Sinonasal Mucormycosis: A Cryptic Paradox

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Abstract

Mucormycosis is one of the sinister variant of fungal rhinosinusitis. An immunocompromised case with mucormycosis requires prompt diagnosis, in order to prevent a fatal outcome. Though the primary cause lies in the nose and sinus, due to lack of awareness, patients usually visits a physician, an ophthalmologist and may be then the otorhinolaryngologist, during this period, patient would have progressed to an advanced stage missing the early intervention. This case report of 70 year old male with mucormycosis of left side of the nose and sinus with paradoxical involvement of right eye and has been successfully treated with repeated endoscopic clearance of the crusts and diabetic control. Since the disease has a stormy course and rapid progression, an early diagnosis and aggressive management is imperative and all the more so when the presentation is paradoxically elusive to avoid complications.

Keywords: Mucormycosis; Amphotericin; Fungal

Introduction

Sinusitis or more accurately rhinosinusitis is a common disorder, affecting approximately there are 20% of the population at some time of their lives. 400,000 known fungal species of which 400 are human pathogens and 50 of which cause systemic or CNS infection. Once a rare disorder, is now being identified and reported with increasing frequency over the last two decades. Increased diagnostic methods enables us for more frequent recognition. It carries a high residual morbidity and mortality due to the angioinvasive property of fungi, causing vascular occlusion and extensive tissue

necrosis.³ Functional endoscopic sinus surgery with repeated suction clearance still remains the main stay of treatment with intravenous antifungal Amphoterecin.

Case Report

We present a 70 year old male, reporting to physician for generalized weakness and was incidentally diagnosed with diabetes mellitus. He was referred to ophthalmologist for right eye swelling and reduced vision since 10 days and was subsequently referred to us for Left sided nose block since 10 days and headache since 8 days. He had associated symptoms of foul smelling discharge.

Nose examination: Revealed thick crusts with foul smelling discharge filling both the nasal cavities.

Diagnostic Nasal Endoscopy: Thick crusts with tenacious, foul smelling discharge in the left nasal cavity, left maxillary and sphenoid sinus and with bony septal perforation.

Right nasal cavity, maxillary and sphenoid sinus normal.

Ophthalmic examination: On the contrary to the presence of disease on left side of nose and paranasal sinuses, in this case on examination, Right eye showed mild proptosis, chemosis, extraocular movement (right lateral) was restricted, with right eye reduced vision. Left eye Normal.

CT Scan: PNS plain with contrast: Showed crusts filling the entire Left nasal cavity, osteomeatal complex and involving entire Maxillary, Ethmoid and Sphenoid sinus with no breach in the lamina papyracea on the left side. Right nasal cavity and sinus appeared normal but there was soft tissue edema around the right orbit with no breach in the lamina papyracea.[Fig. 1]

Management

Under general anesthesia. 0 degree endoscope was used to visualize bilateral nasal cavities



Fig. 1: A,B,C: Preoperative CT scan PNS images of Axial, Coronal and Sagittal views respectively, it shows thick tenacious discharge filling the left nasal cavity, maxillary, ethmoid and sphenoid sinus with no breach in the lamina papyracea. Right nasal cavity- Clear



D,E,F: Postoperative CT scan PNS images of Axial, Coronal and Sagittal views respectively images with bilateral clear nasal cavity and sinus.

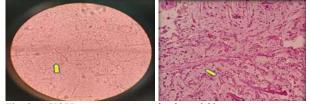


Fig. 3: A-KOH mount - positive for fungal filaments.

B-HISTOPATHOLOGY: mucosa showed edema, congestion and diffuse inflammation with necrotic material showing numerous hyphae branching at right angles suggestive of mucormycosis.

A black eschar was seen involving the left osteomeatal complex, left maxillary sinus and left anterior ethmoidal cells and sphenoid sinus. Underlying bone was necrosed.

Hence, the diseased bone and the unhealthy mucosa were excised, the specimen was sent for KOH and histopathological examination.

He underwent repeated DNE and suction clearance of the crusts formed. His post op repeat CT scan showed clear left nasal cavity left maxillary sphenoid and ethmoid sinus as compared to pre op imaging [Fig. 2]

KOH: Positive for fungal filaments.[Fig. 3]

Histopathology: Mucosa showed edema, congestion and diffuse inflammation with necrotic material showing numerous hypha branching at right angles suggestive of mucormycosis. [Figure 3]

He was treated with IV antifungal of liposomal amphotericin B and fluconazole. Also, nasal douching with fluconazole twice a day.

His diabetes mellitus was managed with insulin.

He was asymptomatic following 2 months and 6 months follow up. His eye swelling was completely resolved [Fig. 4].



A: Shows thick white tenacious discharge filling the entire left maxillary sinus

B: Shows white discharge in the sphenoid sinus with erosion of inter sphenoidal septum

C: Bony septal perforation

D,E: Shows postoperative clear maxillary and sphenoid sinus respectively.



Fig. 4: A: Preoperative right eye swelling.
B: Postoperative reduced eye swelling at 2 month follow up.

Discussion

Fungal infections of the NOSE and PNS have been recognized since the beginning of this century. Rhino/sino-orbital mycosis- most often caused by the saprophytic moulds - aspergillus and Mucorales^{6,7,9-12}. Found worldwide in a variety of habitats in soil, on decaying vegetation, in the air, and in water supplies. Their thermotolerance permits a wide range of suitable host conditions. 4,5,8,9. They are encountered more frequently in patients with immunocompromised status.4 They usually present with unilateral or bilateral nasal cavity involvement with ipsilateral or bilateral orbital involvement respectively, with or without intracranial involvement. Unlike in this case where the patient has presented paradoxically with contralateral eye involvement and ipsilateral eye being normal.

Amphotericin B remains the only antifungal agent approved for the treatment of this infection.⁴ The lipid formulations of amphotericin B are significantly less nephrotoxic than amphotericin B deoxycholate and can be administered at higher doses for a longer period of time. Several case reports of patients with mucormycosis documented successful treatment with up to 15 mg/kg/day of a lipid formulation of amphotericin B.

Concurrent therapy: Azoles and Amphotericin-B better than monotherapy.

Though the primary cause lies in the nose and sinus, due to lack of awareness, patients usually visits a physician, an ophthalmologist and may be then the otorhinolaryngologist, during this period, patient would have progressed to an advanced stage missing the early intervention.

Conclusion

An immunocompromised case with mucormycosis requires prompt diagnosis, in order to prevent a fatal morbidity. Delay in the treatment and risk of associated intracranial and intraorbital complications are major determinants of the

survival outcome in these patients.^{5,6} In these cases along with the ipsilateral eye examination, *it is important to examine the contralateral eye* also for the signs of endogenous spread or systemic spread of the disease, while monitoring it closely for progression, 7. It also needs to be emphasized that, high clinical suspicion from physician, otolaryngologist and ophthalmologist is a must, so that, the patient receives early intervention to avoid the dreaded complications.

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