DOI: httpp://doi.org/10.36611/upjohns/volume11/Issue1/7 Date received: 06.04.2023 / Date published: 30.06.2023

POST-COVID ROCM (RHINO ORBITAL CEREBRAL MUCORMYCOSIS) INVOLVING SKULL BASE – A RARE CASE REPORT

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ABSTRACT

INTRODUCTION

Mucormycosis in the setting of COVID-19 has given rise to the concept of 'epidemic within a pandemic'. It is a life-threatening fungal infection caused by fungi belonging to class Zygomycetes. It has got special affinity for the immunocompromised which is one of the main reasons for the flare up of the disease during COVID. Rhino-orbito-cerebral Mucormycosis (ROCM) is by far known to be the most common form of this disease. This usually presents with nasal dryness and blockage, swelling and numbness of cheek, blackish discoloration of nose, cheek and palate. It has been seen that in late stages of mucormycosis, patients presented with ophthalmoplegia, diminuition of vision, loss of vision, altered sensorium or even facial palsy.

MATERIAL AND METHOD

We here-in report a rare case of a 67-year-old diabetic male who was diagnosed with post-COVID Mucormycosis involving skull base with right abducens nerve and right hypoglossal nerve palsies. Management comprised of endoscopic debridement of the disease followed by administration of liposomal amphotericin. Patient was followed for 6 months with repeated nasal endoscopies and follow-up MRI.

CONCLUSION

There was visible recovery of the sixth and twelfth

nerves. No recurrence has been observed.

Keywords - Rhino-orbito-cerebral mucormycosis, abducens nerve palsy, hypoglossal nerve palsy, endoscopic debridement, dolleros canal

INTRODUCTION

Mucormycosis, also known as Zygomycosis, is a life-threatening angioinvasive fungal infection caused by filamentous fungi of the order Mucorales of class Zygomycetes. 1. The common predisposing factors include uncontrolled diabetes mellitus, prolonged use of steroids, patients on chemotherapy, hematologic malignancies, organ/stem-cell transplantation, iron overload. 2. In a country like India mucormycosis is associated with a high mortality rate due to a delay in diagnosis and the high cost of management.3

Rhino-orbito-cerebral mucormycosis is known to be the most common form of this infection. 1

The major mode of transmission is inhalation of sporangiospores into nasal and paranasal sinuses. The infection spreads rapidly and extensively into adjacent tissues within a period of few hours to days and may lead to intracranial involvement and cranial nerve palsies.4

We have reported a unique case of post COVID mucormycosis involving skull base withabducens and hypoglossal nerve palsy with no nasal

symptoms. Only a few sporadic cases of this nature have been described in literature till date.

CASE REPORT

A 67-year-old diabetic and hypertensive male presented to our emergency with complaints of headache for 4 months and diplopia in right eye for 10 days. Patient had history of hospital admission for COVID-19 infection one month prior to presentation and administration of steroids during the stay. On presentation, patient's general condition was fair. On examination his tongue was deviated to right on protrusion, there was evident palsy of right lateral rectus muscle (Figure I) with diplopia in his right eye.

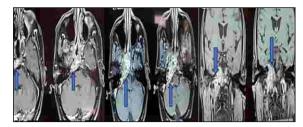
Figure 1 - Pre-op image of the patient showing (a)



right lateral rectus palsy and (b) deviation of tongue to right

There was no facial or peri-orbital swelling. Other cranial nerves were normal. Anterior and posterior rhinoscopy were within normal limits. Contrast-enhanced Magnetic Resonance Imaging (CEMRI) of nose and paranasal sinuses with orbit revealed bony erosion of the right half of clivus and adjoining skull base with extension of lesion to right jugular foramen and hypoglossal canalwith soft tissue enhancement (Figure II).

Figure 2- Pre-operative Contrast-enhanced Magnetic Resonance Imaging (CEMRI) -axial (a and b) and coronal (c) views showing soft tissue thickening and bony erosion in region of right jugular foramen, involving right hypoglossal canal



with erosive changes in adjoining skull base and right half of clivus (marked with blue arrows)

Patient was immediately planned for endoscopic endonasal debridement under general anaesthesia. Endoscopically trans nasal sphenoid sinus was explored and was found filled with necrotic material which were debrided. (Figure III).

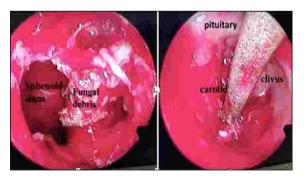


Figure 3 -Intra-operative images of endoscopic debridement

Clivus was found partially eroded. The debrided tissues were sent for histopathology. Microscopic examination revealed irregular broad aseptate hyphae branching at wide-angle which was suggestive of mucormycosis (Figure IV).

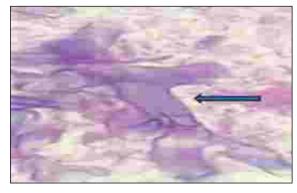


Figure 4- Histopathological picture showing broad aseptate obtuse angle branched fungal

hyphae suggestive of mucormycosis

Liposomal amphotericin-B was started with continuous monitoring of serum electrolytes, creatinine and blood sugar. A total dose of 5.4gm was administered. Patient was discharged in improved condition with recovery of right sixth and twelfth cranial nerve palsies (Figure V) and was adviced oral Posaconazole 300mg twice daily on first day followed by once daily for 6 months.



Figure 5A - Post-op image showing recovery of right lateral rectus palsy

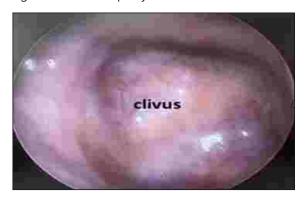
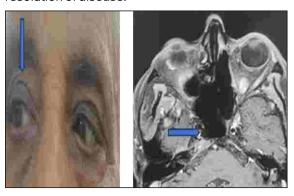


Figure 5B- Follow-up nasal endoscopy showing complete resolution of disease at clivus

He was adviced oral Itraconazole for an additional 3 months. Patient was called for regular follow-ups every 2 weeks for first 3 months and then once a month for next 3 months.On follow-up nasal endoscopy there was complete resolution of disease (Figure V). No recurrence has been observed on a follow-up of 6 months. Follow-up MRI after 6 months also showed disease-free status (Figure VI).

Figure 6: Follow up MRI showing complete resolution of disease.



DISCUSSION

Mucormycosis has now been well-established as a fulminant angioinvasive fungal infection. It affects predominantly diabetics and immunocompromised individuals. There has been an alarming rise of cases as an after-effect of COVID infection. 2, 5Petrikkos et all described the various forms of invasive mucormycosis based on their anatomic location and clinical presentation as 1) rhino-orbito-cerebral 2) pulmonary 3) cutaneous 4) gastrointestinal, 5) disseminated, 6) miscellaneous. Of these forms, our patient was diagnosed with Rhino-orbito-cerebral mucormycosis.

Symptoms of ROCM are most commonly known to be excessive dryness of nose with blackish discharge, numbness over and around nose and cheek, cheek swelling, loose teeth, blackish discoloration of cheek and/or palate, eye lid swelling or proptosis. 6A black necrotic Eschar has been known to be the hallmark of mucormycosis. 1 In our case, there were no nasal complains and on examination no nasal findings, so the infection most probably traversed through the right spenoid sinus to the compact bone, finally affecting skull base. There have been reports of involvement of VII cranial nerve in mucormycosis. 7,8 In our patient the facial nerve was not affected, and the infection spread directly inferiorly to involve the jugular foramen. As the infection progressed, further involvement of hypoglossal nerve at its exit at the hypoglossal

canal was noticed. Involvement of the abducens nerve probably occurred at the Dorello's canal where it passes over the edge of the Petrous pyramid. According to the clinico-radiological classification for ROCM as proposed by Soni et al 9, our patient belonged to stage IV-a.

Radiologically MRI is superior to Computed Tomography scans with respect to delineation of intracranial extent of the disease; it helps to evaluate patients who demonstrate signs of intracranial invasion such as altered mental status, cranial nerve palsies, cavernous sinus thrombosis, orbital apex syndrome, seizures or stroke. 10 Since our patient exhibited multiple cranial nerve palsies we opted for a contrast MRI straightforward.

The first and foremost management of this disease is aggressive surgical debridement of all necrotic tissues with orbital exenteration wherever required. 9,11,12 This should be followed by prolonged administration of liposomal Amphotericin-B. This is a welldocumented first-line medical cure for mucormycosis. 13 For patients with CNS involvement doses as high as 10mg/kg/day can be given.14In our patient we started Amphotericin at a dose of 5mg/kg/day and a total of 18 doses (5.4qm) were given followed by oral Posaconazole300mg BD on day one followed by 300mg OD day 2 onwards for 6 months. Posaconazole has also been used as salvage therapy for Amphotericin refractory cases. 14,15

CONCLUSION

Skull base mucormycosis is a rare entity and carries poor prognosis. Clinical suspicion aided by diagnostic MRI and nasal endoscopy help to achieve a correct and timely diagnosis. Surgical debridement of necrotic tissues and administration of liposomal amphotericin-B with long-term Posaconazole go hand-in-hand in the management of this fungal infection and should be the treatment norm. It is prudent for a rhinologist to be familiar with the disease pattern and be aware of the outcomes.

Conflict of interests

None

Funding

None

Acknowledgement

We are grateful to our Head of Department Prof. Anupam Mishra for his support and encouragement.

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How to cite this article

Patnaik M. etal; Post-Covid Rocm (Rhino Orbital Cerebral Mucormycosis) Involving Skull Base - A Rare Case Report; UPJOHNS; June 23; 11(1); page 44-48

DOI: httpp://doi.org/10.36611/upjohns/volume11/lssue1/7

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