Case Report

Mucormycosis presenting as an oro-antral fistula: A rare case report

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Abstract

Mucormycosis is a rare opportunistic fungal infection of order mucorales. It has a rapidly progressive and fulminant course with even a fatal outcome, if not recognized early or treated inadequately. This fungal infection is usually more common in immunocompromised people. We are discussing here the case of mucormycosis presenting as palatal perforation along with facial palsy.

Keywords: Mucormycosis, oro-antral fistula, diabetes, facial palsy.

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INTRODUCTION

Fungal infections are also called as mycoses, four major types: Superficial and cutaneous, subcutaneous mycoses, endemic mycoses and opportunistic mycoses. Opportunistic mycoses can cause life – threatening systemic diseases in individuals[1]. Mucormycotina are widely distributed in nature and cause no harm to immunocompetent individuals, but they infect immunosuppressed people, causing mucormycosis. Mucormycosis (formerly zygomycosis) is an opportunistic infection caused by bread mold fungi, including mucor, rhizopus, lichtheimia and cunninghamella, A number of conditions can predispose to the development of this disease like diabetic ketoacidosis, renal failure, malnutrition, organ transplant, acquired immunodeficiency syndrome and chronic corticosteroid therapy, this infection if not recognized early or if treated inadequately can lead to fatal outcome.

CASE REPORT

A 65 year old male report ed to the Department of E.N.T, Peoples college of Medical Sciences and Research centre Bhopal, with the complain of purulent nasal discharge from the nasal cavity he was farmer by occupation, he had left sided facial palsy. Patient was hypertensive and had poorly controlled diabetes for last five years. Patient gave history of left side molar tooth extraction two months back. Extra oral examination showed facial nerve palsy. Intraoral examination showed edematous maxillary arch, non-healing extraction of maxillary left molars, escape of water through left nostril was demonstrated on oral intake. Panoramic radiograph of the jaw bone showed breech in the continuity of sinus floor, suggestive of oro-antral fistula. Routine blood examination was done fasting blood sugar was 200mg/dl and post prandial sugar was 300 mg/dl his blood pressure was 140/110 mm hg, urine examination showed presence of sugar ++ and protein and ketone bodies in urine. Rest of the routine investigations were within normal limits. Diagnosis of oro-antral fistula with facial nerve palsy was made. Surgical closure of the oro-antral communication was made and the scrapings from the fistulous site were sent to the histopathology. Computed Tomography (CT) was advised postoperatively, to study extent of skeletal involvement. Gross: received multiple soft tissue pieces

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altogether measuring 1x1cm. Microscopic examination: study revealed presence of fungal hyphae, broad aseptate hyphae with acute irregular branching pattern characteristic of Mucormycosis along with Actinomycotic colonies both were further confirmed by PAS stain. Antifungal treatment was advised.

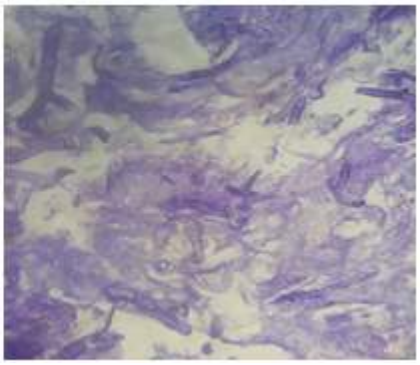

Figure 1: Clinical picture of oro-antral fistula on the hard palate.
Figure 2: HandE stained slide shows fungal hyphae entagled in the necrotic tissue.
Figure 3: PAS positive fungal colonies.

DISCUSSION

Mucormycosis is a rare and opportunistic infection, it was first reported in humans by Paultaufin in 1882[1], it is of the order mucorales. The class "zygomycetes "is subdivided into two orders, which contain agents of human Zygomycosis- the mucorales and the Entomomopthorales. Among the mucorales, Rhizopus is the most common; Mucor, Absidia, Rhizomucor, Cunninghhamell, Saksenaea, Cokeromyces and Apophysomyces [2] have been implicated in causing human disease[3] mucoraceae which includes Rhizopus, Mucor and Absidia genera. These fungi exsists in environment, in soil, air food and animal excreta. The major mode of transmission for the fungi is presumed to be via inhalation of spores from environmental spore. Our patient was a farmer, a paddy field farmer and would have possibly got infected by inhalation of sporangiospores from the atmosphere or contact with contaminated soil. The hallmarks of disease with these organisms are angioinvasion, thrombosis, infarction and finally necrosis of the involved tissue. Mucormycosis is characterized by rapid growth rate and typically cause aggressive and frequently angioinvasive infections, especially in immunosuppressed hosts. Diabetes is the single most common predisposing factor especially when associated with ketoacidosis. Mucor thrives in an acidic ph and glucose rich medium. Hyperglycemia enhances fungal growth and impairs neutrophill chemotaxis, while lactic acidosis decreases phagocytosis[4,5]. Our patient had uncontrolled diabetes along with ketoacidosis for last five year and he took off and on medication for that. Zygomycosis present as spectrum of diseases, depending on the portal of entry and the predisposing risk factors. Five clinical forms are rhinocerebral, pulmonary, abdominal pelvic and gastrointestinal, prunarycutaneous and disseminated forms[6] Rhinocerebral mucormycosis represents one-third to onehalf of all cases of zygomycosis. It manifests itself in a setting of poorly controlled diabetes in about 70 % cases[7]. The process starts from the paranasal sinuses following inhalation of spores disease begins with the symptoms of sinusitis, nasal discharge. Soft tissue swelling, spread along the cribiform plate can result in intracranial involvement can result in multiple cranial nerve palsies. The most common oral sign of mucormycosis is ulceration of the palate, which results from necrosis due to invasion of palatal vessels[8,9]. There are many differentaial diagnosis of palatal perforation like syphilis, leprosy, wegener's granulomatosis. However in our case evidence of diabetes or immunosuppression in a patient presenting with necrotic lesions of the nasal cavity and palate strongly favours the diagnosis of a deep fungal infection. CT and MRI may demonstrate erosion or destruction of bone or sinuses and help delineate the extent of the disease. Diagnosis of zygomycosis is easily made on histopathology sections by the presence of wide, ribbon like aseptate, hyaline, hyphal elements often intermingled with areas of necrosis involved tissue demonstration of aseptate hyphae with wide angle branching with angioinvasion is characteristic of mucor species. The special stains that can be done are Gomori methenamine silver stain (GMS), periodic aacid Schiff (PAS) and Papanicolau stains. Here we have done PAS stain for further confirmation. Mucormycosis disease spreads rapidly and can cause life threatening complications so accurate diagnosis and prompt treatment is required which involves several simultaneous approaches like surgical debribement, antifungal therapy, and medical management.

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