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RHINOMAXILLARY MUCORMYCOSIS IN AN IMMUNOCOMPETENT PATIENT: A CASE REPORT

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ABSTRACT Mucormycosis is an opportunistic fungal infection having a high fatality rate. Mostly it is associated with immunocompromised patients. However, mucormycosis in immunocompetent individuals has been reported in several cases throughout the world with a high incidence of such cases from the Indian subcontinent. Here, we report one such case of rhinomaxillary mucormycosis in immunocompetent patient who presented with nasal obstruction.

KEYWORDS : Mucormycosis, Rhizopus, Immunocompetent

INTRODUCTION

Mucormycosis is a highly aggressive fungal infection that affects the rhino-orbital, respiratory, gastrointestinal or cutaneous systems. [1] Four forms of mucormycosis infection of the sino-nasal tract has been recognized of which two are non-invasive: allergic fungal rhinosinusitis and fungal ball (mycetoma) and two are invasive: chronic invasive (indolent) fungal sinusitis and acute invasive (fulminant) fungal sinusitis.[2]

Infection caused by the fungi of the order Mucorales is known as mucormycosis [3] .The causative organisms are members of the family Mucoraceae, which belongs to order Mucorales of class Zygomycetes. The pathogenic species of the family Mucoraceae, includes the genera Lichtheimia, Mucor, Rhizomucor, and Rhizopus. They are saprophytes which are commonly found in soil, decomposed vegetation, and in healthy human respiratory and digestive tracts, and their distribution is worldwide.[4]

Mucormycosis manifest as one of six different clinical syndromes that are rhinocerebral, pulmonary, gastrointestinal, central nervous system, subcutaneous, and disseminated forms. Among them Rhinocerebral mucormycosis (RCM) is the most common, and has three subtypes: rhino-maxillary, rhino-orbital, and rhino-orbitocerebral [5,6].

The RCM classification has no effect on patient care as the mainstays of treatment are similar regardless of the site of extension. The mainstay of management includes reversal of the underlying cause of immunocompromised state such as diabetic ketoacidosis or neutropenia along with appropriate antifungal therapy and surgical debridement of the involved tissues.[4-10]

Case Report

A 43years non diabetic, non hypertensive male presented to the Department of Otorhinolaryngology with chief complaints of nasal obstruction for three months in right nostril. The obstruction was continuous in nature with no history of trauma, itching, pain, or discharge.

On General physical examination, patient was of average built, afebrile, with vitals (Pulse-82 bpm, regular, BP-162/86 mm Hg). Systemic examination was within normal limits.

Local examination-
Externally there was no visible pathogenic finding. However after Otorhinolaryngological examination there was a mass in the right maxillary sinus which was soft on palpation, non friable, non haemmorhagic and do not bleed on touch.

Diagnostic workup-
Routine investigations revealed normal haematological parameter, non diabetic, serum electrolyte and liver function test was within normal limits.Viral markers (HIV, HCV, HbsAg) – Negative, CD4 count was 721.

Xray PNS -
showed haziness in right maxillary sinus. CECT PNS was in favour of malignant mass involving right maxillary, bilateral ethmoidal, left frontal and left maxillary sinus. Bilateral inferior turbinate hypertrophy was seen with Deviated Nasal Septum to left. There was septal spur with destruction of the meatal wall. Functional Endoscopic Sinus Surgery for right medial maxillectomy with right lateral rhinotomy was done. The biopsy tissue was sent for histopathological examination and Fungal culture. Histopathology revealed polyp lined by respiratory epithelium with dense inflammatory infiltrate in the sub-epithelium. No malignant cells were seen. Features were suggestive of inflammatory polyp.

Fungal Culture -
The tissue was cut into small pieces in Normal saline and was examined in 20% KOH solution and visualized under light microscopy at a magnification of 400x. Branched, non septate, broad hyphae were evident in this preparation. The remainder of the tissue was cultured onto one set of Sabouraud's dextrose agar tube and one set of Sabouraud's dextrose agar with cycloheximide and chloramphenicol. One tube of each set was incubated at 25°C and the other at 37°C.Cotton woolly like white growth appeared in 3 days in the test tube of SDA without antibiotics kept at 25°C. The reverse was pale brownish in colour. After that LPCB mount and slide culture was done that showed broad hyphae with no septa. Numerous stolons among the mycelia, connecting groups of long unbranched sporangiophores. Root like rhizoids are present at the point where the stolons and sporangiophores meet. The sporangiophores are terminate in a dark, round sporangium containing a columella and many oval, colorless or brown sporangiospores. No collarette remains when the sporangial wall dissolves. Large chlamydospores are sometimes seen. So the etiological agent in this case is Rhizopus arrhizus morphologically.

A set of blood culture was also done and both came negative after five days of aerobic incubation.


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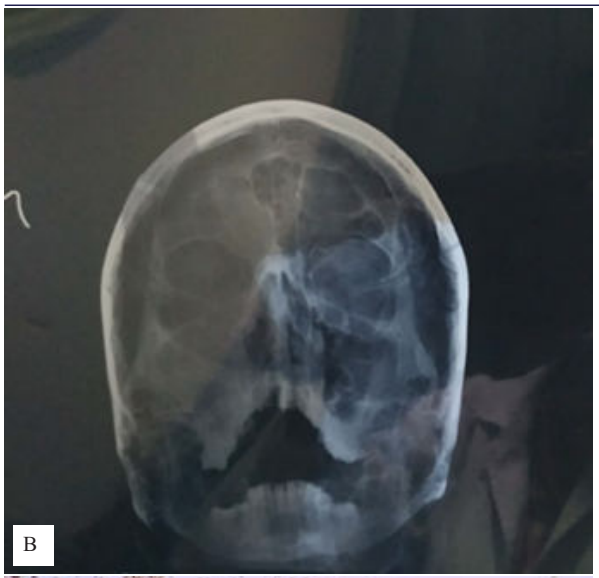

Fig a - Rhizopus arrhizus in SDA culture tube, **b** - X-ray PNS of the patient. **c** - LPCB mount of Rhizopus arrhizus

Patient was treated with Amphotericin B for 4 weeks and made an uneventful recovery.

DISCUSSION-

The risk factors for mucormycosis includes diabetes, corticosteroid therapy, neutropenia, hematologic disorders, renal failure, intravenous drug abuse, immune suppression after organ transplantation and desferroxamine therapy, none of which was present in this case [11,12]. As the fungus is ubiquitous in nature, inhalation of sporangiospores is the most common route for rhino-orbito-cerebral and pulmonary infections, while physical trauma is the main route for cutaneous form. In immunocompetent patients, the nose and/or maxillary sinuses are the predominant site of infection. [13]. In this patient no invasive lesion was seen. As the mucosal/cutaneous epithelium and endothelium is an effective barrier against tissue invasion and angioinvasion, it seems invasive fungal infection in immunocompetent healthy patients might be relatively less. So in our patient there was no invasiveness although allergic manifestations was seen. The chance of developing Mucor infection in immunocompetent patients seems to be related to the virulent capacity of the fungus. [13,14] In this case the exact cause of the infection is not known**.** Our case presented with typical features of mucormycosis which was confirmed by culture. We envisage mucor infection in chronic sinusitis should be taken into consideration. Host agent factors might play a role in infection in immunocompetent person. Thus in immunocompetent patients any polypoidal growth or chronic sinusitis patient even in immunocompetent subjects mucormycosis should be considered.

Limitation-

Diagnosis was done morphologically by culture and microscopy. Antifungal susceptibility test and virulence factor study could not be done as facility was not available. Because of lack of facility molecular confirmation could not be done.

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