CASE REPORT

A Rare Case of Middle Ear Mucormycosis Presenting with Facial Nerve Palsy

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Abstract

Introduction: Mucormycosis is an uncommon emerging fungal infection with high morbidity and mortality among diabetic and immunocompromised patients. The most common clinical manifestation is rhino-orbito-cerebral mucormycosis. Occurrence of middle ear mucormycosis with facial palsy is very rare.

Case description: We report a case of a 59-year-old male patient who was presented with ear pain, ear discharge, and facial asymmetry. On clinical examination House-Brackman grade IV facial nerve palsy was noted and otoendoscopy showed external auditory canal edema, subtotal tympanic membrane perforation, necrotic malleus, lenticular process erosion with pale granulation tissue in anterior epitympanum. High resolution computed tomography (HRCT) temporal bone revealed soft tissue density in middle ear, mastoid, and external auditory canal with mild erosion of tegmen tympani and rarefaction of facial canal at first genu and tympanic segment with thickening and edema of first genu of facial nerve. Based on histopathological diagnosis of mucormycosis, patient was started on liposomal amphotericin B injection and clinical improvement was noted after a total dose of 3500 mg over 16 days.

Conclusion: Early diagnosis and treatment with antifungals, glycemic control, and other supportive treatment with regular facial physiotherapy remain the mainstay of management in mucormycosis. Amid an increased number of COVID-19-associated rhino-orbito-cerebral mucormycosis, we report an unusual case of facial nerve palsy secondary to middle ear mucormycosis emphasizing the need for surgeons to have a broad mind to look for fungal infection in patients presenting with above-mentioned complaints and refractory to antibiotics.

Keywords: Amphotericin B, Facial palsy, Middle ear, Mucormycosis.

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Introduction

Mucormycosis is an uncommon emerging fungal infection with high morbidity and mortality among diabetic and immunocompromised patients. It is caused by a fungus belonging to the order of Mucorales existing in many forms like rhinocerebral, cutaneous, pulmonary, disseminated, etc. Mucormycosis involving the middle ear is quite rare.[1,2] So far, very few case reports on facial nerve palsy secondary to mucormycosis have been noted.[1,3] Malignant otitis externa (MOE) is an intractable life-threatening inflammatory condition, most commonly caused by *pseudomonas aeruginosa* primarily affecting elderly diabetic and immunocompromised patients. Fungi accounts for 5–20% of MOE cases with aspergillus being the most common fungal cause.[4] We report a rare case of middle ear mucormycosis causing right-side facial nerve palsy.

Case Description

A 59-year-old male patient was presented with complaints of right ear pain since 1 month, foul smelling right ear discharge since 3 weeks, and facial asymmetry since 3 days. Patient is a known diabetic since 8 years, not on regular treatment. Patient reported a score of 9 on visual analog scale for his ear pain. On examination patient had loss of right-side facial frowning, incomplete closure of the right eye, deviation of angle of mouth to left, and air escape on blowing cheeks—House Brackmann Grade IV (Fig. 1).

Right ear otoscopic examination showed edematous external canal with discharge. Left ear examination was normal. Discharge from right ear was sent for gram stain, culture sensitivity, KOH. Gram stain showed plenty of gram-negative bacilli. No fungal elements were observed in KOH. Blood investigations revealed increased fasting and postprandial sugars with HbA1C-9.2, CRP-8.07. Culture sensitivity showed the growth of *Pseudomonas aeruginosa* isolate. COVID-19 RT-PCR was negative. Diagnostic nasal endoscopy was normal. High-resolution computed tomography of temporal bone showed homogeneously enhancing soft tissue density in right middle ear with extension into mastoid air cells, EAC with mild erosion of tegmen tympani, and rarefaction of facial canal was noted at first genu and tympanic segment. Edema and thickening were noted at first genu of facial nerve (Figs 2A and B). Magnetic resonance imaging brain showed soft tissue inflammatory changes in right mastoid air cells, middle ear cavity, and EAC with

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soft tissue edema of deep neck spaces of neck and around right temporomandibular joint (Figs 2C and D).

Topodiagnostic testing of facial nerve showed left eye positive Schirmer's test for lacrimation, absent stapedial reflex, absent taste sensation on the right side of anterior two-thirds of tongue. Electroneurography, an objective test for estimating the amount of axonal degeneration, done on day 3 of admission to hospital showed 70% loss of amplitude compared to normal side. Patient was started on Tablet Ciprofloxacin 500 mg and injection Meropenem 1 g twice daily basing on the lower Minimum Inhibitory Concentration scores (<0.25), when compared to other antibiotics with higher MIC scores. Steroid were withheld as the patient had uncontrolled diabetes mellitus.

As there was no improvement in the clinical condition of the patient after 4 days of treatment, patient was taken up for otoendoscopic examination under general anesthesia. Tympanic membrane showed subtotal perforation, necrotic malleus, lenticular process erosion with pale granulation tissue surrounding the malleus in anterior epitympanum was noted. Biopsy of the granulation tissue was sent for histopathological examination. Histopathology showed fibrous connective tissue and dead bone with inflammatory infiltrate chiefly consisting of neutrophils, broad branching aseptate hyphae—suggestive of fungal infection—mucormycosis (Fig. 3).

In view of histopathological diagnosis of mucormycosis, patient was started on liposomal amphotericin B after thorough checkup of renal parameters. After an initial dose of 50 mg in dextrose infusion, daily dose was gradually increased from 50 to 100 mg, 150 mg, 200 mg based on the patient tolerability with regular monitoring of renal function tests. Patient reported reduction in symptoms of pain (visual analogue score—4) after 4 days of initiation of amphotericin; hence patient was not taken up for surgery. A total dose of 3600 mg was given for 16 days along with supportive IV antibiotics and diabetic control with insulin. Standard facial physiotherapy exercises protocol was followed. With all these measures, patient's general condition gradually improved over a period of 20 days. Patient underwent cortical mastoidectomy with type III tympanoplasty after recovery.

Discussion

Incidence of mucormycosis in middle ear is very rare.[1,2] Incidence is more in patients with diabetes mellitus, immunocompromised states. Immunocompetent patients can also be affected after trauma or burns. The causative organisms are ubiquitous hyaline moulds belonging to the order Mucorales under the subphylum mucorycotonia.[5] The airborne conidia produced by these organisms infects various parts of the body and leads to rhinocerebral,

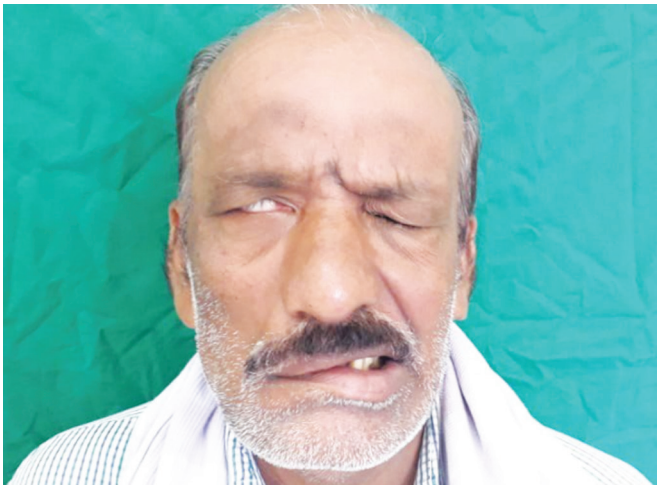


Fig. 1: Picture showing facial nerve palsy HB grade IV at presentation

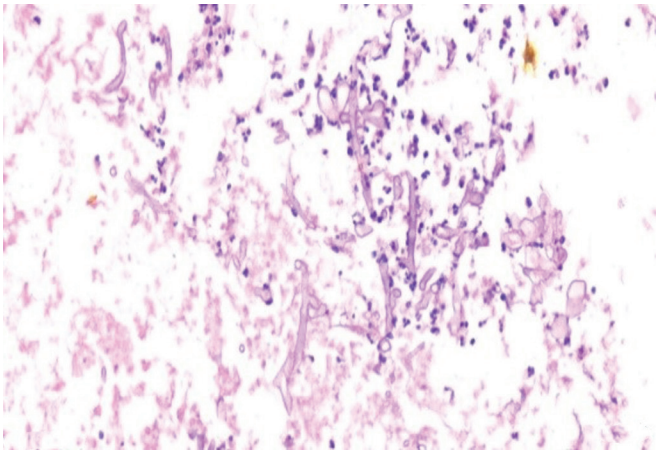
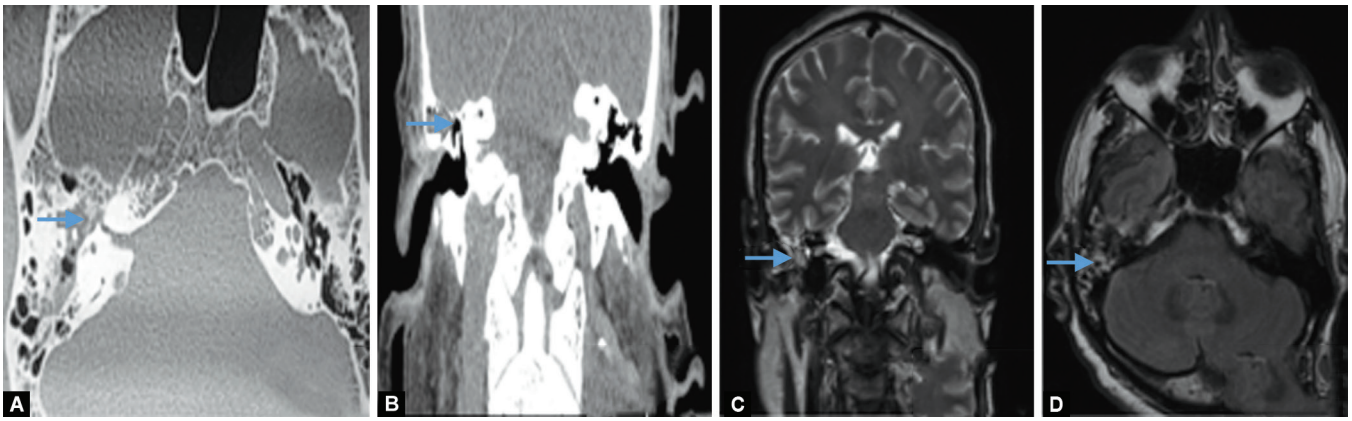


Fig. 3: Histopathological picture showing tangled masses of aseptate hyphae—mucormycosis



Figs 2A to D: (A) HRCT temporal bone showing rarefraction at first genu of facial nerve (right side); (B) Mild erosion of tegmen tympani; (C) MRI Brain—Altered signal intensity in right middle ear; and (D) Right mastoid air cells

cutaneous, pulmonary, disseminated, gastrointestinal, and other subtypes. Huckepo was the first to describe the involvement of ear in mucormycosis in the year 1886.[5]

MOE, first described by James R Chandler in the year 1968,[6] is a life-threatening inflammatory condition of the external ear typically caused by *P. aeruginosa*, a gram-negative bacteria,[5] and is one of the nearest differential diagnosis that is commonly considered in a patient presenting with ear discharge and uncontrolled diabetes.

Till date, few case reports have been published on mucormycosis of temporal bone, mastoid, and middle ear.[1,2,3,7,8] Sathish Kumar et al. reported a case report of mucormycosis of temporal bone where the patient presented with scanty ear discharge, pain, and facial asymmetry, and the histopathological examination after debridement revealed the presence of mucormycosis.[1] Ear pain, discharge, and facial asymmetry are some of the most commonly reported symptoms during the course of this disease. Our patient also presented with similar symptoms.

In a case report of mucormycosis of middle ear by Hazarika et al., where after modified radical mastoidectomy, histopathological examination of the soft tissue debris in attic and antrum showed the presence of tangled masse of aseptate fungus with diagnosis of mucormycosis.[2] The diagnostic histopathological feature of mucormycosis showed the presence of broad, aseptate, thin-walled fungus with irregular branching at right angles which made us to conclude the diagnosis of mucormycosis in our case.

Vaishali et al. reported a case of isolated facial nerve palsy secondary to mucormycosis where patient showed improvement after treatment with Amphotericin B for 24 days followed by step down treatment with Posoconazole for 2 weeks.[3] In our case, facial nerve palsy grade improved after treatment with Amphotericin B for 16 days.

Information about viability of the nerve can be obtained by an objective test electroneurography which could be one of the decision makers in regard to surgical management (facial nerve decompression). The amount of axon loss can be predicted by measuring the amount of evoked action potentials in affected muscles. Surgical decompression is considered in patients showing 90% axonal degeneration. As per the literature patients with less than 90% axonal degeneration may not require surgical decompression.[9,10] Electroneurograpy is repeated at 3–5-day interval until a plateau is obtained.[10] As our patient had 70 % axonal degeneration, surgical decompression was not done.

There are various strategies involved in the treatment of infection. Glycemic control and treatment of the underlying metabolic or immunocompromised conditions are the primary steps to prevent the spread of infection. Review of literature shows effectiveness of timely intervention with liposomal amphotericin B or posoconozole or voriconazole.[1,2,3,8] In our case, liposomal amphotericin B was started at a dose of 1 mg/kg/day after a complete check of renal functioning of the patient. The dose was gradually increased and a total dose of 3600 mg was given over a period of 16 days.

Conclusion

Middle ear mucormycosis presenting with facial nerve palsy is rare. It requires multidisciplinary approach to control the disease. As mucormycosis is one of the rare causes for ear discharge, surgeons also should have a broad mind to look for fungal infection when patients present with persistent ear discharge despite antibiotic treatment. Early diagnosis by sending granulation tissue for fungal stains and histopathological examination with appropriate treatment will improve the survival rate and decreases the mortality and morbidity of the patient. Antifungal agents, diabetic control, and regular facial physiotherapy were the main stay of treatment in our case. Amid an increased number of COVID-19-associated rhino-orbito-cerebral mucormycosis, this is a rare case of middle ear mucormycosis which emphasizes the need for differential diagnosis of mucormycosis in patient presenting with ear discharge refractory to antibiotics.

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Declarations

Availability of data and material: Data transparency has been maintained.
Ethical approval: Appropriate ethical clearance has been obtained from the institute.
Informed consent to participate: Informed consent was obtained from the patient for case report.
Consent for publication: Appropriate consent for publication taken.

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