Rhinomaxillary Orbital Mucormycosis in A Diabetic: A Case Report

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Abstract
Mucormycosis was earlier considered a rare fungal infection, but recently the number of cases reported has increased considerably throughout the years. A 35 year old patient reported with a swelling on right side of face with severe headache and palatal ulcer. After thorough clinical examination and investigations, he was diagnosed with mucormycosis. Need for an early diagnosis and to render the adequate treatment is necessary to combat this disease and thus reduce mortality rates of the affected patients. This article gives an insight into the diagnosis, pathogenesis and management of mucormycosis.

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1. 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 (Auluck A., 2007). Mucormycosis is a saprophytic aerobic fungus commonly found in our environment, for example, in bread moulds or decaying vegetation. This organism is frequently found to colonize the oral mucosa, nasal mucosa, paranasal sinuses and pharyngeal mucosa of asymptomatic patients (Fogarty C et al., 2006). For many years the term mucormycosis has been interchangeably used with zygomycosis in the medical literature. Currently, in line with a recent classification, we prefer to use the term mucormycosis rather than zygomycosis (Mignogna M.D et al., 2011).

This condition was first described by Paltauf in 1885 in human beings (Doni BR et al., 2011). The Mucorales species most often recovered from clinical specimens are those of the genera Rhizopus (the most common genus associated with mucormycosis), Lichtheimia (formerly known as Absidia and Mycocladus), and Mucor (Petrikkos G et al., 2012).

Commonly, this disease occurs in persons with immunosuppression, diabetic ketoacidosis and those whom on antibiotics, steroid (or) cytotoxic therapy. Other predisposing factors include underlying malignancy, burns, malnutrition, blood dyscrasias and renal failure (Doni BR et al., 2011). Based on clinical presentation and the involvement of a particular anatomic site, mucormycosis can be divided into at least six clinical categories: (i) rhinocerebral, (ii) pulmonary, (iii) cutaneous, (iv) gastrointestinal, (v) disseminated, and (vi) miscellaneous.

The most common presentation in the head and neck region is maxillary and orbital cellulitis (Pandey A et al., 2011). The pathophysiology of mucormycosis in normal healthy individuals and immunocompromised patients is given in Fig 1.
2. Case Report

On 4th January, 2015, a 35 year old patient reported to the department of oral and maxillofacial surgery with a chief complaint of swelling and severe headache on the right side of the face along with difficulty of vision. On general examination the patient was febrile with history of diabetes. On extra oral examination it was noted that the patient...
Figure 3: Patient showing palatal ulceration on the right side of maxilla intra orally

Figure 4: Orbital exenteration
had marked facial asymmetry due to right maxillary swelling. The swelling also extended on to the right eye. On intra oral examination it was found that the palate was necrosed. A yellowish patch with tender swelling was seen on right palatal mucosa region extending from second premolar to second molar measuring about 6cm x 4cm approximately. The surrounding mucosa appeared inflammed.

Investigations advised were facial CT scan, blood glucose levels, glycosylated haemoglobin (Hb1Ac) level and incisional biopsy. Hard tissue specimen along with the adjacent soft tissue was excised under local anesthesia and sent for histopathological examination. Histopathological examination with hematoxylin and eosin stain readily identified non-septate mucormycotic hyphae. Grocott’s modified silver methenamine special staining technique further identified these non-septate branching hyphae of mucormycosis. The laboratory reports also revealed that the patient had fasting blood sugar level of 300mg/dl (Normal 60mg/dl-90mg/dl) and post-prandial blood sugar level of 450mg/dl (90mg/dl-140mg/dl). Thus a final diagnosis of Mucormycosis was concluded. The patient was admitted and medical management was immediately started. The patient was administered with intravenous liposomal Amphotericin-B 2gm/day and Lysosomes. Blood glucose level was controlled with insulin and appropriate platelet coverage was also given.
Pre-operative consent about surgical intervention under general anesthesia and possibility of data usage for publishing in scientific journals was taken. The surgical management included extensive debridement with right orbital exenteration along with partial maxillectomy. All necrotic tissue was resected back to viable-appearing bone and the wound was irrigated and closed. A temporalis myocutaneous flap was used for the coverage of the defect. Metal staples were given on scalp region while non resorbable vicryl sutures were given in naso-orbital region. Post operatively the patient was again administered with Amphoterecin B (I.V.) and his blood sugar levels were also monitored with insulin.

3. Discussion

Mucormycosis is a life-threatening fungal infection that occurs in immunocompromised patients. These infections are becoming increasingly common, yet survival remains very poor (Spellberg B et al., 2005). Mucormycosis is one of the most severe, fatal and sudden fungal infections in humans with a high mortality rate (Mohanty N et al., 2012). Management of patients suffering from mucormycosis should be multidisciplinary and initiated without delay (Mimouni O et al., 2010). Mucormycosis is a disease which is more commonly seen in immunocompromised individuals who may be affected as a result of trauma or surgery (Mohanty N et al., 2012).

Spellberg and coworkers reviewed cases of cutaneous form of mucormycosis. In diabetic and immunocompromised patients, cutaneous lesions may also arise at insulin injection or catheter insertion sites. Gredilla et al concluded that the predisposing factors for Mucor infection in HIV disease include low CD4 count, neutropenia, and active intravenous drug use. Mucor infection may present as a devastating fungal infection in HIV immune deficient patient. The association with certain risk factors, such as metabolic acidosis, may help the clinician to suspect and make an early diagnosis and correct treatment. Atypical symptoms such as facial pain, sinus pain, or unexpected odontalgia of otherwise healthy teeth should alert clinicians (Papadogeorgakis N et al., 2010). The initial symptoms of sinus mucormycosis are consistent with those of sinusitis and facial and/or periorbital cellulitis (Nicolaou-Galitis O et al., 2015).

Conventional approaches for treatment include putting the patient on antifungal therapy (Reed C et al., 2008). There are 3 possible mechanisms by which addition of an echinocandin may improve the efficacy of polylene therapy for mucormycosis: (1) disruption of β glucan cross-linking of the cell wall (Bowman JC et al., 2002; Chiou CC et al., 2001) leading to enhanced polylene delivery to the cell membrane; (2) altered virulence of the fungus, either by stunting filamentation (Kurtz MB et al., 1994) or altering cell wall content; (Stevens DA et al., 2006) and (3) enhanced host response to the fungus (Denney KM et al., 2007; Kinoshita K et al., 2006; Taylor PR et al., 2007). Investigation into the mechanism of action of echinocandin combination therapy is ongoing.

Other conservative treatment modalities may include adjunctive treatment with deferasirox or deferiprone and adjunctive treatment with hyperbaric oxygen (Skiada et al., 2013). Surgical procedures by Vironneau were staged and graded I, II and III, when the patients underwent isolated diagnostic biopsy (grade I), debridement of necrotic tissue in the nasal cavity and in the sinus (grade II), or extended surgical resection (grade III) by resection of infected tissues, drilling of underlying bone, exenteration, if necessary, and/or drilling of the skull base (Vironneau P et al., 2014).

Conclusion

This study enlightens us that increased awareness of dentists and maxillofacial surgeons may initiate the early diagnosis of a sinus mucormycosis in high-risk patients who present with dental pain and palatal ulcer. Early diagnosis and prompt treatment is critical for a good prognosis. This is important in view of the increasing numbers of cases of mucormycosis being reported worldwide (Nicolatou-Galitis O et al., 2015).

Since general dental practitioners and maxillofacial surgeons have a great chance to be presented with a patient with symptoms of mucormycosis of head and neck region, it is imperative that they are able to recognize and diagnose the condition and provide the earliest line of treatment.

References


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