

A Case Report of Black Fungus: Mucormycosis Osteomyelitis of Mandible

S M Mahbubul Alam^{1*}, Ahmed Khaled¹, Golam Mohiuddin Chowdhury², Fahmida Ferdousi³, Nikhat Ara⁴

1. Sr. Consultant,
Histopathology,
Evercare Hospital Dhaka.
2. Sr. Consultant,
Dental & Maxillofacial Surgery,
Evercare Hospital Dhaka.
3. Associate Consultant,
Oral & Maxillofacial Surgery,
Evercare Hospital Dhaka.
4. Sr. Consultant,
Microbiology & Infection Control,
Evercare Hospital Dhaka.

ABSTRACT

The case we report, is a rare occurrence of mandibular mucormycosis in a 44-year-old male with poorly controlled Diabetes. The patient presented symptoms of pain and loosening of all lower jaw teeth accompanied by pus discharge. CT scan imaging revealed characteristics indicative of mandibular osteomyelitis. He underwent thorough debridement, and the diagnosis was established on histopathological section of tissue samples. Histopathology showed wide hyphae without septa and branching at the right angle compatible with mucormycosis. Isolated Mandibular mucormycosis is a rare, potentially life-threatening fungal infections. It is imperative to address any cases of mucormycosis in Bangladesh, especially considering the high prevalence of diabetes in the country.

Keywords: COVID-19-associated mucormycosis, Diabetes, Mandibular osteomyelitis.

Address for Correspondence :
Dr. S M Mahbubul Alam
Sr. Consultant, Histopathology
Evercare Hospital Dhaka.
mahbubhistopath@gmail.com

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INTRODUCTION

The sudden upsurge in mucormycosis cases during the second wave of COVID-19 pandemic in India had drawn global attention and raised concern in Bangladesh. Bangladesh detected its first case of COVID-19-associated mucormycosis (CAM) on May 8, 2021, while by Jun 20, 2021, India had reported a total of 40,854 cases¹. There was no additional instance of mucormycosis reported during the intervening period in Bangladesh. This case presented here highlights a recently diagnosed, rare instance of mandibular mucormycosis in the post-Covid-19 period, albeit not as sequel to Covid-19. The case is presented within the context of the endemic variation in mucormycosis between two neighbouring countries.

REPORT OF A CASE

A 44-year-old man with poorly controlled diabetes mellitus, reported at the Dental and Maxillofacial

surgery outpatient department of a tertiary care Hospital in Dhaka in October 2023 with complaints of pain, loosening of all lower Jaw teeth, and pus discharge from lower left teeth for about 3 months. All para-nasal sinuses were normal clinically. His Covid-19 test returned negative. He had no preceding history of Covid-19 infection. He underwent a dental procedure shortly before experiencing symptoms. dental procedure before became symptomatic. His blood sugar level was recorded at 30.9 mmol/L. A contrast CT scan facial bone suggested osteolytic change in mandibular body, angle, ramus and also in the symphysis area (Figure. 1) suggestive of osteomyelitis. He was planned for sequestrectomy and saucerization. After controlling glucose level operation was done (Figure. 2). All teeth were extracted from the lower jaw except 47. Histopathology from debrided tissue

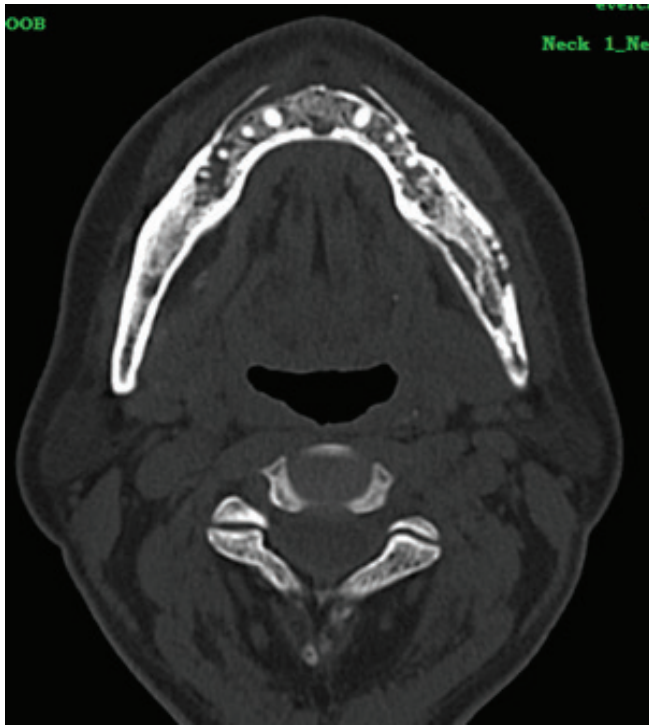


Figure 1: CT Mandible -Multiple irregular radiolucent areas



Figure 2: Intra-operative picture showing mandibular lesion

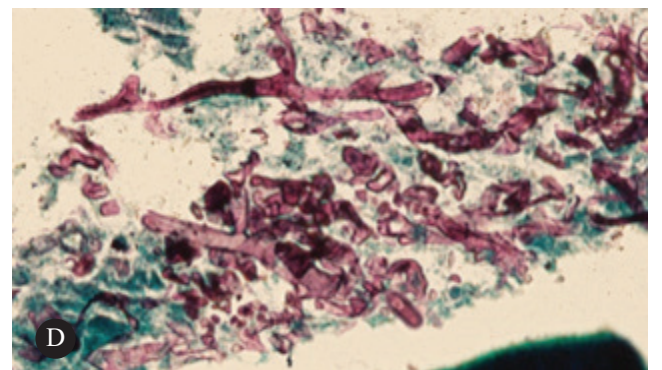
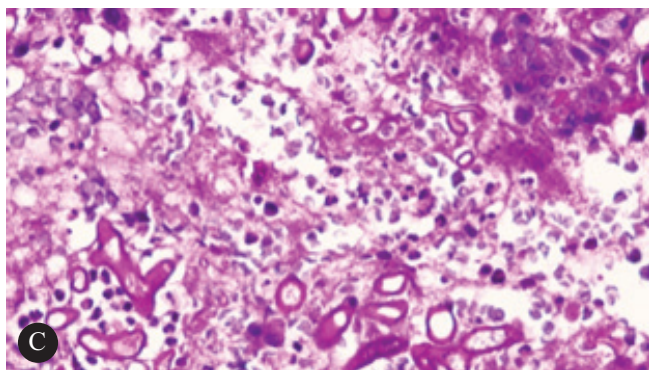
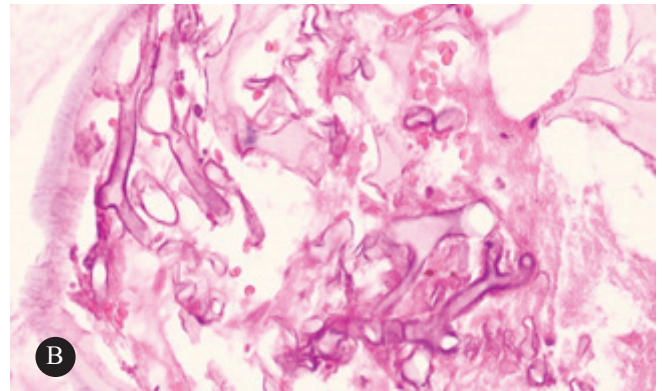
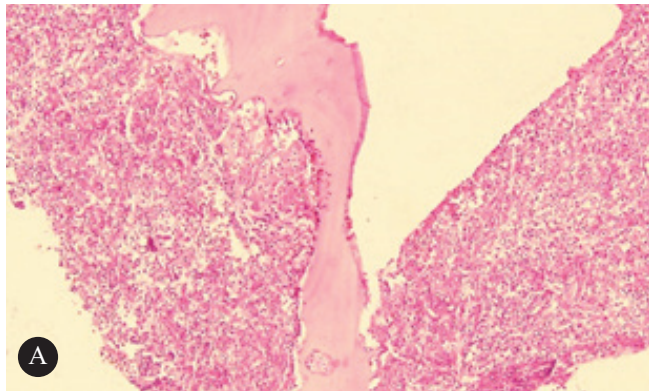


Figure 3: A. Bony and soft tissue studded with fungal hyphae in a necro inflammatory background. B. C. PAS stain x 400: Highlighted the fungus. D. GMS stain x 400: Broad wide angle, non-septate hyphae of the fungus Mucor.

of the mandible showed that bony and soft tissues were studded with fungal hyphae. PAS and GMS stain highlighted the broad, non septate hyphae,

branching at right angles compatible with Mucormycosis. (Fig 3).

The patient was started with systemic anti-fungal (Posaconazole) orally. He has completed his 3-month anti-fungal course. He continued to improve with treatment.

DISCUSSION

During the second wave of the COVID-19 pandemic in the summer of 2021, there was a notable increase in cases of COVID-19-associated mucormycosis. This surge was predominantly observed in India, with an estimated prevalence of mucormycosis nearly 70 times higher than the global data².

The heightened prevalence of uncontrolled diabetes mellitus in India, along with frequent corticosteroid usage and immune dysregulation due to COVID-19, likely contributed to this upsurge. The interplay between COVID-19 and mucormycosis appears to involve complex pathogenesis.

COVID-19 itself can result in impaired immune system as well as aggravate hyperglycaemia. SARS-CoV-2-induced cytokine storm leading to a systemic proinflammatory milieu, resulting in clinically detectable glycometabolic dysregulation that persists long after the post recovery period³. The treatment of COVID-19 with corticosteroids and other immunomodulatory drugs contributes to immune dysfunction. Hyperglycemia and diabetic ketoacidosis make free available iron and, in the presence of lowered host defences, promote specific interaction of host cells with Mucorales hyphae, resulting in CAM.

The case presented here involves a diagnosis of mucormycosis in the post-COVID period, unrelated to COVID infection (the patient had no history of Covid-19 infection). The individual had uncontrolled diabetes, a common factor among COVID-19-associated mucormycosis cases detected in the capital in May 2021, as well as in 85% of cases reported in India².

Mucormycosis, widely known by the misnomer “black fungus”, is an aggressive and potentially life-threatening fungal infection caused by fungi of the Mucorales order. These fungi are ubiquitous in the environment. Transmission occurs through

inhalation, inoculation, or ingestion of spores from the environment. Individuals with an immune-compromised state, such as those with haematology malignancy, or organ transplant recipients and associated with certain independent risk factors like diabetes mellitus (53.6%), long-term steroid therapy, chronic kidney disease, neutropenia, ketoacidosis, etc. are at a higher risk of developing mucormycosis.

It most commonly affects the sinuses or the lungs after inhaling fungal spores from the air. Covid-19 associated mucormycosis (CAM) predominantly involves paranasal sinuses and further spreading to the orbit and cerebral regions presenting most common variant as rhino-orbito-cerebral disease. The spread of infection from the paranasal regions to the caudally placed mandible is very rarely seen. Mandibular involvement without paranasal sinus infection, as seen in this case, is rare. Mucorales can cause infections at various anatomical sites, with associated signs and symptoms. Based on anatomic localization, the clinical form of mucormycosis can be: rhinocerebral, pulmonary, cutaneous, gastrointestinal, or disseminated^{4,5,6}.

Early diagnosis of mucormycosis hinges on maintaining a high index of suspicion rooted in clinico-radiological features. Direct microscopy (by KOH wet mount), histopathological examination and fungal culture with identification are the cornerstones of diagnosis.

In our case, the diagnosis was confirmed through histopathological examination. The histopathological findings revealed broad, aseptate fungal hyphae with branching at right angles, consistent with Mucormycosis. Special stains like GMS and PAS aid in morphological clarity. The histopathological differential diagnosis includes aspergillosis, characterized by septate hyphae that are smaller in width and branch at more acute angles. Immunohistochemistry tests offer a valuable solution to overcome the challenges associated with histomorphologic diagnosis when differentiating between mucormycosis, aspergillosis and other fungal infection.

There is a growing need for the country to prioritize PCR identification and sequencing to

improve early detection and confirmation of Mucorales. The major obstacle in the management of mucormycosis has been lack of a non-invasive, rapid and a reliable diagnostic test.

Successful management of mucormycosis largely depends on early diagnosis, reversal of underlying predisposing factors, prompt and ideally broad surgical debridement of infected tissue and rapid administration of systemic antifungal therapy.

CONCLUSION

The very high incidence rate of mucormycosis in India, coupled with the widespread prevalence of diabetes, raises concerns about the potential for a significant number of undiagnosed cases in neighbouring Bangladesh, where diabetes, widespread corticosteroid use, and other similar factors are also prevalent. The situation poses a looming threat of undetected spread. With the recent increase in cases globally, it is essential for healthcare providers to be vigilant and consider mucormycosis in high-risk individuals.

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