Case Report

Fatal orbitomaxillary mucormycosis presenting with facial numbness in a patient with type 2 diabetes mellitus: A Case Report

Anushan Madhushanka A[1], Pemasiri KACC[2]

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Abstract

Mucormycosis is a potentially life-threatening invasive fungal infection typically occurring in immunocompromised patients. The common route of entry of microorganisms is by inhalation of spores, and the fungi exhibit a strong affinity for angioinvasion. Rhino-orbito-cerebral disease is the most typical manifestation of the infection. The incidence of mucormycosis following dental extraction remains relatively rare.

We report a rare case of orbito-maxillary mucormycosis following a tooth extraction in a young female with diabetes who presented with acute facial numbness and swelling. Facial numbness was an uncommon but crucial early sign that helped point towards the diagnosis. Despite early administration of liposomal amphotericin B and surgical debridement, the patient's condition deteriorated rapidly, leading to death.

A high degree of suspicion and awareness about uncommon clinical signs, prompting early intervention, increases the likelihood of a favourable outcome in mucormycosis.

Keywords: *facial numbness, mucormycosis, tooth extraction, liposomal amphotericin B*

Introduction

Mucormycosis is a life-threatening opportunistic infection caused by saprophytic fungi of the order *Mucorales*.[1,2] First described by German pathologist Paultauf in 1885, it predominantly affects immunocompromised individuals.[1] *Rhizopus* and *Rhizomucor* are the most common genera that cause human infection.[3] Infections are usually acquired following inhalation of fungal spores and rhino-orbito-cerebral disease is the most frequent clinical presentation.[3]

[1] *Ministry of Health, Colombo, Sri Lanka*
[2] *Faculty of Medicine, University of Kelaniya, Sri Lanka*

Address for correspondence: Dr. KACC. Pemasiri, Department of Medical Microbiology, Faculty of Medicine, University of Kelaniya, Ragama,.Sri Lanka Tel: +94718334069

Email: chethana_pemasiri@yahoo.com *https://orcid.org/0009-0000-7284-4817*

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We report a rare case of orbito-maxillary mucormycosis in a young female following tooth extraction, with facial numbness as an early presenting feature of the infection.

Case report

A 29-year-old female was admitted to the oral and maxillofacial unit of a tertiary care centre with progressive left-sided facial swelling for two days. One week before admission, she had undergone extraction of a painful, mobile tooth on the left upper jaw. As the examination findings of the oral cavity favoured a mixed bacterial infection, intraoral incision-and-drainage was performed, followed by administration of intravenous(IV) co-amoxyclav 1.2g 8 hourly. It was unusual to note that the patient did not feel any pain at the site of the incision, nor was there bleeding. Sensory mapping of the face revealed that the left mid-face was completely numb, prompting an urgent request for a computed tomographic scan of the region (performed two days later). Although she did not have a significant past medical history at the time of admission, her HbA1c was 7.9%. In addition, the white cell count was 14.8 x 10^6 µL (neutrophils 81%) and C-reactive proteins were 112 mg/L.

Figure 1: Blister and colour change with a demarcation

On day 2, the patient developed a few blisters and greyish discolouration of the affected area (Figure 1). An urgent multidisciplinary consultation resulted in making a diagnosis of probable orbitomaxillary mucormycosis. A few hours later, she was started on IV liposomal amphotericin B 3mg/kg/day. Later that day, she became febrile with a reduced level of consciousness, necessitating intubation. By the next day (day 3), the left mid-face showed extensive necrosis. An urgent surgical exploration revealed necrotic debris in the ethmoidal sinus with necrosis extending from the left eye to the skull base.

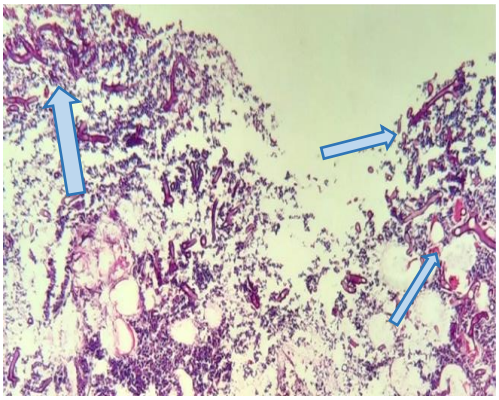

Figure 2: Numerous fungal filaments within necrotic debris (arrows)

Figure 3: Complete necrosis of left side of the face

Histological examination of the tissue specimens taken at debridement reported broad, non-septate fungal hyphae branching at a right angle, suggestive of mucormycosis (Figure 2). CT imaging revealed thrombosis of the left carotid body, ischemic features in the left-brain hemisphere, and involvement of all sinuses.

By day 4, the entire face had gross discolouration due to progressive necrosis (Figure 3). Further imaging revealed infarction of the left brain. Later, the patient went into a state of coma and, unfortunately, succumbed to death on day 6.

The timeline of the disease is shown in Figure 4.

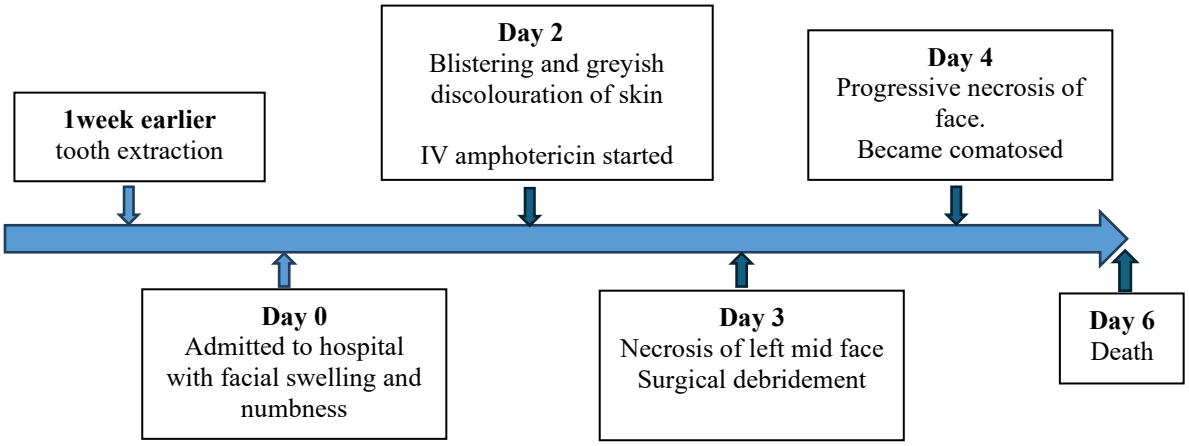

Figure 4: Timeline of the disease

Discussion

Mucormycosis is a rapidly progressive fungal infection with high morbidity and mortality. Uncontrolled diabetes is the main predisposing factor, with others including haematological malignancies, immunosuppressive therapy, and organ transplantation.[3,4]

The infection is usually acquired by inhalation of fungal spores but can also occur following ingestion or contamination of an open wound, albeit rarely. Rhino-orbito-cerebral disease is the most common manifestation, followed by pulmonary, cutaneous, gastrointestinal and disseminated disease.[1,5] However, mucormycosis following dental extraction is uncommon.[2] Extraction of teeth, especially the maxillary molars, can increase the likelihood of infection due to their proximity to

the maxillary sinuses, which are often affected by inhalation of fungal spores.[1] Contamination of the extraction site either during or after the extraction is another possibility.[6]

Mucorales typically begin to proliferate in the nasal mucosa or palate and extend to the paranasal sinuses. Subsequent invasion into the retro-orbital region, eroding through sinuses and skull bones, can predispose dissemination even to the brain.[7] Angioinvasion is the pathologic hallmark of the disease, which leads to thrombosis, followed by tissue infarction and necrosis.

Common symptoms of rhino-orbito-cerebral mucormycosis include headache, malaise, low-grade fever, facial pain, and swelling.[5] Involvement of cranial nerves V and VII may lead to ophthalmic manifestations, facial nerve palsy, and paresthesia.[7] Complications, such as cavernous sinus thrombosis or internal carotid artery involvement, can result in cerebral infarctions.

In our patient, facial numbness followed by greyish discolouration and blistering was the earliest indicator of mucormycosis. Awareness of atypical presentations is crucial, as highlighted in reported cases where a high index of suspicion facilitated early diagnosis and timely intervention. For instance, one case described a woman presenting with facial cellulitis and orbital apex syndrome.[4] Another report involved a man with poorly controlled diabetes who developed unilateral facial numbness, later complicated by hemiparesis and multiple cerebral infarcts, with post-mortem findings confirming extensive intracranial mucormycosis.[8] In addition, radiological findings in the early stages typically show only mucosal thickening, while bone erosions are usually evident in more advanced disease.[9]

Laboratory diagnostic methods include histopathological examination of tissue biopsies and mycological culture methods. Mucorales appear as broad aseptate/pauci-septate ribbon-like hyphae with frequent right-angle branching. Direct potassium hydroxide smears of tissue specimens are helpful for early presumptive diagnosis.

Antifungal therapy, surgical debridement, and effective control of underlying risk factors are the cornerstones of management.[10] Early diagnosis and timely initiation of antifungal treatment are critical for achieving favourable outcomes. Liposomal amphotericin B is recommended as first-line therapy at a dose of 5–10 mg/kg/day. At the same time, posaconazole and isavuconazole are alternative agents, though neither is currently available in Sri Lanka. In this patient, initiating liposomal amphotericin B at 5 mg/kg/day would have been preferable, as higher doses have been associated with improved responses. Prompt surgical debridement aids disease control and provides specimens for histopathological and microbiological diagnosis, while effective management of underlying risk factors, with rigorous glycaemic control optimise overall treatment success.

Conclusion

Mucormycosis is a potentially life-threatening invasive fungal infection that typically affects immunocompromised patients. Its occurrence following dental extraction is relatively rare. In this case, facial numbness and blistering were uncommon but significant early indicators that guided

the diagnosis. Maintaining a high index of suspicion and recognising atypical clinical signs are crucial for early intervention and improving the chances of a favourable outcome.

Declaration

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Author contributions: All authors read the content and agreed

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