 3). Thus a final diagnosis of mucormycosis was made.

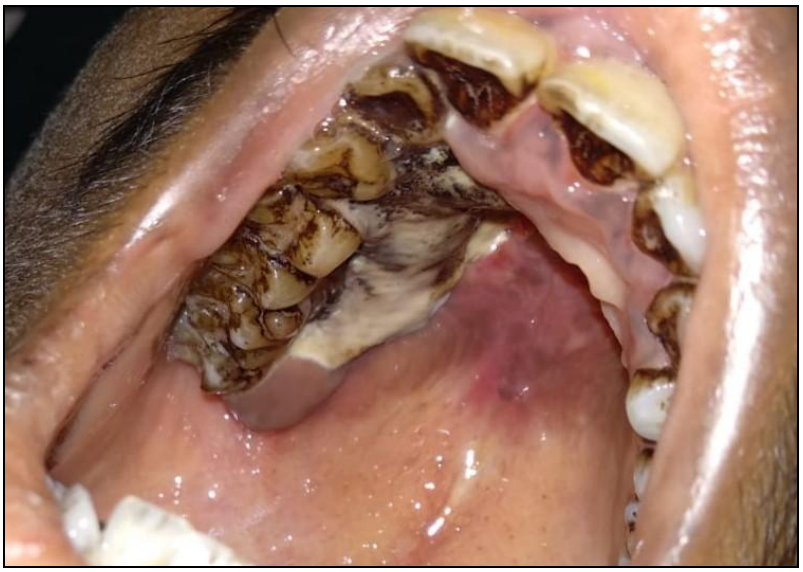

**Fig 1**: Intraoral photograph showing necrotic slough covering palatal mucosa extending from 17 to 24

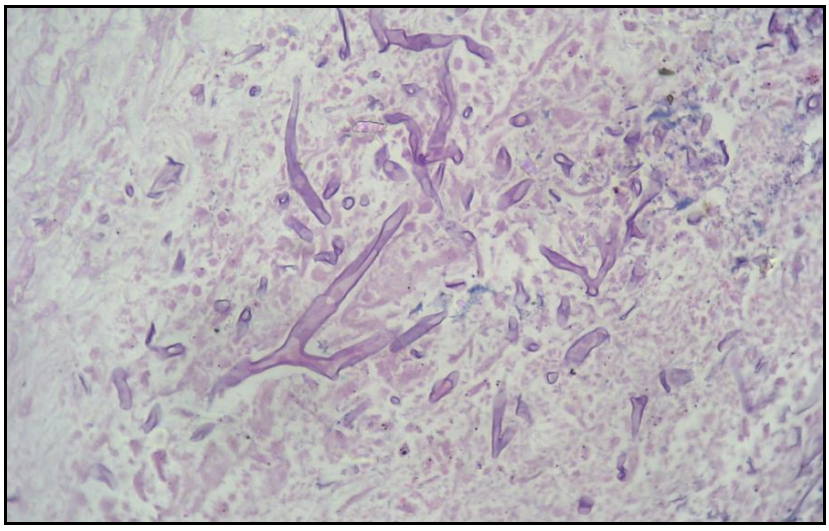

**Fig 2**: H&E showing broad, non-septate hyphae with right angle branching (40x)

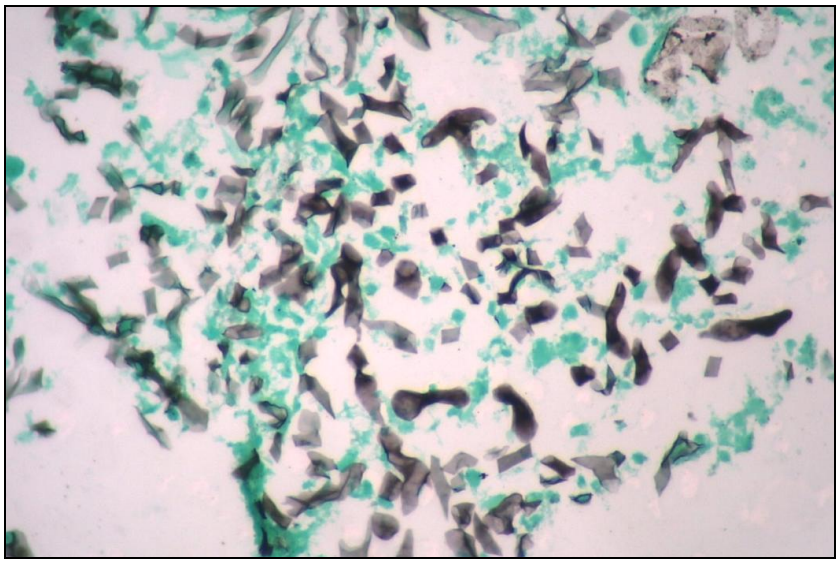

**Fig 3**: GMS showing Broad aseptate hypha with branching at right angles characteristic of mucormycosis (40x)

## III. Discussion

Zygomycosis is an umbrella term, previously named phycomycosis or mucormycosis is an aggressive opportunistic fungal infection most commonly associated with hematologic malignancies or patients with poorly controlled diabetes[6]. The reports of zygomycets affecting human beings were noted since 19th century[7]. Platauf had published the 1st documented case of mucormycosis in upper airway in 1885[2] . In 1943, Gregory et al reported three cases of advanced rhinocerebral mucormycosis in patients with diabetic ketoacidosis[8] . Frequency of Zygomycosis has been increasing over the past decades as reported by Brown et al in 2009,who identified 6.8% of affected patients at autopsy[9].

Mucorales and entomophthorales are the two orders of clinical concern,among which mucorales are most often implicated in human pathologies.These microorganisms causes acute deep mycoses with life threatening disease that mostly affect immunocompromised patients[10] .These opportunistic pathogens get entry , when an underlying disease processes causes impairement in the immune function associated with neutropenia. Eventhough a majority of cases of human zygomycosis occur due to neutrophil dysfunction induced by ketoacidosis, a growing proportion of cases can be seen in the literature where neutropenia occur by bone marrow suppression during chemotherapy in hematologic malignancies[6] .The order of Entomophthorales, causes infection mainly in immunocompetent subjects following trauma, with limited degree of invasiveness compared to Mucorales[10] .

Infection by microorganism occur by inhalation ,burn or trauma causing cutaneous zygomycosis, ingestion or direct inoculation. Inhalation of highly infective dose of spores causes initial localization in paranasal sinuses which further spread to brain leading to fatal outcome. Hematologic or severly neutropenic patients develops pulmonary zygomycosis.[11] . The principal host defense mechanisms against mucormycosis occur through mononuclear and polymorphonuclear phagocytes due to their production of oxidative metabolites and cationic peptide defensins, thus explains the increased incidence of the invasive fungal infections during the prolonged neutropenic period[12] . Decreased phagocytic ability of granulocyte, with increased levels of free ferric ion and acidic environment favours the growth of mucorales in diabetic patients. Higher occurance of mucormycosis caused by Rhizopus oryzae, is due to their abilty to produce ketoreductase enzyme ,by which they make use of patients ketone bodies[3].

Mucormycosis is usually a disease of the immunocompromised patient with a potential predisposition includes: hematologic malignancies, ketoacidosis and uncontrolled diabetes mellitus, other forms of acidosis ,cancers, immunosuppressive therapy even for a short period of time, following bone marrow transplantation, patients with congenital or acquired neutropenia, or anemia such as thalassemia or aplastic anemia, with different degrees of burns and in Individuals with acquired immunodeficiency syndrome (AIDS)[2] .Our case was a known AML patient thus predisposing for mucormycosis.

Literature review showed an unexplained predilection of zygomycosis to the male gender. The male-female ratio was between 2.4:1 and 3:1,which was confirmed by Roden and associates in their review study done in 2005which showed 65% of the reviewed cases were males[13] .The mean age was in the 4th-5th decades ,with a wide spectrum of age ranging: from neonates to the very old[14] . Our case also presented in young male patient with the lesion localized to the palate .Till now in the literature 19 cases were reported with palatal presentation limited to oral cavity, of which 12 cases were seen associated with AML[15].

According to Eisenberg et al mucormycosis can be classified into six clinical variants Rhinocerebral, Pulmonary, Cutaneous, Gastointestinal, CNS and Disseminated. Rhinocerbral being the most common and gastrointestinal being the rarest form. Rhinocerbral form accounts for one third to half of all cases[12] . It is seen primarily in uncontrolled diabetic patients. The primary route being inhalation or hematogenous or lymphatic spread. The infection involves the maxillary sinuses, which spread to adjascent structures including nose, orbits, brain, cranial nerves, hard and soft palates, mandible and the rest of the face[16] .The lesion clinically present as black eschar or frank tissue necrosis. Severe form of rhino-cerebral zygomycosis present with periorbital cellulitis associated with proptosis, ophtalmoplegia, and visual loss . Lesion involving hardpalate may cause palatal perforation.

Second most common form being Pulmonary mucormycosis, present with following symptoms which includes fever, cough, sputum production, dyspnea, hypoxia, chest pain and hemoptysis. Mucormycosis can cause lobar consolidation, multiple disseminated lung nodules, fungal ballor mycotic abcess formation[17] . Pulmonary zygomycosis can disseminate to the pleural space, chest wall, or the mediastinum where it can cause catastrophic rupture of large vessels

Disruption of intact skin caused by skin maceration, burns or trauma cause Cutaneous zygomycosis[18] . It usually carries a better prognosis than other forms until the fungal reaches deeper into muscle, bone or fascia where it causes severe necrosis with very high mortality rate.

Gastrointestinal zygomycosis is the rare form of zygomycosis. It occur either by ingestion of zygospores especially in the malnourished and alcoholics, or can occur secondary to trauma[19].The presenting

symptoms include fever, abdominal pain and bloating, nausea and vomiting, bleeding or bowel perforation. It can be observed in the stomach where it can cause ulceration, bleeding or perforation .

Disseminated zygomycosis has the worse prognosis, with a 100%mortality rate. Disseminated involvement occurs in intravenous drug users and diabetcs with ketoacidosis .The ability of fungal hyphae to invade the blood vessels and there by enter into blood stream can cause subsequent dissemination to other organs such as brain, heart, lungs, and kidneys. The diagnosis is suspected in the presence of disseminated organ infarction and necrosis[20]. Mucor hyphae form thrombi within the blood vessels that reduce vascularity to the tissues and cause infarction and necrosis which forms the clinical hallmark of the disease.[9]

Diagnosis of mucormycosis is made by biopsy of infected tissues, showing wide, ribbon-like aseptate hyphal elements that branch at right angle which distinguish it from Aspergillus hyphae which are septate and bifurcate at acute angles[21] .The treatment of mucormycosis has been proven challenging due to the limited available treatment modalities. The appropriate antifungal therapy along with surgical intervention is considered as a successful management for mucormycosis, eventhough both risk and benefits of surgical treatment should be focused in patients with hematologic malignancies.

## IV. Conclusion

Mucormycosis in the present case represent the one that limited to oral cavity unlike other forms which showed rapid progression . An accurate histopathologic diagnosis with early antifungal theraphy is the cornerstone of successful treatment of zygomycosis thus results in a better outcome.

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