

## Palatal Mucormycosis Masquerading as Bacterial and Fungal Osteomyelitis: A Rare Case Report

### Abstract

Mucormycosis is an acute, fulminating, fungal disease that frequently involves oral, cranial, and facial structures. It is an opportunistic fatal infection which occurs in debilitating and immunosuppressive states. This report documents a rare case of localized maxillary mucormycosis in a patient with uncontrolled diabetes, with emphasis on early and prompt diagnosis of the same.

**Keywords:** *Diagnosis, mucormycosis, palatal ulceration*

### Introduction

Fungi are normally seen in soil, manure, and fruits. The nose and paranasal sinuses are commonly involved by infections caused by organisms entering through inhaled dust.<sup>[1]</sup> Usually, mucor fungi responsible for mucormycosis are nonpathogenic but may present as an opportunistic infection in patients on cytotoxic drugs, diabetes mellitus, renal failure, leukemia, cirrhosis, and severe burns.<sup>[2]</sup> The organisms proliferate in tissues, vessel walls, perineural spaces, and have a predilection for muscle layers of arteries, veins, and lymphatics, causing thrombosis and infarction. This causes invasion of the organism into the orbit and cranial vault leading to meningoencephalitis and cavernous sinus thrombosis, which may present as facial pain, swelling, orbital cellulitis, proptosis, loss of vision, ophthalmoplegia, necrosis of nasal turbinates and palate as well as osteomyelitis of facial bones.<sup>[3]</sup> Most reported cases of mucormycosis affecting facial region have been disseminated with orbital and cranial involvement. This is a rare localized form of mucormycosis.<sup>[4,5]</sup> We report this case to make clinicians aware of the clinical presentation of this disease for early diagnosis and treatment of this fatal infection.

### Case Report

A 57-year-old male patient reported to our department of oral medicine and radiology with a complaint of painful ulcer on the

left side of the palate and unhealed socket for 2 months. History of the present illness revealed pain and discharge from the palate for 2 months. Pain was insidious in onset, dull, gnawing, intermittent in nature, lasts for hours, and relived on its own. Pain was associated with discharge. There was no history of paresthesia or anesthesia in the region of area of interest. Past medical history revealed that the patient was a known diabetic for 5 years and not on any medications. Past dental history revealed that the patient reported to a local dentist 2 months ago with tender swelling of gums and loosening of teeth. The condition was diagnosed as advanced periodontal disease with severe bone loss and periodontal abscess involving the left maxillary canine. The patient had undergone localized oral prophylaxis followed by extraction of the involved tooth under local anesthesia and oral prophylaxis was later completed. He was advised to maintain good oral hygiene and replacement of missing tooth once the socket heals. Personal history revealed that the patient was a smoker and smoked 2–3 bundles of bidi for 35 years. General examination revealed that he had poor built and nourishment with the presence of pallor and signs of anemia. The patient had submandibular lymphadenopathy without any evidence of tenderness or swelling extraorally. Intraoral examination revealed missing left maxillary canine, generalized signs of severe periodontitis, and exposed necrotic maxillary bone in the palatal area with a necrotizing ulcer. The ulcer extends mediolaterally from

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the crest of alveolar bone at canine region toward the midline of hard palate and anteroposteriorly from palatal gingival region of the left maxillary central incisor to the left maxillary second molar. The ulcer was covered by brownish yellow slough, deprived of soft tissues. The ulcer measured around 3 cm × 5 cm in size and had denuded bone in its floor with irregular margins and sloping edges. The slough was scrapable with evidence of bleeding and the surrounding mucosa was indurated. Soft-tissue margins around the necrotic bone appeared normal with no signs of inflammation or purulent discharge [Figure 1]. Based on clinical findings and history, a provisional diagnosis of deep fungal infection and a differential diagnosis of chronic nonhealing ulcer, mucormycosis, and midline lethal granuloma were made.

The history of extraction, uncontrolled diabetic status 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 such as hemoglobin, which was 10.2 gm%, and fasting blood sugar, which was 183 mg/dl. Glycosylated hemoglobin value was found to be 8.2% and was suggestive of diabetes mellitus. Cytological smears were prepared from the lesion which revealed numerous fungal hyphae intermixed with bacterial colonies [Figure 2]. Biopsy under local anesthesia was planned and specimen was obtained from the palatal mucosa and, on histopathological examination, necrosed bony trabeculae showed empty bone lacunae [Figure 3]. However, deeper sections in histopathological examination on lower and higher magnification (×10 and × 40, respectively) revealed cellular connective tissue stroma with large nonseptate (aseptate), with randomly spaced branches and nonparallel sides and broad fungal hyphae along with few areas of necrosis and focal areas of mixed inflammatory cell infiltrate [Figure 4]. Periodic acid–Schiff (PAS) and Grocott's silver methenamine (GSM) staining further showed broad, thick-walled infrequent nonseptate with randomly spaced branches and nonparallel sides indicative of mucormycosis [Figures 5 and 6]. Based on histopathological findings, a final diagnosis of mucormycosis associated with candidal osteomyelitis of the palate was made.

### Sequence of events

First and 2<sup>nd</sup> weeks: Local surgical debridement was planned and done under local anesthesia. The patient was advised for mouth rinse with 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 antibacterial drugs such as amoxicillin 500 mg and potassium clavulanate 125 mg, 2 times a day after meals for 5 days. Anti-inflammatory drug such as aceclofenac 100 mg in combination with

paracetamol 325 mg, 3 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 and treatment of the same.



Figure 1: Intraoral pretreatment picture

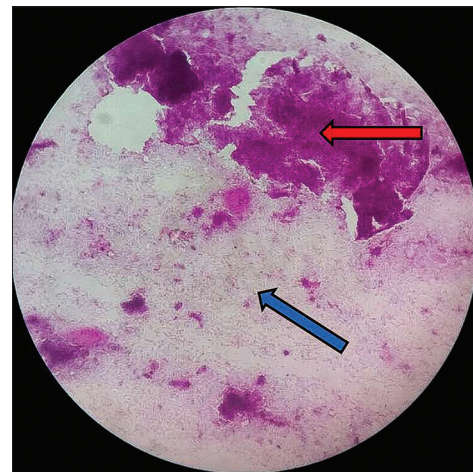


Figure 2: Photo micrograph (×40 magnification) showing bacterial colonies (red arrow) and candidal (fungal) hyphae (blue arrow)

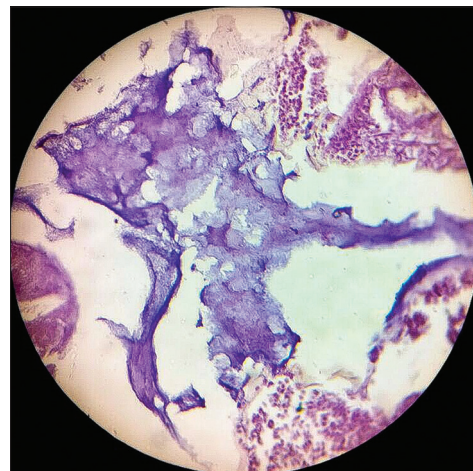


Figure 3: Necrosed bony trabeculae showing empty bone lacunae



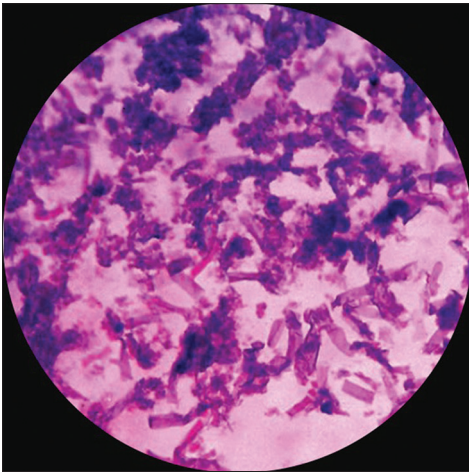


Figure 4: Deeper sections containing cellular connective tissue stroma with large nonseptate (aseptate), broad fungal hyphae suggestive of mucormycosis along with few areas of necrosis and focal areas of mixed inflammatory cell infiltrate

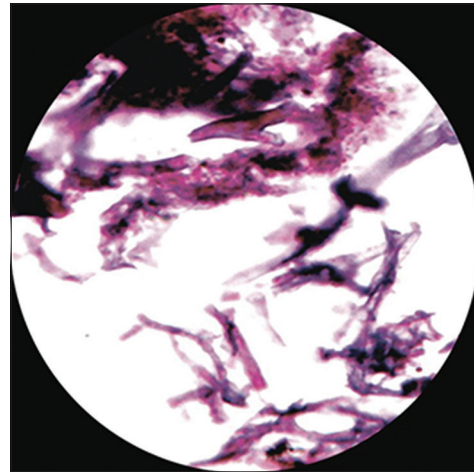


Figure 5: Periodic acid-Schiff staining showing broad, thick-walled infrequent nonseptate with randomly spaced branches and nonparallel sides of fungal hyphae

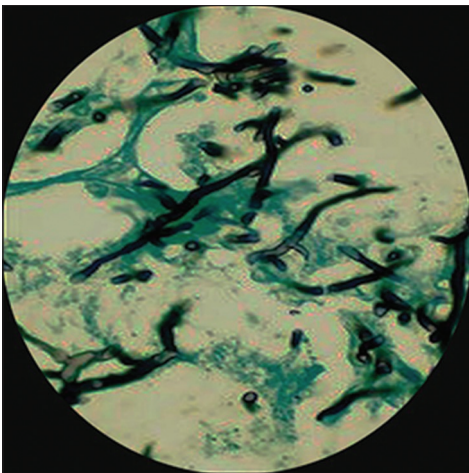


Figure 6: Grocott's silver methenamine staining showing mucormycosis

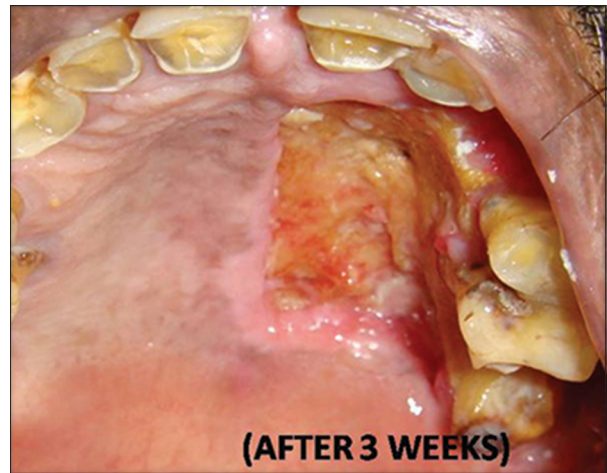


Figure 7: Healing after 3 weeks



Figure 8: Healing after 10 weeks



Figure 9: Healing after 18 weeks

Third–10<sup>th</sup> weeks [Figure 7]: The patient was advised to use clotrimazole 1% w/w mouth paint, 3–4 times a day, for

local/topical application and ketoconazole 200 mg 1 tablet daily for 7 weeks for systemic use. He was also advised to

take supportive therapy for 2 months which included 5 mg folic acid tablet once daily and iron therapy available as “Syrup Haem Up” 30 ml daily before meals. The patient was followed up after 8 weeks.

Eleventh–13<sup>th</sup> weeks: Surgical debridement was repeated and the same treatment was continued for 3 more weeks.

Fourteenth–17<sup>th</sup> weeks: A remarkable healed alveolar mucosa was observed in the regions of 23 and 24 with regeneration of palatal mucosa in the posterior part of the denuded bone and area of denuded bone reduced up to 50% [Figure 8]. The patient was again recalled after 4 weeks.

Eighteenth week: Significant healing of denuded mucosa and bone was seen and the lesion healed up to 80% with no fresh complaints [Figure 9]. The patient was advised to continue the same treatment for the next 2 weeks so that the total duration of treatment becomes 20 weeks (5 months). Unfortunately, the patient was lost to follow-up after this visit.

## Discussion

Paltauf in 1885 was the first to describe mucormycosis.<sup>[1]</sup> Mucormycosis is a rare, rapidly progressive, and often fatal opportunistic infection caused by fungi belonging to the class zygomycetes/phycomycetes, order mucorales. It represents the third most common angio-invasive fungal infection after candidiasis and aspergillosis. These fungi are ubiquitous, usually harmless, and become pathogenic in humans under certain conditions.<sup>[5]</sup> The common predisposing conditions causing mucormycosis are as follows:<sup>[6]</sup>

1. Uncontrolled diabetes mellitus is the single-most common predisposing factor, especially when associated with ketoacidosis. Such patients have decreased granulocyte phagocytic ability with altered polymorphonuclear leukocyte response. *Mucor* thrives in an acidic pH and glucose-rich medium. Hyperglycemia enhances fungal growth and impairs neutrophil chemotaxis, while lactic acidosis decreases phagocytosis
2. Immunocompromised states such as deficient T-cell immunity. Reports have suggested that the ability of serum of immunocompromised patients to inhibit *Rhizopus in vitro* is reduced, which makes them suitable hosts to opportunistic fungal infections
3. Hematological malignancies such as leukemia and lymphoma
4. Bone marrow transplants or organ transplantations
5. Use of drugs such as corticosteroids and deferoxamine therapy
6. Severe and prolonged neutropenia
7. Immature babies or babies with low birth weight
8. The infection may also occur due to inhalation, ingestion, or contamination of traumatized mucosa such as ulcer or extraction socket by fungal spores.<sup>[6]</sup>

Our patient also had a history of extraction of left maxillary canine and uncontrolled diabetes that could have been a possible reason for this infection to occur.

Patients with mucormycosis usually present with malaise, headache, facial pain, swelling, and low-grade fever. Mucormycosis usually occurs in one of the following four clinical forms – rhinocerebral, pulmonary, gastrointestinal, or disseminated. Rhinocerebral form is further subdivided into rhino-orbito-cerebral form which is invasive and may involve the ophthalmic and internal carotid arteries and rhino-maxillary form which involves the sphenopalatine and greater palatine arteries, resulting in thrombosis of the turbinate and necrosis of the palate.<sup>[3,7]</sup> Nearly 40%–70% of all reported cases manifest signs and symptoms involving facial and oral tissues. A black necrotic eschar is the most characteristic and pathognomonic lesion. Other sites of oral lesions include gingiva, lips, alveolar ridge, cheeks, tongue, and mandible. Necrosis of the maxilla is usually rare due to its rich vascularity.<sup>[8,9]</sup> Our case had a similar presentation involving palate and left alveolus in relation to the left maxillary canine region.

Clinical differential diagnosis of the lesion includes chronic granulomatous infection such as tuberculosis, tertiary syphilis, midline lethal granuloma, Wegener’s granulomatosis, other deep fungal infections, carcinoma of palate, and patients on bisphosphonate therapy. Such cases present as chronic ulcers with raised margins causing exposure of the underlying bone. A malignant salivary gland tumor arising from the accessory glands of the palate can also be considered in the differential diagnosis.<sup>[10]</sup>

## Histopathological features

Mucormycosis can be cultured on Sabouraud’s dextrose agar medium but is confirmed by histopathological examination using hematoxylin and eosin (H and E), PAS, and later by Grocott’s Methenamine Silver (GSM) stains.<sup>[9]</sup> Typical histopathological picture of mucormycosis shows the characteristic ribbon-like branching, smaller width nonseptate (aseptate) fungal hyphae which are prominent and have long, acute-angled, or right-angled branching varying from 45° to 90°. As the fungus is angio-invasive, it is commonly found in close proximity with the necrotic vessel walls. Usually, tissue shows nonspecific inflammatory cell infiltrate, with necrosis and granulation tissue along with the hyphae.<sup>[4,9]</sup>

There is a close histopathological resemblance between mucormycosis and aspergillosis. Microscopically, aspergillosis has septate branching hyphae, whereas mucormycotic hyphae have smaller width and prominent acute angulations of branching hyphae.<sup>[3]</sup> In the present case, diagnosis was confirmed by histological examination of biopsy specimen using H and E, PAS, and GSM stains. Mucormycosis presents a nonspecific presentation clinically and radiographically; therefore, histopathology proves

to be the “gold standard” for its diagnosis. However, histopathological identification of mucormycosis in tissue specimens requires significant pathological expertise. Therefore, molecular confirmation by detection of fungal DNA in tissue samples by polymerase chain reaction (PCR) is a novel method that may allow improved diagnosis of mucormycosis. In particular, PCR with sequencing of the 18S ribosomal DNA of *Mucorales* species in order to diagnose this organism in clinical cases of invasive fungal infection has been expressed.<sup>[11]</sup> However, in our case, molecular confirmation through PCR could not be established due to financial constraints faced by the patient who was further lost to follow-up.

Despite early treatment, the mortality rate of patients with mucormycosis is very high, ranging from 16% to 100%. Cutaneous mucormycosis has a mortality rate of 17%, whereas rhinocerebral, pulmonary, and gastrointestinal forms of mucormycosis have mortality rates of 67%, 83%, and 100%, respectively.<sup>[8]</sup>

As the disease progresses with alarming rapidity, prompt and aggressive therapy is essential. Successful treatment of mucormycosis consists of aggressive and repeated surgical debridement of necrotic tissue, systemic antifungal therapy, and immediate control of the underlying systemic diseases.<sup>[12]</sup> Use of amphotericin B, an antifungal agent, is widely advocated in literature for treatment of this lesion, but it should be used with caution due to the risk of development of an amphotericin-induced nephrotoxicity. In case of palatal perforation, rehabilitation of the patient can be achieved surgically using free flaps or by constructing an obturator. However, our case showed no palatal perforation and was treated by surgical debridement and medical management including topical and systemic antifungal agents along with supportive care.<sup>[13]</sup>

#### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

#### Conclusion

Early and prompt diagnosis of this disease is crucial for its subsequent successful treatment. Oral physicians often

have the opportunity to make the correct diagnosis of mucormycosis and its proper referral if involving areas other than oral cavity. Proper history of the underlying systemic disease associated with this disease reduces the mortality and morbidity rate associated with this debilitating disease. This patient survived because of early diagnosis and prompt treatment.

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Nil.

#### Conflicts of interest

There are no conflicts of interest.

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