Medical Science

pISSN 2321–7359; eISSN 2321–7367

Post COVID-19 recurrent rhino ocular cerebral mucormycosis: A curse like no other

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To Cite:
Saldanha C, Daigavane S, Nagendra V. Post COVID-19 recurrent rhino ocular cerebral mucormycosis: A curse like no other. *Medical Science* 2023; 27: e124ms2678.
doi: https://doi.org/10.54905/disssi/v27i133/e124ms2678

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Peer-Review History
Received: 11 December 2022
Reviewed & Revised: 15/December/2022 to 27/February/2023
Accepted: 01 March 2023
Published: 03 March 2023

Peer-review Method
External peer-review was done through double-blind method.

URL: https://www.discoveryjournals.org/medicalscience

ABSTRACT

Coronavirus disease 2019 (COVID-19), caused by SARS-CoV-2, has been linked to a number of opportunistic bacterial and fungal infections. Aspergillus and Candida have been identified as the primary fungal pathogens for COVID-19 co-infection. Recently, several cases of mucormycosis in COVID-19 patients have been reported worldwide, particularly in India. Now, remember that mucormycosis can be of different types like rhino-ocular-cerebral (ROCM), pulmonary, gastrointestinal, etc., here we are dealing with a case of rhino-ocular cerebral mucormycosis which not only occurred once but twice post covid-19 infection. As we recover from the epidemic and the pandemic, it is essential to look back and understand what led to the insane surge of mucormycosis in post-covid-19 patients, especially in our country, India. Mucor is ubiquitous and present constantly around us, so no single factor but an array of elements is responsible predominantly being the incorrect use of corticosteroids in uncontrolled diabetics, which led to severe immunosuppression, injury to the beta cells of the pancreas by the SARS-COV-2 in newly diagnosed diabetics. In such cases, it is important to evaluate the patient thoroughly and orbital exenteration is done depending upon the doctor's judgment and MRI findings. Following is a case report of one such patient with recurrent post COVID mucormycosis predisposed by uncontrolled diabetes mellitus and managed by orbital exenteration.

Keywords: Mucormycosis, recurrence, exenteration, amphotericin B, MRI, Orbital extension.

1. INTRODUCTION

Diabetes mellitus, inappropriate corticosteroid usage, immunosuppressive medicines, p haematological malignancy, haematological stem cell transplantation, etc., all increase the risk of acquring mucormycosis, an angioinvasive and possibly fatal fungal infection. India has seen a surge in mucormycosis cases during the second phase of the covid-19 epidemic (Daga et al., 2022). Recurrence of mucormycosis has been seen in a couple of cases. India has the second-most number of people suffering from diabetes mellitus worldwide (65.1 million), with uncontrolled diabetes accounting for over 70%

of all cases (Cho et al., 2013). The prevalence of mucormycosis is 0.16% among patients with diabetes and 1.2% among kidney transplant recipients, according to data from specific patient groups, with the majority of these cases appearing as the ROC type (Cho et al., 2013). Mucormycosis can be of different types like rhino-ocular-cerebral (ROCM), pulmonary, gastrointestinal, etc., ROCM being the most common among them. Mucormycosis is a progressively invasive, possibly fatal disease, whenever presented late (Peterson et al., 1997). It is of prime importance to treat such a patient as a medical emergency. No single factor is responsible for this disease but an array of factors. The most important parameter in the management is the need for orbital exenteration; it is based on the treating doctor's judgment as this does not necessarily save from mortality. We look into one such case in a patient with uncontrolled diabetes suffering from recurrent ROCM, which was successfully managed by orbital exenteration by curbing the further spread of the disease.

2. PATIENT DETAILS

A 48-year-old male came to the Ophthalmology OPD on 25th May 2021 with complaints of swelling of the right upper and lower eyelids and gradual diminution of vision in the right eye for eight days. He also complained of pain over the right side of the face and head for eight days. He denied any history of trauma to the eye. On enquiring about his medical history, he underwent functional endoscopic sinus surgery for chronic allergic fungal sinusitis in February 2021 after being diagnosed with Covid-19 in January 2021. MRI brain and orbit (contrast) done at that period showed fungal sinusitis of the right maxillary, ethmoid and sphenoid sinus**.**

He was again diagnosed with COVID-19 on 5th May 2021 and was admitted to an Intensive care unit for nine days. During his stay at the hospital, he received a Tablet of Dexamethasone 4mg for nine days in tapering doses and a Tablet of Clopidogrel for ten days. On completion of treatment, he was discharged from the hospital and resumed daily activities. The patient is also a known case of diabetes mellitus on medication but uncontrolled, with no other systemic illness; he also uses a Tablet of Aspirin 75 mg once daily. The patient then gave history of diminution of vision in the right eye for the past three months, which had worsened over the past eight days and nasal congestion on and off. The patient did not give any history of ear discharge/trauma, nasal discharge and loss of sensation over the face, difficulty swallowing or any COVID positive symptoms.

3. CLINICAL EXAMINATION

On ophthalmological examination, it was observed that the patient had the perception of light in the right eye, but projection of rays was inaccurate in the superior, medial and inferior quadrants. Proptosis of the right eye was present. The lid was edematous and ptotic. Conjunctival chemosis was present. The pupil was normal and reacting to light. Lens status was normal and posterior segment evaluation was also within normal limits (Figure 1) Extraocular movements were restricted in every gaze. The left eye was within normal limits, with visual acuity improving to 6/6.

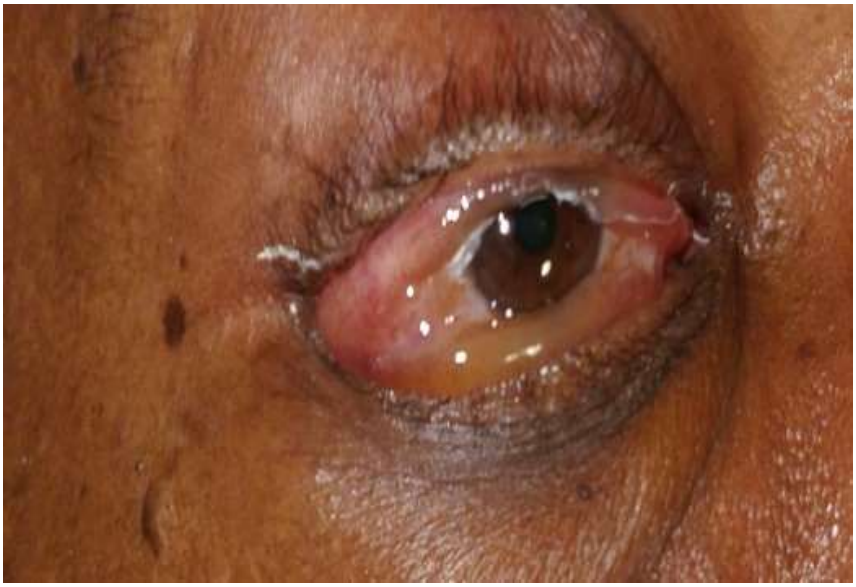

Figure 1 The preoperative image of the right eye

CONTRAST MRI ORBIT-PNS (Figure 2) showed previous post-operative (FESS) findings in the bilateral sino-nasal region with heterogeneously enhancing thickening of the mucosa in the left maxillary and left anterior ethmoid, right maxillary, ethmoid, sphenoid and frontal sinuses. Mild erosions along the posterior part of the right maxillary sinus, medial wall and floor of the right orbit involvement of pterygoid fossa, infratemporal fat, deep head of temporalis and adjacent lateral pterygoid muscle proptosis of the right eye. Abnormal enhancement and bulk in the posterior aspect of the orbital cavity involved an inferior orbital fissure, orbital apex and its contents and superior orbital fissure. Therefore, suggesting right invasive sinusitis (fungal) with right orbital cellulitis.

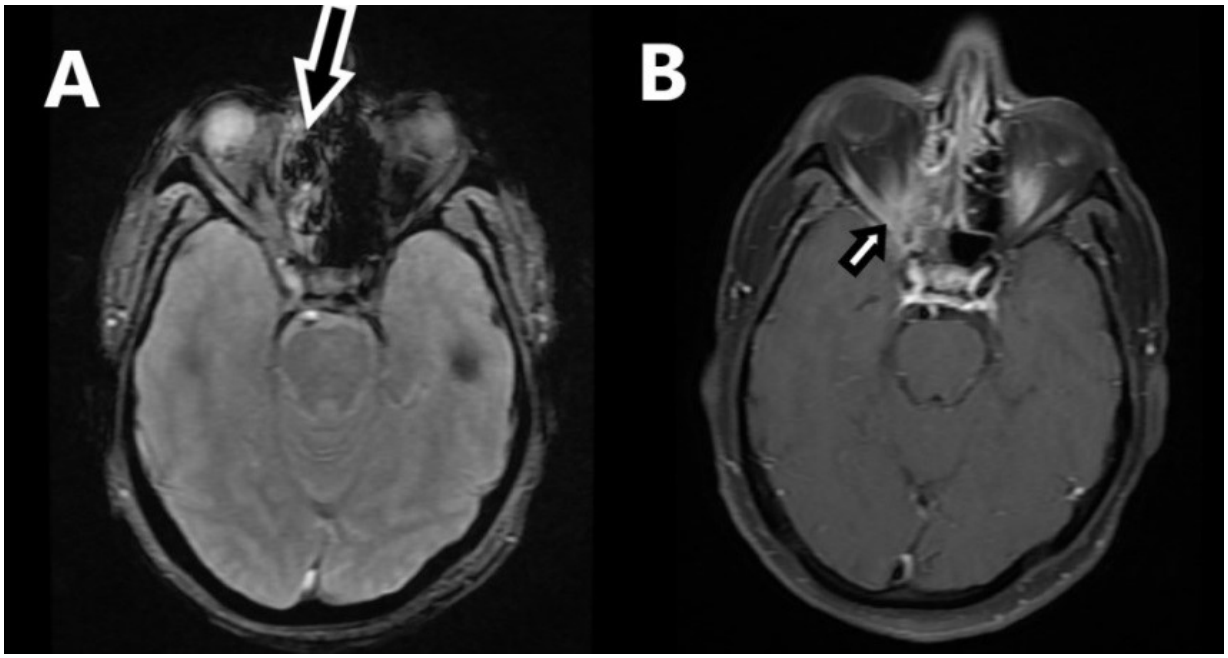

Figure 2 A: Axial GRE PNS section showing mucosal thickening with internal areas of blooming (arrow) in the right ethmoid sinus suggesting fungal elements. B: Axial contrast-enhanced T1 PNS section showing enhancing mucosal thickening in the right ethmoid and sphenoid sinus with involvement of right inferior orbital fissure (arrow)

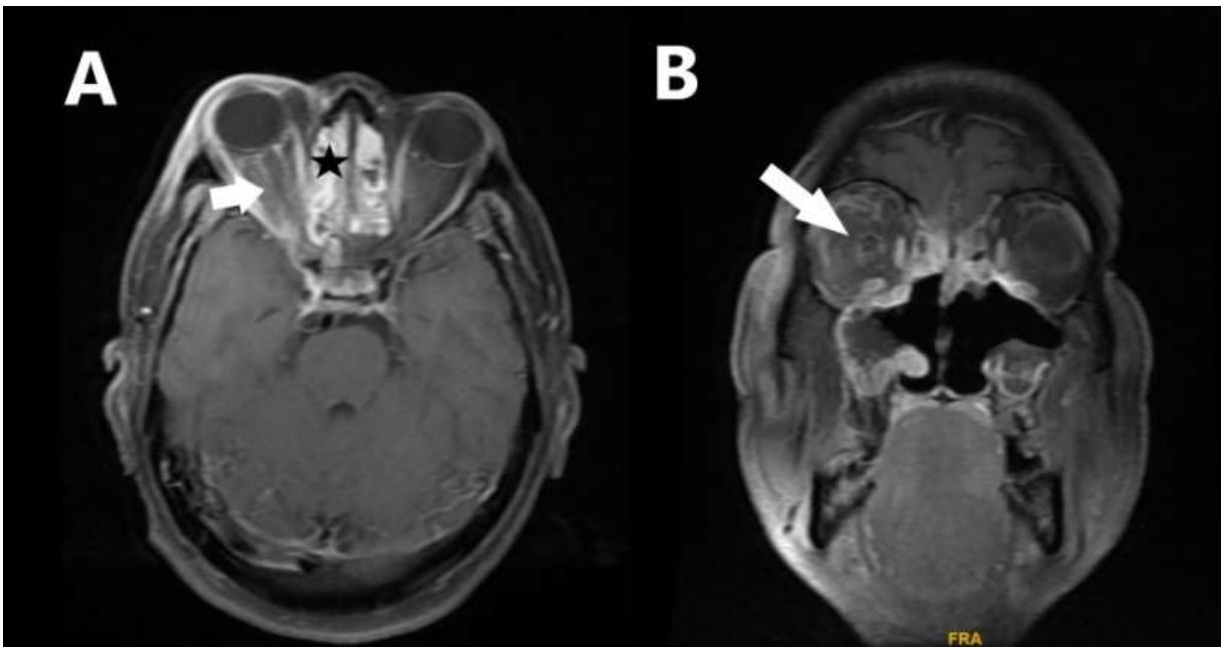

Figure 3 A: Axial contrast-enhanced T1 section of orbit and PNS showing proptosis of the right eye, thickened and enhancing right optic nerve (arrow) and intensely enhancing mucosal thickening in the bilateral maxillary sinus (asterisk). B: Coronal contrast-enhanced T1 section of orbit and PNS showing bulky right orbit, thickened and enhancing right optic nerve (arrow) compared to the contralateral side

He then underwent revision functional endoscopic sinus surgery. He was started on Injection of liposomal Amphotericin B intravenously and Injection of liposomal amphotericin B peribulbar in the right eye. After a few days, a repeat examination showed that the patient had no perception of light, lid edema and ptosis remained the same and conjunctival chemosis increased. The patient was advised for repeat Contrast MRI ORBIT-PNS (Figure 3) with brain screening, which showed previously described above MRI findings with altered signals in both intra and extraconal fat, mainly on lateral aspect extending to the posterior part of the orbital cavity with encasement of the right optic nerve. The orbital apex and its contents are involved and extend posteriorly along the meninges of the lateral aspect of the right cavernous sinus. Right lateral rectus shows increased bulk, altered signals and enhancement, suggesting involvement.

The patient was then planned for lid-sparing orbital exenteration under general anaesthesia to curb the further spread of the infection. This call was ultimately the surgeons keeping in mind the MRI finding and ocular findings to curb the further spread of the disease. The wound was healthy after surgery (Figures 4, 5), with no soakage, discharge and sutures in situ. The patient was discharged the following week and was called for prosthesis after two months.

Figure 4 Postoperative day 1

Figure 5 Postoperative day 15

Timeline of the current episode

January 2021: COVID-19 positive February 2021: Diagnosed and operated on for nasal mucormycosis. May 2021: COVID-19 positive and with eye complaints, so he was admitted to the department of ophthalmology and MRI was conducted. June 2021: Right eye exenteration was successfully performed. August 2021: Follow up.

4. DISCUSSION

Any fungal infection brought on by a member of the order Mucorales is known as mucormycosis. They permeate the atmosphere and are regularly inhaled by humans. As a result of our robust immune system, however, humans rarely fall victim to such illnesses. Diabetes Mellitus is the most prevalent predisposing factor. Hence it primarily affects those who are immunocompromised (Cho et al., 2013; Peterson et al., 1997). Post-covid mucormycosis is most commonly caused by poorly managed diabetes. Airborne fungal spores are the primary vector for the propagation of many diseases. Other transmission routes include direct contact with the skin or nasal mucosa and accidental ingestion following trauma (Peterson et al., 1997). Nasal and paranasal sinus infections are the most prevalent types. The brain and lungs are also vulnerable to the effects of mucormycosis (Peterson et al., 1997). Once within the body via one of the aforementioned entry points, these spores make their way to the bloodstream and eventually the tissues, where they lodge. The presence of this foreign body triggers an inflammatory response, which results in the creation of a thrombus, which in turn gradually causes ischemia necrosis. Because of this, tissues become darkened, as is typical of mucormycosis (Lee et al., 2014). The infection then travels to the orbit from the infra temporal fossa, superior part of orbital fissure, inferior part of orbital fissure and to the orbital apex, ultimately reaching the lamina papyracea. Diplopia, chemosis, congestion and restricted extra ocular movements are early indications, depending on the affected site. Optic atrophy, total blindness and total ophthalmoplegia are all late symptoms (Gillespie et al., 1998).

Tissues are surgically debrided and Injection amphotericin B 1mg/kg/day are administered. A decision on whether or not to do orbital exenteration is made by a team of specialists, including ophthalmologists and otolaryngologists, after carefully considering each patient's condition. Exenteration may cause psychological and social issues in addition to losing sight, which is the most essential human sense. Keeping the eye or stopping the disease from spreading inside the brain and killing the patient is a difficult choice (Hargrove et al., 2006). Amphotericin B, in its newer forms, such as liposomal amphotericin B, is effective in treating mucormycosis. The azoles itraconazole and posaconazole are used as first-line treatments and they come in various forms (oral suspension, delayed-release pills and infusion.

Particularly in our case, uncontrolled diabetes and excessive corticosteroid use could have triggered the occurrence of the disease again as hyperglycemia causes the adhesion and penetration of Mucorales by increasing the bond between endothelial receptor glucose-regulated protein and morale-regulated protein, now being an angioinvasive fungus this adhesion is vital for the spread of the disease (Bhandari et al., 2022).

5. CONCLUSION

Mucormycosis is an uncommon fungal infection owing to its low virulence but becomes common in immunocompromised individuals suffering from long-standing diabetes mellitus before steroid therapy etc. Studying the patient's status thoroughly and carrying out appropriate treatment is of paramount importance. With the help of aggressive resection techniques and correct medical management, it is possible to reduce the recurrence of mucormycosis. The main aim of lowering recurrence is treating the underlying cause and regular follow-ups to improve immunity.

Acknowledgement

We thank the participants who have contributed in this study.

Informed consent

Informed consent was obtained from the patient.

Author's contribution

All the authors contributed equally to the case report

Funding

This study has not received any external funding.

Conflict of interest

The authors declare that there is no conflict of interests.

Data and materials availability

All data sets collected during this study are available upon reasonable request from the corresponding author.

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