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Rhinomaxillary Mucormycosis: Report of a case

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Abstract

Mucormycosis is a rare but life-threatening opportunistic fungal infection, predominantly affecting immunocompromised individuals such as uncontrolled diabetics. Rhino maxillary, a subtype of rhino cerebral form of mucormycosis frequently presents with oral manifestation as ulceration of the palate, which results from necrosis due to invasion of a palatal vessels and sometimes may lead to perforation of the palate. Treatment of mucormycosis must be initiated at the earliest as it progress relentlessly and has a fatal outcome. Successful management consists of treating the underlying disease and aggressive surgical debridement of necrotic tissue along with systemic antifungal therapy. Here we present a case of mucormycosis involving palate and maxillary antrum in a diabetic patient.

Keywords: diabetes, immunocompromised, maxillary sinus, mucormycosis, palatal ulceration

Introduction

Mucormycosis is one of the most rapidly progressing and lethal form of fungal infection in humans which usually begins in the nose and paranasal sinuses [1]. This saprophyticfungus is non-pathogenic for healthy individuals and can be cultured regularly from nose, throat and oral cavity representing opportunistic rather than a true pathogen [2].Mucormycosis, a deep seated mycotic infection is more commonly seen in immunocompromised patients. The infection is acquired by the inhalation of spores or by direct inoculation of the fungus into the

damaged skin or mucosa. This fungus invades the arteries, forms thrombi within the blood vesselsthat reduce blood supply and cause necrosis of hard and soft tissues [1, 2]. Once entered into the arteries, the fungus can spread to orbital, intracranial structures or can disseminate to other organs[3, 4]. Usually presents as an acute infection and manifests as rhinocerebral, pulmonary, gastrointestinal, cutaneous or disseminated form [1-4].

Tissue necrosis is the clinical hallmark of invasive mucormycosis due to angioinvasion and subsequent thrombosis.Rhino maxillary, a subtype of rhino cerebral form of mucormycosis frequently presents with oral manifestations and may lead to considerable dilemma in clinicians unfamiliar with this entity, which in turn may worsen the prognosis for the patient.

Here we present a case of mucormycosis involving palate and maxillary antrum in a diabetic patient.

Case Report

A 65 year old male patient was reported with the complaint of palatal ulceration since a month. Patient had noticed swelling in the same region 3 to 4 months back that was initially small, which slowly progressed and turned to an ulcer of present size. The lesion was painless and there was no history of nasal congestion and regurgitation, fever, purulent discharge, and paraesthesia. Patient was a known case of diabetes mellitus since 7 to 8 years and was on oral hypoglycemics since then. He got admitted in a local private hospital due to increased blood sugar level one month ago wherein he got treated for the diabetes and was referred to our institution for the investigation and treatment of palatal ulceration.

On examination there was no evidence of swelling extra orally. Intraorally, an irregular endophytic ulcerative lesion of approx. size 4 X 6 cm was present involving the complete right side of palate and alveolar ridge, crossing the midline and extending onto left side sparing soft palate. Exposed palatal bone was present and ulcer's base was covered with necrotic yellowish white slough. On posterior aspect of ulcer slight blackish necrotic tissue was noted. The margins were firm and lesion was nontender [Fig. 1]. Bilateral submandibular lymphadenopathy was present and the lymphnodes were firm, mobile and non-tender.

Radiographic examinations[Maxillaryocclusal, OPG and PA Water's view] revealed erosion of maxillary bone, destruction of floor and posterior border of right maxillary antrum as well as destruction of right nasal floor [Fig. 2a and 2b]. PA Water's view showed haziness/opacification in right maxillary sinus [Fig. 2c]. CT scan revealed the erosion of right side of maxilla, palate, nasal floor, and destruction of bony walls of maxillary antrum.

Considering history, clinical and radiographic features provisional diagnosis of deep mycotic infection of maxilla involving antrum and nasal cavity was made. Differential diagnosis of squamous cell carcinoma, lymphoma, salivary gland adenocarcinoma and chronic granulomatous infection were given.

A swab was taken from the lesion for culture and sensitivity. The report revealed the presence of fungal filament with branched septate hyphae. Incisional biopsy was done and histopathology report showed granulation tissue with areas of necrosis which were superimposed by fungi having large non septate hyphae and branching at obtuse angles. Ovoid sporangia were also seen. Special staining with Grocott's modified silver methenamine stain also revealed fungi with non septate hyphae and sporangia indicative of mucormycosis [Fig. 3a and 3b].

The patient was hospitalized for further management where blood sugar levels were brought under control with insulin. The patient was administered Amphotericin-B 0.8mg/kg/day intravenously for two weeks. It was slowly infused over 4-6 hours and blood urea and creatinine

levels were monitored as the drug can cause renal toxicity. The necrotic bone along with 1 cm of adjacent normal bone was excised under general anaesthesia. Two weeks later the area started healing and subsequently after three months, after complete healing an obturator was made for the patient. Now the patient is doing well and is under regular review.

Discussion

Mucormycosis is a rare, often deadly, opportunistic fungal infection caused by saprophytic organisms from mucormycocae family. Although the term phycomycosis and zygomycosis are occasionally used, mucormycosis is the most frequently used term, and it was first described by Paultauf in 1885[3]. It is the third common fungal infection after candidiasis and aspergillosis in diabetics and post-allogenic stem cell transplant patients. Up to 50% of patients presenting with mucormycosis are diabetic [4].It is a highly angioinvasive and a relentlessly progressive condition resulting in high morbidity and mortality. It usually occurs in the presence of some underlying immune compromising conditions like uncontrolled diabetes, malignancies such as lymphomas and leukemias, renal failure, organ transplant, long term corticosteroids and immunosuppressive therapy,cirrhosis, burns, protein energy malnutrition, deferoxamine therpay and AIDS. Ourpatient had uncontrolled diabetes which is a well known predisposing factor for mucormycosis [1-5].

These organisms commonly found in the soil or as a mould on decaying food or organic matter.They are non-pathogenic for healthy individuals and can be cultured regularly from nose, throat and oral cavity representing opportunistic rather than a true pathogen [2]. These fungi can reproduce either sexually or asexually with an incubation period of 1 to 2 weeks making diagnosis late. It can affect any organ in the body once transported through blood. This affects Lungs followed by sinus, liver, kidney and CNS due to either breathing in the spores or by directly coming in contact with the open wound. Such cases most commonly seen during natural calamities like earthquakes or road traffic accidents.

Mucormycosis presents as a spectrum of disease, depending on the portal of entry and underlying predisposing risk factors of the patient. Following are 5 major clinical types: rhinocerebral, the most common type, represents one third to half of all cases as reported by Pillsbury H C et al followed by pulmonary, gastrointestinal, cutaneous and disseminated form [6].

The disease process usually originates in the paranasal sinuses following inhalation of the fungal spores. It begins with symptoms consistent with sinusitis which includes sinus pain, nasal discharge, anaesthesia, headache, fever and soft tissue swelling. Then, it becomes rapidly progressive, extending into neighbouring tissues (cellulitis). Involved tissues become red, then violaceous and finally black as vessels are thrombosed and tissues undergo necrosis. Periorbital oedema, proptosis, tearing and ocular or optic nerve involvement is seen due to extension into the orbital region. Spread along the cribriform plate can resultin intracranial involvement [7].

Jones et al stated that the most common oral sign of mucormycosis is ulceration of the palate, which results from necrosis due to invasion of a palatal vessel [5]. Extension from the sinuses into the mouth causes painful, black necrotic ulcerations in the hard palate which sometimes may lead to perforation of the palate. The lesion is characteristically large and deep, causing denudation of the underlying bone. Ulcers from mucormycosis have also been reported on the gingiva, lip and alveolar ridge[2].

Differential diagnosis of a lesion presenting as palatal ulceration with or without perforation of the palate should include chronic granulomatous lesions like tertiary

syphilis , leprosy, cancrumoris, traumatic ulcerative granuloma, intranasal cocaine abuse , malignancies, especially nasal T cell lymphomas, squamous cell carcinoma, salivary gland adenocarcinomas, plasmacytoma, Wegener'sgranulumatosis, necrotising sialometaplasia and midline non-healing granuloma [2,7-10].

Plain radiography of paranasal sinuses and orbits may demonstrate mucosal thickening, opacificationswith or without fluid levels and irregular destruction of bony walls of nose and paranasal sinuses. CT scan and MRI may demonstrate erosion or destruction of bone and shows the exact extent of the disease. In the present case opacification of maxillary sinus, erosion of right side of maxilla and palate, nasal floor, and destruction of bony walls of maxillary antrum was evident. All these imaging findings are not specific for diagnosis of mucormycosis[7, 8].

There is no specific serological test for its diagnosisand a definitive diagnosis of mucormycosis can only be made by a biopsy that identifies the characteristic hyphae and by culture. Histologically, mucormycosisis characterized by extensive tissue necrosis and the presence of numerous large nonseptate hyphae, with budding and dichotomous branching, giving a ribbon like appearance.

Successful management of mucormycosis consists of treating the underlying disease and aggressive surgical debridement of necrotic tissue along with systemic antifungal therapy. The use of amphotericin B in patients with mucormycosis has a survival rate of up to 72%, which is a widely published and accepted treatment. Other antifungal chemotherapeutic agents such as posaconazole and caspofungin are used as a second line treatment[2]. Fluconazole may be of benefit in treating this infection, although some reports indicate an increase in resistant organisms to fluconazole. More recently hyperbaric oxygen therapy is believed to improve neutrophilic killing by higher oxygen delivery and delaying or totally inhibiting the growth of fungal spores and mycelium [7]. Treatment of mucormycosis must be initiated at the earliest as it progress relentlessly and has a fatal outcome. The mortality rate is 50 to 100 % in spite of therapy [2].

Dentist should be alert and suspicious in cases of palatal ulceration with or without perforation especially in elderly patients who are immunocompromised. Early diagnosis and complete treatment will be of great benefit to the patient in terms of improved survivaland reduced morbidity from extensive disease; dissemination of the disease, wide spread surgical resection, post-surgical complications and loosing physiologic functions.

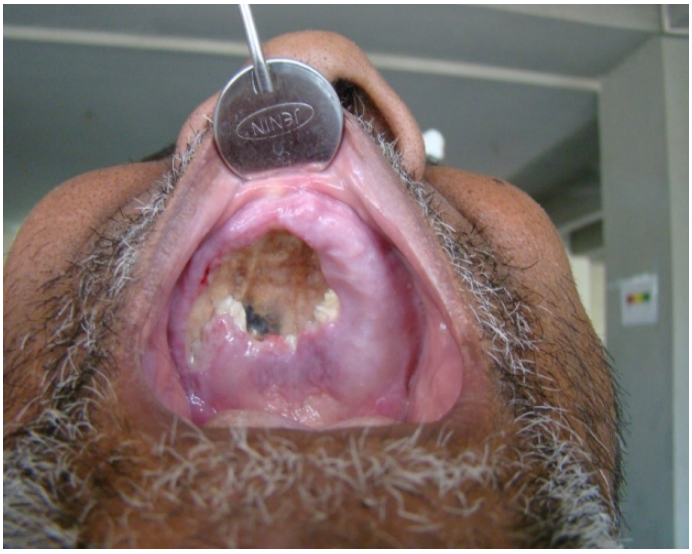

Fig.1: Intraoral picture showing irregular necrotic ulceration of palate with exposed bone.

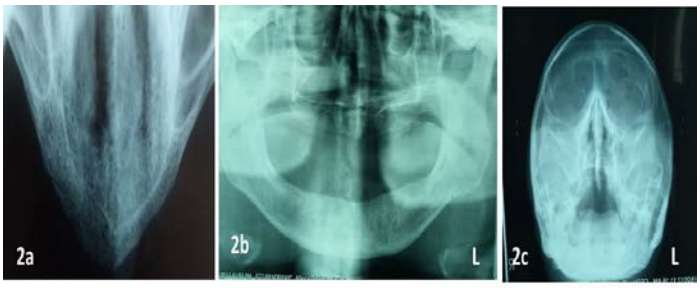

Fig.2a. Maxillary occlusal radiograph showing the erosion of right maxilla and nasal floor.

Fig.2b. Orthopantomograph showing destruction of floor and posterior border of right maxillary antrum.

Fig.2c. PA Water's view showing haziness/opacificationof right maxillary antrum

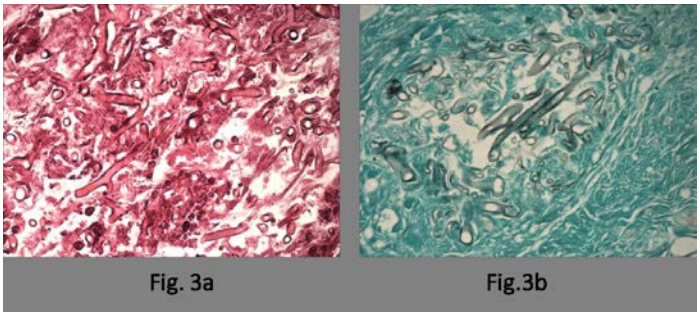

Fig.3a & 3b: Histopathological photomicrograph of H&E stain (3a) Grocorts silver methanamine stain (3b)

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