



Co-existing Mucormycosis with Aspergillosis in a patient with Diabetes Mellitus- First case report

Nitin M Nagarkar¹, Hitesh Verma², RPS Punia²

¹ AIIMS Raipur ²Government Medical College and Hospital, Chandigarh

Abstract

50 year male patient presented with a history of left nasal bleeding with discharge, left facial palsy for the last 15 days with uncontrolled diabetes. Black eschar was seen in nasal cavities with defect in hard palate on the left side (3×3cm). Debridement and cumulative dose (2.3gm) of amphotericin was given.

Introduction

Invasive fungal infections appear to have increased over the past few years, mostly in immunocompromised patients¹. Moreover, the diagnosis of invasive filamentous fungal disease (IFFD) is often difficult in immunodeficient patients. Mucormycosis is an invasive fungal infection caused by various members of the class Phycomycetes, especially

Mucoraceae, subdivided into the genera *Absidia*, *Rhizopus* and *Mucor*. After Aspergillosis, Mucormycosis is the second most common mycosis caused by filamentous fungi. Since a high level of mortality occurs in immunocompromised patients with IFFD², an accurate and rapid diagnosis is required in order to give the appropriate anti-fungal treatment. In addition, depending on the isolated fungal species, some of these treatments can be inefficient³. The combination of two or more IFFD occurring simultaneously or sequentially in immunocompromised patients makes diagnosis of these patients very challenging.

Combined invasive mucormycosis and aspergillosis has been rarely described, its occurrence has probably been underestimated⁴. Predisposing factors and clinical features in association with these mycoses are almost the same⁵.

We report a case of an associated Aspergillosis and Mucormycosis infection responsible for necrotic and extensive involvement of nose and paranasal sinuses in a patient with uncontrolled diabetes. Histopathological analysis identified *Aspergillus* and Mucormycosis in same patient of diabetes mellitus and the patient was treated successfully. There is no such report mention in literature.

Case report

A 50 year male patient presented with a history of left nasal bleeding with discharge (dark colored), discharge from oral cavity (dark colored) and left facial palsy for the last 15 days. He was a case of Diabetes Mellitus (on irregular treatment). On examination black eschar was seen in both nasal cavities with defect in hard palate on the left side (3×3cm). Investigations showed uncontrolled diabetes and fungal smear report showed aseptate hyphae. Insulin, intravenous antibiotics and physiotherapy exercises for facial palsy with eye care was started. Patient was also started on empirical intravenous liposomal

amphotericin (50-75 mg/day) on the basis of clinical suspicion and fungal smear report. The subsequent biopsy report and fungal culture were showed Mucormycois. The Non Contrast CT (NCCT) of nose and paranasal sinuses showed soft tissue density in nasal cavity and maxillary sinus both side with involvement of left ethmoid and sphenoid sinus.

Heterogenous mass with feature of calcification was present in right nasal cavity and maxillary sinus, there was no intracranial involvement (figure 1, 2). The patient underwent Functional endoscopic sinus surgery right side and Pansinusectomy left side (external approach) under general anesthesia and liposomal amphotericin was continued postoperatively till a cumulative dose 2.3gm over 5week period. Final biopsy report was showed Mucormycosis with Aspergillosis (figure 3) The NCCT scan was repeated on completion of treatment which showed disease free nose and sinuses (figure 4).

Check endoscopy showed minimal crusting and subsequent biopsy report revealed no fungal pathology.

Presently the patient is under follow-up with blood sugar control (by insulin) with improvement in left side facial nerve palsy with healthy nasal cavities. Patient uses palatal obturator for palatal defect.

Discussion

Invasive mycoses have long been recognized as significant pathogens, particularly in immunocompromised patients. Rhino-orbital-cerebral form of mycosis (ROCM) which is a fulminant and life threatening invasive fungal infection⁶ is most commonly encountered in patients with uncontrolled diabetes mellitis Most of the invasive mycoses are caused by filamentous fungi, rarely by other fungi. Of the seven different syndromes of Mucormycois namely the pulmonary, gastrointestinal, CNS, subcutaneous, disseminated, miscellaneous

(bones, joints, heart, kidney, mediastinum) and the rhino-orbital-cerebral form, ROCM is the commonest in clinical practice⁷. The term ROCM refers to the entire spectrum ranging from the sino-nasal tissue invasion (limited sino-nasal disease), progression to orbits (limited rhino-orbital disease) to finally central nervous system involvement (rhino-orbital-cerebral disease)⁸. ROCM is generally encountered in the settings of diabetic ketoacidosis, hematological malignancies, severe burns, renal or hepatic disease, AIDS and organ transplant recipients⁹. Smith and Krichner¹⁰ gave the following criteria for the clinical diagnosis of Mucormycosis: (i) a blood tinged nasal discharge and facial pain, both on the same side, (ii) soft peri-orbital or peri-nasal swelling, going on to discoloration, induration and progressive vascular occlusion, (iii) ptosis of the eyelid, proptosis of the eyeball and complete ophthalmoplegia, (iv) multiple unrelated cranial nerve palsies, and (v) black, necrotic turbinates, easily mistaken for dried, crusted blood. Clinical examination in patient of Aspergillosis reveals grayish brown fungal ball or grayish white color cheesy mass in nasal cavities. The typical clinical features of ROCM are as a consequence of invasion, thrombosis and necrosis of the underlying tissue. Pathological identification of the hyphae is based on different well-described criteria: diameter, presence of septa, and branching angle (right or acute branching), pigmentation. It is important to consider the characteristics of the infiltrate at the inflammatory region that is in contact with hyphae: presence of angioinvasion, of giant cells and of eosinophilic necrosis. The most common fungi associated with ROCM are the Mucor, Rhizopus, Rhizomucor and Absidia. In cases of Aspergillosis, most common fungi are aspergillus fumigatus and flavus. The spores of these saprophytic fungi germinate favorably in an environment of low oxygen, high glucose, acidic medium and high iron levels¹¹.

Rhino-orbito-cerebral form of invasive Mycosis with coexisting Aspergillosis in diabetes mellitus is not quoted in literature. In our patient coexisting facial nerve palsy have been due to simultaneous presentation of Bell's palsy or diabetic neuropathy.

Diagnosis of Mucormycosis requires a high index of suspicion in the relevant clinical setting. The differential diagnoses in such clinical setting include septic emboli, atherosclerotic vasculopathy, vasculitis, local streptococcal and clostridial infection, and invasive Aspergillosis.

An early diagnosis, prompt institution of antifungal therapy (amphotericin) with timely surgical debridement is the key to survival in cases of invasive mycosis. Correction of underlying disorder is of paramount importance. Survival rates in ROCM have ranged from 21-70% in the past but are steadily improving with aggressive management strategies. Treatment outcome is good when the disease is anatomically confined to the sinuses. In other cases the disease carries a very high mortality with conservative treatment alone¹².

Simultaneous presentation of mucormycosis with aspergillosis is not mentioned in literature of cases of diabetes mellitus. We are the first to discuss over experience with readers.

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Legends

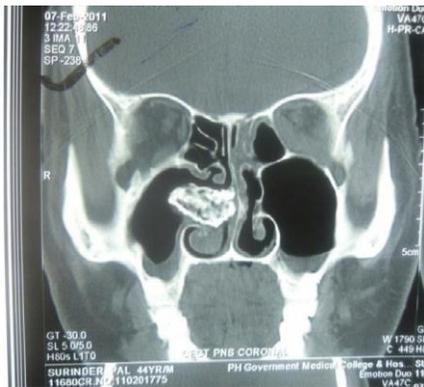


Figure 1 Preoperative NCCT scan coronal view



Figure 2 Preoperative NCCT –axial view

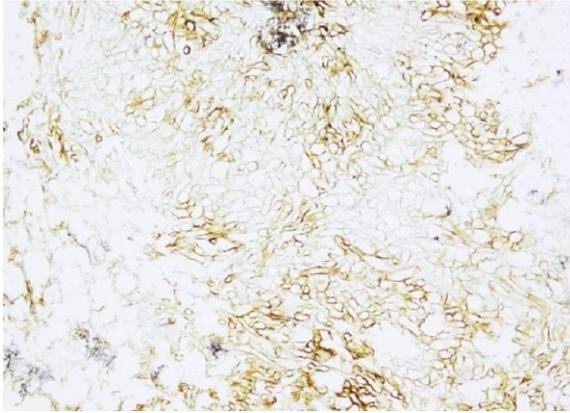


Figure 3 Histopathological view

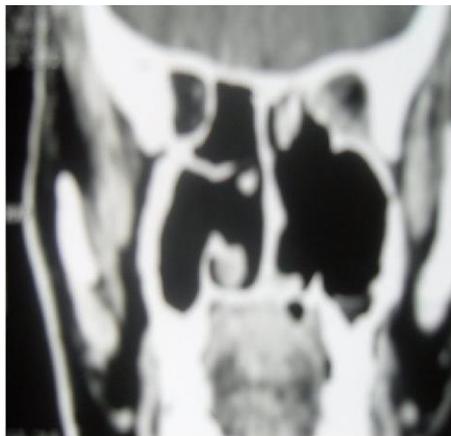


Figure 4 Post surgery coronal view