CASE REPORT

Unravelling invasive fungus: A case report of rhino-orbital mucormycosis in a patient with diabetic ketoacidosis

Muhammad Syazani I[1,2], Nazrina H[1,3], Lizawati RH[4], Julieana M[2]

[1]Department of Ophthalmology Hospital Tuanku Ja'afar Seremban, [2]Department of Ophthalmology and Visual Science, School of Medical Sciences, Health Campus Universiti Sains Malaysia, [3]Department of Ophthalmology, Faculty of Medicine and Health Science, Universiti Malaysia Sabah, [4]Department of Pathology Hospital Tuanku Ja'afar Seremban

SUMMARY

Rhino-orbital mucormycosis is a rare but life-threatening invasive fungal infection, commonly associated with poorly controlled diabetes mellitus, particularly diabetic ketoacidosis. A case of a 52-year-old woman is reported with poorly controlled type 2 diabetes who presented with bilateral periorbital swelling and chemosis, progressing to right panophthalmitis. Computed tomography demonstrated bilateral sinusitis with orbital involvement, while fungal culture and histopathology confirmed angioinvasive mucormycosis. The patient was treated with systemic liposomal amphotericin B, local and retrobulbar amphotericin B, right eye evisceration, bilateral functional endoscopic sinus surgery, and repeated surgical debridement. Radical exenteration and maxillectomy were considered during multidisciplinary discussion. However, the family declined further extensive surgery due to concerns regarding morbidity and facial disfigurement. MRI was not feasible because the patient was critically ill with prolonged intubation and difficult intravenous access. Therefore, serial CT imaging was used to monitor disease progression and treatment response. Despite the inability to achieve complete radical surgical clearance, the patient showed favourable recovery with preservation of left eye vision following aggressive multimodal therapy. This case highlights the importance of prompt antifungal therapy, multidisciplinary management, and the potential role of globe-sparing approaches in selected patients.

INTRODUCTION

Mucormycosis is a highly invasive and potentially fatal infection caused by Mucorales fungi, for example Rhizopus sp. and Mucor sp.[1] and commonly presented as rhino-orbital mucormycosis.[1] It carries a very high mortality rate, ranging from 50–100%.[1] Initial symptoms and signs can be non-specific and may delay diagnosis, leading to devastating consequences. Major risk factors include poorly controlled diabetes mellitus (particularly in the context of diabetic ketoacidosis), haematological malignancies, immunodeficiency, and use of immunosuppressive agents.[2] Orbital symptoms include chemosis, ophthalmoplegia, proptosis, drop in vision as a result of orbital involvement from paranasal sinus.[2,3]

CASE PRESENTATION

A 52-year-old woman with poorly controlled type 2 diabetes mellitus (HbA1c 17.6% at admission) presented with a 2-day history of bilateral periorbital swelling and chemosis, progressing to right eye panophthalmitis. Fever and cough preceded the onset of periorbital swelling. Computed tomography of the brain and orbit showed mucosal thickening with rim enhancing lesion in the bilateral maxillary sinuses, with collections extending to the bilateral infraorbital foramen, bilateral thickened rectus muscles, irregular right globe with thickened optic nerve.

The patient was co-managed with the otorhinolaryngology team; rhinoscopy demonstrated crusted, unhealthy mucosa and eschar in both nasal cavities. Fungal culture and sensitivity of the sample demonstrated hyphae consistent with Mucor. She was then treated with intravenous amphotericin B 45mg OD for 3 days and then changed to liposomal amphotericin B 225mg OD for a total duration of 1 month, as per the infectious disease team. The patient subsequently underwent right eye evisceration and bilateral functional endoscopic sinus surgery (FESS) for extensive fungal infection involving the sinuses with extension into the right orbit.Post-operative histopathology confirmed angioinvasive fungal sinusitis. Postoperatively, daily debridement and desloughing were performed owing to persistent purulent discharge and slough tissue at the eviscerated socket. As she showed no signs of improvement, she underwent bilateral orbit exploration and removal of eschar tissue.

Following a multidisciplinary team meeting involving oculoplastics, otorhinolaryngology, and the infectious disease team, bilateral maxillectomy and exenteration were planned to achieve total eradication of mucormycosis. However, the family members opted for palliative management owing to concerns regarding high mortality risk and facial disfigurement. The patient was then discharged from the ward after completing intravenous amphotericin B. The patient was reviewed in clinic every two weeks, with gradual improvement in general condition.

At the third month post-discharge review, the right orbit demonstrated healthy granulation tissue with no pus or slough. No evidence of mucormycosis extension into the left eye was identified, with preserved visual acuity of 6/9 on the Snellen chart.

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Corresponding Author: Muhammad Syazani Ibrahim
Email: syazani101@gmail.com

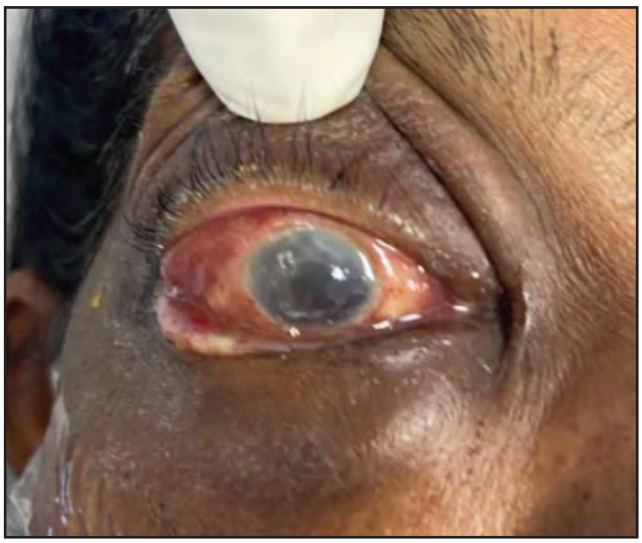

Fig. 1: Clinical photograph demonstrating right eye complete ophthalmoplegia with marked chemosis and corneal melting. B-scan ultrasonography revealed scleral thickening with vitreous opacities

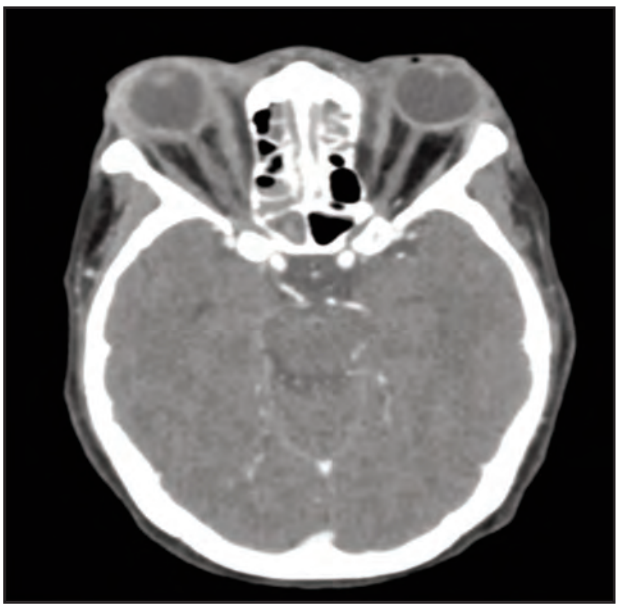

Fig. 2: Computed tomography brain and orbit showed an irregular right eye globe with enhancement, swollen recti muscles, and a thickened optic nerve with intraconal fat streakiness left extraconal collection extended into intraconal region

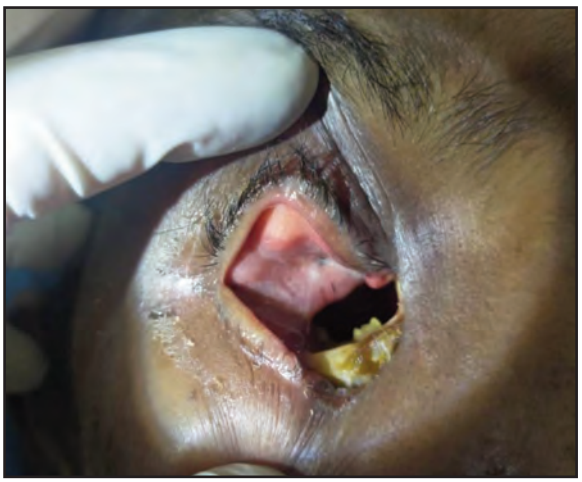

Fig. 3: Patient condition after 3 months post discharge from ward with development healthy granulation tissue and no remnant of necrotic tissues of orbit

DISCUSSION

Diabetic ketoacidosis precipitates the progression of fungal sinusitis in poorly controlled diabetes mellitus, leading to rhino-orbital mucormycosis — diabetes mellitus being the most common predisposing factor.[3] An improved prognosis can be achieved through early diagnosis and prompt initiation of treatment.

Orbital symptoms include ophthalmoplegia (approximately 67%), visual impairment (65%), proptosis (64%), periorbital oedema (43%), and periorbital pain. In this case, arriving at a correct diagnosis presented a challenge, as the only symptoms that could be elicited during the first encouter patient was bilateral periorbital oedema, given that the patient was intubated and sedated for diabetic ketoacidosis.[3]

A significant difference in survival rates exists depending on the timing of treatment initiation. Initiating therapy within 6 days can lead to survival rates of 76-81%.[4] Conversely, delays of more than 12 days may drastically reduce survival rates to 36–42%.[4] In this case, initiation of treatment was delayed in view of delay in arriving at the diagnosis and difficulty in intravenous access that eventually required insertion of peripheral inserted central catheter.

Amphotericin B improves the survival rate by up to 60% in mucormycosis patients, and it has been the gold standard treatment since then.[5] In this case, liposomal amphotericin B was chosen because it has better blood-brain barrier penetration and a safer renal profile.[5-6]

Liposomal amphotericin B was selected for its superior tissue penetration and more favourable renal risk profile compared with conventional amphotericin B. Amphotericin B is a rapid fungicidal treatment, especially in the acute phase of illness.[7] Alternative agents such as posaconazole and isavuconazole are generally reserved as salvage therapy or for patients who are intolerant of amphotericin B.[8]

Clear-margin resection of all infected tissue reduced mortality by up to 49%.[7] and increased it up to 71%[9] with combination of liposomal amphotericin B. Mucormycosis causes angio-invasion, thus leading to vascular obstruction and inadequate penetration of systemic antifungal therapy. To enhance tissue penetration, local irrigation with amphotericin B was performed.[7] The patient underwent several operative procedures to remove infected and necrotic tissues but was unable to achieve adequate removal in view of patient refused exenteration of left eye radical clear margin resection of the face. Local irrigation and retrobulbar with amphotericin B were also performed during dressing of the wound.

Evisceration was chosen because infection was confined to right globe, and no definitive evidence of extensive orbital tissue involvement or intracranial involvement, with good visual acuity in the left eye. Despite MRI providing better delineation of soft tissue involvement, the lesion extent, the patient was critically ill with difficult intravenous access and prolonged intubation, limiting the feasibility of MRI. CT imaging provides more rapid assessment, facilitates earlier medical and surgical intervention. It more feasible to be performed to monitor the progression and treatment response.

Despite not undergoing exenteration, the patient survived the infection, attributable to the combination of systemic and local amphotericin B together with multiple surgical debridements of necrotic tissue. This suggests that, in carefully selected cases, aggressive multimodal therapy without exenteration may achieve infection control whilst potentially preserving a better quality of life.

Given significant morbidity associated with repeated surgical debridement, other successful treatment approaches include hyperbaric oxygen therapy in combination with systemic and local amphotericin B including retrobulbar amphotericin B administration.[10]

Rhino-orbital mucormycosis may present with subtle and non-specific findings, which can lead to diagnostic delay. A high index of suspicion for fungal infection should be maintained in patients presenting with diabetic ketoacidosis and chemosis. Primary infection of orbital mucormycosis may be detected by early, thorough nasal endoscopy with suspicious eye symptoms and signs. A favourable outcome is achievable with early diagnosis, intravenous antifungal therapy, and clear resection margins. Rhino-orbital mucormycosis is a life-threatening orbital infection; the possibility of exenteration should be considered to prevent intracranial spread. Continuous multidisciplinary team co-management and in-depth counselling of family members regarding the high mortality rate are essential in the management of this condition.

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