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RHINO- ORBITO- CEREBRAL MUCORMYCOSIS IN A NON-COVID DIABETIC PATIENT: A CASE REPORT

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ABSTRACT Introduction: Muccomycosis is an aggressive fungal infection in immunocompromised individuals. It can rapidly lead to rhino cerebral ocular involvement with devastating consequences. With such grave involvement, the airway of the patient becomes compromised, and it becomes difficult to secure the airway for surgery. **Case report**: We report a case of 20-year-old diabetic, covid negative male patient who was operated for mucormycosis and subsequently presented for drainage of temporal abcess and debridement. Clinical examination of the patient predicted a difficult airway. It was managed with successful orotracheal intubation while keeping the patient spontaneously breathing. Tracheostomy was kept as plan B in case of CVCI situation. **Conclusion:** mucormycosis is a rapidly progressive disease which requires early and prompt treatment and timely surgical intervention to debride the necrotic tissue keeping in mind the associated comorbidities and presence of difficult airway.

KEYWORDS:

INTRODUCTION

Mucormycosis is a rapidly progressing and fulminant infection caused by members of family mucoraceae, order mucorales and class Zygomycetes.¹ Depending upon the immunological status of the patient and the site of the body that is affected, the disease may manifest as rhinocerebral, pulmonary, cutaneous, gastrointestinal, central nervous system or disseminated forms. Rhinocerebral mucormycosis is the most common type and its extension to the orbit and brain is quite usual making it life threatening.² Since mucormycosis may affect any organ system in an immunocompromised host, it may be accompanied by hemodynamic instability and difficult airway management. The fungal debris gets deposited in the oropharynx and may lead to supraglottic edema, which makes securing airway difficult.

The aim of this case report is to present a patient with rapidly invasive rhino orbital cerebral mucormycosis with a difficult airway and the importance of multidisciplinary management of such patients.

Case report

A 20-year-old male patient presented in the emergency department with left sided facial lesion, nasal obstruction, and decreased vision in the left eye. Five days prior, the patient had a tooth ache for which he underwent root canal therapy from a local practitioner without any complications. On presentation, he had hyperglycemia with blood sugar 530 mg/dl and ketosis and was immediately started on intravenous fluids and insulin infusion. Serum glycosylated hemoglobin was 17.7%.

Patient was not a known case of diabetes mellitus and denied any family history for the same. He had a history of fever of short duration but tested covid negative. Chest x ray was unremarkable and there was no history of steroid intake or oxygen inhalation in recent past. CT paranasal sinus was suggestive of extensive anaerobic cellulitis with multiple abscesses formation and left panophthalmitis with deformity of left ocular globe. An emergency functional endoscopic sinus surgery (FESS) with left maxillectomy, right sided uncinectomy and right sided anterior and posterior ethmoidectomy was done under general anesthesia. Histopathological examination of the biopsy sample confirmed mucormycosis.

After a week of absolute blood sugar control, Liposomal amphotericin b and antibiotics, repeat CT and MRI scan was done. CT paranasal sinus and orbit showed mucosal thickening in ethmoidal, maxillary, bilateral sphenoid sinus, and bilateral frontal sinus with internal CT no. of 40-50 hu in bilateral sphenoid sinuses. Multiple heterogeneously enhancing areas were seen in subcutaneous plane of face and neck and in parotid region and parapharyngeal space.

Rarefaction of right ethmoid air cell wall and cribriform plate was seen (Fig 1). MRI brain showed a large rounded focal lesion showing peripheral enhancement involving left temporal lobe measuring $3.1 \times 2.1 \times 2.3$ cm. Both sphenoid sinuses revealed heterogenous enhancement with extension into skull base. Unfortunately, fungus had also invaded the left cavernous sinus, however, both internal carotid arteries were normal.





An emergency multidisciplinary meet was done with the consulting neurosurgeon, otorhinologist and the anesthetist to decide upon the plan of management for the patient.

With such widespread involvement patient was planned for emergency drainage of temporal abcess and debridement. On clinical examination patient was conscious and oriented but poorly built and emaciated. Heart rate was 86 beats/min and blood pressure were 90/50mmHg. Patient was maintaining a saturation of 95% on room air. Chest was bilaterally clear and heart sounds were normal. Blood sugars were within normal range on regular insulin. Due to extensive debridement in the previous surgery, the oropharynx and nasopharynx were all exposed (Fig 2).



Fig.2

Mask ventilation was difficult in the patient due to absence of the supporting structures for the mask. Thus, it was decided to do an awake laryngoscopic guided intubation while keeping the patient spontaneously breathing as plan A and tracheostomy as plan B in a CVCI situation. After a covid 19 negative result from reverse transcriptase polymerase chain reaction (RT-PCR) test, patient was taken for surgery with a written informed high-risk consent because of the associated comorbidities and a difficult airway. In the operating room, a subclavian central line was inserted, and intravenous fluids started. Standard monitors including ECG, non-invasive blood pressure and pulse oximetry were applied. Injection glycopyrrolate 0.2mg and inj. fentanyl 100 micro gm was given intravenously as premedication. The oropharynx was sprayed with 10% lignocaine. Nebulization was done with 4 ml 4% lignocaine to anaesthetize the airway. Supplemental oxygen was delivered via a catheter continuously which was placed in the oropharynx. All the exposed area was covered with gauge pieces to make a seal for the mask. However, there was a lot of leaks despite the cover and mask ventilation was not a possibility. Inj Propofol 20 mg was given in increments to keep the patient arousable and spontaneously breathing.

Gentle laryngoscopy was done to assess Cormack Lehanes grade. It was found to be grade I. Upon seeing the glottis, lignocaine spray was done. We waited for 10 seconds and then the patient was intubated with endotracheal tube size 7.5mm. The tube was connected to the circuit and bilateral air entry checked. It was found to be adequate. Tube was fixed and inj. vecuronium 5mg was given. Intraoperative period was uneventful, and the patient was extubated on return of spontaneous respiratory efforts. Postoperatively, patient was shifted to intensive care unit for observation.

DISCUSSION

Mucoraceae are ubiquitous saprophytic fungi that can cause mucormycosis. It is an aggressive and rapidly invasive disease.⁴ Within a period of only five days our patient developed facial lesions, severe facial pain and decrease in vision. It was so fulminant that even after doing extensive debridement at first place and aggressive treatment with Liposomal amphotericin b and antibiotics, it invaded intracranially.

The spores of the fungi usually enter the host through inhalation. The healthy individuals effectively clear the fungi from their bodies by the innate immune system. However, people with risk factors like diabetes (particularly in patients having ketoacidosis), hematological malignancies, organ transplant, renal failure, use of steroids or immuno suppressive therapy, cirrhosis and acquired immuno deficiency syndrome (AIDS) are more prone to develop the fungal infection. Decreased phagocytic activity and increased available serum iron in diabetic ketoacidosis has been explained as a potent mechanism for increased fungal invasiveness in patients with diabetes.⁵ Our patient's infection was triggered by his tooth extraction which aggravated by virtue of his diabetes which was incidentally diagnosed after his arrival in emergency room. There have been many case reports of rhinocerebral mucormycosis occurring after dental extractions.

Rhino orbital cerebral mucormycosis is the most common type of mucormycosis. Initial signs of infection are fever, headache, facial pain and swelling. The fungal elements invade the blood vessels in the nasal structures and can extend to the surrounding structures. It can involve the paranasal sinuses and the orbit which may lead to loss of extraocular movements, proptosis, and vision loss.⁵ In our case, the patient presented with facial lesions, swelling and pain and decreased vision in left eye for which he underwent debridement. He was started on amphotericin b and antibiotics but when a repeat CT scan and MRI scan was done after a week, the fungi had invaded intracranially to involve temporal lobe, sphenoid sinus and cavernous sinus.

Treatment of rhinocerebral mucormycosis involves a multidisciplinary approach aimed at reversal of the underlying predisposing factor, initiation of antifungal therapy and surgical intervention when required. Our patient required extensive surgical debridement in the first time to contain the invasive disease. He was started on insulin therapy, systemic amphotericin b and antibiotics to halt the progression of the disease but it spread intracranially for which temporal abcess drainage was planned in the second surgical intervention.

In rhinocerebral mucormycosis, fungal debris may get deposited in the oropharynx obscuring the glottic view. In addition, vascular invasion by the fungal hyphae may lead to inflammation and edema of the supraglottic structures.³ Extensive debridement of the infected tissue of the face and airway leads to loss of supporting structures for holding the mask which makes it difficult to mask ventilate the patient as in our case. In such a scenario, planning for the procedure was important in collaboration with the otorhinolaryngologist, neurosurgeon and the consultant anaesthetist. We proceeded with awake intubation in spontaneously breathing patient with continuous oxygen insufflation via nasal catheter as plan A and tracheostomy was kept as plan B in CVCI situation. Oxygenation of the patient was not a problem in this patient as the oropharynx was open in view. So an oxygen catheter was placed in the oropharynx at all times despite a nearly impossible mask ventilation. A good topical anaesthesia was important in our case for awake intubation which was achieved with nebulisation with 2% lignocaine, lignocaine spray in the posterior pharyngeal wall and instillation of 2% lignocaine over the vocal cords when a gentle laryngoscopy was done. There are risks associated with every approach that we use but they can be reduced by careful planning and multidisciplinary management.

CONCLUSION

This case is an unusual presentation of rapidly progressive rhino cerebral ocular mucormycosis in the backdrop of incidental finding of diabetic ketoacidosis in a young male.

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Presence of an impossible mask ventilation and achieving successful awake intubation with continuous oxygenation in the backdrop makes it a rare entity.

Attempts should be made to diagnose the infection early and initiate prompt treatment to prevent rapid progression. Special consideration should be given to existing comorbidities and presence of a difficult airway for debridement of the infected tissue.

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