Case Report

Post COVID Rhino Orbital Mucormycosis with Pulmonary Mucormycosis : A rare case report

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Mucormycosis is an acute and fulminant fungal infection caused by fungi of family Mucoraceae, seen usually among immunocompromised or decompensated diabetic patients. Post COVID upsurge of Mucormycosis have been a well-documented entity witnessed in last year affecting several states of India after second wave in form of an epidemic. Disseminated form which indicates involvement of two or more Non-contagious Organ System is extremely rare and generally occurs in severely immunocompromised patients with disseminated Mucormycosis, often discovered in autopsy. Pulmonary and Rhinoorbital forms in a same patient without systemic dissemination is rarely reported.

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Mucormycosis is an acute and fulminant fungal infection caused by fungi of family Mucoraceae, seen usually among immunocompromised or decompensated diabetic patients[1]. Post COVID upsurge of mucormycosis have been a well-documented entity witnessed in last year affecting several states of India after second wave in form of an epidemic.

Mucormycosis may manifests in six different forms: Rhino-Orbito-Cerebral (ROCM), Pulmonary (PM), Cutaneous, Gastrointestinal, CNS or others[5]. ROCM is mostly associated with Uncontrolled Diabetes Mellitus (DM). Pulmonary Mucormycosis (PM) is more commonly associated with hematological malignancies rather than DM. Disseminated form which indicates involvement of two or more non-contagious organ system is extremely rare and generally occurs in severely immuno-compromised patients with disseminated mucormycosis, often discovered in autopsy[7]. PM and ROM in a same patient without systemic dissemination is rarely reported.

Case Report

An 18-year-old male was referred to ENT emergency of our Tertiary Care Facility on 08/08/21 with complaints of swelling, redness and dimness of vision of left eye for the last 6 days with Non-productive cough & Irregular fever for the last 20 days (Fig 1). He had no record of COVID positivity in recent past but suffered from COVID like symptoms. There was no other comorbidity except newly developed Diabetes Mellitus (DM).

Editor's Comment :

- Early diagnosis and adequate early treatment leads to better prognosis.
- Judicious use of steroids.
- Screening should be done for diabetic immunocompromised patients with non resolving dyspnoea and ROCM cases with Chest CT and bronchoscopic KOH smear keeping pulmonary mucor as differential diagnoses.
- Imaging, KOH Smear and biopsy proven disease can be treated with aggressive adequate surgical debridement, which prevents disease progression and eslablishes vascularisation for better penetration of anti-fungal medications, which in turn improves prognosis and survival chances.

Patient was alert, conscious and cooperative on admission. Initial laboratory studies showed mild Leukocytosis with low Hemoglobin. Ophthalmic examination showed Proptosis of left eye, drooping of upper eyelid, Chemosis and Edema with loss of Perception of Light (PL). During hospitalization on second day patient developed Dyspnea and productive Cough. Physical examination revealed tachypnea and coarse basal crepitations in both Lungs along with falling Oxygen saturation near about 89%. Sputum for AFB, HIV antibody test and RT-PCR for COVID-19 were negative.

Chest X-ray showed multiple rings like opacities in both the Lungs (Fig 2). HRCT thorax revealed multiple large thick walled cavitary lesions in both Lungs and septae like structures within. Multifocal ground glass opacities were noted in Right Upper Lobe and basal segment of Left Lower Lobe (Fig 3). USG abdomen was normal.

Diagnostic Nasal Endoscopy (DNE) showed black crusts on left middle turbinate. Sample taken for KOH smear, culture and biopsy. Bronchoscopic suction done and lavage material sent for KOH smear and culture. Both the specimens showed broad aseptate hyphae.

MRI of Nose, Paranasal sinuses, Orbit and Brain were done in sequences of T1, T1 with contrast and T2 with

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Fig 1 — Patient at presentation with nasal discharge and facial cellulitis with orbital cellulitis

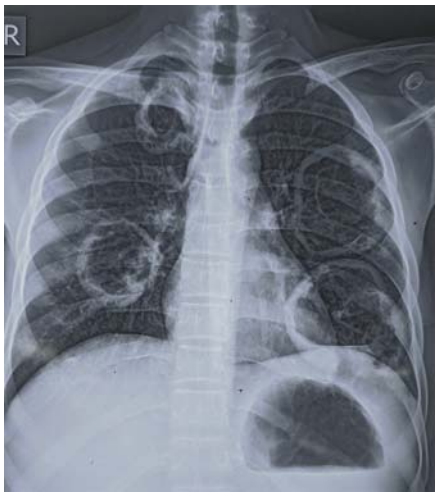

Fig 2 — Chest X-ray showing multiple ring like opacities in both the lungs

FSE, DWI and GRE. The MRI findings are suggestive of – Heterogenous areas with mucosal thickenings involving ethmoids, spheroids, left maxillary antrum and adjacent left frontal sinus. Postcontrast study showed marked heterogenous enhancement. Such heterogenous areas were also seen involving retro-ocular portion of left orbit and adjacent peri orbital region with enhancement in post contrast study. Left ocular bulb showed loss of morphology with extra ocular muscles involvement. Left Optic Nerve also had signal changes. Enhancement of para-seller carotid more on left side was also seen on MRI.

Management was done as per Institutional mucormycosis guideline. On clinical suspicion for Post COVID Mucormycosis, patient was started on Amphotericin B (liposomal) 1mg/kg body weight from day 2 of admission awaiting histopathological confirmation. Adequate antibiotic & anti-inflammatory coverage along with medical therapy was also initiated to stabilize underlying metabolic derangement.

After diagnosis was confirmed, surgical management in form of endoscopic endonasal wide local debridement of all sinuses, pterygopalatine fossa and drilling of pterygoid wedge along with eyelids sparing exenteration of left eye done under General Anesthesia on 4th day of admission (Fig 4). Postoperative period was uneventful. Postoperative histological examination showed inflammatory tissue invaded with broad aseptate hyphae with irregular branching pattern at right angles suggesting Mucormycosis.

Regular endoscopic suction clearance, removal of crusts and amphotericin washes were given to the Postoperative cavity. After 25 days of injection Amphotericin B, he was started on oral Posaconazole 300 mg once daily. Patient was discharged on day 29 on Posaconazole. At the time of discharge patient was hemodynamically stable, CRP level was 20 mg/l and blood sugar were well controlled with insulin therapy. On follow up after 2 weeks eyelids stitches were removed. Postoperative cavity of sinonasal & orbital areas was well epithelized and Chest X-ray became normal. During successive follow up for 8 months no recurrence was seen. Patient is still under our supervision with oral Posaconazole.

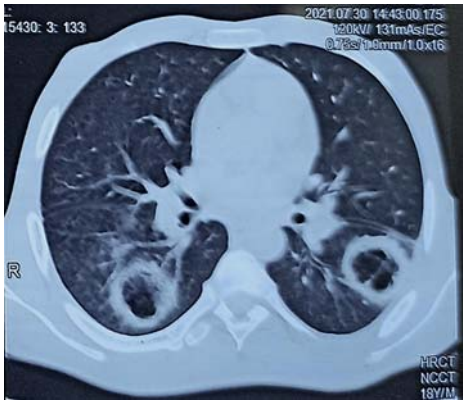

Fig 3 — HRCT thorax revealing multiple large thick walled cavitary lesions in both lungs and septae

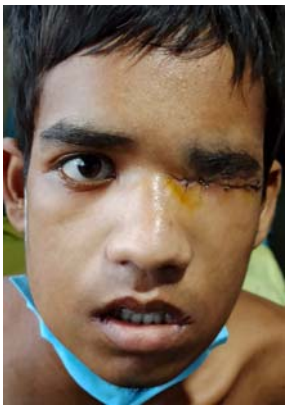

Fig 4 — Postoperative photograph prior stitch removal where patient has undergone endoscopic endonasal wide local debridement of nasal component of mucor along with orbital exenteration

Discussion

Mucormycosis is an opportunistic infection caused by Mucorales. Such fungal infection mainly occurs in patients with immune system deficiency, though it can rarely attack immunocompetent patients. Such a massive involvement in an immuno-competent patient at such a younger age is an extremely rare entity, which is possibly due to post COVID immunosuppression. Still the patient probably survived due to timely interventions and more importantly due to well-maintained immune status.

Rhino-orbital Mucormycosis begins with nonspecific nasal complains like Rhinorrhea, Nasal blockage, Headache, Deep seated retroorbital pain, which rarely alarms clinicians. It's generally the visual compromise, facial cellulitis or check hypoesthesia which warrants the Otorhinolaryngologist for a Nasal

Endoscopy or MRI and possibly a biopsy demonstrating mucorales for diagnosis. Similarly, non-specific symptoms such as Fever, Cough, Dyspnoea, Haemoptysis and Chest pain are presenting features of Pulmonary Mucormycosis. The definitive diagnosis of Pulmonary Mucormycosis also comes from identification of typical hyphae through bronchoalveolar lavage culture, needle biopsy and resected lung tissue biopsy. Variable presentations make it difficult to distinguish Rhino-orbital and Pulmonary Mucormycosis from other sino-nasal or pulmonary pathologies. Mucormycosis should be suspected in patients with normal immune function with fulminant progression, especially when routine anti-infective treatments fail. Invasive procedures in decompensated patients are always dilemmas. Culture takes longer time and culture positivity rates are near about one-third. Such limitations delays diagnosis and systemic antifungal therapy, which worsen prognosis and increase the risk of death.

The Post COVID mucormycosis is still an enigma. The virus spreads from the Nasopharynx to the Lungs or inhaled directly to both separately, where it can cause intense inflammatory response with alveolar edema and may result in the Acute Respiratory Distress Syndrome and to other tissues that expresses the ACE- 2 receptor, including the blood vessels[2]. The locoregional spread pathway of ROCM has not been adequately described. Pterygopalatine fossa acts as a reservoir of the disease through which it can spread to the neighboring structures including the retro-global space of the orbit and Infratemporal Fossa (ITF)[3]. Mucor causes angioinvasion and its ability to cause tissue necrosis and dissemination through bloodstream is well established[4]. Dissemination most commonly affects the Brain (ROCM), but can affect any other organ like Lungs (PM), Spleen, Heart and Skin.

Diagnostic nasal endoscopy shows characteristic eschars, discharge or necrotic mucosa which directs surgeon to collect biopsy and conduct KOH smear. MRI of Nose, Paranasal sinuses & Orbit shows devitalized and involved tissues for surgical planning. The patient developed ptosis, ophthalploplegia and visual loss on left side. Loss of vision probably due to cavernous sinus involvement. Cavernous Sinus Thrombosis usually results from spread of infection from orbital apex or sphenoid sinus. A through Ophthalmic examination is required for deciding fate of the eye. Radiologic signs can suggest Pulmonary Mucormycosis in an appropriate clinical setting[8]. Halo sign on CT, a ring of ground glass opacity surrounding a nodular infiltrate which pathophysiologically represents a region of ischemia. Reverse Halo sign on CT (atoll sign) represents an area of ground glass opacity surrounded by a ring of consolidation are pathognomonic of Pulmonary Mucormycosis (Fig 5).

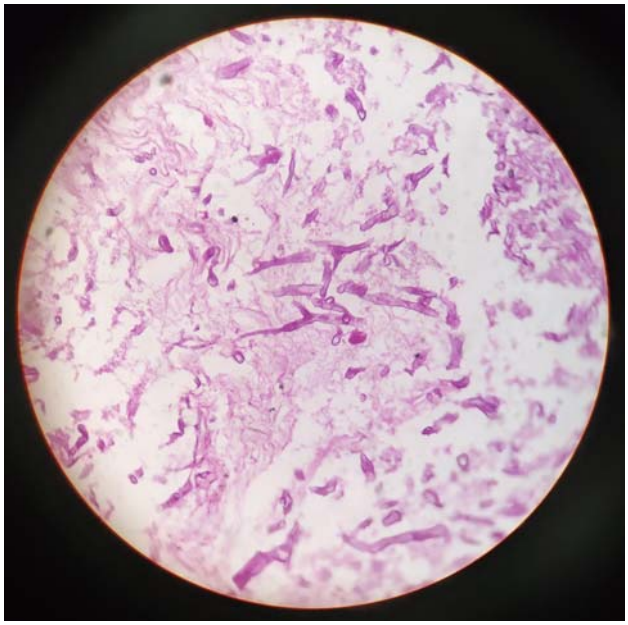

Fig 5 — KOH smeer from tissues obtained from endoscopic nasal biopsy

Conclusion

Key to successful outcome is early diagnosis and initiation of systemic antifungal in form of Amphotericin B based on clinical judgement. KOH smear has role in early diagnosis. Prompt and adequate surgical debridement of involved areas reduces disease load, slows down disease progression and establishes vascularization for penetration of anti-fungal which in turn results in better prognosis & survival chances.

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