

Mucormycosis of Maxilla: A Multifarious Disease

A Report of Two Cases.

S. Jayachandran, Vidya Jayaram

Department of Oral Medicine and Radiology,
TamilNadu Government Dental College and Hospital, Chennai, India

Abstract

Mucormycosis is an invasive fungal infection with fulminant and fatal course and with high mortality rate. Immunocompromised patients like uncontrolled diabetes, renal transplant patient, haematological malignancies etc are more commonly affected by this infection. Rhino cerebral mucormycosis is the most common type.

Keywords : Diabetes mellitus, Immunocompromised, Amphotericin B

Introduction

Mucormycosis is a rare fulminant angioinvasive fungal infection caused by fungi of order mucorales. They enter our body through inhalation causing thrombosis and ischemia in blood vessels leading to necrosis of hard and soft tissues and invade the adjacent tissues rapidly [1]. It is an opportunistic infection common in immunocompromised patients but also several cases have been reported in immunocompetent over the last 30 years [2]. The incidence of mucormycosis is 1.7 cases per 1 million individuals per year [3] Chakrabarti et al in his study reported the overall prevalence of mucormycosis in India was 0.14 cases per 1000 individuals [4] Here we present two cases of mucormycosis reported to our tertiary hospital with varied clinical presentation.

Case Report

Case 1

A 32 year old female patient reported to the Department of Oral medicine and Radiology with complaints of pain and swelling in the right cheek region for one year and nasal congestion since three months. History revealed extraction of right upper tooth one year ago due to pain, mobility and persistent swelling and subsequently developed nasal congestion with pus discharge from extracted socket. Patient is a known diabetic for 1 ½ years

and under insulin with Random blood sugar of 302mg/dl and HbA1c 14.4 revealing poor diabetic control. She was diagnosed with mucormycosis 1 ½ years back but did not undergo treatment due to pregnancy. On examination extraorally a diffuse swelling present on right zygomatic region and intraorally yellowish necrotic bone in 16,17 region with palatal perforation. [figure 1a and b]. Axial and coronal Computed tomography revealed a hypodense mass with erosions involving the sphenoidal, ethmoidal sinuses and floor of the orbit. Based on clinical and radiological findings, provisional diagnosis of chronic suppurative osteomyelitis and deep fungal infection (mucormycosis) were made. Incisional biopsy of necrotic alveolar bone revealed a histopathology of necrosis with aseptate right angled and slender acute angle branching hyphae. Based upon histopathology final diagnosis of invasive mucormycosis was made. Patient was planned for surgical debridement of necrotic tissue along with iv Amphotericin B in 5% dextrose with pre-load and after-load. Patient was in oral hypoglycemic drugs and insulin therapy and the Random blood glucose level at the time of surgery was 58mg/dl, renal functions were monitored with serum urea and creatinin. Patient improved after surgery and is under regular follow-up. (figure 3).

Address for correspondence

Dr. Vidya Jayaram, Assistant Professor
Department of Oral Medicine and Radiology, TamilNadu Government
Dental College and Hospital, Chennai- 3, TamilNadu, India
Email: dr.vidyajayaram@gmail.com

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Figure 1: (a) Extraoral diffuse swelling on right side
b) Intraoral necrotic alveolar bone in relation to 16 17

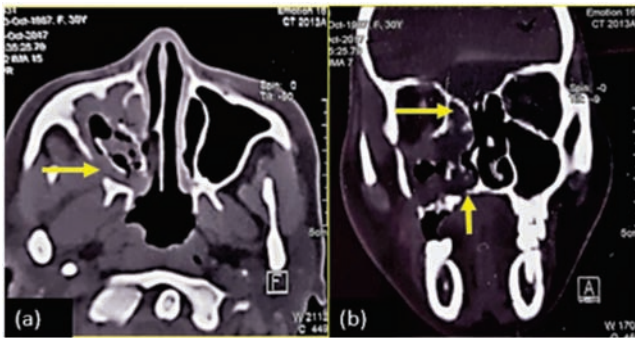


Figure 2: a) CT axial section showing hypodense mass with erosions in right maxillary sinus
b) CT coronal section showing erosions involving the ethmoidal sinus and palate



Figure 3: Post - Operative image

Case 2

A 55 year old male patient was referred to the department from ENT department for obturator. History revealed that patient had pain in left upper back tooth region before 3 months proceeded by swelling in left side of the face involving the orbit with the partial loss of vision, watery discharge and pain in the left eye and loss of consciousness. Patient was hospitalised and newly diagnosed as diabetic with blood sugar values of 480mg/dl and started with oral hypoglycemic drugs and insulin. On examination extraorally a diffuse swelling on left zygoma region extending to infraorbital region with ptosis of left eye, and intraorally necrotic alveolus on left side with palatal perforation were found. (Figure 4a and b). Ophthalmologist opinion was obtained. The visual acuity of left eye is decreased to counting finger at one meter and paralysis of III,IV,VI cranial nerves. Computed tomography in axial and coronal sections revealed complete erosions of left maxillary sinus involving the ethmoidal sinus, floor of the orbit and palate Magnetic Resonance imaging revealed involvement of the left extraocular muscles, left internal carotid artery and left cavernous sinus (Figure 5a,b and c). Endoscopic incisional biopsy of both hard and soft tissue was done from nasal cavity and Histopathology report revealed septate

branching fungal hyphae with necrotic bony spicules and inflammatory cells consistent with invasive fungal infection. The random blood sugar level at the time of surgery was 98mg/dl .Surgical debridement of necrotic bone, (figure 6) along with IV amphotericin B and insulin was given followed by 2.5mgs of amphotericin B for 4 weeks with regular monitoring of renal functions. Patient responded well and is under regular follow up.



Figure 4 : a)Extra oral diffuse swelling on left side with ptosis of left eye
b) Intraoral necrotic alveolar bone in relation to 24 25 26 27 region

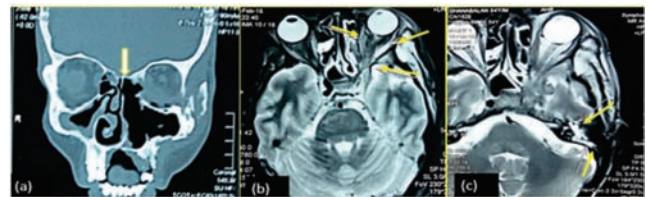


Figure 5 :a) coronal CT showing hypointense mass causing erosion of right maxillary sinus, ethmoidal sinus and palate
b) MRI T1 weighted image showing thickening of extraocular muscles
c) MRI T1 weighted image showing obstruction of internal carotid artery



Figure 6: Post - Operative Image



Figure 7: a) 3 months follow up with intraoral defect
b) With interim obturator



Figure 8 : a) 3 months follow up with intra oral defect b) With interim obturator.

Comparison between two cases

	Case 1	Case 2
Age	32yrs	55years
Gender	Female	Male
Type	Rhinomaxillary	Rhino-orbito-cerebral
Risk factor	Type II diabetes	Type II diabetes
Progression	1 year	4 months
Treatment	Surgical debridement , amphotericin B, insulin	Surgical debridement, amphotericin B, insulin

Both the cases are on regular 3 monthly follow up, and the symptoms have been reduced and both the cases are using interim obturator and planned for permanent obturator and the pictures for case 1 (figure 7a and b), case 2 (figure 8a and b).

Discussion

Mucormycosis in humans was first reported in 1885^[5] and described by Platauf in his paper mycosis mucorina. In India Balasubrahmanyam et al gave the first reference to mucormycosis in 1963 [6]. The last two decades has witnessed an increase in the occurrence of invasive fungal infections because of increase in the size of the population at risk [5] and increased incidence of mucormycosis has been reported after natural disasters [7,8]. The disease can manifest in five major clinical forms based on the involvement of anatomical site as rhinocerebral, pulmonary, gastrointestinal, cutaneous, and disseminated [1] of which Rhino-orbito-cerebral [ROC] is the commonest [5,9] and was first described by Hazarika et al.[6]

The major risk factors include uncontrolled diabetes, diabetic ketoacidosis, prolonged steroid therapy, persistent neutropenia, desferoxamine therapy, haematological malignancies, illicit use of intravenous drugs and autoimmune disorders of which poorly controlled type II diabetes with ketoacidosis is the commonest risk factor, contributing 44–88% of the cases reported from north to south India [4]. In our case reports both are diabetic, however progression in the first case was delayed due to immediate control of blood sugar levels by insulin and in contrary the second case was undiagnosed with diabetes mellitus and hence the rapid

progression of mucormycosis implying control of the predisposing risk factor as an important aspect in treatment planning.

Most commonly, fungi enter the respiratory tract, but in primary cutaneous mucormycosis, fungi enter through abraded skin [10]. ROC commonly initiate symptoms from the paranasal sinuses causing chronic sinusitis, nasal congestion, nasal discharge, soft tissue swelling and orbital involvement causes periorbital oedema and proptosis. Intracranial spread occurs through the cribriform plate of ethmoid. Diagnosis of mucormycosis can be made from clinical presentation and demonstration of aseptate hyphae with wide angle branching (45° to 90°). Imaging modalities like Plain radiographs of sinuses and orbits, CT, MRI help to delineate the extent [11].

The management of mucormycosis include aggressive surgical debridement combined with medical treatment and control of predisposing factors [5]. Amphotericin B is the drug of the choice with dosage of 0.5-1.0 mg/kg body weight once daily for minimum of 4 weeks [1]. Recently lipid-associated amphotericin B is used, due to nephrotoxicity of regular amphotericin with doses as high as 4–8 mg/kg/day [12]. Daily irrigation of the infected site with amphotericin B and packs can give excellent results due to sustained release [13]. After surgical debridement we advised patient to maintain proper oral hygiene and diabetic control and regular follow up. A meta-analysis conducted by Yohaiet al reported that survival rates decline if there is a delay of initiation of treatment by longer than 6 days from the diagnosis [3] and mortality rates as high as has been reported. Hence timely diagnosis and attention to control of predisposing factors plays an important role in the management of this multifarious disease.

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