

Case Report

A report of successfully treated rhino-orbital mucormycosis

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ABSTRACT

Rhino-cerebral mucormycosis is a rare life-threatening infection caused by fungi from the order Mucorales. The disease occurs mostly in association with diabetic ketoacidosis. Because of its rapid progression and high mortality, early recognition and aggressive treatment offer the only chance to increase the survival rate. We report a case of invasive mucormycosis in a 55 year old diabetic male, who presented with diabetic ketoacidosis and sinusitis. The patient came with complaints of swelling in right side of face, pain in right eye, right orbital swelling and numbness right cheek. An ENT examination revealed right facial swelling and proptosis. Nasal swabs were sent to microbiology for fungal culture. Material from the swab was inoculated on Sabaraud's dextrose agar and incubated at 37°C and 25°C. The culture was identified as mucor species. Material sent for histopathology showed presence of chronic inflammatory polyp with broad aseptate hyphae suggestive of mucormycosis. On the third day, bilateral middle meatal antrostomy, bilateral anterior and posterior ethmoidectomy and bilateral wide sphenoidotomy was done. Fungal debris were noticed in both maxillary antrum. In the post-operative period, patient was started on inj. amphotericin B. On the 11th post-operative day, patient developed palatal mucosal necrosis. Under general anaesthesia, extensive surgical 2nd look and debridement was done. The necrotic palatal mucosa was completely removed. In addition to IV amphotericin B, topical douching of both nasal cavities and antrum with amphotericin B was done for 20 days. Patient improved, became completely asymptomatic and was discharged.

Keywords: Rhino-orbital mucormycosis, Diabetic ketoacidosis, Intranasal amphotericin B, Orbital decompression, Proptosis

INTRODUCTION

Mucormycosis is an acute, fatal fungal infection in man. The disease causes a characteristic pattern of clinical signs and symptoms, prompt recognition of which is necessary for the immediate institution of antifungal therapy.¹ Rhinocerebral mucormycosis is an invasive fungal sinusitis with a high mortality rate, especially in immunocompromised patients.² After aspergillosis and candidiasis, mucormycosis is considered to be the third most common invasive fungal infection.³ Opportunistic fungal infections are usually associated with diabetic ketoacidosis, lymphoma, leukaemia, corticosteroid

treatment, radiation therapy, chemotherapy, myelodysplastic syndrome, aplastic anaemia and acquired immunodeficiency syndrome.⁴

CASE REPORT

A 55 year old male was referred from a private hospital with complaints of swelling in right side of face, pain in right eye, right orbital swelling and numbness right cheek. He was a known diabetic for the past ten years on regular insulin and oral hypoglycemic agents. He was not a smoker or alcoholic. On examination, he was conscious, oriented, with right facial swelling (Figure 1),

right proptosis (Figure 2) and anaesthesia over right side V2 (maxillary division of trigeminal nerve). He was provisionally diagnosed with orbital cellulitis and fungal sinusitis.



Figure 1: Patient at the time of surgery with right cheek swelling.



Figure 2: Patient at the time of surgery with right proptosis.

An ENT examination revealed right facial swelling and proptosis. Nasal endoscopy performed revealed bilateral mucosal edema with pale mucosa. On the third day, bilateral middle meatal antrostomy, bilateral anterior and posterior ethmoidectomy and bilateral wide sphenoidotomy was done. Right sided orbital decompression was also done. The findings showed a pale, edematous mucosa, right lamina propria was eroded and the orbital periosteum was intact. The left side showed similar mucosal changes. Fungal debris were noticed in both maxillary antrum.

Nasal swabs were sent to microbiology for fungal culture. KOH wet mount showed presence of broad, aseptate hyphae. Material from the swab was inoculated on Sabaraud's dextrose agar and incubated at 37°C and 25°C. The colonies were rapidly growing, dull white in colour with abundant aerial and substrate mycelia. Slide culture was done and LPCB from the culture showed: Broad, aseptate hyphae with branched sporangiophores, bearing spherical sporangia at the tip of each branch. The hyphae were without rhizoids. The culture was identified

as mucor species. Material sent for histopathology showed presence of chronic inflammatory polyp with broad aseptate hyphae suggestive of mucormycosis.

In the post-operative period, patient was started on inj. amphotericin B and anti-diabetic measures with ICU admission for monitoring of electrolytes and correction of DKA.

On the 11th post-operative day, patient developed palatal mucosal necrosis. A swab taken from that site revealed fungal hyphae. On day 14, under general anaesthesia, extensive surgical 2nd look and debridement was done. The necrotic palatal mucosa was completely removed. Fungal cottony growth with necrotic mucosa was observed in the right maxillary antrum (Figure 3). Fungal cultures sent again revealed mucor species.

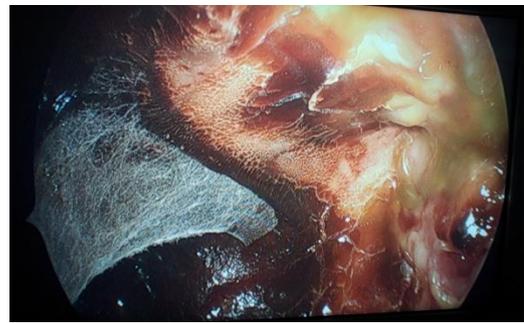


Figure 3: Endoscopic view inside R. maxillary sinus showing necrosis and fungal growth at the time of revision surgery.

In addition to IV amphotericin B, topical douching of both nasal cavities and antrum with amphotericin B was done for 20 days. Amphotericin B soaked nasal pack was kept in the right maxillary antrum and removed after 24 hours, alternating with the topical douching. Total dose of Amphotericin B given to the patient was 2.3gm.

Patient improved, became completely asymptomatic and was discharged on the 23rd day of admission. Swab sent for fungal culture was negative. Follow up was done after 3 months when patient was clinically normal and fungal culture was negative.

DISCUSSION

Rhino-cerebral mucormycosis is an invasive fungal sinusitis with a high mortality rate, especially in immunocompromised patients.² Rhino-orbital-cerebral mucormycosis (ROCM) is an acute and aggressive fungal infection in which diabetes has been recognized as the most common (60%-81%) predisposing factor.⁵ Mucorales, normally saprophytic, can colonize the sinuses and then the orbit, followed by the meninges.⁶ The initial site of infection in ROCM is the nasal mucosa, from where it extends towards the orbits and cranial cavity with or without bony erosion of the skull base.⁴

The conventional management of rhino-orbito-cerebral mucormycosis includes control of metabolic abnormality, administration of amphotericin B and surgery that spans simple sinus clearance, radical debridement and orbital exenteration. Debridement of the sinuses is almost always necessary in all cases of rhino-orbito-cerebral mucormycosis.⁷

When evidence points to a fungal etiology, there has been a trend towards treatment of chronic rhinosinusitis with topical antifungal medication, namely amphotericin B (AmB). AmB is a natural polyene antifungal that is not absorbed through the gastrointestinal tract. AmB binds to ergosterol, a component of cell walls of most fungi, leading to formation of ion channels and cell death; AmB may also act secondarily through oxidative damage to fungal cell membranes through creation of free radicals from its own oxidation.⁸ It is hypothesized that topical intranasal application of AmB can decrease the fungal load in the sino-nasal region, thereby decreasing the local eosinophilic inflammatory reaction to fungal antigens.⁹ In a study of 74 patients with persistent nasal polyposis, along with saline lavage and corticosteroid spray, addition of intranasal AmB was associated with complete disappearance of polyps in 39% of patients.¹⁰ Therefore the combination of intra-nasal and IV amphotericin B along with aggressive surgical debridement could be a better option for treatment of invasive mucormycosis.

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REFERENCES

1. Ferry AP, Abedi S. Diagnosis and management of rhino-orbitocerebral mucormycosis (phycomycosis). A report of 16 personally observed cases. *Ophthalmol.* 1983;90(9):1096-1104.
2. Hadzri MH, Azarisman SM, Fauzi AR, Kahairi A. Invasive rhino-cerebral mucormycosis with orbital extension in poorly-controlled diabetes. *Singapore Med J.* 2009 Mar;50(3):e107-9.
3. Prabhu RM, Patel R. Mucormycosis and entomophthoromycosis: a review of the clinical manifestations, diagnosis and treatment. *Clin Microbiol Infect.* 2004;10(Suppl. 1):31-47.
4. Haliloglu NU, Yesilirmak Z, Erden A, Erden I. Rhino-orbito-cerebral mucormycosis: report of two cases and review of the literature. *Dentomaxill Radiol.* 2008;37:161-6.
5. Bhansali A, Bhadada S, Sharma A, Suresh V, Gupta A, Singh P, Chakarbarti A, Dash RJ. Presentation and outcome of rhino-orbital-cerebral mucormycosis in patients with diabetes. *Postgrad Med J.* 2004;80:670-4.
6. Orsel S, Bessède JP, Sauvage JP. Naso-orbito-cerebral mucormycosis. A more & more common disease. *Rev Laryngol Otol Rhinol (Bord).* 1990;111(3):221-5.
7. Nithyanandam S, Jacob MS, Battu RR, Thomas RK, Correa MA, D'Souza O. Rhino-orbito-cerebral Mucormycosis. A retrospective analysis of clinical features and treatment outcomes. *Indian J Ophthalmol.* 2003;51:231-6.
8. Groll AH, Gea-Banacloche JC, Glasmacher A, Just-Nuebling G, Maschmeyer G, Walsh TJ. Clinical pharmacology of antifungal compounds. *Infect Dis Clin North Am.* 2003;17(1):159-91.
9. Ponikau JU, Sherris DA, Weaver A, Kita H. Treatment of chronic rhinosinusitis with intranasal amphotericin B: a randomized, placebo-controlled, double-blind pilot trial. *J Allergy Clin Immunol.* 2005;115(1):125-31.
10. Ricchetti A, Landis BN, Maffioli A, Giger R, Zeng C, Lacroix JS. Effect of anti-fungal nasal lavage with amphotericin B on nasal polyposis. *J Laryngol Otol.* 2002;116(4):261-3.

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