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Research Article

Successful Treatement of Angioinvasive Complication of Rhinocerebral Mucormycosis – A Case Study

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Abstract:

Saprophytic fungi cause rhinocerebralmucormycosis, an uncommon but lethal opportunistic infection of the nasal passages, oral cavity, sinuses, and brain. It is most frequent in those with diabetes and those who are immunocompromised. This case report involves a 38-year-old man who had recently been diagnosed with type 2 diabetes mellitus and complained of headaches, cough, numbness and tingling in his face, jaw discomfort, and diplopia for many days. Despite a differential diagnosis of tubercular meningitis, rhinocerebralmucormycosis was established by radio imaging methods and histopathological examination (HPE). Even though he had acquired carotid artery blockage, suggesting an angioinvasive infection, the patient fully recovered after taking an amphotericin B infusion for 20 doses. The current report emphasizes the significance of having a high index of suspicion when managing patients with diabetes presenting with facial pain, headache or cough and prompt inception of clinical management to control rhinocerebralmucormycosis.

Keywords: Rhinocerebralmucormycosis, Angioinvasive Nature, Carotid Artery Occlusion, Amphotericin-B

Introduction:

Mucormycosis, the second most common invasive mold disease after aspergillosis, is a rare but fatal fungal sickness caused by the phycomycetes class, order Mucorales, and family Mucoraceae.[1] Mucormycosis is characterized as rhinocerebral, pulmonary, gastrointestinal, cutaneous, or diffuse, depending on where it occurs.[2]

Rhinocerebralmucormycosis is one of them, affecting the nose, paranasal sinuses, and brain. It is more common in those with uncontrolled diabetes, those who are immunocompromised, those who have hematologic malignancies, those who have had organ transplants, and those who use intravenous narcotics.[3] Rhino cerebral mucor mycosis frequently appears suddenly, with symptoms that resemble sinusitis or periorbital cellulitis, and is diagnosed based on clinical suspicion and HPE.[4]

Case History:

A 38-year-old male who was newly diagnosed with diabetes mellitus (HbA1c- 8.9) initially in 2018 presented with complaints of headaches for the past one week, cough for the last 20 days, tingling and numbness of the face, diplopia for one day and jaw pain in the past ten days. Computed Tomography (CT) showed features of mild bilateral maxillary sinusitis with a deviated nasal septum to the right. In contrast, Magnetic Resonance Imaging (MRI) Brain was suggestive of features that showed inflammatory changes involving the paranasal sinuses and were treated symptomatically elsewhere. After one month, he rushed to our institute with complaints of severe headache and jaw pain from the past 20 days, for which MRI-brain revealed acute left MCA-PCA (Middle Cerebral Artery – Posterior Cerebral Artery), ACA-MCA (Anterior Cerebral Artery - Middle Cerebral Artery) watershed infarcts. Neck vessel imaging showed mucosal thickening in bilateral mastoid air cells, maxillary, ethmoidal, and sphenoidal sinuses. Lumbar puncture Cerebrospinal fluid (CSF) was performed and revealed cell counts- 350cells, neutrophils-21%, lymphocytes-74%, monocytes-05%, protein-40, sugar-102, and chloride-123. Therefore, he was differentially diagnosed to have Tubercular Meningitis and was empirically initiated with antitubercular therapy (AKT-4) [Pyrazinamide (750mg)+ Ethambutol (800mg) +Rifampicin (450mg)+ Isoniazid (300mg)] along with Inj. Methylprednisolone, Inj enoxaparin, tab aspirin, atorvastatin, IV fluids, multivitamins, and other supportive medications.

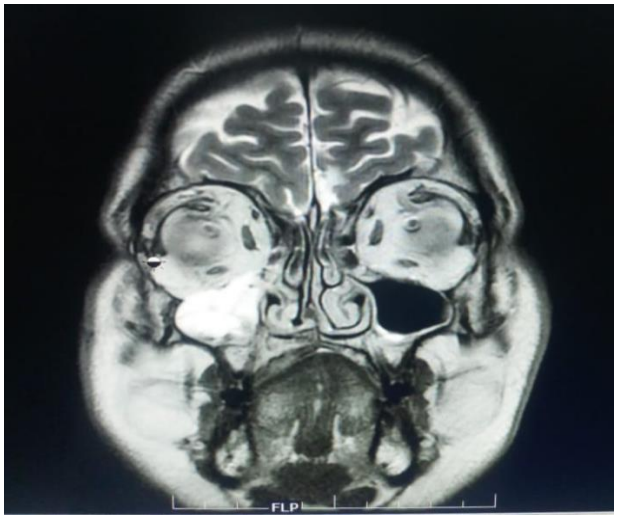

Fig No 1: Magnetic Resonance Imaging showing invasive fungal sinusitis of right maxillary, ethmoid and sphenoid sinuses with extention of the inflammatory process into bilateral carvernous sinus

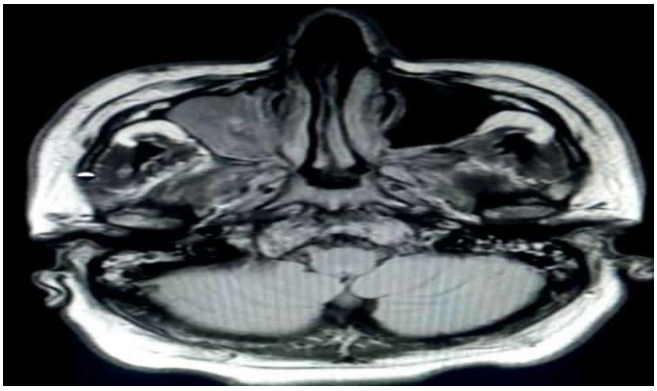

Fig No 2: CT Scan of soft tissue thickening with adjacent bone erosion in the right maxillary sinuses of hard palate

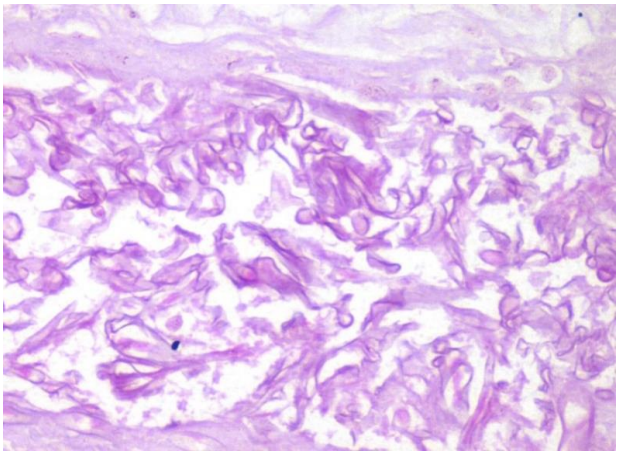

Fig No 3: Histopathological Examination using periodic acid Schiff stain under magnification power of 200x from meatal region bilateral with broad non-septate fungus showing evidence of mucormycosis

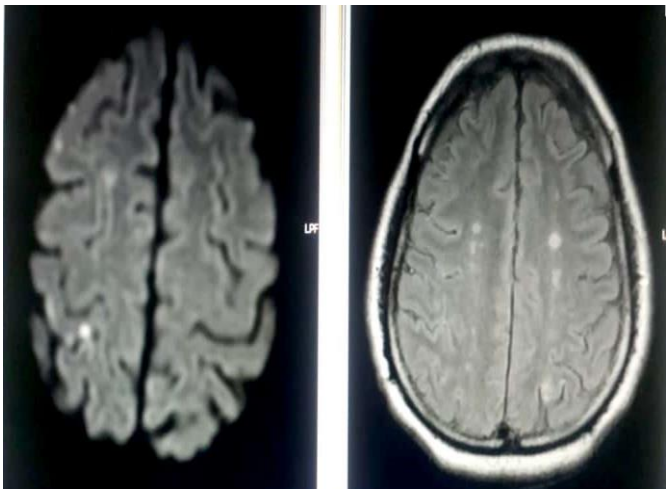

Fig No 4: MRI showing acute right MCA infarcts involving right frontal parietal lobes and right insular cortex, chronic lacunar infarcts on bilateral fronto-parietal centrum semiovale and corona radiata.

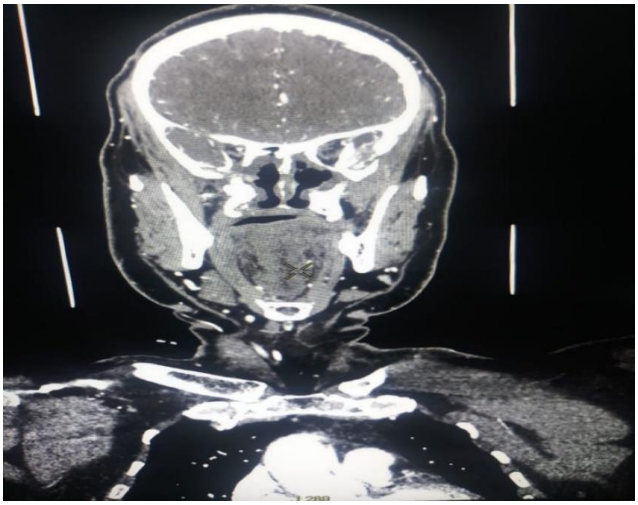

Fig No 5: CT angiogram showing mucosal thickening in bilateral maxillary, ethmoid and visualized portion of sphenoid sinuses, bony erosions in the greater wing of sphenoid. Soft tissue thickening in the parasellar region. Features likely representing infective/inflammatory fungal etiology

Furthermore, after three months, he was admitted with complaints of headache, decreased sleep, and fever over the past three days, and his Random Blood Sugar (RBS) was found to be 307 mg/dl. MRI with contrast showed features favoring invasive fungal sinusitis of right maxillary, ethmoid and sphenoid sinuses with the extension of the inflammatory process into bilateral cavernous sinus (Fig No 1). CT of paranasal sinuses (PNS) concluded mucosal and soft tissue thickening with adjacent bone erosion in right maxillary sinuses (Fig No.2). Accordingly, FESS (Functional Endoscopic Sinus Surgery) procedure was planned and uneventful. CSF analysis showed cell counts-60, neutrophils-21%, lymphocytes-74%, monocytes-05%, protein 61 and chloride-125. For further evidence, Histopathological examination (HPE) from the bilateral middle meatal region showed plenty of broad ribbon-like non-septate fungal hyphae with right-angle branching resembling Mucormycosis (fig No.3). Hence, fungal meningitis- mucormycosis emerged as the final diagnosis. Fungal culture was sent which grew *Rhizopus oryzae*. Amphotericin B infusion (5mg/kg for 10 days), tab isoniazid, and InjClexane were initiated along with other supportive medications for his underlying conditions.

MRI revealed multiple acute right MCA infarcts involving right frontoparietal lobes and complete occlusion of intra and extra cranial portions of left Internal Carotid Artery (ICA) on Magnetic resonance angiography (MRA) (Fig No.4). He was initiated with tab clopidogrel, Inj. Enoxaparin, tab aspirin, tab. atorvastatin, 5-fluoro cytosine, IV fluids, and supportive medications. Approximately after ten months, he was readmitted with complaints of sudden onset of slurring of speech, blurring of vision, left-sided upper limb, and facial weakness. Further, a CT angiogram of the head and neck revealed mucosal thickening in the bilateral maxillary, ethmoid and sphenoid sinuses, with soft tissue thickening in the parasellar region likely representing infective/inflammatory/ fungal infection (Fig No.5). Because of complete left ICA occlusion and fresh stroke in the right ICA, the patient underwent DSA (Digital Subtraction Angiography) and right ICA stenting. Additionally, he has added posaconazole 300mg along with amphotericin B due to persisting invasive fungal sinusitis.

Recurrent hospital admissions due to complications of rhinocerebralmucormycosis, amphotericin B infusion was administered up to 20 doses. However, the patient achieved complete remission with the procedures as mentioned above and medications.

Discussion:

Fungal infections of the central nervous system (CNS) are a rare but life-threatening condition. They can cause granuloma, cerebritis, abscess development, meningitis, and meningeal vasculitis, which can proceed quickly and cause recurring symptoms.[5]. Mucormycosis is an uncommon fungal infection that is less prevalent than Aspergillus and Candida species. [6] The annual incidence of mucormycosis is around 1.7 cases per million.[7]

The most frequent form of mucormycosis, rhinocerebralmucormycosis (RCM), is known to target the skull base and its related blood arteries, resulting in mycotic aneurysms, ischemic infarcts, and intracerebral bleeding. [8] Rhizopus oryzae is

implicated in the cases of rhinocerebral mucormycosis. [15]

Rhinocerebralmucormycosis frequently affects the maxillary and ethmoid sinuses and the brain. Headache, periorbital edema, vision loss, fever, diplopia, rhinitis, and reduced mental performance are all typical symptoms, although none are specific. [9] Initial CT and MRI scans are generally unremarkable, but symptoms of sinusitis, such as clogged sinuses or thickening mucosal lining, appear. It is critical to repeat the tests to keep a careful eye on the disease's course. A histopathological study along with fungal culture confirms the presence of infection.[10] Although the differential diagnosis in our case report was originally tubercular meningitis, further HPE findings indicated strong evidence for mucormycosis, with supported impressions from MRI and CT scans. Fungal culture isolation of *Rhizopus oryzae* clinched the confirmation.

Our patient began with moderate acute sinusitis restricted to the bilateral ethmoid and right sphenoid sinuses, which quickly proceeded to bony erosions in the larger wing of the sphenoid bone, mucosal thickening in the parasellar area, left ICA blockage, and large segments in the right ICA.

Bae M et al. reported a similar case in which the MRI and MRA of a patient with mild ethmoid sinusitis appeared normal at first. However, the sinusitis quickly worsened with extension to the right orbit and anterior cranial fossa within two months, which was associated with the right ICA, indicating the pathological nature of the infection.[11]

Another case by Won-Kwak S et al. reported that a patient diagnosed with rhinocerebralmucormycosis died due to massive infarction on the right cerebral hemisphere, occlusion of the right ICA and severe sepsis thereby indicating the increased mortality risk when carotid arteries get involved. [12]

A combination of surgery and Amphotericin B therapy (5mg/kg IV daily) in patients with rhinocerebralmucormycosis has a better prognosis having a survival rate up to 80%. [13] Concerning this statement, our patient recovered entirely without recurrence of his symptoms.

However, other literature states that the survival rate with brain involvement is 20%. [14] In contrast, our patient recovered utterly, depicting that mortality and morbidity depend on the risk factors, time of initiation of IV antifungal therapy, and surgical intervention.

This paper emphasizes the clinical presentation and pathogenesis of RCM, especially in patients who present with the risk mentioned above factors and the need for a high index of suspicion in diagnosis and initiation of antifungal therapy, which is essential to reduce morbidity and mortality.

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Declarations

Competing interests:

The authors have no conflicts of interest regarding this article**.**

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