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Management of Rhino-Orbital Mucarmycosis- A Case Report

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Abstract: *Considering the present scenario of excessive use of wide spectrum antibiotics and greater longevity of the immunocompromised patients, the incidence of fungal infection is on the rise. The commonest of all fungal infection is the Mucormycosis with as high as 75-80% mortality rate if left untreated. Mucormycosis frequently infects the paranasal sinuses, brain, or lungs. Apart from infection of the oral cavity or brain , this fungus can also infect other areas of the body such as the gastrointestinal tract, skin, and other organ system. The maxilla may also be affected by mucormycosis in rare cases. The disease can be potentially life-threatening in diabetic or severely immunocompromised individuals. The highly vascularised maxillofacial areas usually prevent fungal infections, although more virulent fungi, such as those responsible for mucormycosis, can often overcome this difficulty. This paper presents a case of mucormycosis involving the nasal cavity and the orbital floor to much greater extent and its successful management.*

I. Introduction

At present, the incidence of fungal infections is increasing thanks to chemotherapies, and survival has increased among the immunocompromised patients. These patients receive wide spectrum antibiotics in high doses and for longer period, which has caused increased incidence of fungal infection. Mucormycosis is the second most frequent fungal infection caused by *Mucor* following *Aspergillus*. The most important feature of this infection is that it has a high mortality rate (75-80%) if untreated, even after treatment the mortality rate is 15-34%. The infection begins in the nose and Paranasal sinuses due to inhalation of fungal spores. 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 in Rhinocerebral, pulmonary, gastrointestinal, cutaneous or disseminated form[1]. In the case presented here the infection followed a chronic and indolent form which eventually caused maxillary and orbital plate necrosis.

II. Case Report

A 57 year old male patient reported to the department of oral and maxillofacial surgery with a complaint of nasal regurgitation and change of voice since a month. After taking a thorough history, it was apparent that the patient had a history of sinusitis and nasal stuffiness since 6 years for which he took treatment by homeopathy and ayurvedic doctors. His medical history revealed that he is a known diabetic since 8 years, but undertook treatment for the disease only for 2 years and never did any follow up for it with any physician.
There was no other history of substance abuse. He had undergone extraction of multiple maxillary anterior teeth 6 months ago due to poor periodontal health. Due to financial constraints the patient did not take medications and avoided laboratory investigations.

On general examination vital signs were within normal limits. Extraoral examination revealed generalized flattening on the right maxillary region and the right infraorbital margin and a small cutaneous draining sinus located in the lateral canthus of the right eye (Figure 1). On palpation, no tenderness was present over the right maxillary region and the infraorbital rim was not palpable. The right eye displayed enopthalmos and tethering of the lower eyelid causing increased scleral show (Figure 2). There were no other eye signs, no ecchymosis, swelling, conjunctivitis nor were there any signs of opthalmoplegia or diplopia. There was parasthesia over the distribution of the infra orbital nerve.

Intraoral examination revealed a large perforation 3×4 cms in the anterior midline communicating with the nasal cavity and the maxillary antrum (Figure 3). Enlarged turbinates and a dusky red thickened mucosal lining were visible.

A Paranasal sinus view and Orthopantomograph (Figure 4 A and B) showed non specific haziness of right maxillary sinus with erosion of lateral sinus wall. A CT scan (Figure 5) confirmed the findings of destruction of infraorbital rim. Biochemical investigation revealed an elevated blood sugar levels, postprandial blood sugar level was 386 mg/dl (normal 90-140 mg/dl) and total leukocyte count of 13,600/cumm with neutrophils 78%. A provisional diagnosis of Mucormycosis or Aspergillosis was made. Aspergillosis primarily involves the lungs and airways with symptoms of pleuritic pain, cough, hemoptysis, dyspnea and hypoxia. It is also more common in AIDS patients. Our patient did not have any of the above symptoms and was seronegative for AIDS and HIV. A soft tissue biopsy under local anaesthesia was taken and sent for histological examination. Histopathological examination with Haematoxylin and Eosin stain readily identified non-septate mucormycotic hyphae (Figure 6). Grocott's modified silver methenamine special staining technique further identified these non-septate branching hyphae of mucormycosis.

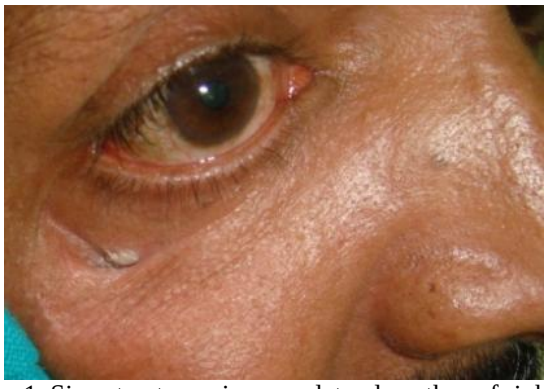

Figure 1. Sinus tract opening near lateral canthus of right eye

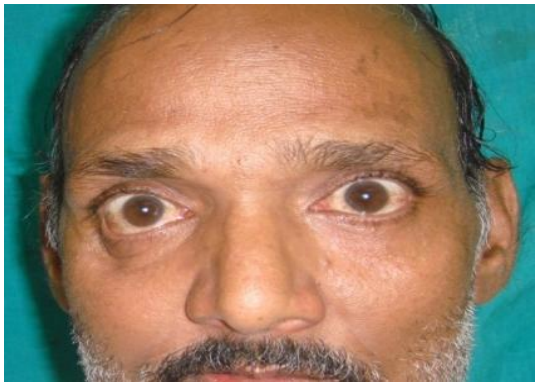

Figure 2. Extra oral photograph Showing right enopthalmos

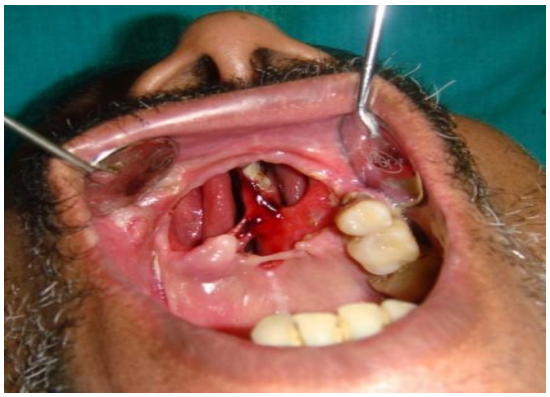

Figure 3. Intraoral photograph showing palatal perforation

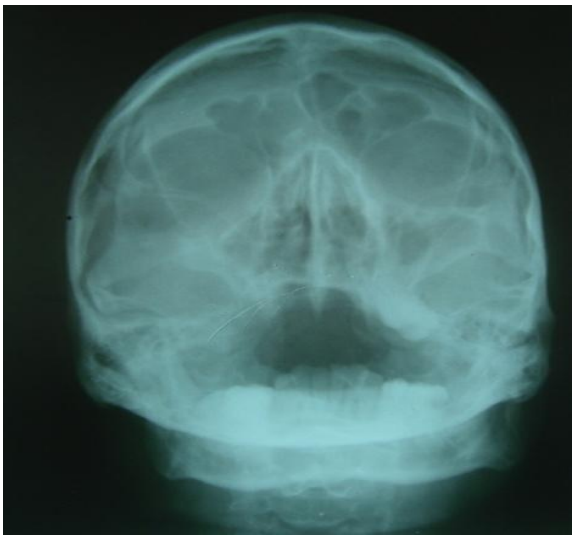

Figure 4 A: Water's view showing resorption of right inferior orbital rim

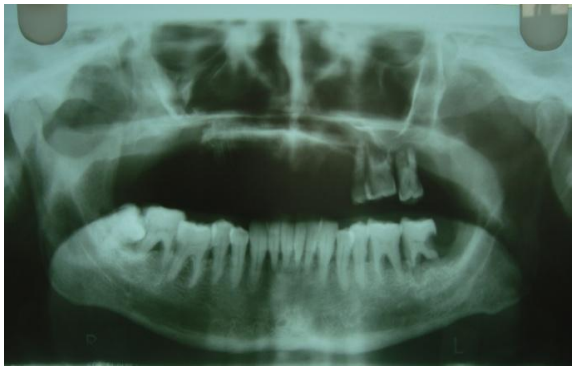

Figure 4 B. OPG showing Haziness of right maxilla antrum

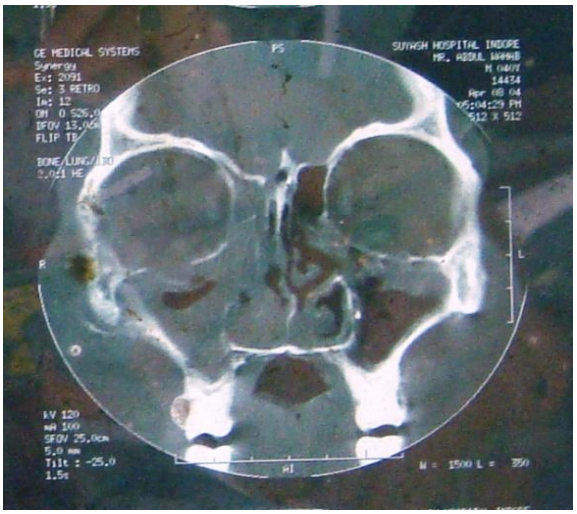

Figure 5: CT scan showing destruction of right infraorbital rim

The patient was hospitalized and the blood sugar levels were controlled with insulin and the patient was administered Amphotericin B 0.8 mg/kg/day I.V for 2 weeks. It was slowly infused over 4-6 hrs and blood urea and creatinine levels were monitored as the drug can cause renal toxicity. The surrounding inflamed tissue, Maxillary sinus lining and necrotic bone was completely excised and irrigated daily with sterile normal saline and packed with iodoform gauze. The remaining periodontally compromised teeth 25, 26, 27 were extracted. The area started healing gradually and subsequently after 3 months an obturator was fabricated.

III. Discussion

Successful treatment of Mucormycosis is based on three principles: First, any accompanying diseases should be kept under control. Second, necrotic tissues should be aggressively debrided or infected tissues should be resected. Last, medical treatment with antimycotic agents should be carried out. Surgical therapy has a great influence on the treatment outcome in cases of mucormycosis. The mortality rate was brought down to 11% in patients with surgical intervention compared to 60% in patients without surgical therapy[2]. Liposomal Amphotericin B can be given in the doses of 3-5 mg/kg/day as it is not as toxic as conventional Amphotericin B.

Organisms of these saprophytic fungi are ubiquitous occurring in soil, bread moulds, decaying fruits and vegetables and in dung. Rhinocerebral mucormycosis is opportunistic; infecting humans whose systemic health is compromised. The tiny spores become airborne and land on the oral and nasal mucosa of humans. In the vast majority of immunologically competent hosts, these spores will be contained by a phagocytic response. If this fails, germination will ensue and hyphae will develop. Because polymorpho nuclear leukocytes are less effective in removing hyphae, the infection then becomes established. It progresses as the hyphae begin to invade arteries, where they propagate within the vessel walls and lumens causing thrombosis, ischemia and infarction with dry gangrene of the affected tissues. Haematogenous spread to other organs can occur (Lung, brain etc) as well as overt sepsis[3]. Rhinocerebral mucormycosis is the most common form of infection and predominantly occurs in patients with poorly controlled diabetes Mellitus. The high iron, glucose-rich, acidic environment favours fungal growth. Other at risk populations include immunosuppressed patients with organ transplants[4], haematological malignancies[5,6], severe burns, patients being treated with chronic corticosteroids and those with end stage renal disease[7]. AIDS patients rarely have been reported with this disease. Invasive mucormycosis in patients without a predisposing condition is also uncommon. Infection is acquired through either the respiratory or gastro intestinal tract and may occur anywhere along their routes. No person-to-person spread has been reported[3].

Rhinocerebral mucormycosis produces a characteristic clinical picture consisting of low grade fever, sinusitis, unilateral facial swelling, black nasal or palatal eschar (seen only in 19-40% of patients) [8, 9], decreased vision or eventually opthalmoplegia. Once established in the Paranasal sinuses , the infection can spread to the orbit via the naso lacrimal duct and the thinness of the medial lamina papyracea and the perforation of the medial wall by arteries and veins, spread to the brain may occur via the orbital apex, orbital vessels or via the cribriform plate. As the disease progresses to the skull the patient may become confused, obtunded and comatose. Superior orbital fissure syndrome (unilateral sensory deficit of the first and second divisions of the trigeminal nerves and opthalmoplegia) chemosis, and proptosis occurs due to vascular compromise. Fungal invasion of the retinal arteries leads to blindness[10]. Our patient had no eye symptoms except enopthalmos due to partial loss of lateral orbital plate.

The nasal turbinates on the affected side are dusky red. Palatal involvement is due to invasion by the fungi into the sphenopalatine, greater palatine and nasopalatine arteries. Tooth ache and extraction of maxillary anterior teeth are associated with onset of acute symptoms. Plain orbit or sinus radiography is non-specific. CT is a sensitive indicator of the extent of orbital involvement. Mucormycosis is a medical emergency. Proper management of the patient by early diagnosis and initiations of vigorous surgical and medical therapy is of great benefit in reducing the mortality and morbidity. Amphotericin B is the drug of choice, but being a highly toxic drug, careful monitoring for renal damage and anaphylaxis has to be done. Over dosage can result in Cardio respiratory arrest. Generally, a total dose of 3-4 gm should be given over 6-12 weeks, which helps to minimize toxicity and side effects. Liposomal Amphotericin B is a less toxic formulation available. (The lethal dose is 10-15 times higher than conventional Amphotericin B). Dose- 5 mg/kg/day prepared as a 1 mg/ml infusion and delivered at a rate of 25 mg/kg/hour[3]. Constant monitoring of the patient's serum creatinine and blood urea is done.Extensive surgical debridement to remove necrotic tissue is essential. Orbital exenteration has to be considered in the presence of actively infected orbit with a blind immobile eye. In our case the orbit did not show active involvement and so was spared. Irrigation with Amphotericin B solution (5 mg/100 ml sterile water) has been advocated to improve delivery to poorly perfused infected tissue. Hyperbaric oxygen has been used as an adjunct to surgical debridement, Amphotericin B therapy. It helps by having a fungistatic effect and by neovascularization. The drawback being that this facility is not available in a lot of centres[11].

The decision as to when to stop treatment is decided on clinical condition of patient and fungal culture obtained from biopsy. Our patient underwent treatment for a month and is well after a follow up of 4 months. But patients need to be followed up for longer periods especially to ensure that the blood sugar levels are maintained adequately. The purpose of this paper is to report a case of rhinoiorbital mucormycosis in an uncontrolled diabetic patient who underwent successful treatment in an otherwise mortal disease (mortality rate as high as 75-80%).

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