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CASE REPORT

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# Auricular mucormycosis complicated by parotid abscess and facial paralysis: A very rare case report

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**Abstract**

Mucormycosis is a serious and relatively rare invasive fungal infection. The rhino-orbito-cerebral localization is the most frequent. Other localizations have been reported including: cutaneous, pulmonary, disseminated, gastrointestinal, and miscellaneous. Mucormycosis of external ear with facial palsy is extremely rare and only few cases have been reported in the literature. We describe a case of mucormycosis of the external and middle in a 44-years-old diabetic patient which had a very aggressive form of necrotic infection of the ear auricle complicated by parotid abscess, facial, and vagal nerve paralysis. Auricular mucormycosis was suspected and the diagnosis was established after biopsy and histological examination. The patient was treated immediately with intravenous amphotericin B followed by extensive surgical debridement of the necrotic lesions. The patient responded well to the treatment despite the persistence of facial palsy.

**KEYWORDS**

amphotericin B, diabetic, facial palsy, Mucormycosis, vagal palsy

## 1 | INTRODUCTION

Mucormycosis is a rare, saprophytic, invasive, and fulminant fungal disease. It is caused by fungi of the order of Mucorales belonging to the class of Zygomycetes. It is a potentially fatal fungal infection that usually affects immunocompromised hosts almost uniformly.[1] Patients at greatest risk of developing invasive mucormycosis have reduced numbers of mononuclear and polymorphonuclear phagocytes, as seen in neutropenia, or disorders that affect the phagocyte function, as in hyperglycemia, acidosis, or glucocorticoid administration. These factors weaken the immune system and allow fungus to grow and spread, resulting in invasive illness.[2] Mucorales are distributed into six families all of which can cause fungal infections. Out of the six families *Rhizopus oryzae* is the commonest cause of infection. Among the other species that are isolated include *Rhizopus microspores*, rhizopodiformis, Absidia corymbifera, Apophysomyces elegans, Mucor species, and Rhizomucor.[3] Clinically, the rhino-orbito-cerebral location is the most common. Other manifestations are pulmonary, cutaneous, disseminated, and gastrointestinal. In the cranio-facial region, a few publications have described mucormycosis that occurred in localized regions such as the tongue, the palate, the mandibula, the maxilla, the infra-orbital region, and the temporal bone.[2] Mucormycosis of the external and middle ear is a rare entity. A review of the literature found very few

cases of facial nerve palsy secondary to mucormycosis of the external ear extended to the temporal bone. The aim of this case is to illustrate the pathogenesis of auricular mucormycosis, its clinical presentation and diagnosis and to discuss its therapeutic modalities and prognosis.

## 2 | CASE REPORT

A 44-years-old diabetic man was hospitalized for the management of a blackish necrotic lesion in the left auricle with significant locoregional extension. The history of his disease goes back to 10 days before hospitalization: He had a pretragal swelling, with a bluish zone, quickly evolving to a blackish necrotic lesion centered on the left auricle associated with a pretragal fistula. This lesion extended anteriorly to the parotid region, superiorly to the temporal region, and posteriorly to the retroauricular region (Figure 1). Purulent otorrhea, otalgia, and left hearing loss were present. Otoscopic examination revealed a blackish tissue filling the external auditory canal and a secretion-covered eardrum that was bluish after aspiration. The rhinological, endobuccal, and cervical examinations were normal. The patient had a left peripheral facial palsy (grade IV of House and Brackmann classification) associated with a left vocal cord palsy. The rest of the neurological examination was normal.

Biologically: the blood count showed a hyperleukocytosis of 21,240 cells/mm$^3$ (92% neutrophils, with lymphopenia: 3.3% lymphocytes, i.e., 700/mm$^3$), normocytic normochromic anemia with a hemoglobin level of 9.2 gr/dl, and platelets of 233,000/mm$^3$. CRP and blood sugar levels were high (respectively 254 mg/L and 260 mg/dL).

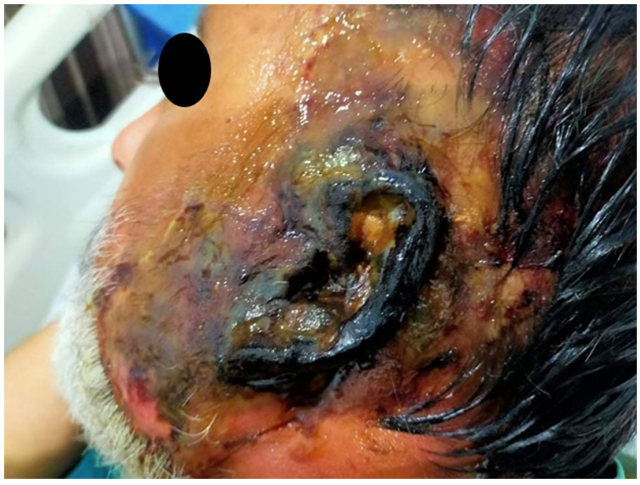

**FIGURE 1** Initial photograph of the patient: Blackish necrotic lesion of the left auricle, parotid, temporal, and left retroauricular region with pus-secreting pretragal fistula.

High-resolution computed tomography of the temporal bone showed thickening of the left peri-auricular soft tissues, filling of the left external auditory canal, middle ear and mastoid cells without bone lysis or nasosinusal involvement or endocranial extension (Figures 2 and 3). Magnetic resonance imaging with gadolinium injection showed infiltration of the subcutaneous soft tissues extending to the homolateral masticatory, parapharyngeal, and parotid spaces with collection in the left parotid region and the masticatory space communicating with the homolateral parapharyngeal space, exerting a mass effect on the pharyngolaryngeal lumen (Figures 4 and 5).

The patient underwent a biopsy of the auricle. Histopathology showed the presence of mucormycosis mycelia filaments. The culture was positive with the presence of *Rhisopus Microspores*. Liposomal Amphotericin B was immediately administered at a dose of 5 mg/kg/d. The collection was drained under general anesthesia. Bacteriological examination of the drained abscess isolated *Klebsiella Pneumonia* confirming bacterial superinfection and the patient was treated by Vancomycin–Imipenem associated with Amphotericin B. However, an immediate surgical debridement surgery was not possible because of poor surgical risk owing to the extensive inflammation around these necrotic lesions.

A regression of the parotid swelling, local inflammatory signs, and biological inflammatory syndrome were noted. However, the necrotic lesion extended inferiorly and superiorly to the temporoparietal region with spontaneous anterior detachment of the necrotic plaque (Figure 6). This plaque was lined with underlying budding tissue which motivated us to perform a surgical debridement of the necrotic lesions with total excision of the left auricle and external auditory canal. (Figure 7). An informed consent was obtained from the patient before surgery.

After several months, a healing of the substance loss was obtained by secondary intention healing (a cover flap was not necessary) (Figure 8). However, facial and left vagus nerves paralysis persisted without any real recovery.

## 3 | DISCUSSION

Mucormycosis is a rare fungal infection caused by filamentous fungi belonging to the class Zycomycetes, in the order of Mucorales. They are not very virulent in immunocompetent patients.[1] They become pathogenic in immunocompromised patients such as those with diabetes and acidosis, immune deficiency with neutropenia, severe malnutrition and during corticosteroid or cytotoxic treatments.[2,4,5]

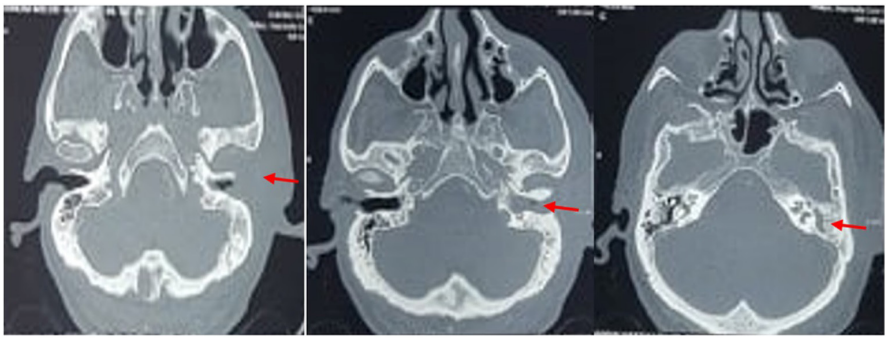

**FIGURE 2** Cerebral and temporal bone CT in non-injected axial section: thickening of the left periauricular soft tissue, filling of the left external auditory canal, middle ear and mastoid cells without bony lysis, or sinonasal involvement.

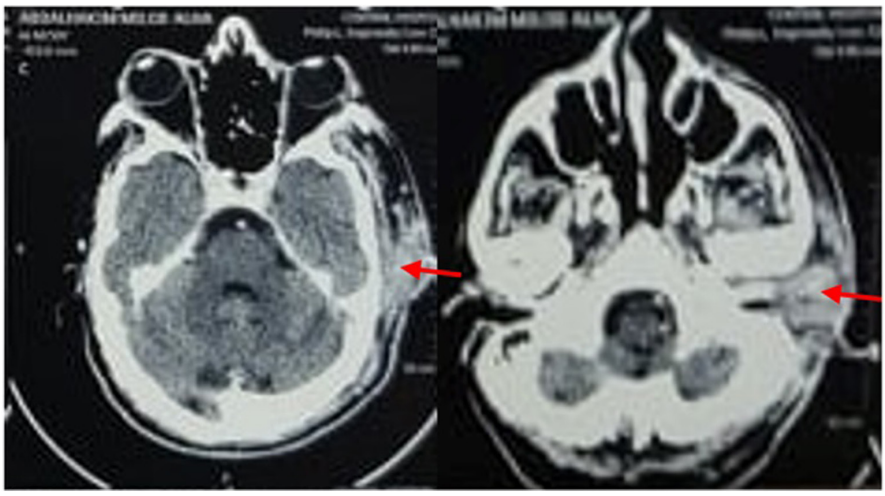

**FIGURE 3** Cerebral and temporal bone CT in injected axial sections: Swelling of the left external temporal muscle and soft tissues with heterogeneous contrast. No endocranial extension.

The rhino-orbito-cerebral localization is the most frequent (40% of cases).[2,6] Other locations have been reported, including cutaneous, pulmonary, disseminated, gastrointestinal, and miscellaneous.[2,7] For the otologic location, several hypotheses have been suggested: either a passage of the fungal spores from the sinonasal cavities to the middle ear via the eustachian tube, or their penetration to the middle ear through a perforation of the tympanic membrane, or an auricular mucormycosis invading the middle ear and the mastoid[8,9] as in our case.

Clinically, otologic mucormycosis may manifest as refractory otitis externa involving the pinna and external auditory canal giving a blackish necrotic appearance with skin scaling.[9] It may be presented as a middle ear cholesteatoma with foul-smelling otorrhea and otalgia.[5,7,10] Endocranial or skull base extension indicates a severe infection with cranial nerve damage, with or without neurological signs. The particularity of our observation consists of the auricular cutaneous starting point with involvement of the external ear and the large extension to the parotid, retro auricular and temporal regions. This extension was accompanied by bacterial superinfection with parotid abscess extending to the parapharyngeal space associated with peripheral facial and vagus nerves paralysis. Among the cranial nerves, the facial nerve is the most commonly affected in auricular mucormycosis.[8–11] Vagus nerve paralysis, presumably related to this parapharyngeal extension, is however unusual and would probably be the first case of this type to be reported so far.

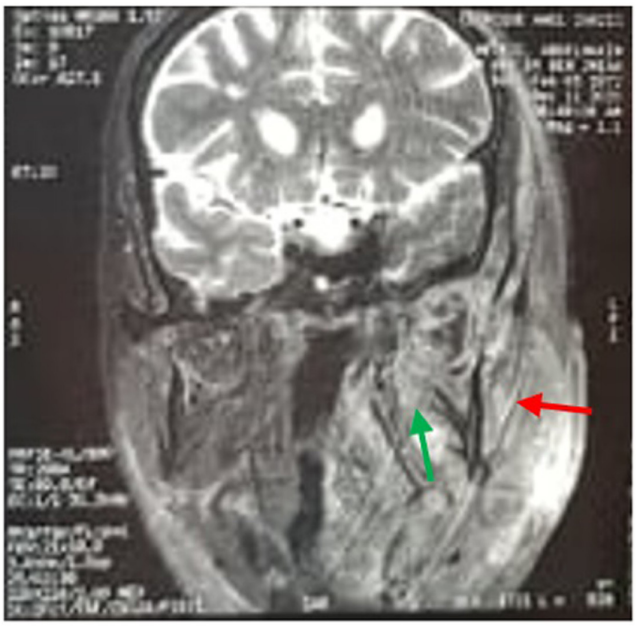

**FIGURE 4** Cerebral and cervicofacial MRI in T2 coronal section: T2 hypersignal of the left masticatory, parotid (red arrow), and parapharyngeal spaces (green arrow).

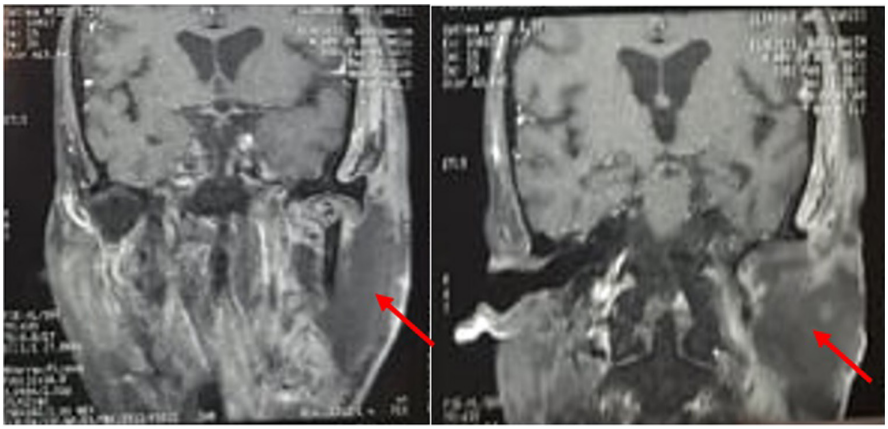

**FIGURE 5** Cerebral and cervicofacial MRI in T1 coronal section: Collection of the left parotid cavity and the masticatory space communicating with the homolateral parapharyngeal space reaching up to the base of the skull and exerting a mass effect on the pharyngo-laryngeal lumen.

Reviewing previously described otologic mucormycosis within the literature, only a few cases of auricular mucormycosis have been reported in the literature. Indeed, Aljehani M[12] reported in 2021 a case of an 18-month-old girl with undiagnosed diabetes presented with an aggressive form of auricular mucormycosis with facial nerve palsy that have fully recovered after a combination of antifungal, antibiotic, and antiviral with extensive surgical debridement. Kumar and Nishan[9] reported in 2015 a case of a mucormycosis of the external ear invading the temporal bone with associated facial palsy. The patient had surgical debridement with a modified radical mastoidectomy and facial nerve decompression combined with systemic liposomal amphotericin B. The patient full recovered with a significant improvement in facial palsy after surgery. M Faruk Oktay[8] reported a case of a 17-year-old female patient with diabetic ketoacidosis presented with auricular mucormycosis and of the external ear canal. Unfortunately, the patient died on the seventh day despite of the extensive debridement associated with systemic amphotericin B.

The diagnosis of mucormycosis is based on histopathological examination. Direct mycological examination is rarely positive, and culture on Sabouraud agar is often negative.[3,4] Pathological examination shows broad mycelial filaments 7–10 microns wide and 100–200 microns long, hyaline, non-septate with right-angled branching.[4,6]

Treatment consists of a combination of antifungal (amphotericin B at high doses of 5-15 mg/kg/day), surgical debridement and systematic control of comorbidities.[6,9] Resection of necrotic tissue reduces the fungal load and improves the penetration of antifungal agents into infected areas.[13] Due to the rarity of cases, the duration of antifungal treatment for otologic mucormycosis has not been established. Besides, various studies have shown the favorable role of hyperbaric oxygen therapy. Increased oxygenation of the affected tissue improves the phagocytic capacity of neutrophils and decreases local acidosis[14] which inhibits fungal spore germination and mycelial growth.

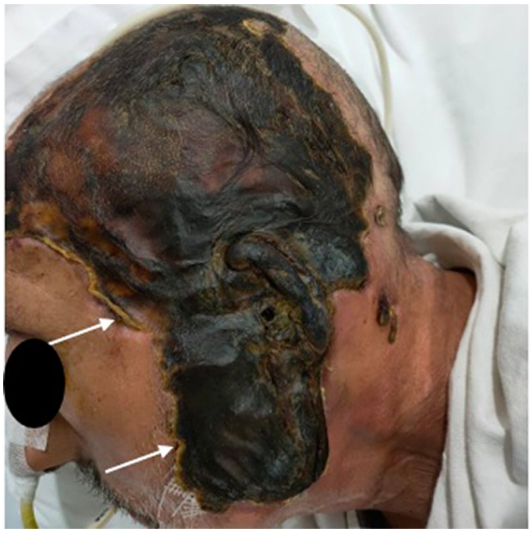

**FIGURE 6** Photograph of the patient showing spontaneous anterior detachment of the necrotic plaque.

Prognosis of any localization of mucormycosis largely depends upon the extent of spread, the aggressiveness of disease and the time of treatment. The immune status of the patient, particularly the presence of any hematologic disease, plays an important role. Overall reported mortality with all forms of mucormycosis range from 40% to 80% with survival rates significantly worse in patients with hematological malignancies and organ transplantation.[2] the mortality of otologic mucormycosis is significantly lower than that of the rhino-orbito-cerebral form.[9] This may be explained by the poor vascularity of the middle ear compared to the paranasal sinuses limiting angioinvasion and spread of infection, differences in the microbiome between the middle ear and paranasal sinuses, or simply a reflection of the small number of cases.[8] Another important prognostic factor is the time at which the disease was diagnosed and treated. If mucormycosis affected patients receive surgical and

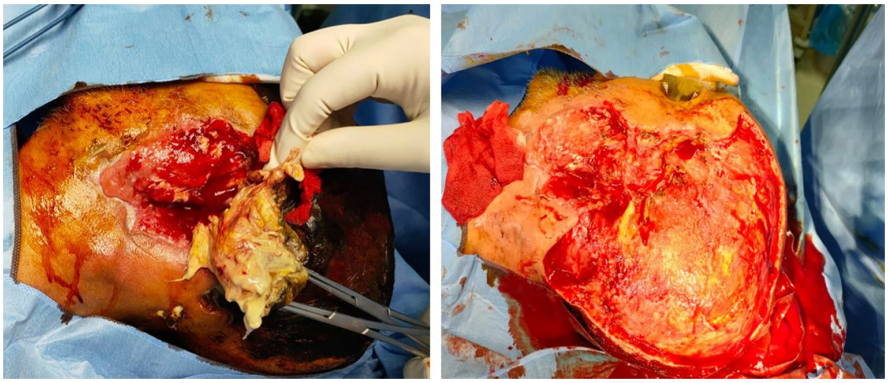

**FIGURE 7** Surgical debridement of necrotic tissue lining an underlying budding tissue with removal of the left auricle and external ear canal.

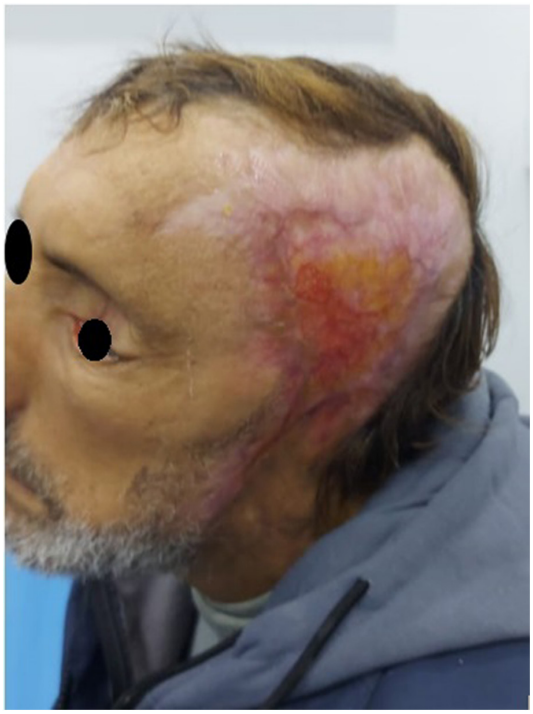

**FIGURE 8** Photo of the patient after several months of treatment: healing of the substance loss and persistence of facial paralysis.

medical treatment on time, there are chances of mortality rate falling down to less than 20%.[2]

## 4 | CONCLUSION

Auricular mucormycosis is a very rare but serious fungal infection affecting mainly immunocompromised hosts. This clinical case is unique in its presentation with an initial extensive involvement of the auricle with locoregional extension associated with involvement of the facial and vagus nerves. Early diagnosis, control of risk factors, surgical debridement of necrotic tissue, and administration of antifungals are the keys to successful treatment.

### AUTHOR CONTRIBUTIONS

**Bouthaina Hammami:** Data curation; investigation; methodology; project administration; supervision; validation; writing – review and editing. **Imen Achour:** Supervision; validation; visualization. **Omar Walha:** Conceptualization; investigation; methodology; software; writing – original draft. **Ghada Yousfi:** Conceptualization; validation. **Malek Mnejja:** Software; supervision; validation; visualization; writing – review and editing. **Ilhem Charfeddine:** Supervision; validation; visualization.

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All authors: Approved the final version and have the agreement to be accountable for all aspects of the work in ensuring that questions related to the accura.

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### CONFLICT OF INTEREST STATEMENT

The authors have no conflict of interest to declare.

### DATA AVAILABILITY STATEMENT

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

### CONSENT

I do confirm that the institutional ethical approval was obtained. A written informed consent was also obtained from the patient to publish this report in accordance with the journal's patient consent policy.

### ETHICS STATEMENT

Obtained from the patient in written.

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