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Case Report

Mucormycosis uncommon encounters: Pediatric and geriatric perspectives in a case report

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ABSTRACT

Mucormycosis, an angioinvasive fungal infection, typically originates in the nose and paranasal sinuses following inhalation of fungal spores. It poses a potentially life-threatening risk, primarily caused by opportunistic, saprophytic aerobic fungi of the Zygomycetes subdivision and Mucorales order. Common risk factors associated with mucormycosis include uncontrolled diabetes, hematological disorders, immunosuppression, underlying systemic diseases, and symptoms associated with COVID-19.

The infection presents various clinical manifestations, including rhino-orbit-cerebral, pulmonary, gastrointestinal, and mucocutaneous symptoms. We observed two cases of mucormycosis precipitated by classical and atypical risk factors, affecting individuals of different age groups. Despite the differing presentations, a common underlying systemic cause i.e. diabetes ketoacidosis was evident. The diagnosis was confirmed through either histopathological examination or positive culture.

Mucormycosis ranks as the third most common invasive fungal infection. Treatment entails rapid diagnosis, correction of risk factors, surgical resection or debridement, and appropriate antifungal therapy.

1. Introduction

Mucormycosis is an angioinvasive fungal infection typically originating in the nose and paranasal sinuses after inhalation of fungal spores. While diabetes mellitus stands out as the primary predisposing factor in adults, it accounts for only approximately 15% of cases in children.[1] Other risk factors in children include immunosuppression, neutropenia, and prematurity.[2] Conversely, in elderly patients, diabetes mellitus assumes a significant role as a risk factor, alongside untreated underlying systemic conditions.[3]

2. Case Presentation

2.1. Case 1: Pediatric mucormycosis

A 12-year-old boy was diagnosed with a chief complaint of pain and swelling on the right side of his face for 2 months. The patient was asymptomatic 2 months back and subsequently developed pain. His blood parameters at the time of admission were was BP (blood pressure) 130/82 mmHg, Creatinine 0.7 mg/dL, FBS (fasting blood sugar) 160 g/dL, HB (hemoglobin) 15.1 g/dL, HBA1C (haemoglobin A1C) 6.5 %, HBsAg (hepatitis B surface antigen) Negative, HCV Ab (hepatitis C antibody) Negative, Lymphocytes 37%, Neutrophils 68 %, Platelet count 26400 cells/mm^, Potassium 4.3mmol/L, Sodium 134 mmol/cc, Urine ketone bodies Negative.

He presented with symptoms suggestive of respiratory infection due to SARS-CoV-2 and severe diabetic

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ketoacidosis, necessitating admission to the intensive care unit (ICU) for multi-organ support. During his stay, changes in skin color were observed over time in the nasal pyramid region, indicative of skin necrosis. Additionally, a crusty lesion measuring 4×2 cm was noted in the hard palate region of the oral cavity on the right side with no paresthesia. [Figure 1]. On examination necrosis in the oral cavity, extended to the nasal septum, nasal turbinates, mucosa of the hard palate, skin of the nasal pyramid, upper & lower eyelids and the right eyeball.

Radiographic investigations, including CT scans, revealed hyperdense areas surrounding hypodense halos in the palatal region, ethmoid cells, and sinus membranes. The KOH mount was negative for fungal elements. The functional endoscopic surgery was performed and a tissue sample was sent for histology showing numerous aseptate hyphae branching at 90 degrees. Following comprehensive investigations, a definitive diagnosis of invasive mucormycosis was established.

Treatment involved initiating daily deoxycholate amphotericin B at 1.5 mg/kg/day. Functional endoscopic sinus surgery (FESS) was performed, followed by surgical debridement of the necrotic tissue with hemi-maxillectomy on right side from 23 to 18 under general anesthesia [Figure 2]. The surgical cavity was packed with a roller gauge soaked in BIPP (Bismuth iodoform paraffin paste) and the patient received antibiotic therapy for the subsequent three weeks [Figure 3]. The patient was discharged with oral antibiotics and analgesics.

2.2. *Case 2: Geriatric mucormycosis*

A 64-year-old man presented to the department displaying an asymptomatic ulcer on the hard palate, coupled with a history of uncontrolled type 2 diabetes from the past 10 years and was under medications (unknown) which he stopped taking a year back. The patient was asymptomatic 4 months back and subsequently developed an ulcer. His blood parameters at the time of admission was BP (blood pressure) 110/62 mmHg, Creatinine 0.8 mg/dL, FBS (fasting blood sugar) 180 g/dL, HB (hemoglobin) 12.1 g/dL, HBA1C (haemoglobin A1C) 7 %, HBsAg (hepatitis B surface antigen) Negative, HCV Ab (hepatitis C antibody) Negative, Lymphocytes 34%, Neutrophils 65 %, Platelet count 25400 cells/mm^, Potassium 3.3mmol/L, Sodium 132 mmol/cc, Urine ketone bodies Negative. Over two weeks, the ulcer evolved to a diameter of 1.5 cm, exhibiting a necrotic background, bony exposure at the center, an irregular erythematous edge and emitting a fetid odor [Fig. 4]. Radiographic investigations, including a brain CT scan with mandible impression, revealed hyperdense areas in the palatal process and maxillary bone. Soft tissue thickness along the paranasal sinuses and complete sinus opacification extending toward the infra-temporal fossa were also observed [Fig. 5].

An incisional biopsy was performed, and histology showed numerous aseptate hyphae branching at 90 degrees, the report suggested a mucormycosis infection of the maxilla. Treatment for diabetes mellitus was adjusted using a sliding scale [Table 1] and liposomal amphotericin B was initiated at a dose of 5 mg/kg/day. Thorough debridement and a complete maxillectomy were performed [Fig. 6]. The sinus was cleaned using Functional Endoscopic Sinus Surgery (FESS) under supervision under the Ear, Nose, and Throat (ENT). A roller gauge soaked in BIPP was inserted, and the patient remained under observation, receiving antibiotic therapy to prevent infection. Medical discharge was granted two weeks after the surgery with analgesics and oral antibiotics.

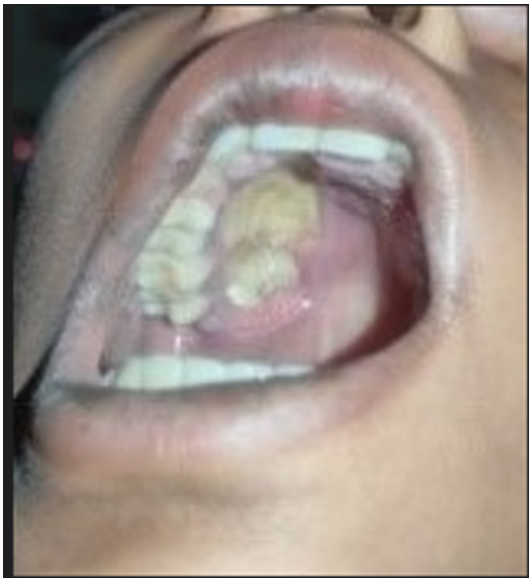
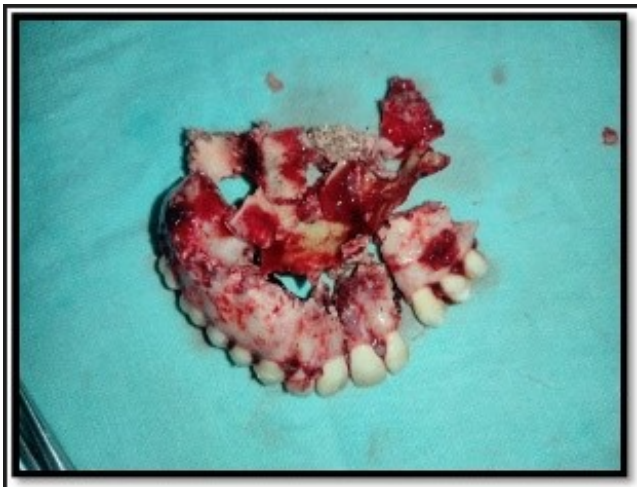


Figure 1:



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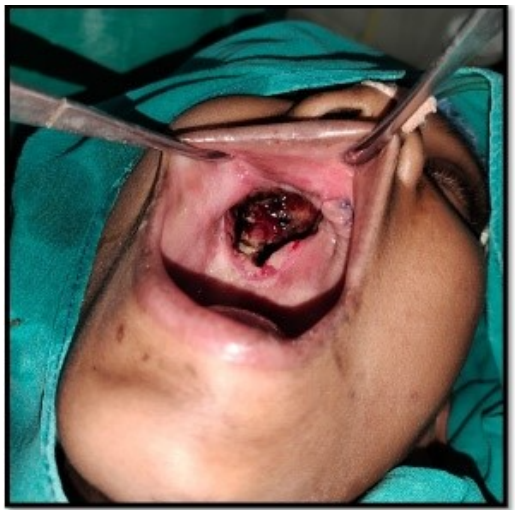

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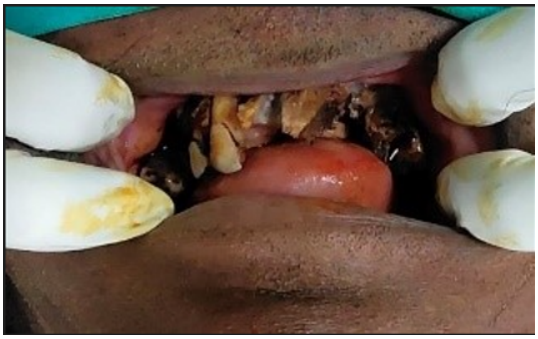

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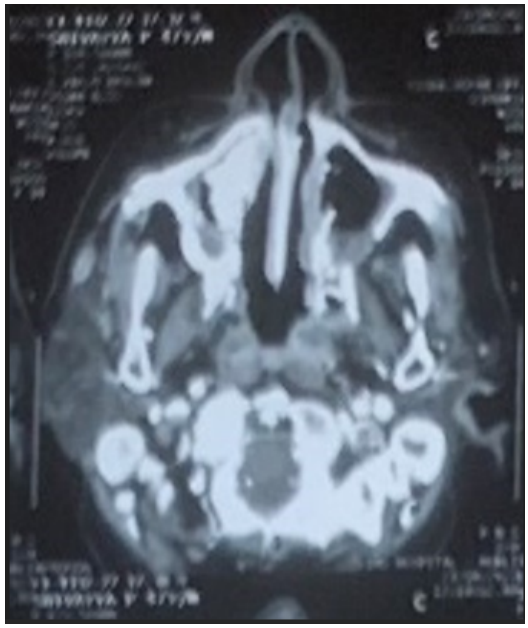

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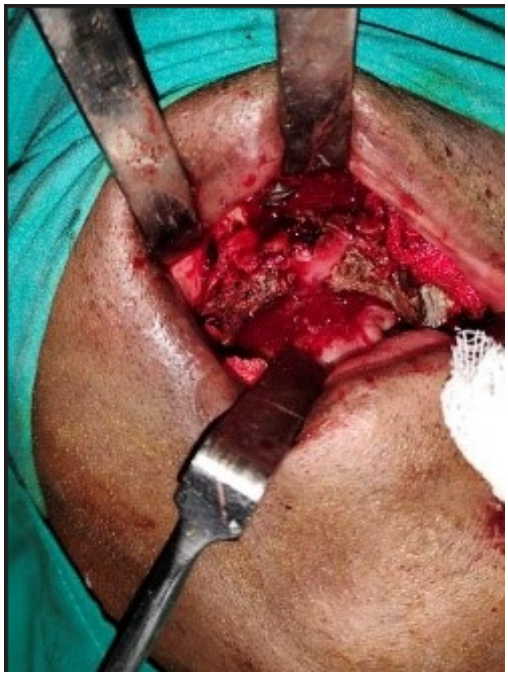

Figure 6:

3. Discussion

Mucormycosis was originally described by Paultauf in 1885. It stands as the third most common invasive mycosis caused by fungi of the zygomycetes class, trailing behind candidiasis and aspergillosis. Rhizopus arrhizus (Oryza) is the most frequently occurring species.[4] Key risk factors include ketoacidosis, immunosuppression, corticosteroid usage, blood malignancies, and deferoxamine use. Mucormycosis affects various parts of the body, including the sinuses (39%), lungs (24%), skin (19%), brain (9%), gastrointestinal system (7%), disseminated form (6%), and other areas (6%).[4] Treatment involves prompt diagnosis, correction of underlying co-morbidities, surgical enucleation or resection, and adequate antifungal therapy. Liposomal amphotericin B combined with surgical intervention constitutes the primary line of therapy. Individuals treated with amphotericin B have shown significantly higher survival rates than untreated individuals. Treatment options for mucormycosis must be tailored to each patient's unique characteristics, disease progression, and severity, necessitating a multidisciplinary approach.

Traditional medication delivery to the infection site is often insufficient due to arterial thrombosis and tissue necrosis, rendering conservative therapy ineffective. Extensive debridement, thorough removal of necrotic tissue, and, in severe cases, orbital evisceration may be necessary. A combination of medical and surgical therapies improves survival rates. While itraconazole has been used in several studies, it has a high failure rate, and voriconazole

is ineffective against mucorales in vitro. Posaconazole and ravuconazole have shown promising results in vitro. The overall fatality rate ranges from 40% to 80%, depending on the involvement of critical structures.[5,6] The acidic environment and free ferric ions in circulation during diabetic ketoacidosis promote the proliferation of Mucorales. These conditions also facilitate invasion and adhesion to the human body.[7] COVID-19 patients are often treated with high-dose steroids, excessive oxygen masks, or ventilator usage. Corticosteroid use reduces inflammation and immune system activity by decreasing white blood cell and T-helper cell production, resulting in neutropenia, which makes the human body susceptible to foreign body invasion and infections. Corticosteroids can induce a hyperglycemic condition in the human body, allowing Mucorales species to invade and proliferate rapidly.[8] According to a retrospective analysis by Mulakavalupil et al., no cases of mucormycosis were reported among the 4221 patients treated for COVID-19 therapy between March 2020 and May 2021. There were no incidences of mucormycosis detected in hospitalized patients or during the six-month post-discharge follow-up. Tight low-dose steroid management, careful glycemic control, and diligent nursing care can reduce the prevalence of mucormycosis during the COVID-19 pandemic.[8]

4. Conclusion

In conclusion, mucormycosis represents a formidable fungal infection, with the prognosis heavily influenced by the underlying disease status. Therefore, the initial focus should be on emergency care for underlying conditions and stringent management to achieve a favorable outcome.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Mishra N, Mutya VS, Thomas A, Rai G, Reddy B, Mohanan AA, et al. A case series of invasive mucormycosis in patients with COVID-19 infection. *Int J Otorhinolaryngol Head Neck Surg*. 2021;7(5):867–70.
2. Roushdy T, Hamid E. A case series of post-COVID-19 mucormycosis-a neurological perspective. *The Egyptian Journal of Neurology*. 2021;57:1–8.
3. Yadav S, Sharma A, Kothari N, Bhatia PK, Goyal S, Goyal A. Mucormycosis: A case series of patients admitted in non-covid-19 intensive care unit of a tertiary care center during the second wave. *Indian Journal of Critical Care Medicine*. 2021;25(10):1193–1193.
4. Bouza E, Munoz P, Guinea J. 2006.
5. Maini A, Tomar G, Khanna D, Kini Y, Mehta H, Bhagyasree V. Sino-orbital mucormycosis in a COVID-19 patient: A case report. *International Journal of Surgery Case Reports*. 2021;82:105957–105957.
6. Rodriguez-Morales AJ, Sah R, Millan-Oñate J, Gonzalez A, Montenegro-Idrogo JJ, Scherger S, et al.
7. Chandra S, Rawal R. The surge in Covid-related mucormycosis. *Journal of Infection*. 2021;.
8. Mulakavalupil B, Vaity C, Joshi S, Misra A, Pandit RA.) under strict protocol-driven management care in a COVID-19-specific tertiary care intensive care unit. *Diabetes & Metabolic Syndrome*. 2009;.

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