Case Report

Mucormycosis of left maxilla: A case report and review

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ABSTRACT

Mucormycosis is an angioinvasive infection caused by fungi of the Zygomycetes order. *Rhizopus* is the main pathogen responsible for 90% of cases of cerebral mucormycosis. The term rhinocerebral mucormycosis should be used only in the face, palatal, orbital, paranasal sinus, or brain area. Here, we present the case of a 42-year-old man who presented with complaints of pain and swelling in relation to the left upper back tooth region for the past 20 days. In this case, the patient was immunocompromised due to type II diabetes, as well as COVID hospitalization. This case enlightens the investigation, as well as the management protocol followed by post-operative rehabilitation. Early diagnosis and prompt treatment can significantly reduce the morbidity and mortality of these deadly fungal infections.

Key words: Amphotericin B, Debridement, Maxilla, Mucormycosis

Mucormycosis is a rare opportunistic fungal infection caused by fungi belonging to the Mucorales and Mucoraceae families. It was first described in 1885 by Paultauf [1]. The disease usually affects immunocompromised people and rarely occurs in apparently healthy people [2]. Mucormycosis infection in cells causes rapid growth and proliferation of fungal organisms in deep tissue as a result of an altered immune response [3,4]. Mucormycosis is one of the most common and fatal fungal infections in humans, usually involving the nose and paranasal sinuses. Various predisposing factors to mucormycosis include uncontrolled diabetes (especially in patients with ketoacidosis), malignancies such as lymphoma and leukemia, kidney failure, organ transplants, long-term corticosteroids and immunosuppressive therapy, cirrhosis, burns, protein-energy deficiency, and immunosuppression deficiency syndrome [5]. Pathophysiology consists of either nasal or oral ingestion of spores or skin lesions. People with cellular and humoral defense mechanisms may not be able to respond adequately. The fungus can then spread to the paranasal sinuses and finally to the orbit, meninges, and brain. However, risk factors cannot be identified in patients with mucormycosis. If the infection extends to the nasal turbinates, the orbit can become involved. Infection can lead to proptosis, periorbital edema, chemosis, ophthalmoplegia, and loss of vision if the orbital apex becomes involved. Infection of the central nervous system (CNS) is usually attributed to the direct extension of the nose, paranasal sinuses, vascular channels, the supraorbital fissure, or the cribriform plate. If the disease invades the mouth, a black, necrotic eschar is often found on the palate, and ischemic, necrotic turbines may be found in the nose [6]. Successful management of this deadly infection requires aggressive and urgent medical and surgical intervention to detect the disease early and prevent the high morbidity and mortality associated with this disease process [7].

Here, we report a case of mucormycosis of the maxilla in a 42-year-old male with diabetes. The infection took a chronic course and resulted in extensive necrosis in the case presented here.

CASE REPORT

A 42-year-old male patient reported to the Department of Oral and Maxillofacial Surgery with a chief complaint of pain and mobile teeth in the upper left back region for 1 month. The pain was moderate to severe in intensity, gradual in onset, throbbing type, and continuous. The pain aggravated on palpation of the left side of the face-and-cheek region. The patient had a history of hospitalization for COVID-19 2 months back and was admitted for 15 days.

On examination, the patient was conscious and was well-oriented with time, space, and person. Vitals were stable

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with a blood pressure of 130/80 mmHg, heart rate of 73, respiratory rate of 14, and SpO_2 of 99%. Extraorally, there was no obvious facial asymmetry (Fig. 1a). Intraorally, a single diffuse swelling was present in the left maxilla extending from 12 to 28 region and palatally involving the middle one-third of the palate. The size of the swelling measures approximately 5×1 cm, erythematous in color along with the presence of multiple sinus openings. The swelling was tender on palpation, firm in consistency, and pus discharge was evident. Grade I mobility was present with 12, 21, 22, 23, 24, 25, 26, 27, and 28 teeth (Fig. 1b).

The patient underwent computed tomography facial bone that showed multiple bony erosion involving the maxillary sinus walls with minimal adjacent soft-tissue thickening and fat stranding (Fig. 2).

The patient underwent an incisional biopsy and the histopathological report revealed acute on chronic non-specific inflammation with suppurative changes and granulation tissue likely to be mucormycosis (Fig. 3). All the mandatory investigations were done, and anesthesia fitness was obtained as the American Society of Anesthesiologists II, following which the patient was planned for the surgical procedure under general anesthesia. The patient underwent the surgical procedure under nasoendotracheal intubation, general anesthesia was induced and maintained, and local anesthesia with adrenaline in filtration was given in to 12–28 region. A crevicular incision was placed from 13 to 28 region using 15 size blade. Trapezoidal mucoperiosteal flap was elevated using the molt periosteal elevator. Necrotic bone was visualized and complete debridement of the bone was done with bone scoop and bone rongeur. Adjacent healthy bones were drilled until a fresh bleed got induced from the site, and complete debridement of necrotic bone was done. Following this, thorough irrigation was done and the primary closure was done with Vicryl 3-0 (Fig. 4). The patient was extubated immediately after the surgery and the recovery was uneventful.

The patient was advised to take post-operative oral hygiene care. Injection amphotericin B (Amph-b) deoxycholate was given for 15 days post-operatively with the periodic nephrologist and diabetologist opinion. The patient was followed up for 6 months wherein the outcome was uneventful (Fig. 5). The patient was referred to the Department of Prosthodontics for rehabilitation with a maxillary arch with the prosthetic obturator.

Figure 1: (a) Pre-operative and (b) intraoral pictures of the patient

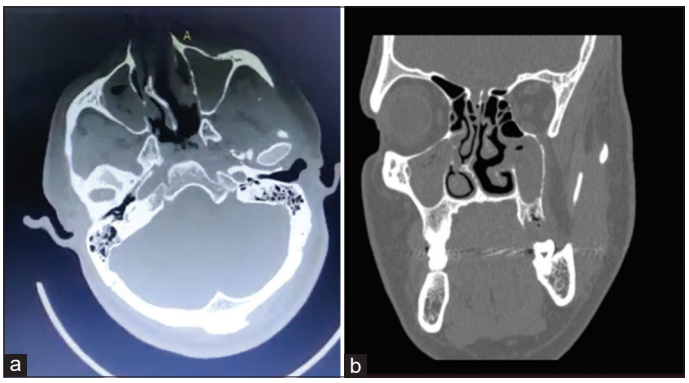

Figure 2: Pre-operative computed tomography picture of the patient

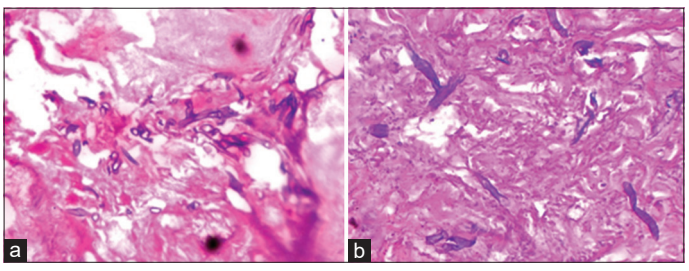

Figure 3: Histological pictures of the patient

DISCUSSION

Maxilla is an essential bone with a rich vascular supply forming the roof of the oral cavity. Mucormycosis is one of the most common fungal infections, which affects the maxilla, especially in diabetes and immunocompromised patients. Hingad *et al.* confirmed that patients with uncontrolled diabetes have a higher risk of developing mucormycosis due to immunosuppression and the use of ketone bodies in such patients [8]. In most

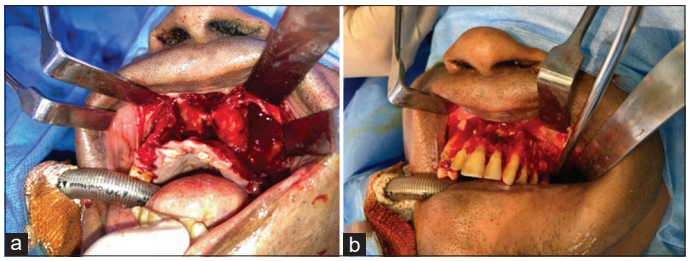

Figure 4: Intraoperative picture of the patient

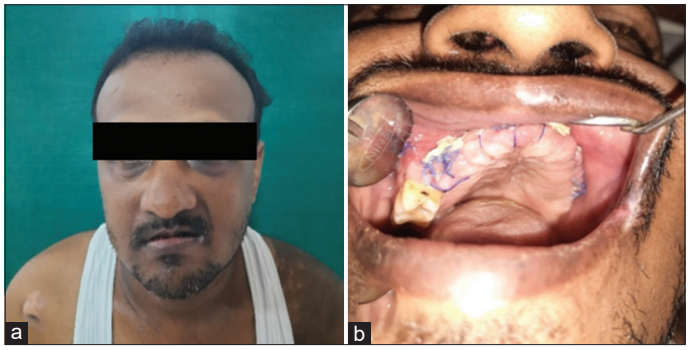

Figure 5: Post-operative picture of the patient (a) Clinical picture; (b) Intraoral picture

immunologically competent hosts, these spores were present with a phagocytic response. If the phagocytic response fails, the inflammation continues, leading to the development of hyphae; thus infection occurs.

Oral mucormycosis can have two origins. One is the spread of infection through the inhalation entry portal (usually through the nose) and the other is from direct wound contamination. When it occurs in the nose and paranasal sinuses, the infection appears black and leads to the development of necrosis. In case of direct wound contamination, the clinical findings may appear anywhere in the oral cavity, including the mandible. Cavernous sinus thrombosis is a serious complication of maxillary infections. Another difference is the rarity of mandibular infections as compared to the maxillary. If the infection extends to the nasal turbinates, the orbit can become involved. Infection can lead to proptosis, periorbital edema, chemosis, ophthalmoplegia, and loss of vision in case of orbital apex involvement. Infection of the CNS is usually attributed to direct extension of the nose or paranasal sinuses or through vascular channels, the supraorbital fissure, or the cribriform plate. If the disease invades the mouth, a black, necrotic eschar is often found on the palate, and ischemic, necrotic turbines may be found in the nose. As mucormycosis often invades blood vessels, infarction, necrosis, and thrombosis are the major characteristics.

Mucormycosis has been reported in patients with severe COVID-19 or those recovering from the disease and have been associated with severe illness and death. The use of systemic corticosteroids/other immunomodulating drugs for mild or moderate patients with COVID-19 should be avoided. Imaging helps in assessing the extent of disease, and identification of complications such as internal carotid artery thrombosis which is indispensable for surgical planning.

Imaging techniques including magnetic resonance imaging (MRI) show only non-specific features such as mucosal thickening during the early stage of the disease which may delay diagnosis. Therakathu *et al.* have shown that MRI can detect cavernous sinus invasion and vascular complications such as thrombosis and ischemia [9]. According to Razek, the involvement of cavernous sinus appears hypointense on T1 and T2 with intense inhomogeneous post-contrast enhancement [10]. Biopsy of infected tissue remains the gold standard for the diagnosis of mucormycosis [11].

Various treatment modalities were used for the management of COVID-19-associated rhino-orbital-cerebral mucormycosis (ROCM) patients among which, a few of them were treated by using only antifungal drugs either alone or in combination with other antifungal drugs. The common antifungal drugs used were Amph-b, liposomal Amph-b, posaconazole (POS), itraconazole (ITC), voriconazole, caspofungin, vancomycin, and meropenem. Various combinations followed were Amph-b with caspofungin, Amph-b with posaconazole, Amph-b with posaconazole and caspofungin, meropenem with vancomycin, and liposomal Amph-b infusion with posaconazole. The use of amphotericin in patients with mucormycosis has been widely published and accepted, with a survival rate as high as 72%. Although treatment combined with surgery and Amph-b achieves a cure rate of 80%, many of the survivors will experience some form of functional deficit (e.g., blindness or cranial nerve palsy). Although Amph-b formulations remain the mainstay of mucormycosis treatment, recent studies have demonstrated the efficiency of posaconazole, a broad-spectrum triazole agent for patients with serious invasive fungal infections. Posaconazole may be an attractive alternative for patients who are intolerant to Amph-b or whose use is limited by nephrotoxicity [12]. As part of surgical debridement, the majority of patients underwent functional endoscopic sinus surgery. The approach used for the treatment of mucormycosis was to go as invasive as possible in the first attempt depending on the extent of the lesion. This approach seemed to be the most effective and decreased the mortality rate but the quality of life was questioned upon. The procedures such as maxillectomy, orbital exenteration, and orbital decompression were done as a form of surgical management. Even though these procedures add to decreased quality of life, they surely give reasons to believe that early antifungal drug therapy along with invasive surgical management has reduced the mortality rate in such patients. This should include the debridement of all infected and necrotic dead tissue [13].

An unprecedented mucormycosis outbreak occurred in India during the 2nd COVID-19 wave in the spring of 2021. COVID-19-associated mucormycosis (CAM) was observed, mainly ROCM, in patients with poorly controlled diabetes and treated with inappropriate doses of glucocorticoids. Moreover, in India, the patients with CAM were more likely to have diabetes mellitus and ROCM; conversely, mortality rates were lower. The reasons for such a localized epidemic in India have remained unclear, but some hypotheses can be put forward, particularly the combination of the high prevalence of uncontrolled diabetes mellitus and frequent indiscriminate corticosteroid utilization in a country that already had a high mucormycosis burden before the COVID-19 pandemic.

CONCLUSION

Mucormycosis has been considered an infectious disease but with early medical and surgical management, the survival rate is now thought to be over 80%. Early recognition of mucormycosis is important to limit the spread of infection, which can lead to high morbidity and mortality. Therefore, health-care professionals should be familiar with the signs and symptoms of the disease and should be very suspicious of people with diabetes. Therefore, early diagnosis, the use of Amph-b, aggressive surgical intervention, and modification of the underlying disease are the keys to improved outcomes for patients with mucormycosis.

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