VOLUME-8, ISSUE-9, SEPTEMBER-2019 • PRINT ISSN No. 2277 - 8160 • DOI : 10.36106/gjra

Original Research Paper

Dental Science

# MUCORMYCOSIS OF THE MAXILLARY ANTRUM: A CASE REPORT

| | |
|---|---|
| **Dr. Anuradha Yadav*** | Department of Oral Medicine and Radiology CDER, All India Institute of Medical Sciences. New Delhi India. *Corresponding Author |
| **Dr. Upasana Sethi Ahuja** | I.T.S Centre for Dental Studies and Research, Ghaziabad India. |
| **Dr. Niraj Kumar** | Department of Neuro-anaesthesia, All India Institute of Medical Sciences. New Delhi India. |

**ABSTRACT** Mucormycosis, an invasive fungal infection is one of the life threatening infection for the immuno-compromised patients. Fungi of this family is ubiquitously present in the environment but is avirulent unless host-response is favorable for its growth and survival. In the present case report, An old male patient with uncontrolled diabetes developed an exophytic bony outgrowth on palate and left half of the maxilla with oro-antral communication over period of time, which after radiological and histopathological investigation turn out to be fungal infection involving maxillary antrum.

**KEYWORDS :**

## INTRODUCTION -:

Mucormycosis is a rare but potentially threatening fungal infection caused by the fungi of the sub-species zygomycetes. Fungi of the sub-species zygomycetes comprised of Mucorales and Entomopthorales. Mucorales are causative for mucormycosis, a life threatening fungal infection.

Mucormycosis was first described by Paultauf in 1885 [1]. Invasive fungal infections are most commonly caused by Aspergillus and Candida species followed by Mucormycosis. The four genera of the family Mucoraceae frequently involved are Rhizopus, Absidia, Mucor and Rhizomucor. Rhizopus arrhizus species are the most commonly identified cause of mucormycosis in humans [2].

Fungi of this family is ubiquitously present in environment but normally they are avirulent unless the host response is extremely low [3]. Risk factors which predispose to this disease are diabetes, immune-compromised individuals, long term steroid therapy, blood dyscrasias and organ transplant.

Based on the involvement of the particular site and its presentation it can be divided into – rhino-cerebral, pulmonary, cutaneous, gastro-intestinal and disseminated [4]. Rhinocerebral mucormycosis is a fatal disease if not managed in its early stage due to involvement of central nervous system.

## CASE REPORT -:

A 65 year old male patient reported to the department with the presenting complaint of dull aching pain in left posterior maxillary region since past one year. The nature of the pain was dull aching pain which aggravates on chewing food. Apart from this there was presence of nasal regurgitation on fluid intake. Patient's medical history revealed uncontrolled type II diabetes mellitus since last 10 years and patient found to be on unmonitored insulin intake without physician's approval. His vital signs were within normal limits.

Extra-orally there was diffuse puffiness over the middle one-third on left side of the face causing obliteration of the nasolabial fold. (fig.1).On opthalamic examination there was blurred vision. Intra-orally necrotic bone growth measuring 3X2 cm in size was evident with foul discharge, putrification (fig.2) and tenderness on left side of posterior maxilla and presence of oro-antral fistula posteriorly.

Because of the necrosed bony growth, overlying mucosa was perforated and due to the pressure effect of the exophytic growth bone was shifted towards the midline. Depending upon the clinical presentation differential diagnosis of invasive fungal infection and avascular necrosis of the maxillary alveolar bone was considered.

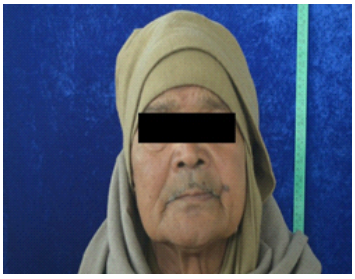

**Figure 1 Diffuse puffines on left side of face**

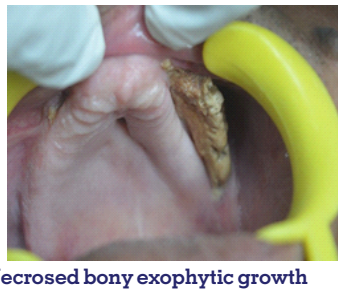

**Figure 2 Necrosed bony exophytic growth**

Radiological investigations revealed presence of opacification in the left maxillary sinus (fig.3). On Contrast Enhanced Computed Tomography (CECT), the postero-lateral and medial walls of the left maxillary sinus revealed areas of sclerosis along with bony erosion. Sclerosis and erosions were also evident in the hard palate and the inferior part of the nasal septum, resulting in passage linking the oral cavity and the nasal cavity. Left inferior, middle turbinate and posterior orbital wall also showed erosion. There is soft tissue thickening seen in the left maxillary sinus with extension into the middle meatus of the nasal cavity on left side. The soft tissue is also extending posteriorly into the pterygopalatine fossa. It was also noted that there was erosion of the medial pterygoid plate on the left side. It was observed that there was sclerosis of the walls of the sphenoid sinus and the greater wing of sphenoid on left side (fig 4a and 4b).

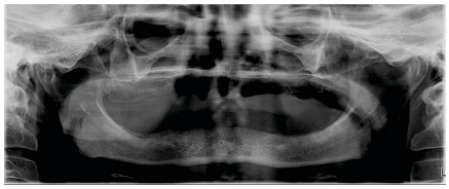

**Figure 3**
Orthopantamograph showing diffuse opacification on left side of maxillary edentulous region

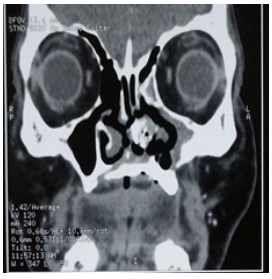

**Fig 4. a**

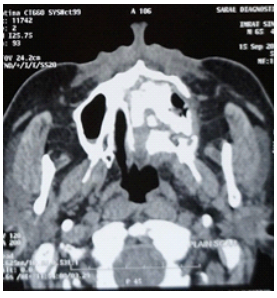

**Fig.4.b**

CECT examination Fig 4a) Coronal section showing opacification of left maxillary antrum along with involvement of nasal wall and floor of the palate. Fig.4 b) Axial section showing sclerosis of left palatal half with erosions of postero-lateral walls.

Laboratory investigations for blood culture showed no growth whereas specimen of nasal discharge when grown on Sabouraud's dextrose agar (SDA) without cycloheximide at 37 degrees celsius showed fungal growth (fig.5a). Histological examination showed fungal hyphae which were non-septate obtuse angles with sporangiophores containing spores, characteristic of Rhizopus species, suggestive of Mucormycosis (fig.5b).

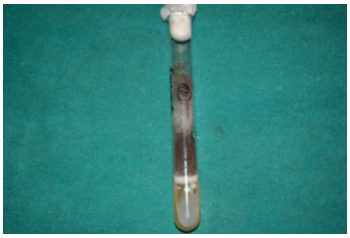

**Fig. 5a**

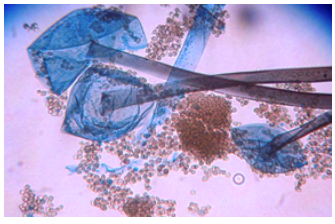

**Fig. 5b**

Fig.5a) Flocculent black growth of the fungi on Sabouraud's Dextrose medium. b) Non-septate fungal hyphae at obtuse angles with sporangiophores containing spores, characteristic of Rhizopus species

Alongwith restoring the patient's glucose level within normal limits, monotherapy with liposomal-Amphotericin B at a dose of 3mg/kg/day parentrally was started. After a week when glucose level was corrected, aggressive surgical intervention followed by prosthetic rehabilitation.

## DISCUSSION –

Mucormycosis is the third leading cause of invasive fungal infection after Aspergillus and Candida species [5]. The characterstic of mucormycosis is invasion of blood vessels resulting in thrombosis and necrosis. It was seen that mucormycosis occurs in patients with reduced defense mechanism and/or available free serum iron [6].

Among the different types of mucormycosis, rhinocerebral mucormycosis accounts for one-third to one-half of all the cases. About 70% of mucormycosis are found in diabetic ketoacidosis patients [7,8]. Diabetes ketoacidosis typically develops the rhinocerebral form of the disease, and rarely develop other form of the disease. Ketoacidosis creates acidic envoirnment which increases available iron in serum and this iron enhances the growth of the fungi. Apart from this, in diabetes host response by mononuclear and polymorp honuclear phagocytes is also reduced.

Initially rhinocerebral mucormycosis is associated with either sinusitis or cellulitis around the orbit followed by facial pain and blurry vision. Infection usually spreads from paranasal sinus towards orbit resulting into proptosis, loss of vision and opthalmoplegia. Involvement of contralateral eye results when there is thrombosis into cavernous sinus. Infection sometimes progresses from the sinuses and produce painful, necrotic ulcer of the hard palate in the oral cavity [9]. Auluck et al in 2007 studied that bone necrosis after tooth extraction in immunocompromised patient should be an alarming possibility for invasive mucormycoses infection [10]. As seen in our case report also bone necrosis in edentulous patient having diabetes ketoacidosis resulted due to invasive fungal infection caused by species Rhizopus arrhizus, which has the ability to produce enzyme ketoreductase, allowing to use patient's ketone bodies for their nutrition [11].

Radiological investigations such as CT scan and MRI can help in defining the extent of the lesion by showing periosteal, mucosal thickening and bony disruption. Confirmation of the clinical diagnosis can be done by histological examination of the specimen showing right-branching aseptate hyphae suggestive of Mucor species. Fungal culture of the specimen in Sabouraud's dextrose agar (SDA) further confirms the diagnosis.

Since rhinocerebral mucormycosis is a life threatening disease and it warrants immediate attention, management of the disease requires four critical points -: i) Early diagnosis ii) recognizing and reversal of the underlying immuno-compromised disease iii) Surgical debridement and iv) Appropriate antifungal therapy. Necessary early diagnosis should be done as the disease progression is very rapid and if possible the underlying debilitating condition should be controlled. Surgical debridement on urgent basis is necessary because of massive amount of tissue necrosis [12]. Classical drug of choice for antifungal therapy includes members of polyenes group. Various species of mucormycosis group have wide range of senstivity to Amphotericin B deoxycholate but due to nephrotoxicity, now lipid derivatives of Amphotericin B given at higher doses are being tried. Increasing the dose of lipid formulations also increases the cost enormously [13].

Recent retrospective reviews now have supported the choice of liposomal amphotericin B 67% more effective than amphotericin B deoxycholate at higher dose [14].

We therefore emphasize early diagnosis, histopathological confirmation and prompt treatment to reduce the risk of mortality and morbidity associated with this destructive disease.

## REFERENCES -:


1. S. Viterbo, M. Fasolis, P. Garzino-Demo et al. Management and outcomes of three cases of rhinocerebral mucormycosis. Oral Surgery, OralMedicine, Oral Pathology, Oral Radiology and Endodontology 2011,112:e69–e74.
2. Ribes JA, Vanover-Sams CL, Baker DJ. Zygomycetes in human diseases. Clin Microbiol Rev. 2000,13:236-301.
3. Prabhu RM, Patel R. Mucormycosis and entomophthoramycosis: a review of the clinical manifestations, diagnosis and treatment. Clin Microbiol Infect 2004,10:31–47
4. SPELLBERG B, EDWARDS J JR, IBRAHIM A. Novel perspectives on mucormycosis: pathophysiology, presentation, and management. Clin Microbiol Rev 2005,18(3): 556-569.
5. Eucker J, Sezer O, Graf B, Possinger K. Mucormycoses. Mycoses 2001;44:253-60.
6. Larsen, K., C. von Buchwald, B. Ellefsen et al. Unexpected expansive paranasal sinus mucormycosis. ORL J. Otorhinolaryngol. Relat. Spec. 65:57–60.
7. McNulty, J. S. Rhinocerebral mucormycosis: predisposing factors. Laryngoscope 1982, 92:1140–1143.
8. Pillsbury, H. C., and N. D. Fischer. Rhinocerebral mucormycosis. Arch. Otolaryngol. 1977,103:600–604
9. Petrikkos, G., A. Skiada, H. Sambatakou et al. Mucormycosis: ten-year experience at a tertiary-care center in Greece. Eur. J. Clin. Microbiol. Infect. Dis. 2003, 22:753–75.
10. Ajit Auluck. Maxillary necrosis by mucormycosis. A case report and literature review. Med Oral Patol Oral cir bucal 2007,12 E: 360-4
11. Skiada A, Pagano L, Groll A. Zygomycosis in Europe: Analysis of 230 cases accruded by the registry of European Confederation of Medical Mycology (ECMM) Working group on Zygomycosis between 2005 and 2007. Clin Microbial Infect 2011,17: 1859-67.
12. Ibrahim, A. S., V. Avanessian, B. Spellberg, and J. E. Edwards, Jr. Liposomal amphotericin B, and not amphotericin B deoxycholate, improves survival of diabetic mice infected with Rhizopus oryzae. Antimicrob. Agents Chemother. 2003, 47:3343–3344.
13. Ibrahim, A. S., B. Spellberg, V. Avanessian, Y. Fu, and J. E. Edwards. Rhizopus oryzae adheres to, is phagocytosed by, and damages endothelial cells in vitro. Infect. Immun. 2005, 73:778–783.
14. Spellberg, B., M. D. Witt, and C. K. Beck. Amphotericin B: is a lipid-formulation gold standard feasible? Clin. Infect. Dis. 2004, 38:304–307