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A Rare Occurrence of Primary Breast Mucormycosis, A Case Report

Dr. Sanjeev Shah, Dr. Kamlesh Patel, Dr. Chetna Bhullar, Dr. Nidhi Barot

Sterling Accuris diagnostic laboratory, Ahmedabad.

ABSTRACT

Zygomycosis occurs primarily in immunosuppressed patients and those with diabetes mellitus. Incidence of zygomycosis has increased among transplant recipients, patients with hematologic malignancy and Covid-19 associated pulmonary complications. Soft tissue zygomycosis may be localized, extend to deep underlying tissues, or may be disseminated. The most common clinical presentation is induration of the skin with surrounding erythema, rapidly progressing to necrosis. Histological examination and culture of soft tissue are important for the diagnosis of cutaneous zygomycosis. Treatment consists of surgical excision or debridement, reversal of predisposing factors and administration of antifungal agents (amphotericin B formulations, Posaconazole/or Isavuconazole).

Keywords: Zygomycosis, immunosuppressed, histopathology, culture, antifungal agents

*Correspondence to Author:

Dr. Nidhi Barot

Microbiology Laboratory, Sterling hospital, Gurukul, Ahmedabad-380052

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Introduction

Zygomycosis constitutes group of infections caused by members of the class Zygomycetes, which is divided into two orders, Mucorales and Entomophthorales. [1]

The fact that mucormycosis is a rare human infection reflects the effectiveness of the intact human immune system. This is further supported by the finding that almost all human infections, due to the agents of mucormycosis, occur in the presence of some underlying immunocompromised condition. Mucormycosis in patients with haematological malignancies is severe infection with high mortality rate. [2]

To date, only two cases of primary mucormycosis of breast have been reported. [10,11] We describe a case of mucormycosis that was localized to the breast in a patient with underlying hematological malignancy. The patient was treated successfully with surgical debridement and reversal of predisposing factor.

Case Report

A 74 years old female presented with gradually increasing left side breast pain, swelling with black discoloration for 20 days without any history of trauma, diabetes or Covid-19 infection. She was a known case of multiple myeloma and

was on chemotherapy for last 2 months. She had history of hemodialysis one month prior to admission. On examination, she was afebrile and had pulse rate of 88 beats/minute and blood pressure of 140/60 mmHg. Her left breast was swollen, tender and warm with black discoloration of the skin. Investigations revealed a hemoglobin concentration of 9.1 gm% with a white cell count of 10,690/cu.mm. Renal and liver function tests were within normal limits. A provisional diagnosis of breast abscess was made and drainage of the abscess was performed.

Oblique incision on medial aspect of left breast yielded thick black colored aspirate. Necrotic tissue with multiple lumps were excised and debrided. Excised tissue was sent for histopathology examination, direct microscopy and culture. Histopathological examination showed near total parenchymal destruction with necrosis, mixed inflammatory cell infiltration and presence of many fragments of pauciseptate, right angled branching, ribbon like folding hyphae. (Figure 1)

Mycelial filaments were positive for GMS stain morphologically consistent with Mucor species. (Figure 2)

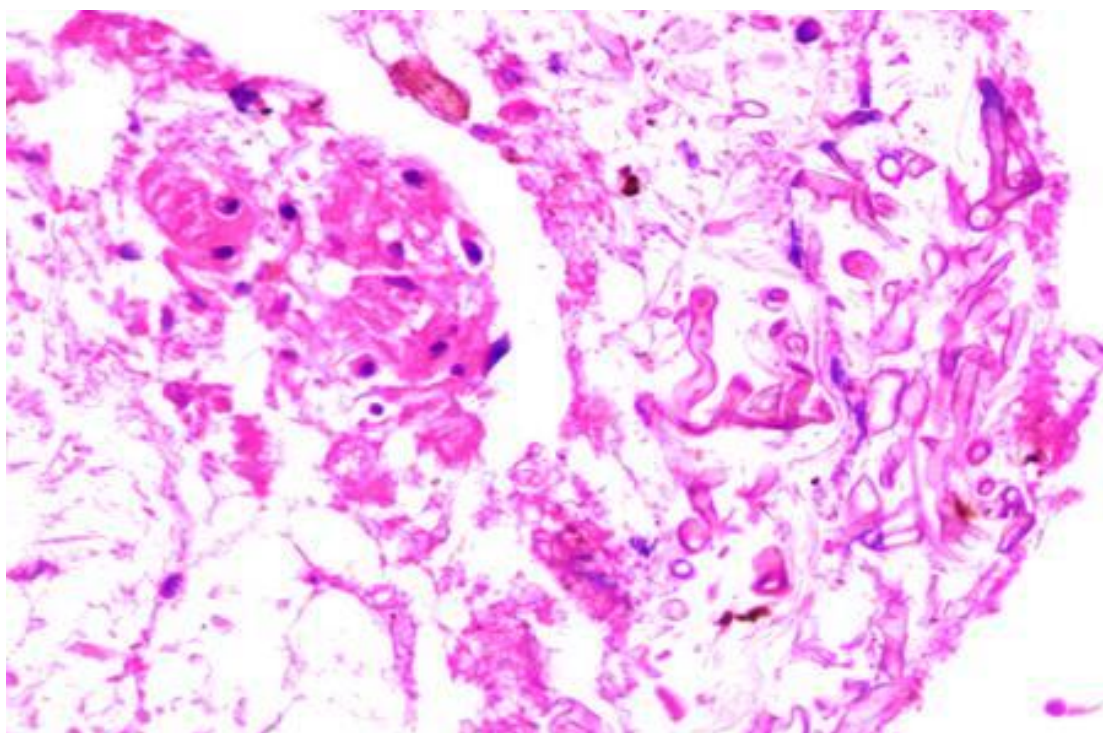


Figure 1: Histopathological findings(H&E): Microscopic view of the biopsy specimen shows several folded, non-septate, broad and right angle branched hyphae which are characteristic for mucormycosis.

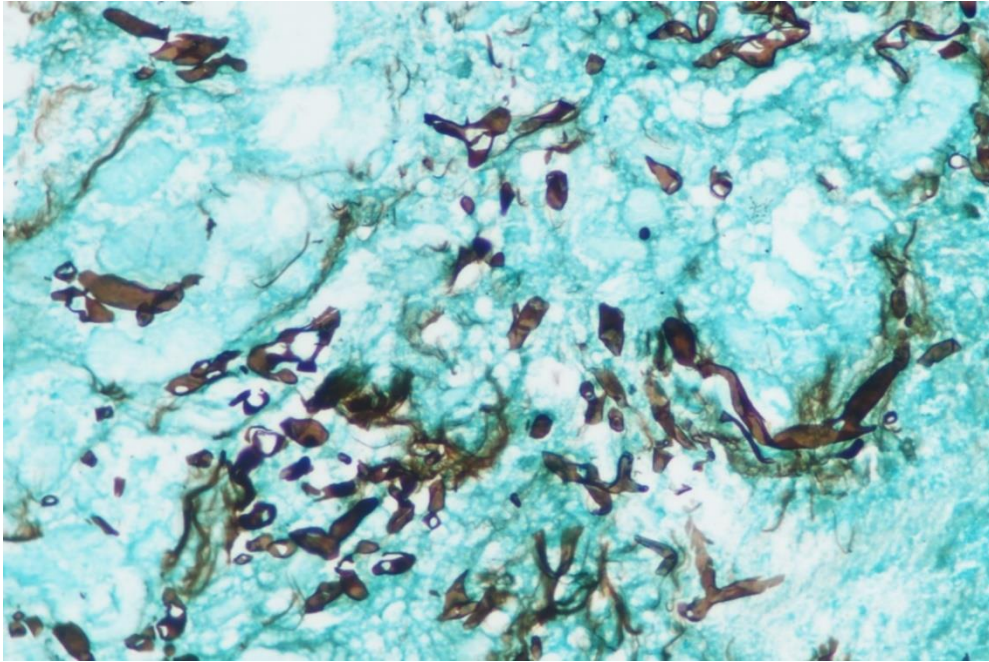


Figure 2: GMS stain: Mycelial filaments were positive for GMS.

KOH preparation of tissue revealed presence of broad aseptate hyaline fungal hyphae branching at right angle suggestive of mucormycosis. (Figure 3)

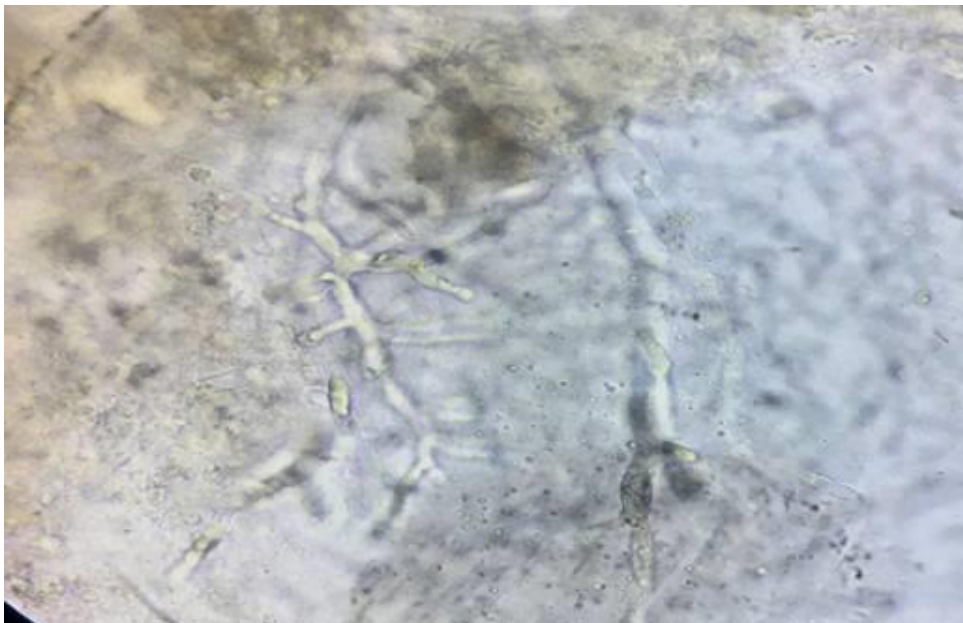


Figure 3: KOH preparation: Tissue shows many right angled branched aseptate hyphae.

Based on direct microscopy and histopathological confirmation, patient was advised for intravenous Amphotericin B therapy to which she refused. So her immunosuppressive chemotherapy was stopped and aggressive dressing was done. She has completely recovered and asymptomatic during her follow-up visit.

Discussion

There are six different manifestations of

mucormycosis based on clinical presentation and involvement of a particular body site: (1) rhinocerebral, (2) pulmonary, (3) cutaneous, (4) gastrointestinal, (5) central nervous system, and (6) miscellaneous.^[5] A hallmark of these infections is the presence of extensive angioinvasion with resultant infarctions and tissue necrosis.^[4] This angioinvasion is associated with the ability of the organism to hematogenously disseminate from the original

site of infection to other target organs.

Both mononuclear and polymorphonuclear phagocytes of healthy hosts kill Mucorales by the generation of oxidative metabolites. [6] Therefore, immunocompromised patients are at increased risk of developing mucormycosis. Recent European studies have demonstrated that haematological malignancies were underlying diseases in 58–60% cases of mucormycosis. [8, 9]

Early diagnosis of zygomycosis is essential to improve the outcome. [7] A necrotic lesion in an immunocompromised host should raise suspicion of zygomycosis. The diagnosis of mucormycosis relies upon the identification of organisms in tissue by histopathology with culture confirmation.

The hyphae of the Mucorales are distinct and allow for a presumptive identification from clinical specimens. The hyphae are broad (5 to 15 micron diameter), irregularly branched, and have rare septations (Figure- 1) Mucorales are angioinvasive; thus, infarction of infected tissues is a hallmark of invasive disease. [3]

The lack of regular septations may contribute to the fragile nature of the hyphae and the difficulty of growing the agents of mucormycosis from clinical specimens.

Four factors are critical for eradicating mucormycosis: early diagnosis, reversal of the underlying predisposing factors (if possible), appropriate surgical debridement of infected tissue, and appropriate antifungal therapy. So discontinuation of immunosuppressive therapy should be strongly considered for remission of such case.

Surgical debridement is necessary due to the massive amount of tissue necrosis occurring during mucormycosis, which may not be prevented by killing the organism.

Conclusion

Mucormycosis is an increasingly common infection in immunocompromised patients. Invasive surgical debridement with discontinuation of chemotherapy (reversal of predisposing factor) led to recovery in our patient. To our knowledge, only two other

reports of primary breast mucormycosis have been reported. [10,11]

Prompt diagnosis, reversal of predisposing conditions, and aggressive surgical debridement remain cornerstones of therapy for this deadly disease.

Conflict of interest: None declared

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