

## CASE REPORT

# Mucormycosis of the Middle Ear: A Case Report

<sup>1</sup>Poonam K Saidha, <sup>2</sup>AM Balasubramanya, <sup>3</sup>Ntaashya H Sima, <sup>4</sup>Jithu Zacharia

## ABSTRACT

*Mucor* is a saprophytic organism and commonly invades the nose and paranasal sinuses of immunocompromised and diabetic patients. Middle ear involvement is extremely uncommon. Early diagnosis based on strong clinical suspicion with radical debridement in the early setting under amphotericin cover offers a suitable management option.

**Keywords:** Amphotericin, Middle ear, MRM, Mucormycosis

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## INTRODUCTION

*Mucor* is a saprophytic organism that commonly invades the nose and paranasal sinuses of immunocompromised patients. The initial presentation may be with pain and foul smelling discharge that is unresponsive to regular treatment. The invasive variety of mucormycosis is seen mostly in diabetic patients.<sup>1</sup> Middle ear/mastoid/temporal bone involvement is extremely rare.<sup>2</sup>

We report one such case of mucormycosis of the middle ear in a diabetic patient at our hospital.

## CASE DESCRIPTION

A 57-year-old male patient reported to ENT clinic with history of a fall in the office 20 days earlier following which he developed purulent discharge from left ear, deviation of angle of mouth and weakness of all limbs. He also complained of dull pain in the left postauricular region.

He was a known case of diabetes mellitus, hypertension and rheumatic heart disease (RHD) having undergone mitral valve replacement surgery in 2005.

On clinical examination, there was mucopurulent discharge in external auditory canal; perforation in tympanic membrane with flakes in attic (Fig. 1).

There was facial nerve paresis left and hemiparesis grade II/IV.

Audiogram showed moderate conductive loss in the left ear.

An initial clinical diagnosis of CVA with left hemiparesis, facial palsy left, ASOM left, and old RHD was made and treatment started accordingly.

Ear swab pus culture grew *Klebsiella*, and based on sensitivity, ceftazidime was started.

The discharge from ear reduced but otalgia persisted.

He developed ear bleed after 3 days that required pressure dressings and adjustments of anticoagulants dosage that were started earlier for CVA.

X-ray skull showed left mastoid sclerosis.

CT scan skull also reported sclerosis of left mastoid and an ill-defined soft tissue lesion the middle ear cavity and mastoid antrum with bony widening and thinning of tegmen tympani (Fig. 2).

Owing to enhanced clinical suspicion, a KOH preparation of the purulent discharge was examined but it did not establish the characteristic fungal hyphae; however, in the discharge culture for fungus, *Rhizopus* growth was seen.

In view of fungal growth and persistence of symptoms the management plan was revised to mastoid exploration.

Modified radical mastoidectomy was done. On exploration, the attic and antrum were seen filled with greenish soft tissue debris. There was discoloration of bone containing the tympanic segment of the facial nerve. The tympanic segment of facial nerve was further traced up to first genu and part of it was found to be exposed and covered with blackish debris (Figs 3 and 4).

Wound closed after conchomeatoplasty.

The debris along with excised portion of tympanic segment sent for histopathology and fungal culture.

Histopathology of the lesion showed masses of thin walled fungus with broad nonseptate hyphae, branching at right angles, evidence of angioinvasion and a neutrophilic inflammatory response; morphologic characteristics of mucormycosis (Fig. 5).

The organisms were visible with hematoxylin and eosin and periodic acid Schiff staining, but stained weakly with Gomori's methenamine silver.

Fungal culture grew *Mucor*.

Amphotericin B intravenously was started immediately at a dose of 1 mg/kg body weight/day and a

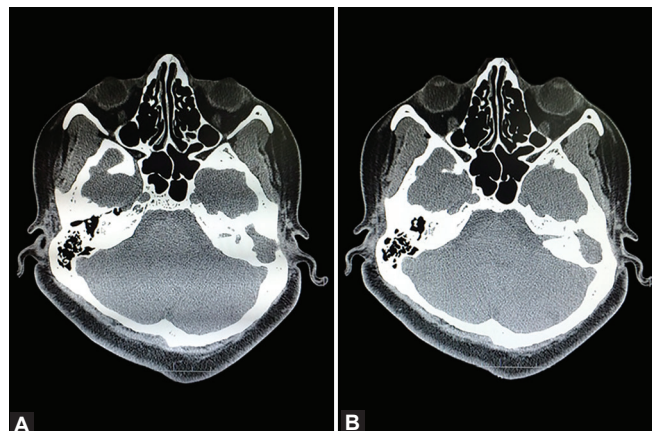
<sup>1</sup>Consultant, <sup>2</sup>Professor, <sup>3,4</sup>Resident

<sup>1-4</sup>Department of ENT, St Johns Medical College Hospital, Bengaluru, Karnataka, India

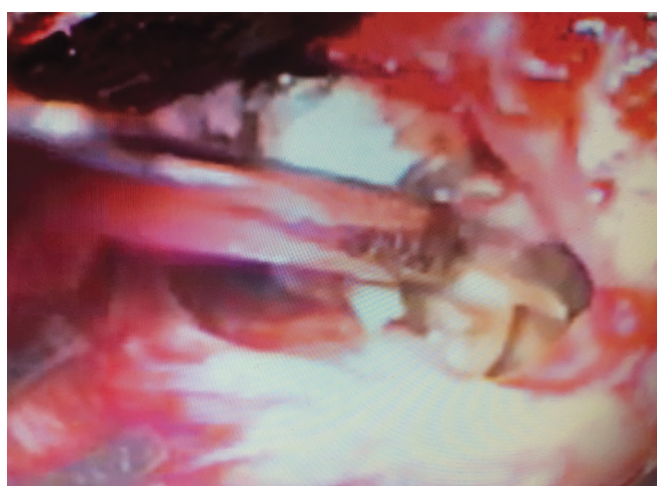
**Corresponding Author:** Poonam K Saidha, Consultant, Department of ENT, St Johns Medical College Hospital, Bengaluru, Karnataka, India, e-mail: nksaidha@hotmail.com



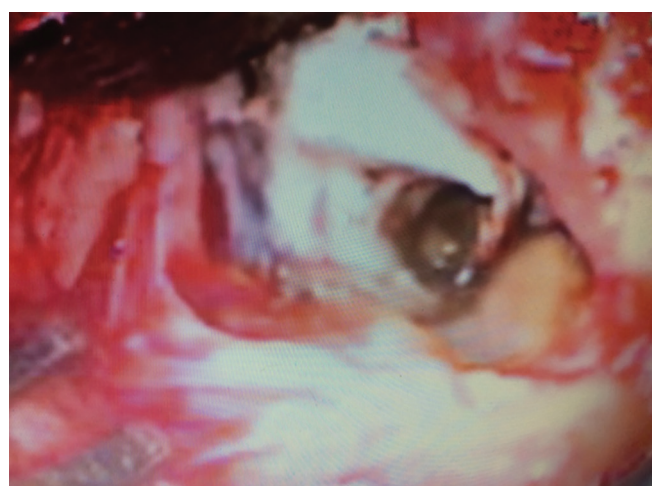
**Fig. 1:** Mucopurulent discharge in external auditory canal; perforation in TM with fakes in attic



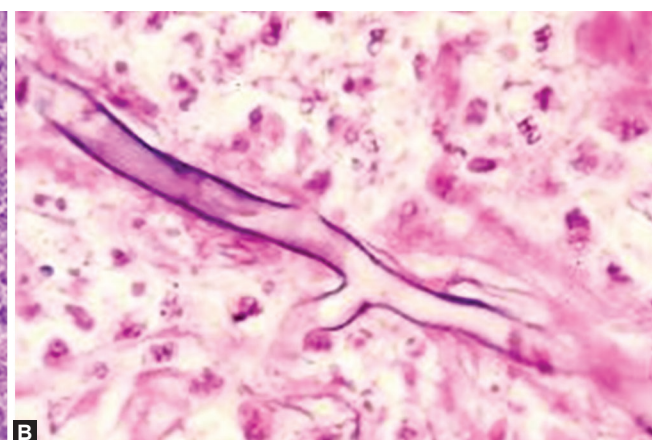
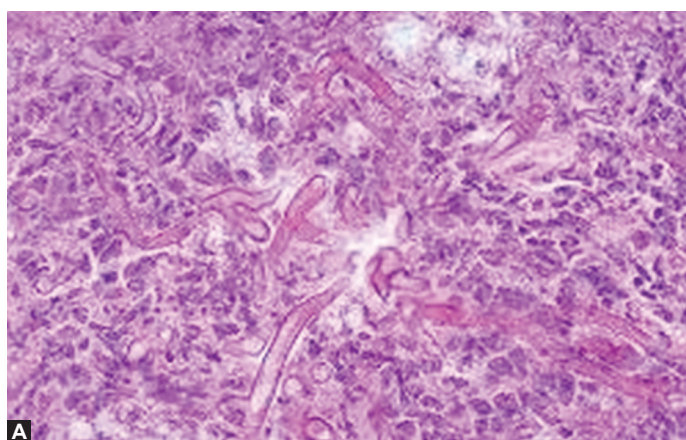
**Figs 2A and B:** CT scan skull: sclerosis of left mastoid; ill-defined soft tissue lesion involving the middle ear, mastoid antrum, with bony destruction



**Fig. 3:** Attic and antrum filled with greenish soft tissue debris



**Fig. 4:** Tympanic segment of the facial nerve which was covered with black debris



**Figs 5A and B:** Histopathology: Tangled masses of fungus, thin walled hyphae, broad, aseptate, and branching at right angles resembling zygomycetes (*Mucor*)

cumulative dose 2 g was administered. He also received gabapentin for the facial pain.

Last follow-up done showed that the mastoid cavity remained well epithelialized, with no evidence of recurrence of infection. The facial palsy persisted.

## DISCUSSION

Mucormycosis (zygomycosis, phycomycosis) is acute opportunistic infection caused by saprophytic fungus belonging to the class of Phycomycetes. Most common forms are *Rhizopus*, *Rhizomucor* and *Absida*.<sup>3</sup>



*Rhizopus* is the predominant pathogen accounting for 90% of the cases of rhinocerebral mucormycosis.

Incidence of mucormycosis with life-threatening mortality is increasing. Despite aggressive surgical and polyene antifungal therapy, overall mortality for MCM infection remains high, with figures ranging from 20 to 50% and it rises markedly in immunocompromised and diabetic patients, nearing 70–90% for cases of disseminated mucormycosis.<sup>4</sup> Diabetes mellitus is the most commonly reported independent risk factor; particularly in the oral and maxillofacial areas. DM is associated with impaired neutrophil function, microvascular insufficiency, and in the case of ketoacidosis, other metabolic abnormalities that promote fungal growth.<sup>5</sup> Hematologic malignancy is the most fatal factor of mucormycosis.

Incidence of fungal infection in middle ear and mastoid is rare. Only a few cases of temporal bone mucormycosis have been reported.<sup>6</sup>

Clinical and radiological aspects of mucormycosis are indistinguishable from ordinary otitis media. Atypical pain, swelling, and loss of function in the oral and maxillofacial areas occur, and they do not have particular discriminative points to suggest fungal infection. Repetition of the investigations is often necessary.

Similarly, CT scan findings are also equivocal and typically show a sequestrum such as amorphous, high attenuated bony fragments in the affected structure and increased bony sclerosis in the medullary portion adjacent to the lesions with erosion and destruction of the bony sinus wall and extrasinusoidal spread of the infection.

These resemble the ordinary OM.

Mucormycosis can infect the nose and the paranasal sinuses through inhalation of sporangiospores and this is the most common route of transmission. Ingestion of spores, direct implantation into injured skin (burns), trauma with contaminated soil, or intravenous (drug users) transmission has also been described.<sup>7</sup> Portal of entry to the middle ear and mastoid in a healthy individual is difficult to explain. Probable portal of entry to the middle ear is either from nasopharynx through eustachian tube to antrum or mesotympanum or through the perforated tympanic membrane. The fungus might have entered through the preexisting perforation in the tympanic membrane.<sup>8</sup>

The late occurrence of bony involvement is explained by the angioinvasive nature of the fungus and characteristically deep extension of infection through perivascular channels that precedes frank bony destruction.

The rarity of this condition makes it difficult for the clinician to suspect middle ear mucormycosis as a principal diagnosis.

Management of invasive form of mucormycosis will require the use of IV amphotericin B, radical debridement of the affected tissue, as well as control of the predisposing factors such as diabetes.

The efficacy of amphotericin B has been established. Delayed therapy results in two-fold increase in mortality. The liposomal AmB formulation has better penetration into cerebral tissue; hence carries a potentially better efficacy in cases of rhinocerebral involvement.

Posaconazole, combination therapy with echinocandins and deferasirox, and hyperbaric oxygen can be used as adjunctive therapy. The treatment has been supplemented with granulocyte transfusion and bone marrow transplantation in select cases. Prince and Stevenson have explained the role of hyperbaric oxygen therapy on presumption that increased the oxygenation of the affected tissues distal to the occluded vessel and decrease local acidosis and enhance the activity of the fungicidal medication.<sup>9</sup>

## CONCLUSION

Invasive mucormycosis of middle ear is a rare but serious condition that requires a high index of suspicion for diagnosis, and prompt therapy.

Radical debridement of the fungal debris with wide conchomeatoplasty for adequate ventilation and drainage is fundamental for the successful management. Shortening the time to start antifungal medication accounts for better survival.

## REFERENCES

1. Hazarika P, Ravikumar V, Nayak RG, Roa PS, Shivnanda PG. Rhinocerebral mucormycosis. *Ear Nose Throat J* 1984; 63:100–106.
2. Yun MW, Lui CC, Chen WJ. *Am J Otol* 1994;15(3):413–414
3. Spellberg B, Walsh TJ, Kontoyiannis DP, Edwards JJ, Jr., Ibrahim AS. Recent advances in the management of mucormycosis: from bench to bedside. *Clinical Infectious Diseases* 2009;48(12):1743–1751.
4. Zilberberg MD, Shorr AF, Huang H, et al. Hospital days, hospitalization costs, and inpatient mortality among patients with mucormycosis: a retrospective analysis of US hospital discharge data. *BMC Infectious Diseases* 2014;14
5. Turunc T, Demiroglu YZ, Aliskan H, Colakoglu S, Arslan H. Eleven cases of mucormycosis with atypical clinical manifestations in diabetic patients. *Diabetes Research and Clinical Practice* 2008;82(2):203–208

6. Gussen R, Canalis RF. Mucormycosis of the temporal bone. *Ann Otol Rhinol laryngol* 1982;91:27–32
7. Petrikos G, Skiada A, Lortholary O, Roilides E, Walsh TJ, Kontoyiannis DP. Epidemiology and clinical manifestations of mucormycosis. *Clinical Infectious Diseases*. 2012;54 (supplement 1).
8. Bergstrom L, Hemenway WG, Barnhant RA, et al. Rhinocerebral and otologic mucormycosis. *Ann Otol Rhinol Laryngol* 1970; 79:70–81.
9. Price JC, Stevans DL. Hyperbaric oxygen in the treatment of rhinocerebral mucormycosis.