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Angioinvasion of anterior cerebral artery by rhinocerebral mucormycosis leading to intraparenchymal hemorrhage: A rare case report

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ABSTRACT

Rhinocerebral mucormycosis leading to intraparenchymal bleed is a rare entity. The objective of this case report is to highlight this unusual incidence in a case of mucormycosis. We present a case of a 50-year-old man with typical signs and symptoms like headache, diplopia, nasal congestion, swelling and congestion of right eye, cerebral sinus venous thrombosis diagnosed as rhinocerebral mucormycosis and on further imaging of brain showing erosion of anterior cerebral artery leading to fatal intra parenchymal bleed. The clinician must be quick to diagnose and aggressively manage the condition with both medical and surgical therapies to salvage the life of the patient.

Keywords: Rhinocerebral Mucormycosis, anterior cerebral artery, Intraparenchymal hemorrhage.

1. INTRODUCTION

The incidence of mucormycosis is increasing globally, but the increase among uncontrolled diabetes mellitus patients is high in India and China. A recent analysis, however, recorded (34 percent) in Europe, Asia (31 percent) and South or North America (28 percent), Africa (3 percent), Australia and New Zealand (3 percent) provide a different indication that the burden of disease in Europe is greater than in Asia (Prakash and Chakrabarti, 2019). Mucormycosis incidence is increasingly being reported in patients suffering from or recovered from COVID-19.

Mucormycosis associated with high morbidity and mortality, is an angioinvasive fungal infection. In recent years, there has been a shift in the epidemiology of mucormycosis, with an increase in prevalence, novel causative agents and a susceptible population. In patients with diabetes mellitus, the rhino-cerebral type of mucormycosis is most commonly seen, while in patients with haematologic malignancy and transplant recipients, pulmonary mucormycosis is most commonly seen. Cutaneous mucormycosis



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is usually seen in immunocompetent hosts following trauma (Kashyap et al., 2019).

Mucormycosis causative agents differ across numerous geographic locations. Although the most common isolated agent worldwide is *Rhizopus arrhizus*, *Apophysomyces variabilis* is predominant in Asia and *Lichtheimia* species in Europe. Rhinocerebral mucormycosis is a life-threatening condition with mortality rate of between 30-70% depending on the site of involvement. Saprophytic fungi (aerobic) of class *Phycomycetes* are responsible for rhinocerebral mucormycosis. The three genera responsible for most cases are *Rhizopus*, *Absidia*, and *Mucor*. These are commonly found in decaying vegetation, mold, and soil. They grow rapidly and are airborne as spores hence, they colonise mucosa of nose, paranasal sinuses, oral cavity and throat. The persons at risk of infection are immunocompromised individuals, as it generally cannot cause infection in healthy individuals and is found as a commensal. After infection of the host, it spreads along the blood vessels and nerves and infiltrates the walls of blood vessels. Infection can erode the sinus and can spread into the orbit, retroorbital area, thereby extending into brain. The ability of these species to be angioinvasive may result in direct infiltration into the blood vessels and thrombosis or emboli presenting as an acute ischaemic stroke (Sabobeh et al., 2018). Most cases are acute surgical emergencies and require immediate surgical excision of infected tissue. This case report highlights about a 50 year old patient who presented with rhinocerebral mucormycosis invading anterior cerebral artery leading to intraparenchymal haemorrhage, probably first case report from India.

2. CASE HISTORY

A 50-year-old male patient presented to the emergency department on 3rd December 2020, with complaints of headache, diminution of vision in right eye with swelling of right eye and lid, nasal blockage for 3 days. Patient also had history of high-grade fever, not associated with chills 1 day prior to admission. Patient was initially taken to a private hospital where his magnetic resonance angiography (MRI) of brain was done which was suggestive of fungal sinusitis with right cavernous sinus thrombosis.

On examination, patient was afebrile with pulse rate of 84 beats/min regular, blood pressure was 110/70 mm of hg in right arm supine position, other general examination was normal, cardiovascular, respiratory and abdominal examination was also normal. Patient had black mycotic debris in external auditory canal of right ear and right nostril; he also had tenderness over frontal sinus, ethmoid sinus and maxillary sinus. Upon examination of his eye, conjunctival congestion, chemosis and corneal oedema was noted. Upon further history taking it was found that patient was recently diagnosed as a case of COVID-19 pneumonia and recovered from the same. His complete blood picture was suggestive of raised total leukocyte count of 16,000 cells/cumm. Kidney and renal function tests were within normal limits. Nasal swab sent for culture and sensitivity showed growth of *Rhizopus* species (Figure 1).

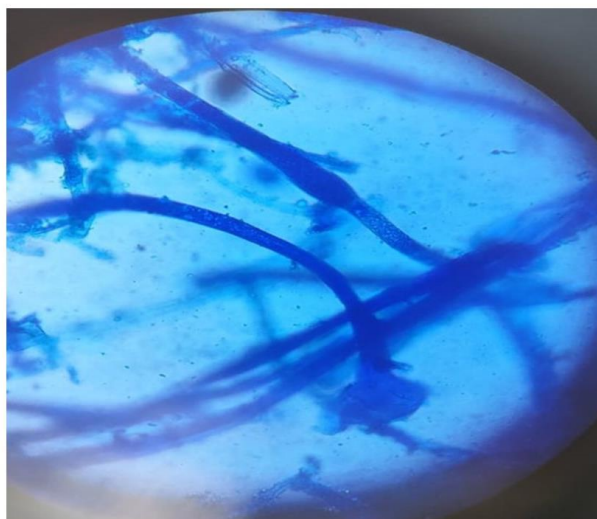


Figure 1 Slide showing growth of *Rhizopus* species with branching hyphae.

Hence the patient was diagnosed as having rhino cerebral mucormycosis with orbital involvement. MRI brain was repeated at our hospital, it was suggestive of angioinvasion of right and left anterior cerebral artery evident as a hyperintense plaque on both sides with right maxillary and ethmoid sinusitis causing right panophthalmitis with proptosis shown in figure 2.

Neurophysician opinion was taken patient was started on injectable amphotericin B (liposomal) and antibiotics along with other supportive management. Patient developed weakness over left side during 11th day of hospital stay, immediate computed tomography (CT) scan of brain was done which was suggestive of acute infarct in right gangliocapsular region with

intraparenchymal haemorrhage in right parieto-temporal region of 25 x 25 x 10 mm size with perilesional oedema (figure 3). Patient was managed conservatively in view of poor general condition with osmotic agents and other supportive care. Patient ultimately succumbed to his disease despite aggressive medical management on 12th day of hospital stay.

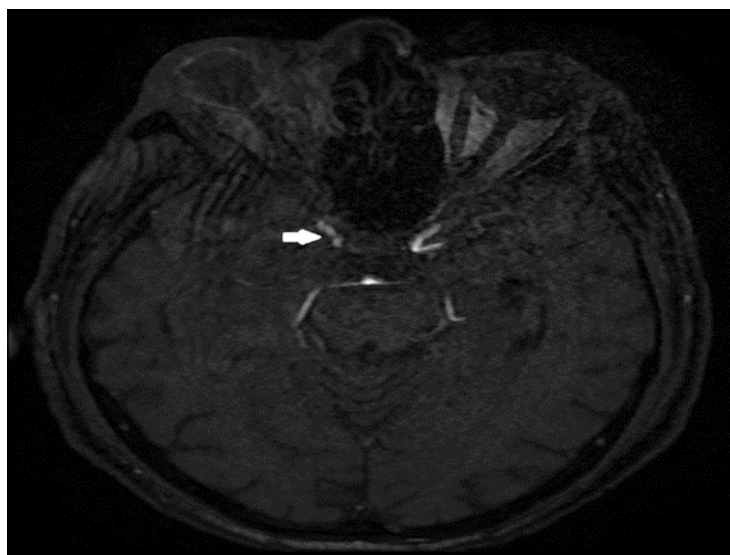


Figure 2 MRA suggestive of angioinvasion of anterior cerebral artery



Figure 3 CT brain suggestive of intraparenchymal haemorrhage in right parieto temporal region

3. DISCUSSION

The most prevalent type of systemic mucormycosis is rhinocerebral mucormycosis. It is common in diabetics and other immunocompromised patients, but the disease, especially zygomyces fungi, is documented even in normal individuals (Jain et al., 2011). Angioinvasion by irregular long, nonseptate hyphae that branch at right angles with a predilection for the internal elastic lamina of blood vessels is the histopathological hallmark of the disease (Singhania et al., 2020). Culture is not the investigation of choice for the diagnosis of mucormycosis. As the fungus is widespread, may even be present in normal individuals as a commensal,

or maybe a laboratory contaminant (Munir et al., 2007). Moreover, during the preparation of tissue specimens for culture, the organism may be destroyed.

Rhino-orbito-cerebral infection usually starts in the paranasal sinuses and spreads to involve the orbit and the brain (Chakrabarti et al., 2001). Complications of this disorder have been identified in the internal carotid artery and cavernous sinus thrombosis (Rajeshwari and Gangadhara Somayaji, 2012). The involvement of anterior cerebral artery in a case of rhino cerebral mucormycosis leading to haemorrhage has not been reported yet in literature. In our patient MRI brain with MRA and MRV was repeated on admission at our hospital. It was suggestive of involvement of bilateral anterior cerebral artery. Patient was started on broad spectrum antibiotics, antifungals and other supportive management. He was planned for orbital exenteration and functional endoscopic sinus surgery of nose in view of extensive involvement of orbit and maxillary sinus but was deferred in view of financial constraints.

On 11th day of admission, when patient developed acute onset left sided weakness CT brain plain was repeated which was suggestive of intraparenchymal haemorrhage in right temporoparietal region. Patient was managed conservatively in view of poor general condition with osmotic agents and other supportive care. Patient ultimately succumbed to his disease despite aggressive medical management.

4. CONCLUSION

Rhinocerebral mucormycosis is a life-threatening condition associated with high mortality, which needs to be treated aggressively with both surgical and medical treatment options. Failure to do so will result in death of the patient.

Conflict of interest

The authors have no conflicts of interest that are directly relevant to the content of this clinico-pathological case.

Financial Resources

There are no financial resources to fund this study.

Informed consent

Informed consent was obtained from the patient.

Author contribution

SD, SA and SK-Concept and writing of the manuscript, SN-Data collection, Sm D- microbiological report of Rhizopus and image.

Data and materials availability

All data associated with this study are present in the paper.

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