

Rare Opportunistic Bread Mold Fungal Infection of Maxillary Sinus in a Diabetic Patient

Abstract

Mucormycosis is a rare potentially fatal opportunistic fungal infection that affects human beings. Normally, a healthy is immune to such infections but there are some risk factors which predispose a person to mucormycosis, of which malnutrition and diabetes mellitus are the most common risk factors. India is the most commonly affected country by mucormycosis because of high proportion people with low socioeconomic status and diabetes mellitus. In diabetes mellitus, mucormycosis is more aggressive and fatal due to an impaired host defense mechanism. In spite of being a rare and fatal fungal infection, early diagnosis and prompt multidisciplinary treatment comprising control of underlying predisposing factor, surgical management, and medical management can be helpful in such cases.

Keywords: Diabetes, Mucorales, mucormycosis, rhinocerebral

Introduction

Mucormycosis is a rare fulminant, fatal opportunistic mycotic infection that affects the human beings.^[1,2] According to analysis, India is the most affected country by mucormycosis contributing to 44.3% of the entire cases reported worldwide, followed by the USA and Australia. The predisposing risk factors for mucormycosis in India may include socioeconomic conditions, poor hygienic conditions, and diabetes mellitus.^[1] Early and prompt diagnosis is very crucial in such cases because any delay in the initiation of treatment can be life threatening due to the ability of these fungi to invade the adjacent blood vessels and to embolize itself at distant organs including the brain.^[3]

Case Report

A 52-year-old female patient reported with a chief complaint of nasal obstruction with facial swelling and facial pain on the right side of the face and also complained of a yellow-colored thick nasal discharge for the past 2 months. She was recently diagnosed with diabetes mellitus with blood sugar levels of 300 mg/dl and is being treated by oral hypoglycemic drugs. Extraoral clinical examination revealed diffuse swelling in the right maxillary

region. On intraoral examination, prominent swelling was seen in the buccal vestibule extending from the infraorbital margin to the right angle of the mouth and extending posteriorly till second molar region. On palpation, the area was tender and the first and the second molars were mobile. To rule out the involvement of the maxillary sinus, computed tomography (CT) scan was advised. On CT scan of nose and paranasal sinus, she had the features of sinusitis with the destruction of walls of the right maxillary sinus as shown in Figure 1. Ear, nose, and throat (ENT) consultation was taken. A strong suspicion of fungal sinusitis was made. They advised anterior rhinoscopy. Rhinoscopy revealed a thick yellow-colored mucopurulent discharge in the right middle meatus. A pinkish mass was seen in right middle meatus during fungal sinusitis of mucormycosis. Before endoscopic debridement, the patient's blood sugar level was controlled with human insulin 12 units twice a day. Endoscopic sinus surgery was performed, and debridement and clearance of right maxillary sinus was done. The patient was treated with an intravenous

infusion of liposomal amphotericin B (5 mg/[kg body weight]) for 4 weeks. The dose was started with 1 mg/kg body weight and slowly increased to 5 mg/

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kg body weight depending on renal function which was continuously monitored by serum urea and creatinine levels. Serum potassium levels were also monitored and replenished accordingly. There was no adverse effect of antifungal treatment during the entire course. The patient responded well to surgical debridement and antifungal therapy. The patient was put on recall after every 1 month. The postoperative CT scan was taken at the recall as shown in Figure 2.

Discussion

Fungi of class Zygomycetes consist of orders Mucorales and Entomophthorales.^[4] Mucorales are ubiquitous fungi that are found worldwide on fruits, air, bread, compost pile, animal excreta, and soil as saprophytes and cause a life-threatening rare fungal infection known as mucormycosis.^[2,5] Paltauf in 1885 gave the first histologic description of generalized mucormycosis.^[5,6] The fungi and spores of Mucorales have minimal pathogenicity in normal individuals; however, they can cause fulminating opportunistic infection (mucormycosis) in immunocompromised individuals.^[5] Since these fungi are ubiquitous in nature inhalation of its sporangiospores is the most common route for rhinocerebral mucormycosis.^[1,5] It usually starts in the nasal mucosa or palate and extends to involve the paranasal sinuses.^[2,3] From the sinuses, it then involves the retro-orbital region after spreading through either direct extension or through the lacrimal, angular, and ethmoidal vessels.^[2] This once it invades the arteries leads to thrombosis of the blood vessels and causes necrosis of the hard and soft tissues.^[7] The pathophysiology of necrosis is depicted in Flow Diagram 1. Upon entering the arteries, the fungus can also spread to orbit, lungs, and intracranial structures which can be fatal for the patient.^[7]

The various predisposing factors for mucormycosis are diabetes mellitus, blood dyscrasias, renal failure, long-term steroid therapy, cirrhosis, burns, AIDS, malnutrition, neutropenia, iron overload, organ transplant, and immunosuppressive therapy.^[2,6] Almost 40% of mucormycosis cases are associated with diabetes mellitus and out of these almost 70% cases are of rhinocerebral mucormycosis.^[2] Diabetic patients have a high incidence of mucormycosis as these Mucorales produce ketoreductase enzyme which allows them to utilize the patient's ketone bodies.^[5] Hyperglycemia also impairs the ability of the phagocytes to move toward and kill the fungi by oxidative and nonoxidative methods which in turn stimulates fungal growth.^[4,5]

It has very high morbidity and mortality rate with an incidence of 62.5% for the rhinocerebral form.^[1] It is attributed to its capacity for rapid vascular invasion and subsequent tissue necrosis and infarction. The survival rate of rhinocerebral form of mucormycosis is higher than that of pulmonary and disseminated forms as the rhinocerebral form is diagnosed at an earlier

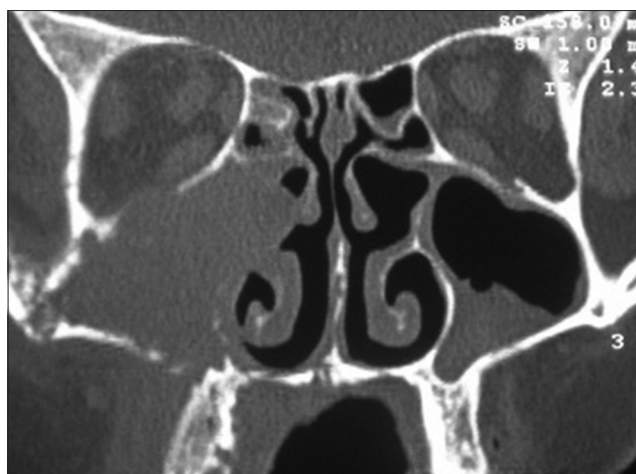


Figure 1: Preoperative computed tomography scan

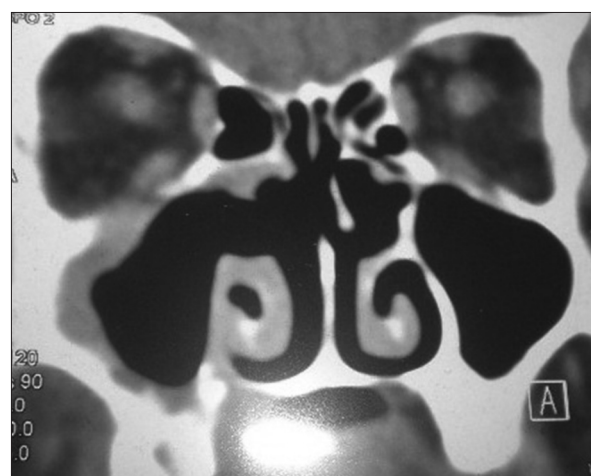
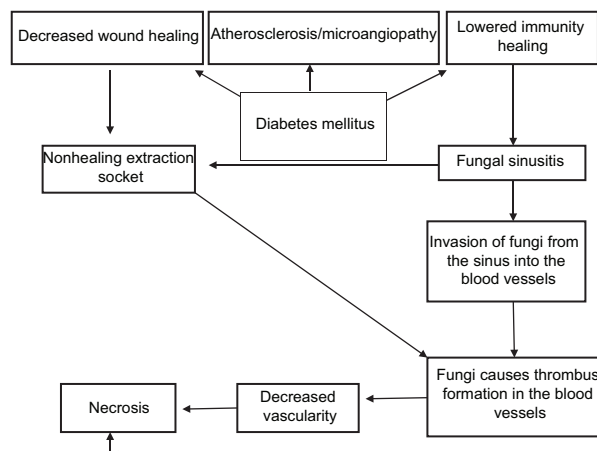


Figure 2: Postoperative computed tomography scan



Flow Diagram 1: Depicting pathophysiology of necrosis secondary to infection by mucormycosis

stage, and the underlying risk factor can be treated. The dentists should be well versed with mucormycosis, especially in diabetic and immunocompromised patients. Atypical symptoms such as sinus pain, facial

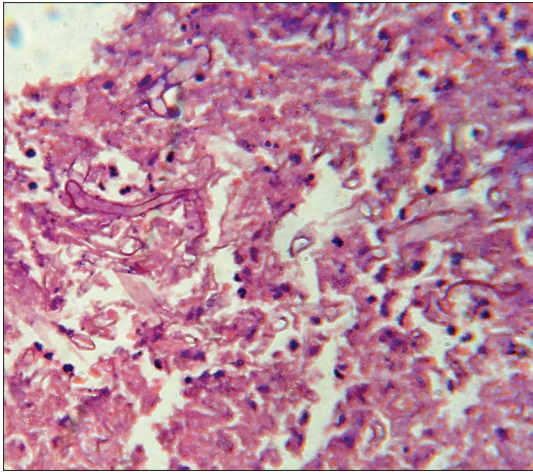


Figure 3: H and E staining showing hyphae

pain, and unexpected odontalgia of otherwise healthy teeth should alert the dentist. Moreover, when the condition deteriorates even after dental treatment, one should consider rare conditions such as mucormycosis and seek medical advice. A definitive diagnosis of mucormycosis can only be made by tissue biopsy and to identify the characteristic hyphae. A histopathological diagnosis is considered more precise and accurate than simple culture as the culture may be unyielding due to the depth of invasion of the infection. It is very important to rule out aspergillosis as there is a lot of resemblances histologically between them.^[7] Histologically, in aspergillosis, hyphae are septate and branching, whereas in mucormycosis, the hyphae are nonseptate, have a smaller width, and they branch at right angles^[2,7] [Figure 3]. CT is 100% sensitive and 78% specific in the diagnosis of sinonasal mycosis. In CT, it usually presents as an aggressive enhancing soft tissue mass that extends beyond the confines of the sinus cavity and shows extensive bone destruction.^[3]

Conclusion

Early diagnosis is vital in these infections because delay in initiation of treatment can be life threatening due to the propensity of the fungi to invade adjacent blood vessels and embolize to distant organs including brain. Any diabetic patient who has clinical and radiographic features of rhinosinusitis should be suspected as having mucormycosis unless proven otherwise. A multidisciplinary approach consisting of dental specialists and ENT surgeons is very important for the successful management of mucormycosis patients.

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Conflicts of interest

There are no conflicts of interest.

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