

Rhino-Orbito-Cerebral Mucormycosis Following Tooth Extraction in a Diabetic Patient: A Case Report



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ABSTRACT

Mucormycosis is a rare, aggressive, and often fatal fungal infection that primarily affects immunosuppressed and diabetic patients, causing soft-tissue lesions. Early detection and prompt management are crucial. We present a case of mucormycosis following dental extraction in a patient with diabetes. A 48-year-old diabetic Iranian Baluch woman with multiple dental caries developed left eye exophthalmia and headache one week after an Afghan unlicensed dentist in Iran extracted four teeth from her left maxilla to alleviate pain and inflammation in her maxillary premolars and molars. Diagnostic tests confirmed that mucormycosis had spread from the extraction site to the surrounding soft tissue, sinuses, left eye, and brain. Diabetes significantly increases the risk of mucormycosis infections. In patients with tooth caries and infections, mucormycosis should be considered as a potential diagnosis. Timely diagnosis and treatment are crucial prior to dental procedures to prevent spread to the sinuses and brain.

Introduction

M

ucormycosis is a rapidly progressing opportunistic infection caused by Mucorales molds, with a high mortality rate (>40%). Infection occurs through inhalation of spores, ingestion of contaminated food, or skin wounds [1].

In developed countries, mucormycosis mainly affects the severely immunocompromised, whereas in underdeveloped countries, it primarily affects those with poorly controlled diabetes

or immunocompetent individuals post-trauma. Mucormycosis tends to invade blood vessels, leading to thrombosis, necrosis, and tissue infarction [2]. Mucormycosis affects 40%–50% of diabetic patients. They have a high mortality rate, and the key to success in treatment is early diagnosis, prompt administration of antifungal drugs, and extensive surgical debridement [3].

Case Presentation

A 48-year-old Iranian Baluch woman presented to

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the Emergency Department at Khatami Al-Anbia Hospital in Zahedan (Sistan and Baluchestan province, Iran) with maxillary inflammation, left eye proptosis, and headache. The patient's history showed that the patient had type 2 diabetes for two years and was receiving twice-daily oral treatment with SYNORIPA tablets (empagliflozin 12.5 + metformin 500). The patient also had multiple tooth decays, which caused mild inflammation and pain in the patient's left maxilla. She sought affordable dental care from an unlicensed Afghan dentist illegally practicing in a private Iranian residence, who extracted four decayed teeth from the affected area. One week post-tooth extraction, severe maxilla pain and inflammation developed, along with eye pain and a headache.

Clinical examination revealed vesicular lip lesions, cheilitis, extensive dental caries, left buccal and periorbital cellulitis, and left eye proptosis with halitosis. The movements of both eyes were normal, but there was a positive Marcus Gunn pupil in the left eye (Figure 1).

The patient was hemodynamically stable and afebrile (BP = 110/70 mmHg, HR = 121 beats/min, RR = 17 breaths/min, T = 37.5 °C, and SpO₂ = 98%) and had a Glasgow Coma Scale score of 14/15. The patient's blood sugar level was measured at 295 mg/dL via a glucometer.

The results of the laboratory tests were as follows.

WBC= $12/4 \times 10^3$ / μl , RBC= 4/1 mil/ μl , HB= 10/6 g/dL, HCT= 34/9 %, PLT= 223×1000 , PT= 14/sec, PTT= 45/sec, INR= 1/1, BS= 250 mg/dl, BUN= 22 mg/dl, CR= 1/2 mg/dl, K= 4/3 meq/l, Na= 136 meq/l, HBA_{1c}= 6/4

Maxillofacial and brain CT with and without contrast revealed vasogenic edema in the basal ganglia and left frontoparietal region, optic nerve edema, and enlarged left eye rectus muscles with left eye proptosis. Bilateral sinusitis was evident in the maxillary and ethmoid sinuses. However, no obvious thrombosis in the cavernous sinus or other vessels was seen on contrast-enhanced CT (Figures 2 and 3).

Rhino-orbito-cerebral mucormycosis was suspected based on symptoms, clinical examination, and diagnostic tests. The patient was empirically treated with amphotericin B liposomal 150 mg daily, vancomycin 1 g twice daily, ampicillin-sulbactam 3 g three times daily, and metronidazole 500 mg three times daily. Bromhexine 4 mg four times daily and pantoprazole 40 mg daily were initiated as adjunctive therapy.

Consultations with an intensivist, an ophthalmologist, an internist, and an otolaryngologist were performed for appropriate interdisciplinary management of the patient's treatment.

To manage the patient's blood sugar, the internist prescribed regular (5 units) and NPH (8 units) insulin twice daily (morning and evening). The ophthalmologist prescribed artificial tears every 2 hours, levofloxacin drops every 4 hours, and erythromycin ointment at night. The treatment team recommended that an otolaryngologist perform a nasal and sinus biopsy to confirm the diagnosis. Family members were informed that if the diagnosis was confirmed, the treatment plan would include left eye enucleation and endoscopic sinus surgical debridement. However, the patient's legal guardian



Fig. 1. The patient's image showed oral lesions and facial and periorbital cellulitis.

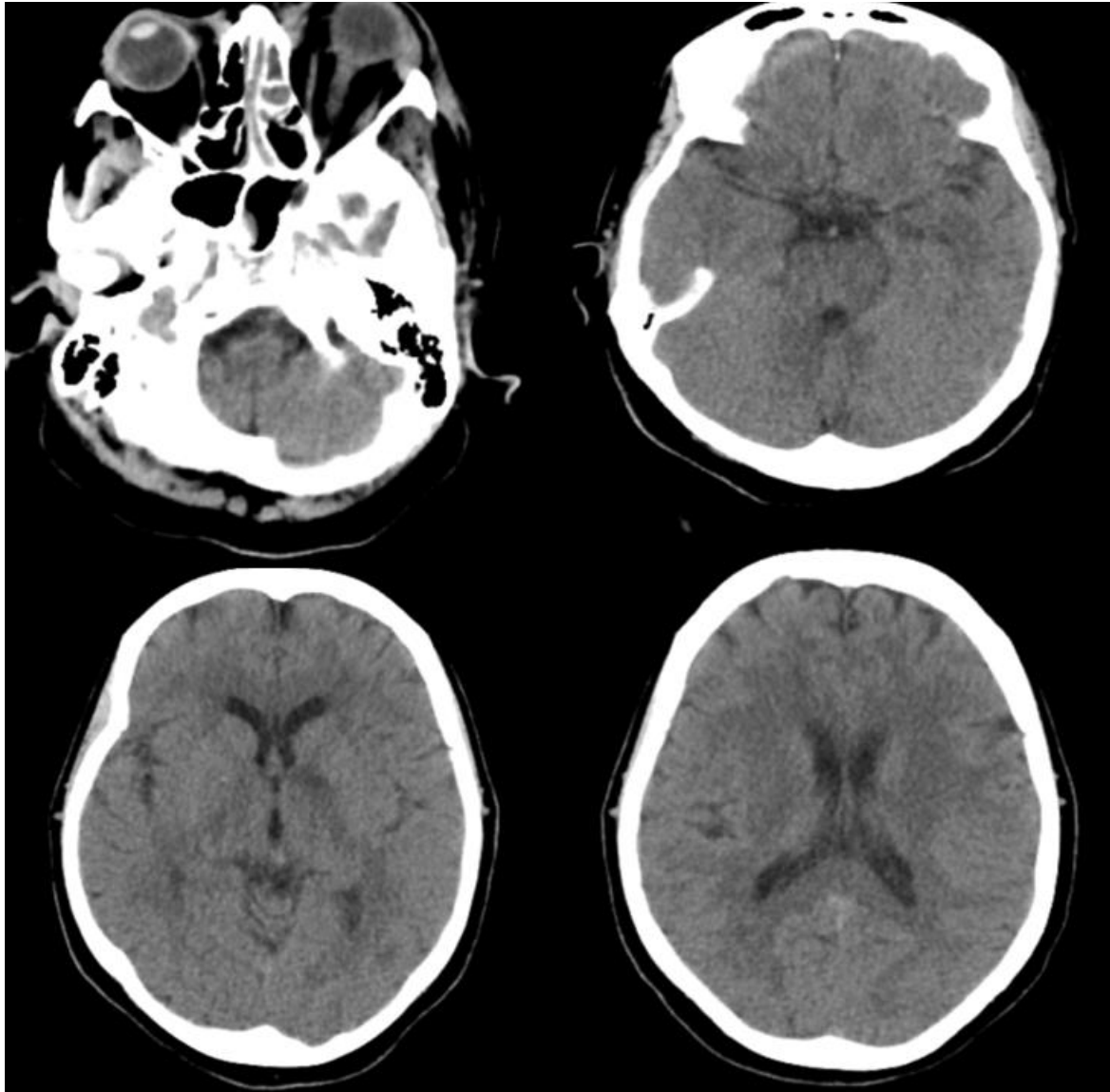


Fig. 2. Brain CT showed normal ventricles and sulci, without midline shift. But vasogenic edema was present in the basal ganglia and frontoparietal lobe.

declined consent for the biopsy and surgery, so the patient was transferred to the ICU for hospitalization and medical management.

During the first week of hospitalization in the intensive care unit, the patient's general condition worsened. Her level of consciousness decreased, and arterial blood gas analysis showed severe respiratory acidosis. Therefore, we intubated the patient and placed her on mechanical ventilation with AC/VC+ (PRCV), TV = 450 mL, RR = 13, Ti = 1.2, PEEP = 5, and FiO₂ = 100-40%.

Given the patient's severe condition, their legal guardian consented to a biopsy. Biopsy results from

the anterior, middle, and inferior nasal and sinus regions confirmed mucormycosis. Follow-up CT scan showed ischemic stroke in the middle cerebral artery (MCA) territory (Figure 4).

The patient's persistent loss of consciousness and unstable hemodynamics precluded an MRI to locate the arterial thrombosis. Due to her condition, the otolaryngologist deemed surgery unbeneficial. Ultimately, the patient died of ventilator-associated pneumonia (VAP) and subsequent multi-organ failure.

Discussion

In this case, a diabetic patient with gingivitis and tooth

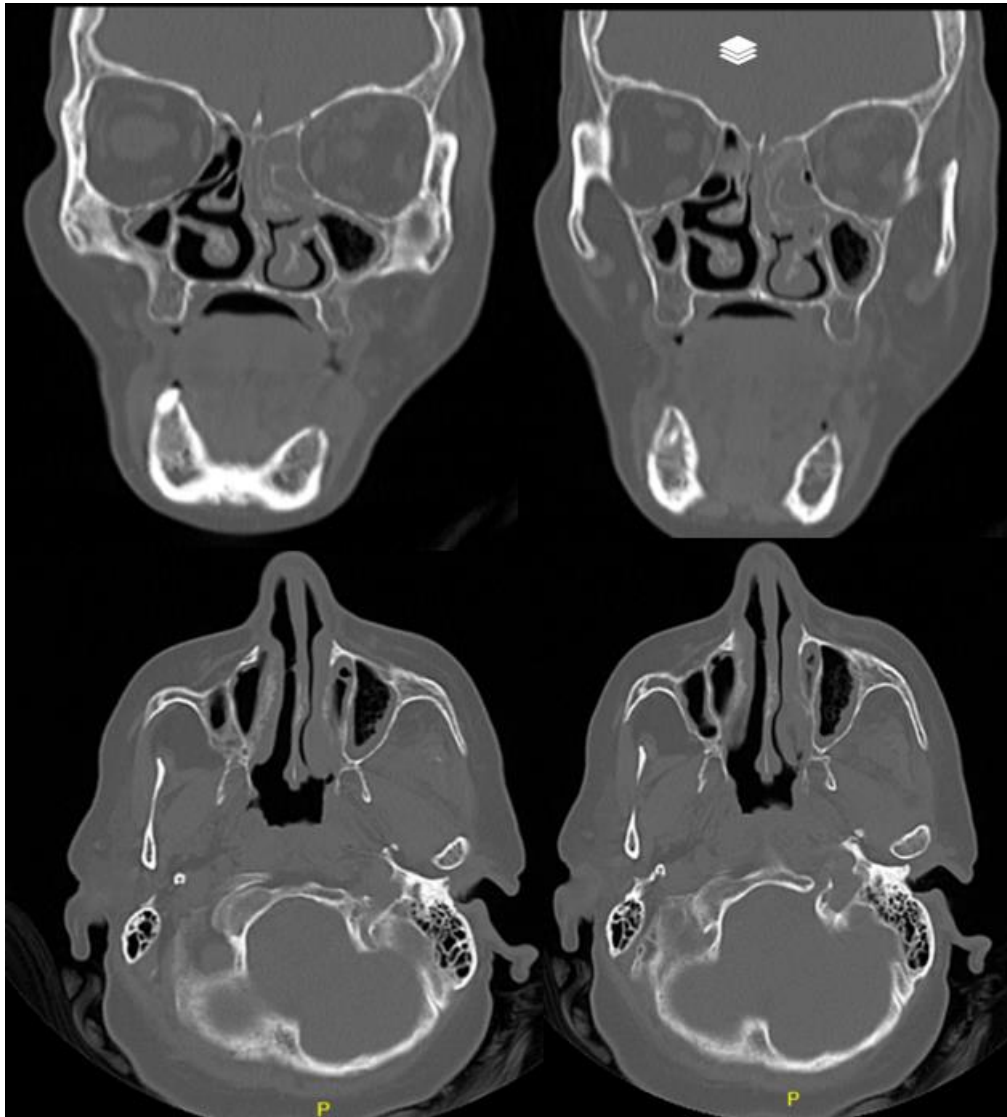


Fig. 3. Maxillofacial CT showed clouding of the bilateral maxillary and ethmoid sinuses, with severe hypertrophy of the left nasal turbinates.

decay received substandard dental treatment from an unlicensed provider, leading to a fatal mucormycosis infection that spread to the sinuses, eyes, and brain. This case highlights the serious risks associated with tooth extraction in the presence of inflammation and infection. If the patient had consulted a specialist dentist, mucormycosis would have been considered, and diagnostic testing and treatment would have been prioritized over tooth extraction. This would have prevented the spread of mucormycosis and the patient's death.

Diabetes is a predisposing factor in 60-80% of mucormycosis cases [4]. Increased blood glucose, lower pH, reduced blood flow, decreased serum inhibition, increased host receptor expression, abnormal phagocytosis, and impaired cell-mediated immunity compromise host defense against

mucormycosis [3]. Mucormycosis begins with facial pain and periorbital swelling, then progresses from the sinuses and nasal mucosa to the orbit and brain. The fungus can spread from the nasal passages and sinuses to the mouth, causing black discoloration of the palate (palatal mucosal necrosis), potentially leading to palatal perforation. Nasal findings may include black eschar discharge, thickened mucosa, and turbinate/septal necrosis. Black discoloration and necrosis indicate advanced disease and a poor prognosis. Disease spread from the nasal cavity and maxillary sinus to the orbit, via the nasolacrimal duct or ethmoid air cells, can cause chemosis, proptosis, vision loss, and external ophthalmoplegia, potentially leading to orbital apex syndrome [5]. The disease spreads to the brain from the orbit via arteries (retinal, ophthalmic, or carotid) or directly through skull fissures/plates. Brain involvement causes

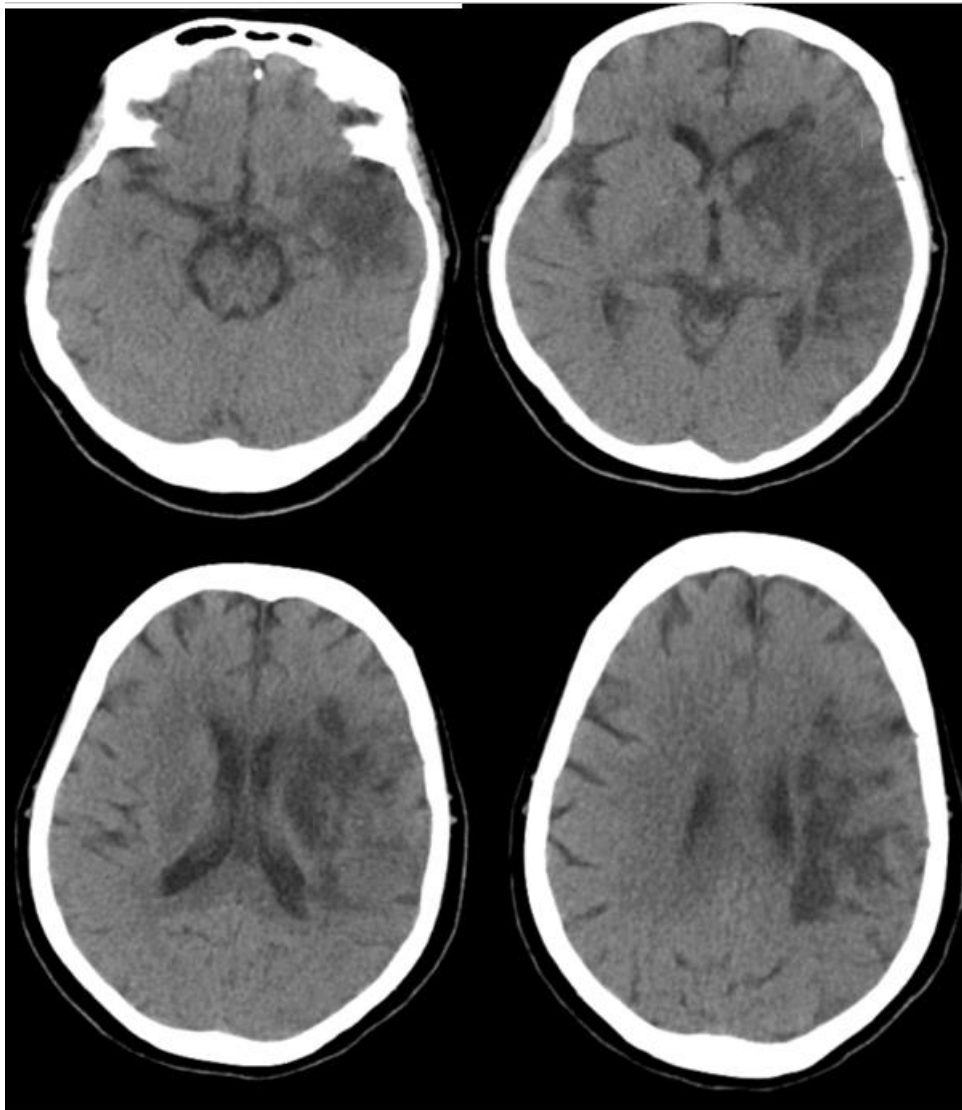


Fig. 4. Brain CT scan showed a left temporal-parietal ischemic stroke with minimal midline shift.

declining consciousness and coma due to thrombosis and infarction [6]. Vascular spread can lead to cavernous sinus thrombosis and brain infarctions, thus worsening the patient's prognosis [7]. As the pathophysiology indicates, in this case, we observed the spread of mucormycosis from the patient's maxilla after tooth extraction to the nose, sinuses, left eye, and brain, ultimately leading to cerebral ischemia. However, on clinical examination, we did not observe a necrotic, black, ulcerated lesion with exudate in the mouth or nose. Following tooth extraction, another case report documented mucormycosis spreading to the nose, sinuses, eyes, and brain, resulting in occipital and frontal ischemic stroke. This patient also presented with necrotic lesions on the hard palate, despite undergoing surgical debridement [5]. Both patients died of ischemic stroke, ventilator-induced pneumonia, and multi-organ failure. Our patient was

in stable condition upon admission. Had the legal guardian consented to the biopsy and debridement surgery on the first day, the outcome might have been improved. In another case report, mucormycosis following tooth extraction had spread to the nasal sinuses and facial bones, the skull base, and the temporal lobe of the brain. Endoscopic debridement of the sinuses and maxillary bone, and treatment with amphotericin, led to the patient's recovery [3]. This indicates the importance and the appropriate effect of surgical debridement in the recovery of patients with mucormycosis and sinus and facial involvement.

Rhino-orbital-cerebral mucormycosis requires immediate medical and surgical intervention. Delaying treatment increases morbidity and mortality. Empiric antifungal therapy and aggressive surgical debridement are crucial when rhino-orbital-cerebral

mucormycosis is suspected based on risk factors, clinical presentation, or radiographic evidence. For effective treatment of mucormycosis, consider a multi-pronged approach involving blood sugar control, reversal of immunosuppression, IV antifungals, and extensive surgical debridement [8].

Conclusion

Prompt diagnosis is crucial for effective treatment of mucormycosis. In patients with diabetes, consider mucormycosis as a primary cause of oral or nasal inflammation and infection. In patients with mucormycosis risk factors (e.g., diabetes, immunodeficiency, corticosteroid use, cancer, AIDS), prompt intravenous antifungal therapy and surgical debridement are crucial upon clinical suspicion and radiological confirmation to prevent mortality.

Abbreviations

VAP=Ventilator-Associated Pneumonia, PEEP=Positive End-expiratory Pressure, ICU= Intensive Care Unit, AC/VC+ (PRCV)= Assist Control/ Volume Control+ (Pressure Regulated Volume Control), RR= Respiratory Rate, TV= Tidal Volume, Ti= Time Inspiration, MRI= Magnetic Resonance Imaging

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Authors Contribution

AK: Providing care, following up on the patient's treatment process, writing case reports, and final editing.

AA: Follow up on the patient's treatment progress and edit the case report.

HRN: Follow up on the patient's treatment progress and edit the case report.

Ethical Considerations

Ethics approval and consent to participate

The study protocol was approved by the ethics committee of Zahedan University of Medical Sciences

(ethical code: IR.ZAUMS.REC.1404.334).

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Conflict of Interests

The authors declare that they have no competing interests.

Availability of Data and Materials

All patient file data and CT scans will be available upon request by contacting the corresponding author of the case report.

Consent for Publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Generative AI Disclosure

No generative AI was used in the writing of this case report.

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