Unravelling the Complexity of Mucormycosis-A Rare Case Report

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Abstract

Rationale: Mucormycosis is a rare, life-threatening, invasive fungal infection often referred to as black fungus. It has gained significant attention due to its increasing incidence during the coronavirus pandemic of 2019. **Patient Concern:** An 8-month-old child, whose identity is being withheld, arrived at our clinic with her parents with the chief complaint of swelling in the upper lip and cheek. **Diagnosis:** Various laboratory procedures, including blood cultures and imaging scans were performed to determine the presence of mucormycosis. **Treatment:** Under general anaesthesia, decortication and resection was done surgically, followed by an intraoral elastomeric impression made over the resected region. Soft silicone splints as oral seals for the suckling reflex were made postoperatively within a week. Immediate post-operative therapeutic low-level laser therapy was done. **Outcome:** Wound healing has been achieved. **Take-away Lessons:** Multidisciplinary intervention provides the best outcomes for the successful treatment and rehabilitation of paediatric patients with mucormycosis of the facial region.

Keywords: Black fungus, Coronavirus pandemic of 2019, Integra Dermal Template, Low-level laser therapy, Mucormycosis

Introduction

Mucormycosis is an invasive fungal infection caused by a *Mucorales* fungus.[1] During the pandemic, there was a rise in mucormycosis cases among coronavirus disease 2019 (COVID-19) patients.[2,3] Mucormycosis can manifest in the gastrointestinal system, skin, lungs, and other facial structures.[4,5]

Diagnosing mucormycosis is challenging and laboratory testing and radiographic imaging can be helpful. With a diagnosis, the affected tissues should be surgically debrided and antifungal therapy administered.[6] Mucormycosis in children has particular problems in terms of diagnosis, management and results. Vigilance and early action are critical for improving patient outcomes. This case study reports the presentation, diagnosis and treatment for an 8-month-old child with a positive history of COVID-19 infection.

Case Report

An 8-month-old child was referred to our hospital with an erythematous swelling involving the upper lip and right cheek for the past month. Initially, the patient had redness and a painful swelling with a foul smell [Figure 1a], which gradually increased in size and led to ulcerative necrosis of the pre-maxilla and right cheek region in 2 months [Figure 1b] from the first symptom. Before admission, the disease rapidly progressed and caused perforation in the pre-maxilla and right cheek region. The child had a positive history of COVID-19 for 1 month, and the parents also had a post-COVID phase for 15 days. There was no history of facial trauma, dental treatment, recent surgery or known sinusitis. The patient had a normal nutritional status without any known underlying conditions.

A multidisciplinary approach is usually required due to the complexity of the diagnosis of mucormycosis in children. Various laboratory tests were performed, including blood cultures, histopathology reports [Figure 2], and imaging scans.

Access this article online

Quick Response Code:

Website: https://journals.lww.com/aoms

DOI: 10.4103/ams.ams_204_23

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Received: 18-10-2023 **Last Revised:** 08-04-2024
Accepted: 17-04-2024 **Published:** 19-07-2024

How to cite this article: Deenadayalan N, Pandyan PD, Satheesh C, Aparnaa M, Samyo GS. Unravelling the complexity of mucormycosis-A rare case report. Ann Maxillofac Surg 2024;14:96-8.

Reverse-transcriptase polymerase chain reaction for severe acute respiratory syndrome coronavirus - 2 (COVID-2) from a nasopharyngeal swab was positive. Histopathology findings from the necrotic tissue showed an inflammatory smear with a chronic granulomatous inflammatory process and a suspicion of fungal infection. Head computed tomography (CT) results showed soft-tissue defects and bony erosion on the pre-maxilla and right buccal region. In addition, a biopsy of the affected tissue was also conducted to confirm the diagnosis. The suspicious crust or black necrotic tissue was sent for microbiology testing by calcofluorescence white stain, which confirmed that it was a fungal infection caused by *Mucorales*. After the confirmation, further management techniques and the procedure of limited debridement were explained to their parents, and consent was given from their side. Hence, we proceeded with the surgical management of the diagnosed disease.

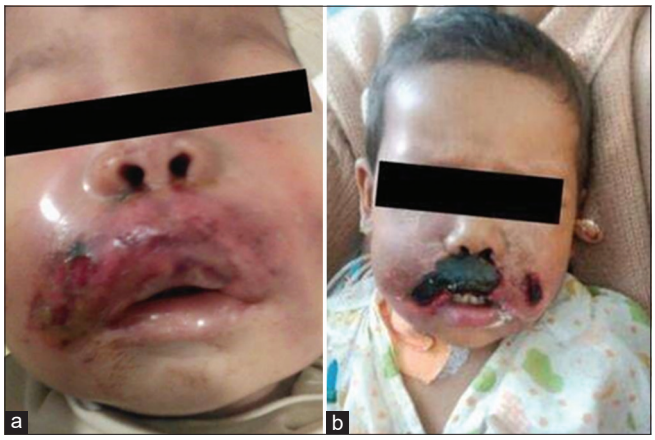

Figure 1: (a) Swelling over the upper lip and cheek (b) Upper lip and left lateral cheek had an avascular necrosis by 2 months

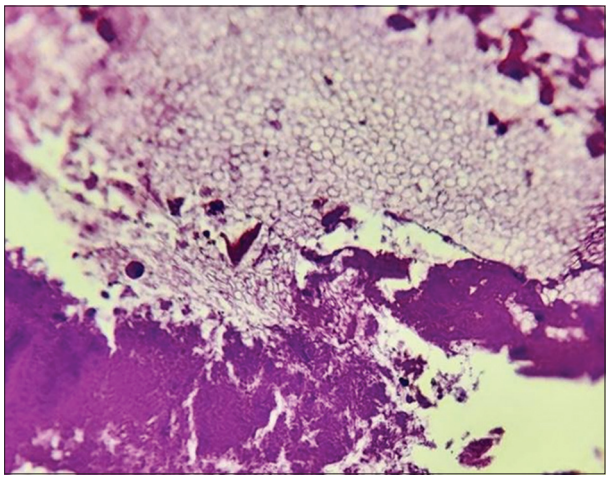

Figure 2: Histopathology picture - mucormycosis (H&E Staining with 10 x magnification)

Treatment for mucormycosis in paediatric patients requires a meticulous and targeted approach due to the unique challenges posed by the infection and the vulnerability of children. In this case, the treatment strategy did not involve antifungal therapy, which led to surgical intervention. While antifungal medications like amphotericin B are crucial in combating fungal growth in adults, their use in children comes with risks, such as hepatomegaly. Therefore, surgical intervention was opted for to address the underlying cause of mucormycosis.

The surgical approach included excision, decortication and the placement of dermal regeneration templates. Excision involved the removal of necrotic tissue and a portion of the upper lip under general anaesthesia with oral intubation. Decortication of the labial cortex was performed to accelerate bone regeneration and prepare the area for further procedures [Figure 3a]. Post-surgery, intraoral elastomeric impressions were taken to create a mould of the surgical site, which could be used for prosthetics.

Dermal regeneration templates were used to encourage the regeneration of the skin's dermal layer. An Integra Dermal Template was placed over the upper lip and cheek region [Figure 3b]. A percutaneous endoscopic gastrostomy was also performed to administer enteral nutrition, as the patient was unable to eat or swallow.

Immediate post-operative care involved the dislodging of dermal regeneration templates and the creation of soft silicone splints to protect the surgical site and aid in oral feeding. Low-level laser therapy was used for biostimulation [Figure 3c], which helped in reducing pain and inflammation, improving tissue repair and increasing circulation. It was used for the child in noncontact mode, 0.5 volts split for 200 ms for 15–20 min twice daily for about 21 days (Dr.N.Deenadayalan's Criteria). It facilitated the removal of the feeding tube and the resumption of oral feeding.

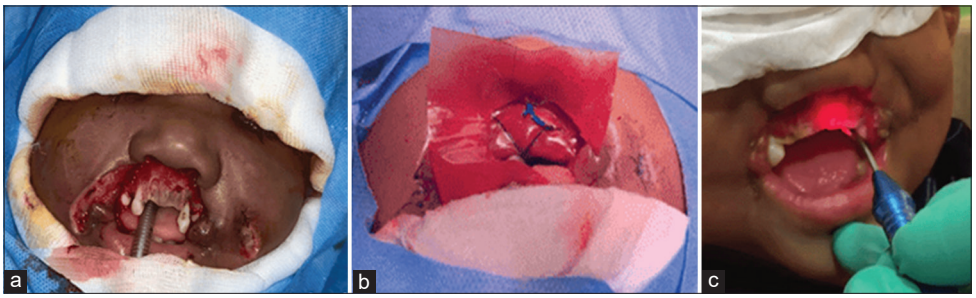

Figure 3: (a) Excision and decortication done both intraoral and extraoral, along with removal of two primary incisors (b) Dermal regeneration templates were placed. (c) Protocol of low level laser therapy followed 0.5 mV/200 ms for 21 days

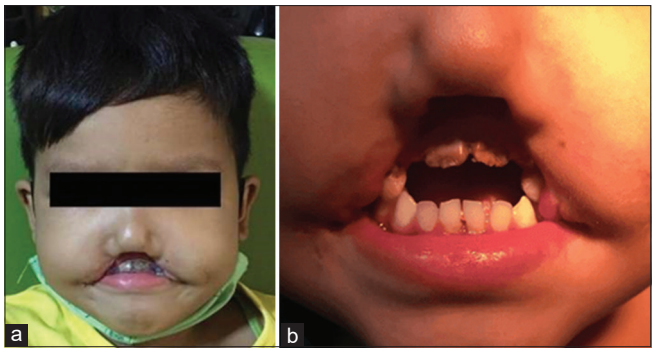

Figure 4: (a) Silicone splint fabrication; (b) Follow-up within 3 years (note: presence of permanent incisors, which may be due to removal of the buccal cortical bone during surgical debridement and retention of palatal shelf, triggering the early eruption)

Follow-up examinations showed significant wound healing with no further signs of mucor. A soft silicone splint was placed on the maxillary arch to protect the healing area and maintain occlusion. Regular follow-up appointments were scheduled to monitor the healing process and adjust the splint if necessary.

After 3 years, the patient showed complete wound healing, and the team is planning to restore the upper lip's form and function using Abbe's flap technique, which is a versatile procedure for lip reconstruction that offers good functional and aesthetic results [Figure 4a and b].

This case highlights the complexity of treating mucormycosis in paediatric patients and the importance of a multidisciplinary approach involving oral and maxillofacial surgeons, paediatric surgeons and other health-care professionals to achieve optimal outcomes.

Discussion

Mucormycosis, also known as zygomycosis, is a serious and potentially life-threatening fungal infection caused by a group of moulds known as mucormycetes. These fungi are commonly found in soil, decaying organic matter and in the environment. Risk factors for mucormycosis include uncontrolled diabetes, cancer, renal failure, organ transplantation, long-term corticosteroid and immunosuppressive therapy, cirrhosis, burns, protein–energy malnutrition and acquired immune deficiency syndrome. Mucormycosis can manifest as gastrointestinal, disseminated, pulmonary or rhinocerebral infections, but in this case, it was limited to the maxillary region.[7]

Numerous cases of oral mucormycosis have been documented in the literature. A 65-year-old male patient with diabetes and immunological impairment was reported to have mucormycosis of the maxilla, which was treated with a detachable prosthesis by Gupta *et al.*[8] There are additional reports of two cases by Bakathir,[5] one of which involved a 14-year-old boy who was undergoing chemotherapy for acute myeloid leukaemia and had oral mucormycosis of the maxilla and mandible.[9]

Patients with the COVID-19 infection may experience hypoxia, weakness and impaired immunity. Malaria, COVID-19, rheumatoid arthritis and lupus are mostly treated with hydroxychloroquine, an anti-inflammatory and immunosuppressive medication.[10] As a result, it can lower patients' immune systems, which in turn promotes fungal development.[9]

The patient underwent labial cortex decortication and resection, followed by intraoral elastomeric impressions and soft silicone splints for the suckling reflex. Abbe's flap was chosen for upper lip deformities affecting up to two-thirds of the upper lip and lateral defects, as long as the commissure or philtrum is intact. This two-stage lip-switch flap is effective for philtral reconstruction and is ideal for female patients. Silicone splints can be used as a temporary therapeutic option for individuals with decortication to stop food or liquid aspiration, phonetics, swallowing and mastication. In this study, wound healing was achieved, with no further mucor seen intraorally or extraorally.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the legal guardian has given his consent for images and other clinical information to be reported in the journal. The guardian understands that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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