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Multifaceted Challenges in Recurrent Rhino-Orbital Mucormycosis: Concomitant Oronasal Myiasis and Lichtheimia Co-Infection in a Post-COVID-19 Patient

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Abstract

Post-COVID-19 rhino-orbital mucormycosis poses significant challenges, particularly when complicated by co-infections. We present the case of a 60-years-old male with Type II diabetes, who developed recurrent rhino-orbital mucormycosis with concomitant oronasal myiasis and Lichtheimia co-infection. The patient presented with facial swelling, vision loss, and oral maggots. Imaging revealed necrotic bone exposure and residual mucormycosis in the paranasal sinuses. Fungal culture confirmed Lichtheimia species. Multidisciplinary management included manual maggot removal, necrotic tissue debridement, retrobulbar Liposomal Amphotericin B injections, and glycemic control. A total of 72 maggots were extracted from the maxillary antrum.

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The patient received 6600 mg of Liposomal Amphotericin B and retrobulbar injections over six months, resulting in complete recovery with no recurrence. This case underscores the importance of early diagnosis and a comprehensive therapeutic approach for complex post-COVID-19 infections. It highlights the potential for preserving vision and avoiding radical procedures, such as eye exenteration, through timely intervention. Effective management of co-infections and meticulous surgical care are crucial in preventing severe complications and improving outcomes in immunocompromised patients.

Keywords

Rhino-Orbital Mucormycosis; Oronasal Myiasis; Lichtheimia; COVID-19 Patient.

Introduction

The emergence of rhino-orbital mucormycosis as a significant complication following COVID-19 has raised considerable concern, particularly in patients with underlying comorbidities such as diabetes mellitus. Mucormycosis is a rare but severe fungal infection that predominantly affects immunocompromised individuals, frequently leading to substantial morbidity and mortality if not diagnosed and managed promptly. The COVID-19 pandemic has exacerbated this risk, primarily due to the widespread use of glucocorticoids and other immunosuppressive therapies employed to manage severe cases of COVID-19, thereby further compromising the immune response of affected patients.

This case report details an unusual presentation of recurrent rhino-orbital mucormycosis complicated by concomitant oronasal myiasis and co-infection with Lichtheimia in a post-COVID-19 patient. Myiasis, characterized by infestation by dipterous larvae, adds a significant layer of complexity to the clinical presentation, necessitating timely identification and intervention. The simultaneous occurrence of these conditions emphasizes the necessity for a multidisciplinary approach to ensure comprehensive patient care.

Through this case report, we aim to enhance awareness regarding the potential complications associated with post-COVID-19 infections, underscoring the urgent need for early diagnosis and targeted therapeutic strategies. This serves as a pertinent reminder of the multifaceted challenges encountered by healthcare providers in managing complex infectious scenarios in immunocompromised patients.

Case Report

A 60 years old male patient presenting with significant physical impairments, underwent Functional Endoscopic Sinus Surgery (FESS) for post-COVID-19 rhino-orbital mucormycosis. He reported a one-week history of generalized facial pain, thick greyish nasal discharge, and recent onset of vision loss in the right eye. Additionally, the patient noted the presence of multiple maggots within his oral cavity. His medical history was notable for Type II diabetes mellitus, managed pharmacologically for the past seven years.

Extraoral examination revealed diffuse swelling on the right side of the face, extending from the ala of the nose to the preauricular region, and from the infraorbital rim to the lateral commissure of the mouth. The swelling was non-tender, non-compressible, and non-fluctuant, with no localized warmth upon palpation.

Right-sided periorbital edema and ptosis were also evident (Figure 1).

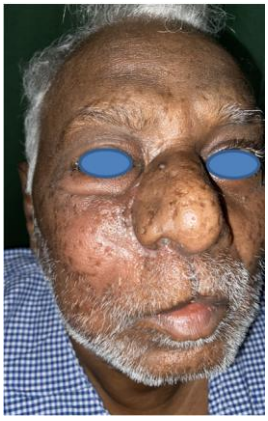

Figure 1: Extraoral view showing diffuse right facial swelling with periorbital edema and ptosis, extending from the infraorbital rim to the lateral commissure and from the ala of the nose to the preauricular region.

Multiple live maggots were observed migrating towards the skin surface from a 3x3 mm puncture wound located on the right ala of the nose. Nasal examination showed thick, foul-smelling greyish discharge, with no evidence of extraoral sinus tracts or purulent discharge, and cervical lymphadenopathy was absent.

Intraoral examination disclosed exposed necrotic bone measuring 3 cm x 1 cm in the right maxillary buccal vestibule, extending from the maxillary canine to the second molars. Perforation of the anterior wall of the right maxillary sinus was noted, with numerous motile larvae present within the necrotic tissue. The gingival and palatal examination revealed signs of inflammation, edema, and generalized bleeding upon probing, indicative of poor oral hygiene and chronic periodontal disease.

Routine laboratory investigations indicated leukocytosis, an elevated erythrocyte sedimentation rate (ESR) of 40 mm/hour, a random blood sugar level of 350 mg/dL, and a Glycated Hemoglobin (HbA1C) of 8 %. An endocrinologist was consulted to manage the hyperglycemia, resulting in the initiation of Regular Insulin therapy. Furthermore, an ophthalmological consultation was sought due to the patient's vision loss in right eye, which led to a recommendation for five weekly retrobulbar injections of Liposomal Amphotericin B, thus averting the need for eye exenteration.

MRI of the brain, orbit, and paranasal sinuses revealed persistent mucosal thickening in the right frontal, ethmoidal, sphenoid, and maxillary sinuses. Short tau inversion recovery (STIR) hyperintensity was detected along the floor and medial wall of the orbit, with diffuse thinning of the right optic nerve and extraocular muscles. Contrast-enhanced computed tomography confirmed the presence of residual mucormycosis in the right paranasal sinus.

Further diagnostic evaluations included potassium hydroxide (KOH) testing of necrotic tissue samples, which revealed broad, branched, aseptate fungal hyphae consistent with mucormycosis. Postoperative fungal cultures identified Lichtheimia as a co-infecting pathogen within the necrotic tissue, alongside Rhizopus, thereby confirming its role in the mucormycosis. Diagnosis of myiasis was established through

clinical evaluation and identification of maggots. Thus, a definitive diagnosis of myiasis with Lichtheimia co-infection in the right maxillary sinus post-COVID-19 rhino-orbital mucormycosis was confirmed based on clinical, histological, fungal culture, and radiological findings.

Management encompassed comprehensive counseling and psychological support for the patient and his relatives. The integrated medical-surgical approach involved manual removal of maggots using blunt tweezers and turpentine oil-soaked cotton pellets within the necrotic debris, followed by debridement of necrotic tissue. A treatment protocol was implemented comprising Ivermectin tablet in conjunction with parenteral and local administration of Liposomal Amphotericin B. Furthermore, retrobulbar administration of Liposomal Amphotericin B was conducted by an ophthalmologist.

Application of turpentine oil facilitated maggot expulsion, and antibiotic prophylaxis was administered under aseptic conditions. Maxillary nerve anesthesia was achieved through an extraoral approach, followed by transoral right-sided infrastructure maxillectomy, which entailed thorough debridement of necrotic tissue. A total of 56 larvae, measuring approximately 1 x 0.3 x 0.3 cm, were meticulously excised during the procedure (Figures 2 and 3). The integrity of the maxillary sinus roof was preserved, and it was packed with gauze soaked in turpentine oil.

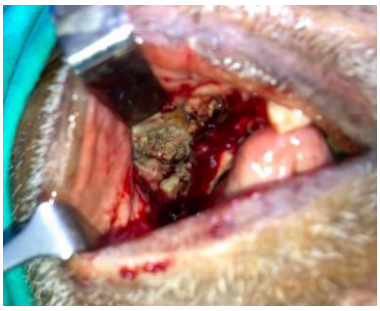

Figure 2: Perforation of the anterior wall of the right maxillary sinus showing multiple motile larvae embedded within necrotic tissue.

Figure 3: Live maggots retrieved from the right maxillary sinus following surgical debridement.

Postoperatively, the patient was initiated on a daily dose of Ivermectin 12 mg and a course of intravenous antibiotics for five days. Liposomal Amphotericin B injections commenced on postoperative day one to address residual mucormycosis, with careful monitoring of hepatic and renal functions. Regular Insulin was continued for ongoing hyperglycemia management. To ensure complete eradication of larvae, the

surgical site underwent re-exploration for three consecutive days, confirming the removal of 72 maggots from the necrotic right maxillary antrum, including 16 larvae expelled preoperatively.

The patient continued antifungal therapy, receiving a cumulative dose of 6600 mg of Liposomal Amphotericin B, alongside five retrobulbar injections at weekly intervals. Daily wound care was performed utilizing Acriflavin-soaked gauze over three weeks. Throughout treatment, the patient adhered to a high-protein liquid diet via a Ryles tube.

Six months post-treatment, the patient exhibited complete recovery, with no signs of disease recurrence, as confirmed by Direct Nasal Endoscopy conducted by the ENT team and an MRI of the head. Improvements in vision were noted during the ophthalmological examination of the affected eye. An impression of the intraoral residual tissue defect was obtained to facilitate the fabrication and delivery of a definitive obturator aimed at restoring oral function and aesthetics (Figure 4). The collaborative efforts of the medical and OMFS teams, coupled with the patient's compliance with the treatment regimen, culminated in a successful outcome and sustained recovery.

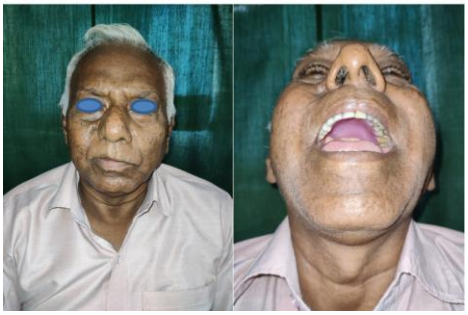

Figure 4: Extraoral and intraoral views demonstrating complete patient recovery and restoration of oral function, with the residual maxillary defect effectively rehabilitated using a definitive obturator one-year post-treatment.

Discussion

Following the emergence of the COVID-19 pandemic, there has been a notable escalation in the frequency of opportunistic infections, with mucormycosis being a prominent example. This heightened susceptibility to opportunistic fungal infections can be attributed to the presence of comorbid conditions, such as diabetes mellitus, coupled with the concurrent administration of glucocorticoid therapy and other immunomodulatory agents for the treatment of moderate to severe COVID-19 cases. This unique case report details the successful treatment of a complex presentation of Rhino-orbital mucormycosis with concomitant Lichtheimia coinfection and Oronasal Myiasis in a post-COVID-19 patient. This report describes the comprehensive approach that included manual maggot removal, necrotic tissue debridement, Liposomal Amphotericin B injections, and the management of hyperglycemia [1].

Myiasis is an ectoparasitic infestation of viable or necrotic tissues by the dipterous larvae of higher flies. Prevention of Myiasis requires a comprehensive approach, which includes maintaining good oral hygiene, controlling the fly population, educating parents and caretakers and providing regular access to primary health services.

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Mucormycosis is usually found in the head and neck, and the first symptoms often emerge in the oral cavity. Therefore, a dental surgeon may be the first healthcare expert to identify symptoms of this dangerous and potentially fatal condition. A prompt diagnosis and appropriate therapy are necessary for the patient's life.

A multicentric, Retrospective, observational study of patients with COVID-19-associated rhino orbital mucormycosis was conducted in various hospitals in India from January 1, 2020, to May 26, 2021. Of the 2826 patients, the mean age of patients was 51.9 years, with a male predilection (71%). 87% of the patients were treated with corticosteroids (21% for > 10 days). Diabetes mellitus (DM) was present in 78% of all patients [2].

Mortality was high at 49%, which was particularly due to patients with pulmonary or disseminated mucormycosis or cerebral involvement. Furthermore, a substantial proportion of patients who survived had life-changing morbidities, e.g., loss of vision in 46% of survivors [3].

Reversal of immunosuppression, systemic Amphotericin B antifungal chemotherapy, and surgical debridement are the pillars of mucormycosis treatment. The use of high-dose Liposomal Amphotericin B as a first-line treatment is strongly suggested, whereas intravenous Isavuconazole and intravenous or delayed-release tablet Posaconazole are somewhat indicated [4].

In cases of orbital mucormycosis where severe orbital debridement is not preferred and the burden of orbital illness is insignificant, retrobulbar injection of amphotericin B may be a viable therapeutic alternative. Lakshmi Ramamurthy et al. evaluated the outcome of transcutaneous retrobulbar injection of liposomal amphotericin B in post-COVID-19 rhino-orbito-cerebral mucormycosis. They found it a successful and safe treatment strategy in mild to moderate rhino-orbito-cerebral mucormycosis [5].

Recovery of lost vision gives scope to further research and hope to Rhino-orbital Mucormycosis patients with vision loss and thus morbidities associated with eye exenteration can be avoided. This Case report highlights the need for assessing the presence of and management of Oral or Nasal Myiasis and coinfection with other pathogens in post-COVID-19 Mucormycosis patients who are immunocompromised, diabetic, or are mentally or physically challenged who are not able to maintain good oral or surgical site hygiene on their own.

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Conflict of Interest

The authors declare no conflict of interest.

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