Case Report

A rare case of rhino-orbito-maxillary mucormycosis having no common signs and laboratory findings but no visible morbidity

Aarti Mahesh Khare[1], Sachin Tukaram Nemane[2], Prashant Shridhar Javade[2], Yogesh Pralhad Narkhede[2], Punita A Parti[3]

From [1]Dental Surgeon, Department of Dental, [2]ENT Surgeon, Department of ENT, [3]Ophthalmologist, Department of Ophthalmology, FRU, NMMC Hospital, Navi Mumbai, Maharashtra, India

ABSTRACT

Mucormycosis is an opportunistic fungal infection caused by omnipresent fungi called Mucorales of class Phycomycetes. It mainly occurs in immunocompromised patients, and only early diagnosis with medical and surgical intervention remains the gold standard in managing it. Here, we present the case of a patient contracted with mucormycosis post his COVID-19 infection involving the rhino-orbito-maxillary area. The patient presented to our hospital with dental pain a month after his discharge. Since the mucormycosis cases were at a peak in this period, our team of doctors did a thorough examination of the patient, which revealed dental and ophthalmologic abnormalities. No clinical necrotic eschar in the palatine or nasal cavity was diagnosed, but magnetic resonance imaging (MRI) revealed a typical COVID-19 mucormycosis infection. Accordingly, prompt treatment with systemic amphotericin B was started. However, as the patient declined surgical intervention, we feared the worst outcome, which to our surprise showed no adverse progression.

Keywords: COVID-19-associated mucormycosis, Periodontal health, Post-COVID-19, Rhinomaxillary mucormycosis

Mucormycosis (earlier zygomycosis) is a serious but rare fungal infection was first described by Platauff in 1885 [1,2]. This fungus lives throughout the environment, especially in moist and humid places and even dust. The infection seldom occurs in an immuno-intact person because macrophages phagocytize the fungal spores, but affect people with underlying health problems or immunocompromised patients [3]. The mucor produces spores which rupture, get dispersed, and become airborne. People get infected after getting exposed to fungal spores [4].

Rhino-orbito-maxillary (ROM) mucormycosis, a subtype of rhinocerebral mucormycosis, is the most common of the five types of mucormycosis having a case fatality rate (CFR) of 30–70% [1,5,6]. The prevalence of COVID-19-associated mucormycosis (CAM) in India was 0.27% among patients managed in the hospital ward and 1.6% among patients managed in ICUs [5]. However, there was a 2.1-fold rise in mucormycosis cases since 2020 as compared to 2019, and research suggests that the increase is attributable to the COVID pandemic, hence the name CAM [4,5].

Here, we present the case of ROM mucormycosis in a post-COVID-19 scenario where the patient's compromised oral hygiene in a partially edentulous jaw led to an easy entry.

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CASE REPORT

An 82-year-old male patient was referred from Urban Health Primary Centre (UHP) of Chinchpada to FRU, Vashi, with complaints of toothache in the left posterior maxillary region for 1 month, eye swelling (left) for 20 days, and blurry vision for the past 10 days (Fig. 1a). Dental pain was chronic and continuous all day long, relieved temporarily only after taking painkiller (Tab: Dolo-650), whereas, there was periorbital pain in the left eye. Notably, the patient had no history of diabetes mellitus, hypertension, or asthma but had a prolonged stay in the hospital for COVID-19. The patient reported to FRU after a month of discharge.

On intraoral examination, the patient was a partially edentulous case with generalized periodontitis of the remnant teeth (17, 26, 34, 35, and 46) which were Grade 2 mobile having Miller's Class 2 recession. On the maxillary alveolar ridge mesially to 26, there was a pedunculated granuloma (polyp) of 4–5 mm, pink in color, soft to touch, and fixed but has fluctuant growth, the differential diagnosis for which was an epulis (Fig. 1b). Its presence can be suggestive of the mucosa reacting to what's going on inside the body. There was no evidence of cellulitis, or facial palsy, but had mild left-sided facial swelling. The patient did not have toothache

Correspondence to: Aarti Mahesh Khare, Ekta Society, D/1, 03, Sector-18, Nerul, Navi Mumbai - 400 706, Maharashtra, India. E-mail: aartik23@yahoo.in

before his hospitalization. An ophthalmic evaluation revealed upper and lower lid edema of the left eye, no proptosis, and no ptosis (Fig. 1a). Corneal sensation was normal and the cornea was clear. The right eye examination was normal. ENT scrutiny by anterior rhinoscopy was normal but there was maxillary sinus tenderness. There were no epistaxis, nasal obstruction, or nasal discharge. Based on these clinical findings and history, a provisional diagnosis of deep fungal infection was considered and the patient was admitted for further assessment.

Biochemical investigations for fasting and post-prandial blood sugar levels showed a normal glucose range. Besides elevated creatine on D1 (1.44 mg/dl), other biochemical parameters such as urea and electrolytes were also normal. Diagnostic 0 degree nasal endoscopy showed no nasal congestion, with a nasal septum and nasal turbinates' appearing normal (Fig. 2). Naturally, the KOH wet mount and fungal culture came negative (Fig. 3).

Magnetic resonance imaging (MRI) of brain, orbit, and paranasal sinus (Plain + Contrast) exhibited acute fungal invasion involving the left maxillary and ethmoidal sinuses with extension into the left orbit, left masticator space, and infratemporal fossa with bony erosion of the left maxilla, left zygomatic bone, and left half of the hard palate. Radiological findings confirmed the final diagnosis of CAM (Fig. 4).

The treatment now comprised of inj. amphotericin B (125 gm in 250 ml normal saline) to be administered for 3 weeks with optimum hydration and kept under follow-up.

On day 4 of admission, the patient underwent an extraction of 26 after complaining of no relief in pain. To rule out mucormycosis by dental path, swab and the histopathological specimen were taken from the dental socket, which had no blackish or crusty features. By day 7, the patient's toothache had reduced and the epulis showed signs of regression (Fig. 1c), but surprisingly, the histopathology report was inconclusive of mucormycosis. It showed aseptate pseudohyphae which was indicative of candidiasis. On day 21, the second MRI showed no signs of further progression of Mucormycosis, which could have been otherwise in the presence of DM and absence of Inj. Amphotericin B.

DISCUSSION

Mucormycosis generally develops secondary to immunosuppression or debilitating disease [7]. Throughout the history of mucormycosis, from the first case in humans reported by Platauf, in 1885, through the publication of the first observation of rhino-orbito-cerebral mucormycosis in 1943 by Gregory *et al*., to the report of the first known survivor in 1955 by Harris, little has changed in the diagnosis and outcome of this disease [3,8].

Clinical manifestations include five major types of mucormycosis – rhinocerebral, pulmonary, cutaneous, gastrointestinal, and disseminated. Out of which, rhinocerebral mucormycosis is the most common type though very little is known about it since it was not a reportable disease before [8]. Rhinomaxillary mucormycosis, a variant of rhinocerebral mucormycosis, is the most common, whose symptoms include proptosis, loss of vision, nasal discharge, sinusitis, palatal necrosis, and perforation [7,9]. In this case, however, all these symptoms were evasive. Even after 7 days of admission, the patient was conscious and valid with no further deterioration in the eyesight. As the nasal point of entry was ruled out, the portal of fungal exposure was possibly through reduced periodontium (periodontal pocket) provoked by steroid therapy in the hospital setting [10]. In the absence of predisposing factors like diabetes mellitus or others, and no near history of dental extraction, this route of ingress was probably novel though not unheard of.

Ordinarily, vascular invasion is the key pathophysiological feature of Mucorales infection but the normal blood sugar level of the patient did not instigate vascular progression [10,11]. In our patient, aseptate pseudohyphae was indicative of candidiasis, though pseudohyphae could also mean candidiasis-Mucorales infection.

This is a curious though infrequent case where despite confirmed radiological findings typical of mucormycosis, there was no histological conclusive proof of the Mucorales fungi. Factors of advanced age, fragile immunity (after fighting COVID-19), and unfavorable periodontal health (often the cause and effect of systemic disease) had not shown the progression of morbidity [12]. Thus at a normal sugar level, the risk of

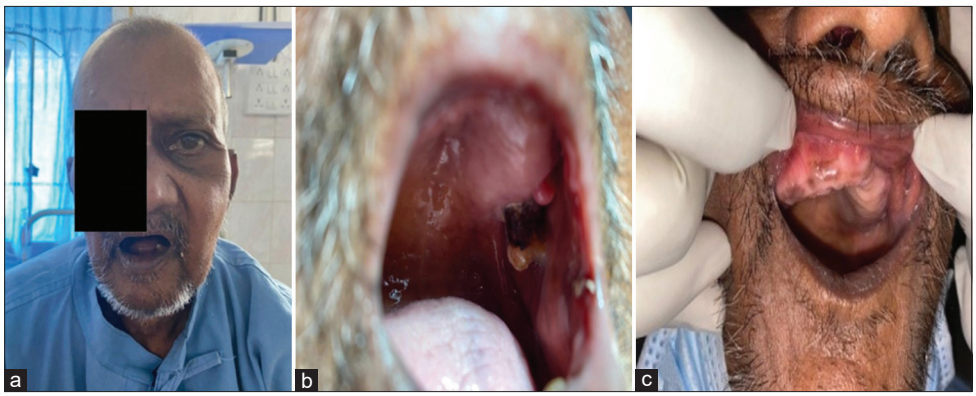

Figure 1: (a) Pre-operative photograph showing left eye infraorbital swelling; (b) maxillary left quadrant showing grade to mobile 27 and an epulis; (c) post-extraction of 27 showing no sign of epulis

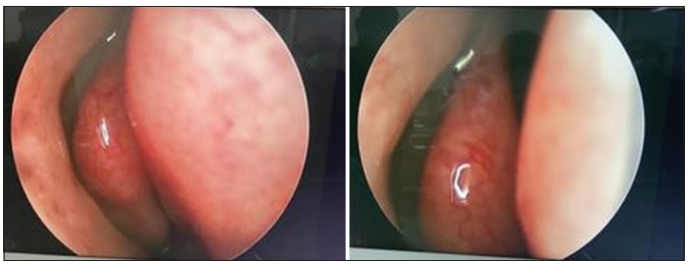

Figure 2: Nasal endoscopy showing normal nasal septum and nasal turbinates

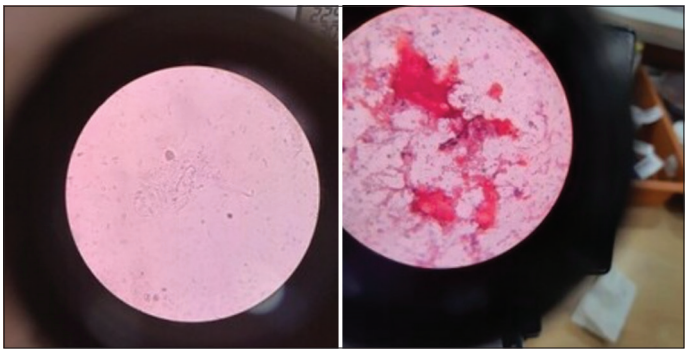

Figure 3: Dental socket of 26 scraped for histopathological diagnosis showed no fungal elements on KOH wet mount (left). Plenty budding yeast with pseudohyphae stained with H and E seen (right)

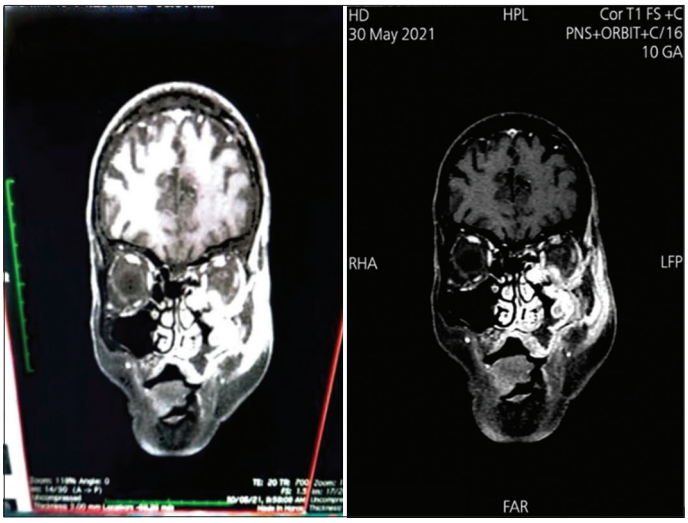

Figure 4: MRI showing fungal invasion of the left maxillary and ethmoidal sinuses with extension into the left orbit, left masticator space, and infratemporal fossa with bony erosion of the left maxilla, left zygomatic bone, and left half of hard palate. MRI taken on day 1 (left) and day 21 (right)

vasculature involvement and resultant fatality is highly reduced and the patient maintains the status quo if not surgically intervened [13]. Administration of amphotericin B, surgical debridement of infected tissue, correction of the underlying cause, and use of adjunctive hyperbaric oxygen (HBO) therapy remain the standard treatment [14]. However, the patient's sound systemic health prompted him to decline surgical procedure, who was just administered inj. amphotericin B until 3 weeks and followed up regularly.

CONCLUSION

Rhinomaxillary mucormycosis presents a diagnostic dilemma in terms of its clinical manifestation, especially when there are no classical signs and symptoms. This is especially crucial to understand and diagnose in a dental setting, wherein, the aftermath of the COVID-19 pandemic and discontinuation of the Epidemics Act (under which mucormycosis was a "Notifiable" disease), patients could easily be missed for a normal dental/periodontal case unrelated to any systemic findings. Since both nasal and periodontal pathways of exposure were ruled out in this case, at least from histopathological findings, it suggests undertaking further research to delineate course entry of Mucorales fungi.

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