

Fungal osteomyelitis involving the maxilla with concurrent mycotic infections: A Case Report

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Abstract

Fungal osteomyelitis is a rare but potentially fatal opportunistic infection. Filamentous fungi such as *Aspergillus* and members of the *Mucorales* group primarily affect individuals with compromised immune systems. Both types of infections are marked by aggressive spread into nearby tissues. A co-infection involving both *Aspergillus* and *Mucorales* in the rhinocerebral region is extremely uncommon and typically occurs in immunocompromised patients, often resulting in high mortality. This case report discusses a rare instance of concurrent aspergillosis and mucormycosis in a patient with type 2 diabetes mellitus and also had a history of tooth extraction at site 27 with necrosis of the left palatal mucosa. Comprehensive diagnostic and interventional approaches led to a final diagnosis of mixed fungal osteomyelitis. The main objective of presenting this case is to emphasize its rarity, clinical features, and the importance of timely management for a favourable prognosis.

Keywords: *Aspergillus*, concurrent, fungal, *Mucormycosis*, osteomyelitis.

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Introduction

Fungal osteomyelitis is a rare condition that typically exhibits a slow clinical course. Nevertheless, once these infections become invasive, they can cause significant tissue destruction and morbidity.¹ *Aspergillosis* and *mucormycosis* are the second and third most common opportunistic fungal infections following *candidiasis*.² Although involvement of the orofacial region by *Aspergillus* and *Mucorales* species is rare, such infections are notably aggressive and may lead to life-threatening complications. Individuals with compromised immune systems are particularly susceptible to these opportunistic pathogens.³

The mortality rate of mucormycosis varies widely, ranging from 10% to 100%, depending on the infection site and the patient's underlying risk factors. Maxillary sinus involvement in mucormycosis is associated with a poor prognosis and a high mortality rate of approximately 46%. *Aspergillosis* carries an overall mortality rate of around 30%. Prompt diagnosis and immediate management are critical for improving patient outcomes. Effective treatment involves addressing predisposing conditions, administering appropriate antifungal medications, surgical debridement, and supportive care. Additionally, surgical or prosthetic

reconstruction plays a vital role in restoring the patient's quality of life to its pre-illness state.²

We present a case of a 52-year-old male with medical history of Diabetes Mellitus and a recent tooth extraction but no prior COVID-19 infection, who developed severe maxillary necrosis resulting from a co-infection with *Mucorales* and *Aspergillus* species.

Case Report

A 52-year-old man presented with intermittent, sharp shooting pain in upper left back tooth region since 2 months, for which he visited private dental clinic where extraction of 27 was done. Approximately one month later, he began to notice nasal regurgitation and blackening of the palatal mucosa. His medical history includes diabetes mellitus for the past five years, with a fasting blood sugar level of 281 mg/dl prior to surgery. He also has a long-standing habit of chewing pan masala (3–4 packets daily) for 10–15 years but no prior COVID-19 infection. On examination, his mouth opening was restricted to 30 mm (interincisal distance). Clinical evaluation revealed necrosis of the left palatal mucosa, nasal regurgitation, and exposed bone extending from tooth 21 to the maxillary tuberosity, crossing the midline in the anterior region. A mesiodens was noted between teeth 11 and 21. The lesion surface was rough, covered with necrotic slough, surrounded by erythematous tissue, and tender to palpation (Fig. 1). A provisional diagnosis of chronic osteomyelitis was made.

Fig 1: Necrosed left palatal mucosa



CT imaging revealed thickening of the bilateral nasal cavity walls and nasal septum. Surgical management included sequestrectomy and saucerization of the affected region under general anaesthesia (Fig. 2). A 7 x 3 cm, greyish-black, hard tissue sample was received for histopathological examination (Fig. 3).

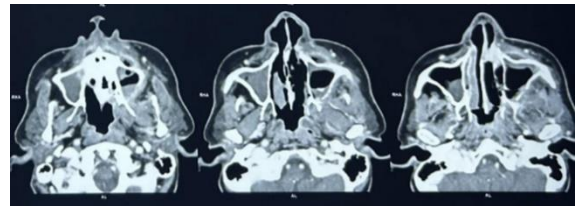


Fig 2: CT image showing thickening of the bilateral nasal cavity walls and nasal septum



Fig 3: Gross specimen

The tissue was decalcified with 5% HNO₃ and stained with Haematoxylin and eosin (H&E). Sections showed multiple tissue fragments with inflamed stromal tissue and necrotic debris. The stroma contained thick collagen bundles, a few spindle-shaped fibroblasts, and a chronic inflammatory infiltrate composed of lymphocytes, plasma cells, and macrophages. Eosinophilic hyaline fungal structures were identified, with septate hyphae branching at acute angles indicative of *Aspergillus* (red arrow in Fig. 4 a, b) and non-septate hyphae branching at obtuse angles suggestive of *Mucormycosis* (yellow arrow in Fig. 4 a, b). Fungal spores were also scattered throughout the stroma. Periodic acid–Schiff (PAS) staining confirmed the presence of

fungal elements. Most of the bony trabeculae lacked osteoblastic rimming and osteocytes. The marrow spaces were filled with necrotic debris. The final diagnosis of mixed fungal osteomyelitis involving both *Aspergillus* and *Mucor* species was made.

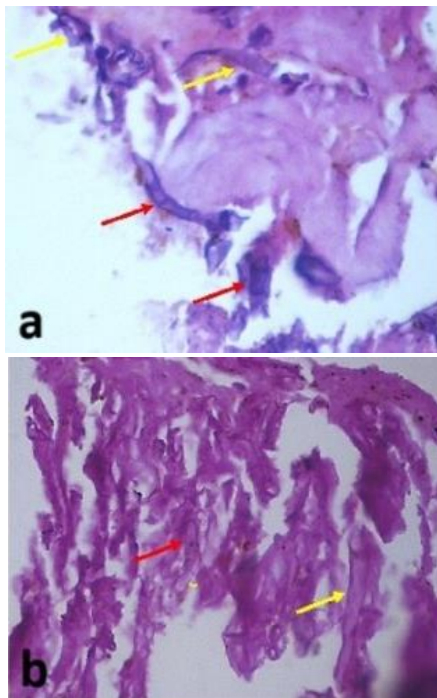


Fig 4: Photmicrograph (X100 magnification) showing eosinophilic hyaline fungal structures with *Aspergillus* septate hyphae branching at acute angles (Red arrow) and *Mucormycosis* non-septate hyphae branching at obtuse angles (yellow arrow), a. Hematoxylin & Eosin staining, b. Periodic Acid Schiff staining.

Discussion

Osteomyelitis is an inflammatory condition of bone and bone marrow content that typically arises from chronic infection. Etiopathogenesis includes surgical procedures like dental extractions, as in the current case, the patient has a history of tooth extraction in the same area as the lesion, which may have served as a potential entry point for fungal invasion, bloodstream infections, fungal infection or a contiguous infectious sites and systemic diseases. One contributing factor is reduced blood flow or stasis, which creates a favourable environment for infection.

While fungal osteomyelitis is rare and typically progresses slowly, in this case, the lesion developed unusually quickly—within just two months following the extraction. However, when fungal infections become invasive, they can be highly destructive to affected individuals as in present case.¹

Aspergillosis and *mucormycosis* affecting the orofacial region are uncommon but they are highly aggressive opportunistic infections that can have life-threatening consequences. Immunocompromised individuals are more vulnerable to such opportunistic infections. Key predisposing factors are uncontrolled diabetes and, in this case, the patient also had a medical history of poorly managed diabetes, lymphomas, leukemia, renal failure, immunosuppressive or long-term steroid therapy, organ transplant, renal failure, protein-energy malnutrition, and AIDS.³

Aspergillus and *Mucorales* are filamentous fungi capable of rapidly invading tissues and disseminating both locally and systemically in affected individuals.⁴ These organisms are widespread in nature, commonly present in decaying organic matter, including plant debris and animal waste. Multiple species exist within each genus, with *Aspergillus fumigatus* and *Rhizopus* spp. being the most frequently encountered. Infection typically occurs through inhalation of spores, though cutaneous or percutaneous transmission can also arise following skin barrier disruptions such as trauma, burns, direct inoculation, or catheter insertion and tooth extraction site.^{2,4} Infections caused by *Aspergillus* species result primarily from direct tissue invasion and the inflammatory response they provoke. In contrast, *Mucorales* infections are characterized by angioinvasion, leading to tissue necrosis as a hallmark pathological feature.⁴

In a study conducted by Niranjana et al., the highest occurrence of fungal osteomyelitis

was observed in individuals over the age of 40, which aligns with the present case involving a 52-year-old patient.³ Fungal osteomyelitis was found to occur more frequently in the maxilla than in the mandible, as in present case.^{1,3}

Typically, necrosis in the maxilla is rare due to its rich blood supply. However, in fungal osteomyelitis, the fungus infiltrates blood vessels and spreads through them. The fungal hyphae create clots (thrombi) within these vessels, reducing blood flow to the tissues and ultimately leading to necrosis. Additionally, diabetic patients with peripheral vascular disease—caused by microangiopathy and atherosclerosis—experience local tissue ischemia, which further increases their vulnerability.³ Hence, diabetes is a major factor in exacerbating symptoms, and in this case, both the patient's diabetic condition and the recent tooth extraction likely contributed to the swift and abrupt development of the lesion within a two-month period.

In the orofacial region typically present with symptoms such as nasal discharge, facial swelling (cellulitis), runny nose, and necrosis of the nasal turbinates, often accompanied by fatigue, fever, and headache. In more advanced stages, the infection may spread to the eyes, leading to symptoms like eye pain, blurred vision, redness of the conjunctiva, drooping eyelids (ptosis), bulging of the eye (proptosis), and swelling (chemosis). In some cases, vision loss may occur due to retinal artery blockage.⁴ However, none of these features were observed in present case with the exception of nasal regurgitation.

Direct histological examination continues to be the definitive method for diagnosis. *Mucor* species are identified by their broad approximately 6-20 μm in size, ribbon-like, non-septate hyphae with right-angle branching, while *Aspergillus* species display narrower, septate hyphae that typically dichotomous and measure

approximately 3-6 μm in diameter, branching at 45 and 90-degree angles, concomitant with the present case.^{1,2,5}

Mucormycosis and aspergillosis are typically severe infections, with mortality rates between 20% and 40% in immunocompromised individuals, and as high as 80% to 90% when the infection spreads or involves the brain. Management involves surgical removal of infected bone (sequestrectomy) along with thorough wound debridement. Antifungal treatment commonly includes medications such as amphotericin B, voriconazole, and posaconazole. In cases where surgical removal results in tissue defects, reconstruction using soft or hard tissue, or the use of a prosthetic obturator, may be necessary for rehabilitation.⁶ In the current case, sequestrectomy was also carried out, an antifungal regimen was subsequently prescribed, and a maxillary obturator was advised for rehabilitation.

Conclusion

Immunocompromised patients especially those with diabetes—are more prone to fungal infections, as diabetes poses a concealed risk that heightens susceptibility to conditions affecting the jawbones and soft tissues of the oral cavity. Early identification of these aggressive lesions is essential to improving patient outcomes. Prompt and accurate diagnosis, followed by appropriate treatment, is crucial to prevent serious complications. Nevertheless, the uncommon nature and limited reporting of these infections often make diagnosis challenging for healthcare providers. As such, fungal osteomyelitis should be included in the differential diagnosis when encountering comparable clinical symptoms.

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