Mucormycosis and Myiasis in Uncontrolled Diabetes: A Double Whammy

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ABSTRACT

Mucormycosis is a rare, often fatal opportunistic fungal infection that is caused by an aerobic saprophytic fungus belonging to the order mucorales and class zygomycetes. Myiasis is caused by the members of the Diptera fly family that lay eggs or larvae on food, necrotic tissue, open wounds, and unbroken skin or mucosa. We report a rare case of mucormycosis coexisting with oral myiasis in a 50-year-old woman with uncontrolled diabetes mellitus.

Keywords: Palatal ulcer, Mucormycosis, Myiasis.

INTRODUCTION

Mucormycosis is a rare, often fatal opportunistic fungal infection that is caused by an aerobic saprophytic fungus belonging to the order mucorales and class zygomycetes. It is a common inhabitant of soil, decaying vegetation and can also be cultured from human nose, throat, oral cavity and stools on a routine basis. The most common predisposing factor for mucormycosis is uncontrolled diabetes mellitus, which favors fungal growth.1

Myiasis is caused by the members of the Diptera fly family that lay eggs or larvae on food, necrotic tissue, open wounds, and unbroken skin or mucosa.2 When tissues of the oral cavity are invaded by the parasitic larvae of flies, this condition is named as oral myiasis.2

The purpose of this paper is to report a rare case of mucormycosis coexisting with oral myiasis.

CASE REPORT

A 50-year-old female reported to the department of oral medicine and radiology with a chief complaint of a painful swelling on the left middle third of face and bleeding from the left nostril since 24 hours. Patient was from a low socioeconomic background, residing in a rural area. Her past medical history revealed that she is a known diabetic and hypertensive since 5 years and was not under regular medications. She also complained of nasal congestion and headache since 15 days. General physical examination revealed that the patient was febrile with an increased pulse rate. A diffuse cellulitis involving the left middle third of face, causing periorbital edema and inferiorly extending up to the angle of the mouth, was seen. Skin overlying the swelling appeared stretched, glossy and erythematous. Epistaxis from the left nostril was noted (Fig. 1).

Intraoral examination revealed poor oral hygiene, caries, lack of several teeth and severe halitosis. Necrosis and ulceration of the posterior hard palate in the mid-palatal region, extending up to the junction of hard and soft palate was seen, causing palatal perforation. The lesion measured approximately 3 × 1.5 cm and was roughly oval in shape with well-defined borders. Tenderness on palpation was present and the surrounding mucosa presented with yellowish-white necrotic slough and erythema (Fig. 2). Irrigation of the lesion was carried out to clear off debris. This revealed the presence of live wriggling larvae raising a strong suspicion of a communication with the nasal cavity where these maggots would have harbored.

Fig. 1: Diffuse cellulitis of left middle-third of face causing periorbital edema
Surprisingly, the patient was unaware of the presence of maggots. Based upon the clinical findings of the presence of maggots, a diagnosis of oral myiasis was made.

Radiographic examination included an orthopantomogram which revealed an irregular haziness in the lower part of the left maxillary sinus revealing a thickened sinus mucosa. A break in the continuity of the hard palate in the midline at the root apex of left maxillary central incisor was seen (Fig. 3). CT scan of the maxilla and paranasal sinuses revealed sequestrated bone in the hard palate. There was an irregular asymmetrical dense mucosal thickening in the maxillary and ethmoidal sinuses without any intracranial involvement. Extensive destruction of the soft and hard tissues of the palate, which had caused an oronasal communication, was also seen (Fig. 4).

Biochemical investigations revealed a blood glucose level of 474 mg/dl and an erythrocyte sedimentation rate of 36 mm/hr.

A smear from the palatal lesion was made and histo-pathological examination with hematoxylin and eosin stain and PAS revealed the presence of aseptate, branched mucormycotic hyphae. A final diagnosis of oral myiasis with mucormycosis secondary to uncontrolled diabetes mellitus was made.

The patient was hospitalized immediately and the physicians controlled the elevated blood glucose levels with insulin. The palatal wound was packed with turpentine soaked gauge and the maggots were removed manually using clinical forceps. They measured approximately 9 to 10 mm in length and 3 to 4 mm in diameter, and had a yellowish-white body with a darker head portion (Fig. 5). The patient was administered cefazolin 1 gm 6-hourly and metronidazole 400 mg 8 hourly for 5 days. The larvae were sent for entomological analysis which were verified to be Musca domestica. The necrotic bone along with 1 cm of adjacent bone was excised and debrided with betadine. The patient was administered Amphotericin-B 0.8 mg/kg/day intravenously for 2 weeks. It was slowly infused over 4 to 6 hours and blood urea and creatinine levels were monitored as the drug can cause renal toxicity. Postoperatively, the patient was advised an obturator to prevent oronasal regurgitation.

**DISCUSSION**

Mucormycosis is caused by saprophytic fungi of the order Mucorales. Rhizopus, Rhizomucor and Absidia are the most common isolated organisms from the patients with mucormycosis.3

Mucormycosis usually begins in the nose and paranasal sinuses. This fungus invades the arteries, forms thrombi within the blood vessels that reduce blood supply and cause necrosis of hard and soft tissues. Once entered into the arteries, the fungus...
can spread to orbital and intracranial structures. Usually mucormycosis presents as an acute infection and manifests as rhinocerebral, pulmonary, gastrointestinal, cutaneous or disseminated form. Mucorales may cause severe, often fatal infection in immunocompromised conditions, like diabetes mellitus, which is the most common predisposing factor, especially when it is associated with ketoacidosis. This relationship has been attributed to impaired neutrophil function, neuropathies and vascular insufficiency has been seen in diabetes. In addition, the acidic environment, increased glucose levels and increased levels of free iron may enhance fungal growth. In our case, the predisposing factor was uncontrolled diabetes, though ketoacidosis was absent.

Rhinocerebral mucormycosis is the most recognized form of this disease because of its severity, and oral manifestations are usually the first clinical signs to arise. Progression is very rapid and dissemination to the brain may be fatal.

In the early stages of the disease, patients exhibit facial cellulitis, anesthesia, nasal discharge, necrotic turbinates, fever, headache and lethargy. In this patient, facial cellulitis, fever, nasal congestion and headache were present. Periorbital edema indicative of involvement of nasal turbinates and orbit were also seen.

In the oral cavity, mucormycosis generally presents as an ulceration of the palate with a characteristic black eschar which may be due to a necrotic infarction of the palate. The lesion is usually deep and causes denudation of the palate along with osteomyelitis of the affected bones. Necrosis of the palate may be the result of thrombosis of the sphenopalatine or internal maxillary artery. In the present case, the oral manifestation was the presence of a palatal ulcer. The maxillary antrum was involved and the involvement of nasal cavity resulted in an oro-nasal communication.

Differential diagnosis of a lesion presenting as a palatal perforation should include squamous cell carcinoma, tertiary syphilis, cancerum oris, midline lethal granuloma, mechanical trauma, malignancies, Wegener’s granulomatosis, aspergillosis and other systemic mycoses.

Radiographically, rhinocerebral mucormycosis demonstrates nodular thickening of the sinus mucosa, sinus opacification without fluid levels and spotty destruction of paranasal sinuses.

Definitive diagnosis is easily established by the detection of broad, irregularly shaped, nonseptate hyphae with right angled branching invading the tissue on hematoxylin and eosin stain. These fungi can be better visualized using PAS or methenamine silver stains. Treatment of mucormycosis requires an aggressive surgical debridement of necrotic tissue followed by systemic antifungal therapy and immediate control of underlying disease process. Amphotericin B is the first drug of choice but is associated with a side effect of renal toxicity. Thus, careful monitoring of serum urea nitrogen and creatinine are essential during Amphotericin B therapy. Liposomal Amphotericin B should be used when the renal function is impaired. Closure of the oroantral/oronasal communication can be done surgically by using free flaps or by construction of a prosthetic appliance.

The term myiasis (from the Greek “Myi” meaning fly) refers to the infestation of living tissues of animals or humans by Diptera larvae. Human myiasis is a rare condition. It is more common in regions with a warm and humid climate and in areas in which people live close to livestock, mostly in rural zones and villages. Based upon the degree of host dependence, myiasis can be: Obligatory, where the maggots require living tissues for larval development and facultative, where flies opportunistically use necrotic wounds as a site in which to oviposit and incubate their larvae. According to the mode of infestation, myiasis can be: ‘accidental myiasis’ in which larvae ingested accidentally with food produce infection, ‘semi-specific myiasis’ wherein the larvae are laid on necrotic wound, or ‘obligatory myiasis’ in which larvae affect undamaged skin. Depending upon the anatomic sites affected, myiasis can be classified clinically as: Cutaneous myiasis, myiasis of external orifices (oral, nasal, ocular, aural, anal, genital) and myiasis of internal organs (intestinal, urinary).

Oral myiasis was first described by Laurence in 1909. Poor oral hygiene, ulcerated lesions, severe halitosis and alcoholism are the pathological factors associated with it.

Musca domestica (Family: Muscidae), the common housefly, is the most common of all flies fluttering in homes and is one of the main causes of wound infestation known as ‘facultative myiasis’. Each female fly can lay approximately 500 eggs in several batches. Fruit and vegetable cull piles, partially incinerated garbage, incompletely composted manure, human excrement, horse and cow manure are highly favored sites for its breeding. Human myiasis occurs accidentally with direct inoculation when the eggs are laid at the edges of wound or on the mucous membranes of natural body orifices. The legless maggots emerge from the eggs in warm weather within 8 to 20 hours, and immediately feed on and develop in the material in which the eggs were laid. Larvae feed voraciously on the tissues, destroying them and inducing serious hemorrhage.

Our patient presented with semispecific oral myiasis since there was a necrotic palatal lesion of mucormycosis secondary to uncontrolled diabetes mellitus. Factors, like rural Indian background, where sleeping outdoors is common, poor dental hygiene, and fetid odor from an open palatal wound, favored the direct inoculation, and the eggs may have been laid at the edges of the necrotic wound. The standard treatment options include maintenance of nutrition, antimicrobials for secondary infection and manual removal of larvae with or without topical asphyxiating drugs (ether, chloroform, olive oil, turpentine oil) that make the larvae to come out. The wound usually heals without further treatment once it is cleansed of the larvae and...
secondary infection is prevented.\textsuperscript{10} Topical nitrofurazone 0.2% has also been used in some cases.\textsuperscript{13} Systemic ivermectin can produce favorable results in severe cases.\textsuperscript{10}

This case report is unique, because to the best of our knowledge, concomitant involvement of mucormycosis and oral myiasis in uncontrolled diabetes has not been documented in the literature before.

To conclude, dentists should be aware of the importance of early diagnosis and management of mucormycosis, because oral signs are usually the first clinical manifestations of this infection especially, if the host is immunocompromised.\textsuperscript{1}

REFERENCES