

## Case Report

# An atypical case report of extensive mucormycotic osteomyelitis of maxilla as a consequence of post-COVID complication

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## ABSTRACT

Saprophytic molds such as *Mucor*, *Rhizopus*, and *Absidia* cause mucormycosis, a fungal infection. These saprophytic fungi are common in the environment and have a strong proclivity for invading major blood arteries, causing tissue ischemia, necrosis, and infarction. They have been linked to immunocompromised individuals with a history of diabetic ketoacidosis, corticosteroid medication, HIV infection, malignant lymphomas, and patients currently receiving and recovering from COVID-19 treatment. The foregoing is the case of a 78-year-old COVID-19 recovered male who presented with a primary complaint of upper tooth movement for 1 month and maxillary segmental mobility. The maxillary alveolar process was resected, and histopathological reports revealed mucormycosis, which was treated with antifungal medication and nasolabial flap surgery. For the past 6 months, he has been disease-free. Early detection and treatment may offer a higher chance of successfully minimizing this debilitating condition.

**Key Words:** COVID-19, fungal infection, maxilla, *Mucor*

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## INTRODUCTION

Mucormycosis is a fungal infection caused by saprophytic, pervasive fungi that are mundanely found in a dormant form in the nasal passages and oral cavities of salubrious people.<sup>[1]</sup> This is a debilitating opportunistic infection that is increasingly being perceived in patients with post-COVID infection, malignant lymphomas, renal failure, organ transplantations, AIDS, diabetic ketoacidosis, and cirrhosis.<sup>[2]</sup> According to a recent systematic analysis, 8% of the patients suffered from fungal or bacterial coinfection while in the hospital. With certain occurrences of an unusual fungal infection linked to an elevated death rate, the present COVID-19

pandemic has thrown up yet another additional problem for health care. COVID-19 patients are prone to fungal infections such as mucormycosis, which has been reported in patients who are currently struggling and those who have just recovered, particularly those who are medically impaired. Coinfections in COVID-19 patients are a growing issue, owing to their difficult diagnosis, severity, and increased mortality.<sup>[3]</sup> We present a case of maxillary mucormycosis in a 78-year-old man who was post-COVID recuperated.

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## CASE REPORT

A 78-year-old male patient (SMIDS IRC Ref. No. 08.01.2021) presented to our outpatient sector with pain and sensitivity in the upper front and back tooth region. Difficulty in speech, mastication, and foul smell were the chief complaints of the patient [Figure 1]. The patient revealed a medical history of diabetes mellitus for the past 15 years and COVID-19 infection within the previous 5 months.

On intraoral examination, there was a generalized gingival recession with segmental movement of the maxilla from 16 to 26 regions, along with a brownish discharge from the sockets of the necrotized maxillary alveolus [Figure 2].

On palpation, the bone surface was nonbrittle, rough, and sensitive. An incisional biopsy was performed, and the decalcified hard tissues after histopathological

tissue processing revealed necrotic marrow with broad aseptate fungal hyphae forms, indicating mucormycosis of the maxillary alveolar processes. The treatment strategy included resection of the infected maxilla and functional endoscopic sinus surgery under general anesthesia [Figures 3 and 4]. The procedure was done, and the entire specimen was sent for histopathologic examination.

Histopathological examination of the decalcified hard-tissue sections showed the existence of trabeculae of necrotic bone with numerous fungal organisms with big nonseptate hyphae branching at obtuse angles. Within the marrow gaps, ovoid sporangia resembling mucormycosis were spotted [Figure 5]. The presence of nonseptate branching organisms with necrotic thrombi was also revealed by special staining using Grocott's methenamine silver staining [Figure 6]. A definitive diagnosis of mucormycosis was made based on these

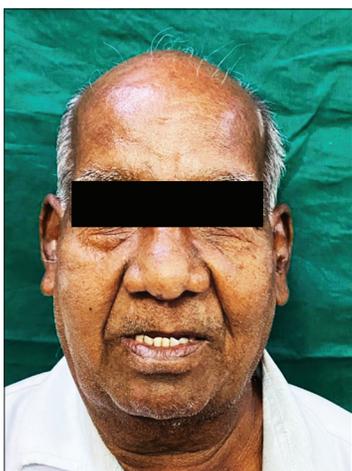


Figure 1: Extra-oral photograph of the patient



Figure 3: Photograph showing Intra-oral surgery



Figure 2: Intra-oral photograph showing sloughed mucosal tissues and exposed alveolar compartment of the maxilla



Figure 4: Photograph showing resected maxillary segment

histopathological findings. The mucoperiosteal flaps were closed first [Figure 7] and then 15-day interval follow-ups were performed. Wound healing, maxillary architectural retention, food intake, and speech were all closely examined. After 4 months of good wound healing, a nasolabial flap procedure was completed to close the oroantral interface [Figures 8 and 9]. On later follow-ups, the patient was confirmed to be fine and healthy.

## DISCUSSION

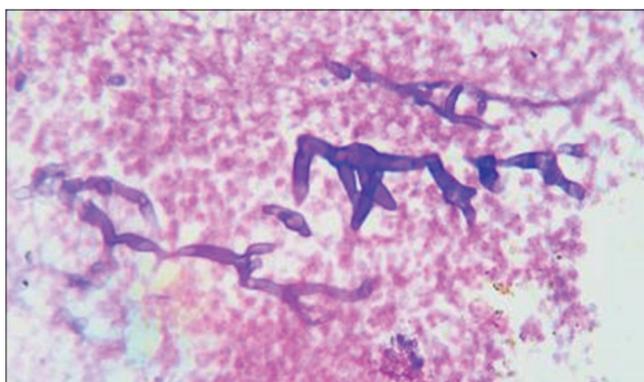
Mucormycosis is a cluster of fungal diseases caused by saprophytic organisms of the Zygomycetes class, which are widespread and thermotolerant organisms that thrive in decaying materials, bread, vegetables, soil, compost piles, and animal excrement.<sup>[3]</sup>

They are commonly found in our nasal passages and respiratory tract, where they can cause an opportunistic illness. In leukemic patients, nosocomial outbreaks

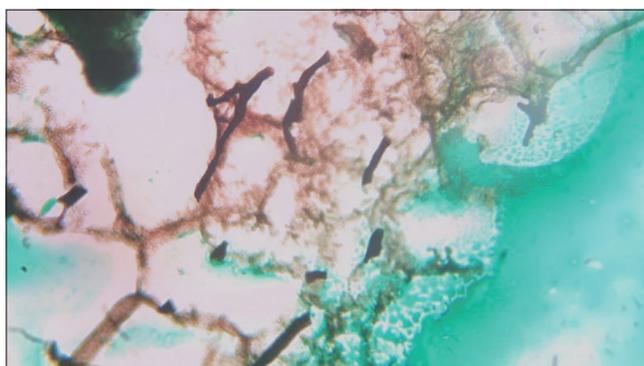
of mucormycosis can occur. Uncontrolled diabetes, metabolic acidosis, treatment with corticosteroid medicines for COVID-19 infection, organ or bone marrow transplantation, malignant hematological illnesses, and deferoxamine therapy are all substantial risk factors for mucormycosis.<sup>[4]</sup>

Mucormycosis enters the body through the respiratory tract and gets disseminated, causing thrombi in the blood vessels and infarcts. Spores can also be injected directly through abraded tissues, where they can multiply and spread to other organs.<sup>[2]</sup>

The extensive use of COVID-19-fighting steroids, monoclonal antibodies, and broad-spectrum antibiotics may result in the development or exacerbation of preexisting fungal infections. The secondary infections can be induced by a complicated interaction of variables such as preexisting illnesses such as diabetes mellitus,



**Figure 5:** Photomicrograph showing colonies of broad ribbon shaped, fungal organisms with large non septate hyphae branching at obtuse angles and ovoid sporangia resembling mucormycosis, H&E X 400



**Figure 6:** Photomicrograph showing colonies of fungal organisms with large non septate hyphae branching at obtuse angles resembling mucormycosis, Grocott's Methenamine Silver Staining X 400



**Figure 7:** Photograph showing primary closure of mucoperiosteal flap



**Figure 8:** Photographs showing removal of nasolabial flap for closing the oro-antral communication



**Figure 9:** Photographs showing closure of oro- antral communication using nasolabial flap

prior pulmonary pathology, immunosuppressive treatment, the risk of hospital-acquired infections, and systemic immunological changes caused by COVID-19 infection itself.<sup>[5]</sup>

The rapid onset of tissue necrosis, with or without fever, is a characteristic clinical symptom of mucormycosis. The other features include sinusitis, facial pain, unilateral headache, drainage, and soft-tissue inflammation. This necrosis is caused by blood vessel invasion and subsequent thrombosis.<sup>[2]</sup> As the blood vessels get thrombosed, and tissue infarction ensues, infected tissues become erythematous, then violaceous, and eventually black. Infections can spread from the sinuses into the mouth, resulting in painful, necrotic ulceration of the hard palate. A bloody nasal discharge might be the first symptom that an infection has spread to the brain through the turbinates. Oral manifestations include ischemia, followed by necrosis, bony denudation, tooth mobility, ulcers of gingiva, lips, cheeks, tongue, bad breath, and pain. Internal maxillary artery thrombosis is also possible.<sup>[6]</sup> A case series by Ahmed *et al.* reported 21 post-COVID-19 patients with oral mucormycosis and suggested that nonspecific palatal ulcer could be considered the presenting sign of mucormycosis.<sup>[7]</sup>

It is difficult and a delicate procedure to culture organisms from a possibly contaminated location. For mucormycosis, there is no effective serologic or skin test. Hence, a biopsy of infected tissues will be the gold standard method for diagnosis.<sup>[8]</sup>

Computed tomography scanning and magnetic resonance imaging are intended for people with rhinocerebral mucormycosis.

The distinctive broad, ribbon-like aseptate hyphal components branch at right/obtuse angles may be seen in histopathological sections. The majority of sporangia are oval in form. Invading the lumen of the blood arteries, fungal particles may be detected. Tissue necrosis is commonly found throughout the fungal-infected areas.<sup>[2]</sup>

When it comes to diagnosing mucormycosis, terms such as orbital cellulitis, aspergillosis, cellulitis, ecthyma gangrenosum, fusariosis, and nocardiosis might be confusing.<sup>[8]</sup>

Mucormycosis was found to be more common in males (78.9%) who were either active (59.4%) or recovered (40.6%) from COVID-19. The survival rate for rhinocerebral illness is around 75% in individuals without systemic disease, 60% in those with diabetes, and 20% in patients with additional underlying disorders.<sup>[4]</sup>

Dhande *et al.* reported a similar mucormycosis case which was treated with surgical debridement, adjuvant antifungal medication, and prosthetic rehabilitation to restore form and function.<sup>[6]</sup> Following the surgical debridement of necrotic tissue, significant reconstructive surgery may be required if the patient survives the acute phase of the illness.<sup>[7]</sup>

The use of liposomal amphotericin B (5 mg/kg) in combination with surgery is the recommended first-line treatment which was followed in this case. The second-line treatments include isavuconazole and posaconazole in intravenous or delayed-release tablet form.<sup>[9]</sup>

## CONCLUSION

A tissue sample that identifies the distinctive hyphae, a positive culture, or both can be used to provide a conclusive diagnosis of mucormycosis. Modern comprehensive devitalizing surgical techniques can be as effective as or more effective than timely medicinal therapy. Antifungal medications and decreased corticosteroids are given at the right time may result in a favorable prognosis.<sup>[10]</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.

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## Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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