ORIGINAL RESEARCH PAPER
Volume - 11 | Issue - 01 | January - 2022 | PRINT ISSN No. 2277 - 8179 | DOI : 10.36106/ijsr
INTERNATIONAL JOURNAL OF SCIENTIFIC RESEARCH

FATAL BILATERAL ORBITAL INVOLVEMENT IN CASE OF ROCM IN A YOUNG DIABETIC PATIENT WITH COVID-19 INFECTION: A RARE CASE REPORT.

Ophthalmology

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ABSTRACT

The second wave of COVID-19 in India was associated with higher incidence of Mucormycosis. We report a rare case of 40-year-old Asian male, with Type 2 diabetes mellitus who presented with bilateral sudden loss of vision, ptosis, proptosis, total ophthalmoplegia and central retinal artery occlusion.

Computed tomography followed by magnetic resonance imaging was suggestive of bilateral sinusitis, orbital cellulitis, optic neuritis and gross intracranial involvement.

Considering his general condition and guarded prognosis for survival, exenteration was deferred. After detailed literature research, we found very few reports of bilateral orbital involvement in cases of COVID-19 associated ROCM.

Even after early diagnosis and prompt treatment of ROCM, patient succumbed to the illness. Covid 19 associated bilateral rhino-orbital-cerebral mucormycosis may be a marker of mortality in the affected population.

KEYWORDS

Bilateral, ROCM, COVID-19, Amphotericin B, Mucormycosis, Diabetes mellitus, CAM.

INTRODUCTION:

Mucormycosis caused by filamentous Mucorales fungi is an angio-invasive infection. Rhino-orbital-cerebral Mucormycosis (ROCM) is rare but life-threatening fungal infection that occurs in immunocompromised, diabetic and COVID-19 survivors. Bilateral involvement in ROCM has been reported in only a few cases.[1] Moreover bilateral involvement in ROCM associated with COVID-19 is rare.[2] The second wave of COVID-19 in India was associated with higher incidence of mucormycosis when compared with the first wave.[3] Owing to its epidemic proportions, mucormycosis has been declared a notifiable disease in India.[4]

Clinical Findings:

A 40 year old Asian male non vaccinated against Covid 19 infection with controlled Type 2 diabetes mellitus (DM) and history of COVID-19 pneumonitis was treated with systemic antivirals and steroids. He presented to our tertiary care centre with sudden loss of vision, inability to open and move both eyes. On detailed ophthalmic examination, there was bilateral severe ptosis, proptosis, conjunctival chemosis, and total ophthalmoplegia with fixed mid dilated pupil. (Figure 1) He had bilateral Central Retinal Artery Occlusion (CRAO) with no light perception. He had malar swelling associated with infraorbital anaesthesia on both sides of his face. Events has been explained in time-line. The patient was not suffering from any other known systemic illness.

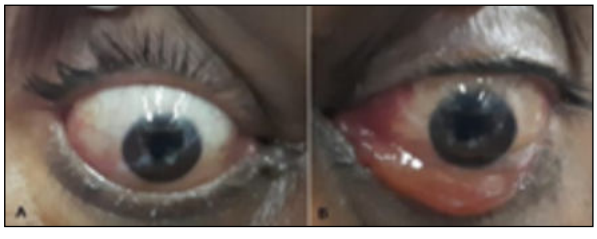

Figure 1. A. Right eye B. Left eye. Showing conjunctival chemosis and congestion.

Time-Line

Days	*Events*	*Findings*
D-0	HRCT	COVID-19 pneumonitis CT severity score of 8 (mild) COVID-19 Reporting and Data System (CO-RADS) – 06
D-6 – 8	*Development of Ophthalmic symptoms*	
D-8	*CT* (PNS+ORBIT) Plain	Bilateral Sinusitis, bilateral orbital cellulitis with possibility of fungal aetiology
D-10	FESS	Necrotic tissue in all paranasal sinuses bilaterally with skull base involvement and exposure of dura matter.
D-11	*Post FESS MRI with Angiography*	• Bilateral orbital cellulitis, optic neuritis, • Pan sinusitis • Acute non haemorrhagic infarct in left frontal subcortical region and mild pneumocephalus along bilateral frontal convexities.
D-12	Referred to Tertiary care centre.	
D-12	RTPCR -SARS-CoV-2	Negative
D-13	Histopathology report-Nasal tissue	Aseptate, stout fungal hyphae branching at right angles diagnostic of Mucormycosis
D-13	MRI (BRAIN +PNS+ORBIT) Plain	• Advanced intracranial extension to left frontal subcortical region, focal encephalitis, and pneumocephalus. • Bilateral orbital cellulitis and optic neuritis (left>right) • Recurrent sinusitis
D-15	Diagnostic and Therapeutic Nasal Endoscopy	• Gross bilateral orbital involvement, Pan sinusitis, • Intracranial extension
D-28	Patient succumbed	

Diagnostic Assessment:

Computed tomography (CT) and Magnetic Resonance Imaging (MRI) was suggestive of bilateral sinusitis, orbital cellulitis, optic neuritis and gross intracranial involvement. (Figure 2) FESS showed necrotic tissue in all paranasal sinuses bilaterally with skull base involvement and exposure of dura matter. Histopathology analysis of nasal tissue was diagnostic of mucormycosis. His disease stage was 4c as per the proposed staging of ROCM.[5]

Therapeutic Intervention:

Patient was diagnosed with Covid 19 pneumonitis and was treated with

injection Remdesivir 200 mg for first day followed by 100 mg for next 5 days, intravenous injection Methyl prednisolone 40 mg twice a day for 5 days followed by once a day for 2 days. Low molecular weight heparin and systemic antibiotics were administered. Blood glucose level was kept under control by injectable insulin. After diagnosis of bilateral ROCM, he was referred to our tertiary care centre.

On admission, he was started on intravenous liposomal Amphotericin B 300 mg /day in 5% dextrose over 3 hrs and Posaconazole 300 mg twice a day for 1 day and then 300 mg once a day, in conformity with the local protocol, along with general supportive care.

Figure 2. MRI Showing Bilateral PNS And Orbital Involvement.

Follow Up And Outcomes:

Patient had bilateral orbital involvement with intracranial extension. Hence, his general condition was bad with guarded prognosis. Considering the above scenario, retrobulbar Amphotericin B as well as exenteration was deferred. Even with systemic antifungal therapy, patient showed no improvement and succumbed after 2 weeks of hospitalisation.

DISCUSSION:

We report a high rate of ROCM cases presenting to our tertiary care centre during the second wave of COVID-19 pandemic. Multidisciplinary approach, timely intervention and standardised treatment protocols were our key strengths.

Even though this patient underwent timely intervention with Functional Endoscopic Sinus Surgery (FESS) and Diagnostic nasal endoscopy (DNE) the aggressive nature of disease with faster angioinvasion and intracranial extension led to a terminal outcome.

Management of ROCM involves control of high-risk factors like uncontrolled DM, immunosuppression along with optimal medical management with antifungal agents. Whenever indicated, aggressive surgical debridement with complete removal of necrotic tissue is performed. Some reports have concluded that Code Mucor stage 3c or worse has a poor prognosis for survival. Yet, in stage 4, surgical intervention in the form of FESS or exenteration has shown some improvement and reduced disease progression and mortality.[5] Orbital exenteration is considered lifesaving in cases with diffuse ROCM with extension limited to the orbit without or minimal extension to the cavernous sinus. [6, 7] Contrary to these reports, ROCM with cerebral involvement was found to be rapidly progressive with high mortality rate of around 30-90%. [8] In our case disease was bilateral with extensive intracranial skull base involvement with exposure of dura matter. Few series have shown orbital exenteration could be detrimental in such cases by further dissemination of the disease.[9,10]

Ultimate decision lies with the treating clinicians since there is no clearly accepted guideline for indications and timing of orbital exenteration. In our patient, we decided against exenteration. Studies haven't demonstrated significant difference in survival, with or without orbital exenteration. [6,7]

We conducted a literature search on PubMed (www.pubmed.gov) with the search terms 'bilateral mucormycosis' "COVID-19 associated mucormycosis" to identify any reported cases. Only a limited number of cases of bilateral ROCM have been previously reported. Out of 12 reported cases, all cases developed orbital infiltration with only two cases (16.7%) having bilateral involvement[2] A recent large collaborative multicentre COSMIC study has shown just 8.6% of cases with bilateral orbital involvement.[7]

To conclude, there should be a high index of suspicion of ROCM in all COVID -19 patients especially in presence of factors like diabetes, use of systemic steroid or immunosuppressant's, prolonged hospital or ICU stay, oxygen therapy and in ventilated patients. All physicians including ophthalmologists and ENT surgeons should be mindful of the probability of development of Mucormycosis in COVID-19 infected patients and screening of these patients should be done routinely. Education and awareness of recovered COVID-19 patients regarding clinical picture of ROCM can help in early diagnosis and management.

Summary:

High index of suspicion of ROCM in all COVID -19 patients with high risk factors to be kept in mind. Education and awareness of recovered COVID-19 patients regarding clinical picture of ROCM can help in early diagnosis and management.

REFERENCES

1. De La Paz MA, Patrinely JR, Marines HM, Appling WD. Adjunctive hyperbaric oxygen in the treatment of bilateral cerebro-rhino-orbital mucormycosis. *Am J Ophthalmol* 1992;114:208-11.
2. Fouad YA, Abdelaziz TT, Askoura A, et al. Spike in Rhino-Orbital-Cerebral Mucormycosis Cases Presenting to a Tertiary Care Center During the COVID-19 Pandemic. *Front Med (Lausanne)*. 2021;8:645270. Published 2021 May 28. doi:10.3389/fmed.2021.645270.
3. *Akshay Raut, Nguyen Tien Huy:* Rising incidence of mucormycosis in patients with COVID-19: another challenge for India amidst the second wave? *Lancet Respir Med* 2021, Published Online June 3, 2021 https://doi.org/10.1016/ S2213-2600(21)00265-4
4. Singh P. Black fungus: here is a list of states with highest number of mucormycosis cases. Hindustan Times. May 21, 2021. https://www. hindustantimes.com/india-news/black-fungus-states-with-highest-number-of-mucormycosis-cases-101621559394002.html (accessed May 28, 2021)
5. Honavar SG. Code Mucor: Guidelines for the Diagnosis, Staging and Management of Rhino-Orbito-Cerebral Mucormycosis in the Setting of COVID-19. *Indian J Ophthalmol* 2021;69:1361-5.
6. Songu M, Unlu HH, Gunhan K, Ilker SS, Nese N. Orbital exenteration: A dilemma in mucormycosis presented with orbital apex syndrome. *Am J Rhinol* 2008;22:98-103.
7. Sen M, Honavar SG, Bansal R, Sengupta S, Rao R, Kim U, *et al*. Epidemiology, clinical profile, management, and outcome of COVID-19-associated rhino-orbital-cerebral mucormycosis in2826 patients in India – Collaborative OPAI-IJO Study on Mucormycosis in COVID-19 (COSMIC), Report 1. Indian J Ophthalmol 2021;69:1670-92.
8. Prakash H, Chakrabarti A. Epidemiology of mucormycosis in India. *Microorganisms* 2021;9:523.
9. Hargrove RN, Wesley RE, Klippenstein KA, Fleming JC, Haik BG. Indications for orbital exenteration in mucormycosis. *Ophthalmic Plast Reconstr Surg* 2006;22:286-91.
10. Nithyanandam S, Jacob MS, Battu RR, Thomas RK, Correa MA,D'Souza O. Rhino-orbito-cerebral mucormycosis. A retrospective analysis of clinical features and treatment outcomes. *Indian J Ophthalmol* 2003;51:231-6.