



TRAUMA INDUCED MUCORMYCOSIS OF RIGHT MAXILLA: CASE REPORT

Surgery

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ABSTRACT

Background: Mucormycosis is a fungal infection commonly affecting structures in the head and neck, such as air sinuses, orbits and the brain. Common predisposing factors include diabetes and immunosuppression, although it can rarely occur in people who are otherwise healthy. Mucorales possess virulence factors that enable the organism to cause disease. One such trait is the ability to acquire iron from host. The rich vascularity of maxillofacial areas usually prevents fungal infection although more virulent fungi such as those responsible for mucormycosis can often overcome this difficulty. Rapid diagnosis and initiation of therapy is critical due to acute fulminant nature of infection. In most cases, the infection is relentlessly progressive and results in death unless treatment with a combination of surgical debridement and antifungal therapy is initiated promptly. One such case of mucormycosis associated with diabetes mellitus reported to our department with an old maxillary fracture on right side.

KEYWORDS

Mucormycosis, Diabetes, Amphotericin

INTRODUCTION

Mucormycosis is the name ascribed to infections caused by usually nonseptate fungi belonging to the class Zygomycetes (Phycomycetes) [1]. The patient generally has uncontrolled diabetes mellitus with ketoacidosis, may have leukemia, or may be on immunosuppressive therapy [2,3]. Usually mucormycosis presents as an acute infection and manifests in rhinocerebral, pulmonary, gastrointestinal, cutaneous, or disseminated forms,[4] rarely affecting otherwise healthy people[5]. The infection begins in the upper turbinate or paranasal sinuses, [6,7] or less commonly in the palate or pharynx. The most common presentation in the head and neck region is maxillary and orbital cellulitis in a person with inadequately controlled diabetes mellitus [7,8]. Since mucormycosis occurs infrequently, it may pose a diagnostic and therapeutic dilemma for those who are not familiar with its clinical presentations. The purpose of this paper is to draw attention to the clinical presentation and pathogenesis of mucormycosis and to emphasize the need for high index of suspicion in its diagnosis and management.

CASE REPORT

A 40-year-old female patient reported to our department with a complaint of pain over the right side of upper jaw and paresthesia over lateral part of nose and lip on right side since 6 months. On taking proper history patient revealed hit from door 6 months back and had fracture of right maxilla with associated facial nerve paralysis on the same side, for which she didn't receive any treatment. Patient is known diabetic since 5 years, and is on Tab Glycomet 500mg once daily. On extra oral examination, facial asymmetry is noted due to signs of Bell's palsy on right side, loss of vision in the right eye, tenderness on palpation over right cheek region is noted. Mouth opening is around 35mm. On intraoral examination, mobility of right maxilla is noted with no signs of bleeding or discharge. OPG and CT scan of facial bones revealed old fracture of anterior and lateral wall of right maxilla, soft tissue densities in right sphenoid and maxillary sinus with osteoporotic changes [Figure 1].

Patient was planned for right maxillary sinus curettage under general anesthesia. Under standard towel preparation, a vestibular incision was given from lateral incisor to second premolar on the right side [Figure 2]. On exposing the site, whitish material was noticed in the maxillary sinus region. This was curetted out [Figure 3] and thorough debridement was done and cavity was packed using 2 ceptigras and tissue was sent for microbiological and histo- pathological

examination. Postoperatively, antibiotics were given and cavity was thoroughly irrigated and pack was changed every alternate day till the cavity has become very small. Histology of the received specimen showed stratified squamous epithelium which was ulcerated with intraepithelial split formation; underlying connective tissue showed numerous minor salivary glands, ducts and muscle tissue. Large amounts of necrotic tissue were evident with cellular degeneration and debris. Fungal hyphae were seen with neutrophil infiltration and generalized chronic inflammatory cell infiltrate within connective tissue. Hyphae were aseptate, branched and resembled mucormycosis [Figure 4]. Microbiology report revealed fungal elements with aseptate hyphae on KOH preparation suggesting mucormycosis. Once the diagnosis was confirmed as mucormycosis, patient was kept on Amphotericin B therapy 50mg IV once daily diluted in 500ml of 5% dextrose over 4 hrs, others antibiotics and Tab Glycomet 500mg BD by taking care of patient's serum creatinine and glucose levels for 10 days. Later, hyperbaric oxygen therapy was given for 1 week (60 dives every day for total of 360 dives). Patient developed sensation over the lateral part of the nose after 3 months of HBO therapy. Patient had a follow up for 6 months after surgery [Figure 5].



Fig 1: OPG



Fig 2: Cavity after curettage (Intraoperative pic)



Fig 3: Cured tissue

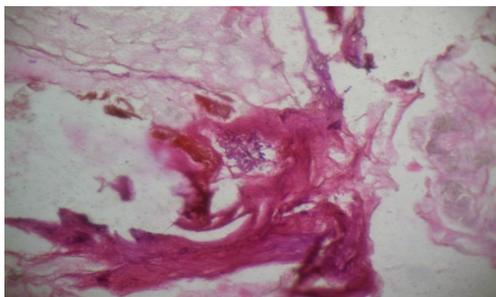


Fig 4: 10x10 H/E, Intraepithelial split and aseptate, branching hyphae



Fig 5: 6 month postoperative pic

DISCUSSION

Mucormycosis is a rare fulminating opportunistic fungal infection caused by a fungus of the order Mucorales [6,7]. These fungi are ubiquitous, found throughout the world on fruit and bread, in air and in soil, where they exist as saprophytes [3,5]. Depending on the immunological status of the host, the disease may manifest in six different ways depending on the affected site: rhinocerebral, pulmonary, cutaneous, gastrointestinal, central nervous system or miscellaneous. For example, patients with diabetes mellitus usually have the rhinocerebral and pulmonary forms of disease. Patients who are malnourished usually have the gastrointestinal forms of disease [9]. High fatality with diabetes is due to the angioinvasive character of the Mucor, thereby causing thrombosis of blood vessels and tissue necrosis [10].

In the diabetic ketoacidotic patient, there is a high incidence of mucormycosis caused by *Rhizopus oryzae*, because they produce the enzyme ketoreductase, which allows them to utilize the ketone bodies [11]. It is also likely that the hyperglycemia stimulates fungal growth, and the diabetic reduction in chemotaxis and phagocytic efficiency permit these otherwise innocuous organisms to proliferate [5]. Mucormycosis of head and neck region results from inhalation of airborne spores. In a study done by Ferry et al.[8] and by Yohai et al.[12] on mucormycosis cases, they reported sinus involvement in 69% and 79% respectively.

Differential diagnosis include Aspergillosis, T-cell lymphoma, Wegner's granulomatosis, nasopharyngeal carcinoma and tertiary syphilis and fungal sinusitis [5].

Suspicion of mucormycosis requires a CT scan of the maxilla, orbits and brain. Routine blood studies will show a leukocytosis. If the patient is diabetic, a full workup of serum glucose, electrolytes, blood chemistries and blood gases is required. Serological assays for Mucor antigens have been developed. Confirmation of the diagnosis is best obtained with a tissue specimen from the junction of necrotic and nonnecrotic tissue. Staining with methenamine silver or a PAS stain, in addition to H and E, will often show the organism in vessel walls or nerve bundles [13]. Mucormycosis is appropriately diagnosed histologically when broad, irregularly shaped, nonseptate hyphae with right angle branching are seen invading the tissue with H and E; but are better visualized with PAS or silver stains [5,6].

Medical management alone is not effective because of poor drug delivery to the infection due to extensive vascular thrombosis [14]. Mucormycosis is a severe tissue-loss and life-threatening disease. Aggressive debridement and control of the underlying disease is required. Patients should undergo a resection-type debridement as soon as they are physiologically stable. The subsequent wound is best left open for care and irrigation but may be obturated with a removable prosthesis-obturator to support speech and feeding [5]. Amphotericin B therapy should also be initiated as soon as possible. It is usually given in Dextrose 5% in water intravenously at a dose of 1.0-1.5 mg/kg daily. Use of HBO therapy has better prognosis, probably because it reverses the hypoxia in local tissues and enhances neutrophil and macrophage killing ability [5]. Magnetic resonance imaging of the head is useful in determining the extent of disease involvement, so that surgical margin can be planned [15]. The prognosis is directly related to the severity of the underlying disease, the extent of the disease when treatment begins and the aggressiveness of the treatment. Death is a common outcome of the disease (30-50%).

CONCLUSION

Mucormycosis is an uncommon frequently fatal fungal infection, which rarely arises in otherwise healthy people. An underlying disease, frequently diabetes mellitus, is almost always present. Tissue invasion by the hyphae of mucormycosis must be seen microscopically to establish the diagnosis, but culture is required to identify the fungal species involved. Hence the general approach is to treat early, aggressively and with all modalities available.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

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