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CASE REPORT

SUBCUTANEOUS MUCORMYCOSIS IN IMMUNOCOMPETANT OLD MALE: A CASE DIAGNOSED ON FNAC

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ABSTRACT:

Zygomycosis is an opportunistic fungal infection with a high mortality rate. It is known to cause invasive disease in immunocompromised hosts but it may produce only cutaneous/subcutaneous infections in immunocompetent hosts. Treatment is difficult due to its fulminant course and lack of effective anti-fungal drugs. Here, we report a rare case of subcutaneous zygomycosis caused by Mucor in an immunocompetent patient without any debilitating illness. The patient was successfully treated by aggressive surgical debridement and anti-fungal therapy.

KEY WORDS: Immunocompetent, subcutaneous mucormycosis, Zygomycosis

INTRODUCTION

Zygomycosis is a rare, devastating and life threatening fungal infection involving cutaneous, sub-cutaneous tissue and systemic infection. Mortality is high because of misdiagnosis, lack of appropriate treatment and the fulminant course of disease.[1,2]

Mucormycosis is caused by molds belonging to the subphylum Mucoromycotina in the order Mucorales.[3] The infection is more common among people with suppressed immune systems, but it can rarely occur in immunocompetent people, since the causative fungus is nonpathogenic for healthy individuals. Cutaneous involvement is less common than rhino-cerebral involvement and mainly occurs in two forms- a “benign” well-localized subcutaneous form and a more fulminant cutaneous infection with necrotizing fascitis, systemic sepsis and a fatal outcome if the diagnosis and consequently the appropriate treatment is delayed.[4] It represents an opportunistic rather than a true pathogen. These opportunistic pathogens are ubiquitous organisms existing in the environment, soil, air, food, composite piles, animal excreta and play a vital role in the cycle of decomposition in natural world.[5] Known risk factors for developing mucormycosis comprise uncontrolled diabetes mellitus, metabolic acidosis, high dose of corticosteroids, prolonged neutropenia, organ transplantation, skin trauma (cuts, scrapes, punctures, or burns) and catheter infection. However, some patients with mucormycosis may have no identifiable risk factors.[6]

The typical presentation of cutaneous mucormycosis is a necrotic eschar accompanied by surrounding erythema and induration. However, a nonspecific erythematous macule may be the cutaneous manifestation of disseminated disease in an immunosuppressed patient.[7] When cutaneous mucormycosis presents with necrotic eschars, these lesions may mimic pyoderma gangrenosum, bacterial synergistic gangrene, or other infections produced by bacteria or fungi.[8]

CASE REPORT:

70 year old male patient presented with painless swelling over dorsum of right hand at the base of 3rd and 4th web space since 1 year. There was history of gradual increase in size of the swelling. The swelling was not subsiding after taking repeated medicines for that. Patient was a farmer by occupation and had a history of unnoticed trauma during working in the farm.

On general examination, patient was averagely built, conscious oriented with no any palpable lymph nodes. Local examination revealed a well- defined swelling of size 3x2 cm non-tender, soft in consistency, freely mobile with overlying normal appearing skin.

Routine laboratory tests including liver function test, kidney function test, lipid profile, fasting and postmeal blood sugar were within normal limit. Serological examination for HIV and HBsAg was negative. The patient was having no any underlying debilitating disease, history of chemotherapy or steroid treatment or organ transplantation.

Fine needle aspiration cytology was performed from hand swelling which revealed aseptate hyphae branching at acute angle along with inflammatory cells; polymorphs, lymphocytes with necrotic material in the background on hematoxylin and eosin staining (H & E) (Figure No 2). Periodic Acid Schiff (PAS) stain revealed magenta coloured aseptate fungal hyphae with acute angle branching. (Figure No 3) Aspirate material was examined by KOH mount for presence of fungal hyphae (Figure No 4) and also sent for culture on Sabourad dextrose agar which confirmed the diagnosis of mucormycosis. Lactophenol cotton blue showed mature sporangia of mucormycosis having aseptate hyphae. (Figure No 5). Incision and drainage of the wound was done and treated with Itaraconazole for 1 month.

Figure No 1: Clinical photograph showing lump in right dorsal aspect of hand.

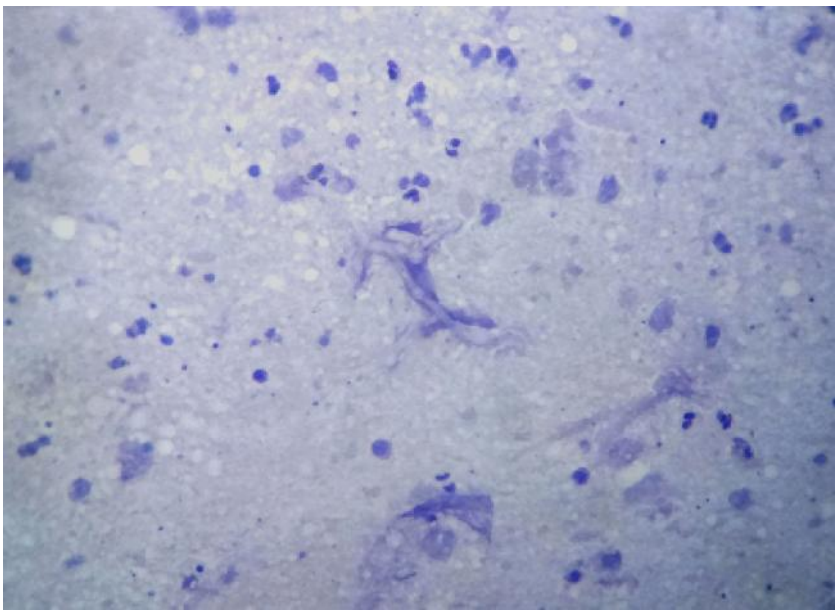

Figure No 2: Microscopic photograph showing aseptate fungal hyphae surrounded by inflammatory cells and necrotic material on FNAC. (H & E) X400

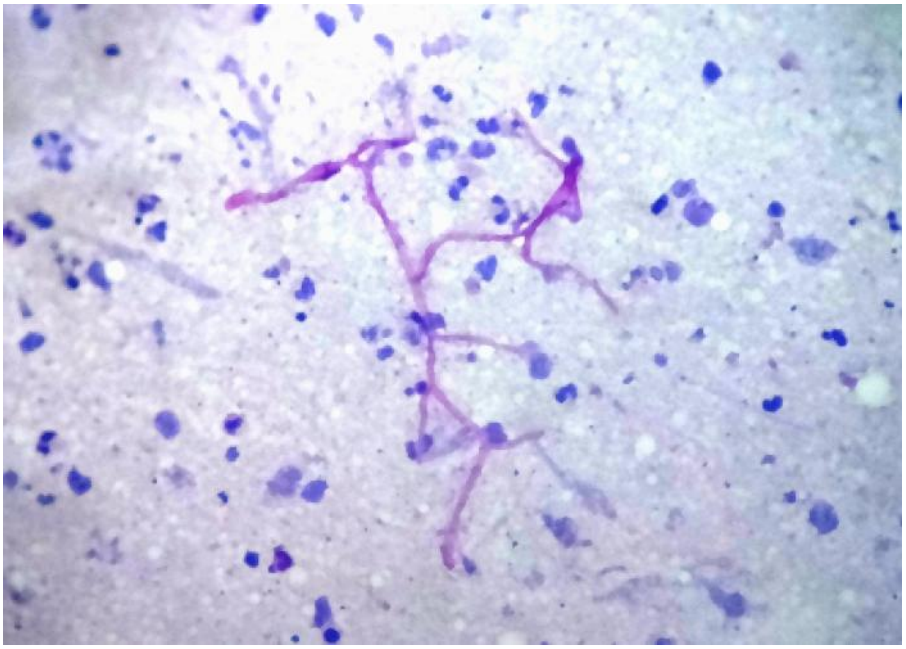

Figure No 3: Microscopic photograph showing magenta coloured aseptate fungal hyphae with branching at acute angle on PAS staining. X400

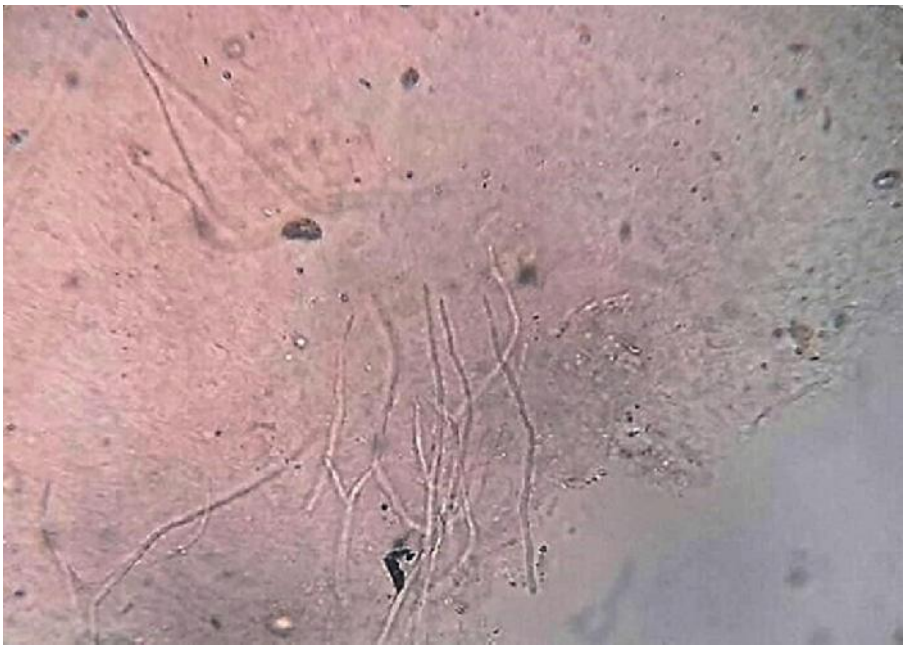

Figure No 4: KOH mount showing aseptate fungal hyphae branching at acute angle.

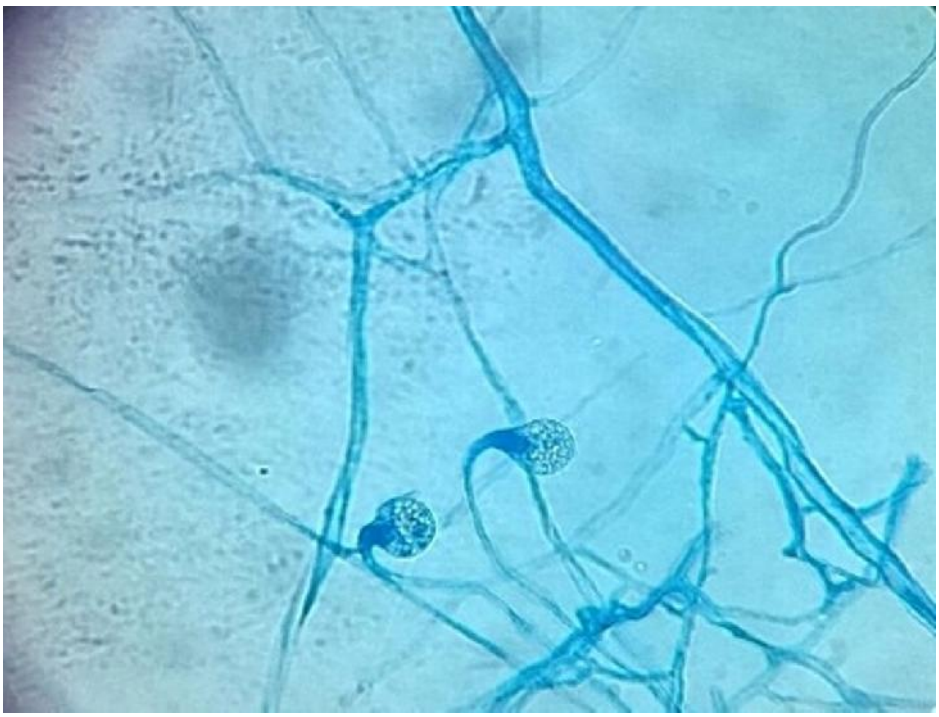

Figure No 5: Lactophenol cotton blue showing mature sporangia of mucor species.

DISCUSSION:

The agents causing human zygomycosis belong to the class Zygomycetes, which is subdivided into two orders, Mucorales and Entomophthorales.[1] Members of the order Entomophthorales such as Basidiobolus ranarum and Conidiobolus coronatus are associated with chronic cutaneous and subcutaneous infections and usually do not

disseminate to internal organs.[1,2] The order Mucorales are usually involved in rhino-cerebral, pulmonary, cutaneous/subcutaneous and gastrointestinal infections in immunocompetent and immunocompromised individuals and is characterised by a tendency to disseminate. Rhizopus and Absidia species are the most common pathogens in the order Mucorales, while Mucor species are only rarely implicated in causation of zygomycosis.[1]

Our patient presented with a chronic subcutaneous infection in the 3rd and 4th web space of right hand without dissemination to internal organs, suggestive of zygomycosis caused by mucor. This is an unusual presentation, as the infections caused by the Mucorales are usually rapidly spreading and tend to disseminate. Moreover, this patient was apparently immunocompetent with no risk factors such as diabetes, haematological malignancy or steroid therapy.

Although Mucor species tend to cause invasive disease in immunocompromised hosts, it produces only cutaneous/ subcutaneous infections in immunocompetent hosts. Traumatic implantation is the common mode of transmission of mucormycosis.[1]

It is often difficult to grow this fungus in culture even from a biopsy specimen, as the zygomycetes due to their coenocytic (aseptate) hyphae, will often be damaged and become nonviable during the biopsy procedure or tissue grinding processes in the laboratory. This emphasizes the importance of direct microscopy in diagnosing this condition as they may often be missed in culture. The presence of broad, infrequently aseptate, thin-walled hyphae with focal bulbous dilatations and irregular branching in KOH mount is diagnostic of zygomycosis.[9] Similarly, tissue sections stained with hematoxylin and eosin stain may clearly reveal the hyphae. Since mucormycosis is fatal in most cases when therapy is delayed, more efforts should be done by the medical community caring for immunocompromised patients to better understand its epidemiology. The availability of advanced culture and molecular techniques would help in correlating environmental fungi to human diseases.[10]

CONCLUSION:

The fact that India turned out to be more affected because of climatic, socioeconomic, scarce hygienic conditions and also diagnostic delay. A high level of clinical suspicion with appropriate use of cytology, tissue biopsy, prompt management and close follow-up are essential elements in reaching recovery for mucormycosis patients. It is recommended to raise the awareness of clinicians toward this rare but fatal fungal infection in order to manage it properly and reach a good outcome.

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