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Cutaneous mucormycosis with maxillary sinus fistula as a presenting feature of COVID-19: A rare case report

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ABSTRACT

Deadly secondary infections caused by Rhizopus species leading to mucormycosis have taken the world by storm. Most common presenting features of COVID-19 remain congectival congestion, headache and blurring of vision. However mucormycosis with a fistula manifesting as the primary symptom of COVID-19 is rare and this is the first case report of its kind to best of our knowledge. We report a case of 61 year old female who presented with a pus draining fistula on right side of her face. A Nasopharyngeal swab came positive for COVID-19 by Reverse Transcriptase polymerase chain reaction method. MRI para nasal sinus was suggestive of fungal sinusitis. Debriedment was done for the same and a swab was sent which showed growth of Rhizopus species on culture.

Keywords: COVID-19, Mucormycosis, Maxillary sinus fistula

1. INTRODUCTION

There has been a drastic increase in secondary infections seen in COVID-19 with increasing use of glucocorticoids in the treatment of COVID-19 as well as predisposition to COVID-19 in diabetic patients. Use of immunomodulatory drugs such as monoclonal antibodies against inflammatory markers which play vital role in COVID-19 also lead to dysregulation of immunity which predisposes to opportunistic infections in COVID-19. These opportunistic infections can be bacterial or fungal. COVID-19 pandemic has been a testing time for physicians all around the world with its everchanging presentations and vast group of organ systems involved. Infection with severe acute reparatory syndrome Coronavirus 2 might range from an asymptomatic patient to mild upper respiratory tract infection to life threatening pneumonia. While there is no single proven treatment for COVID-19, increasing group of medications are being tested for efficacy in this lethal disease. There has been a new twist in the tale of this pandemic by emergence of seconday fungal infection by rhizopus species leading to Mucormycosis. There has been

increased reporting of "Black Coloured Growth" which were seen in patients with COVID-19 which upon investigations turned out to be mucormycosis. While the chief presentation has been rhino orbital mucormycosis there has been rare reporting of pulmonary mucormycosis. A dermatological manifestation with isolated fistula dragning pus from sinus cavity is an even more rare presentation of COVID-19.

Mucormycosis is a highly infectious as well as progressive disease which can be life threatning at times. It is caused by mucormycetes moulds (Radulesco et al., 2020). It is known to infect patients with immunocompromised state. Diabetes is a leading cause for predisposition to mucormycosis. Use of immunomodulatory drugs such as monoclonal antibodies in COVID-19 along with use of high dose corricosteroids might lead to immunosuppression thus predisposing an individual to develop mucormycosis as a consequence of COVID-19. However use of corticosteroids in COVID-19 is a necessary ingredient of the treatment protocol owing to its great anti-inflammatory effects (Talwar et al., 2021).

We report a rare case of a 61 year old female with no prior comorbidities who presented to the out patient department with fistula draining pus on the right side of her face which upon investigations turned out to be a case of COVID-19 associated Mucormycosis treated with antiviral and antifungal conservatively.

2. CASE REPORT

A 61 year old female presented with the chief complaint of a pus dragning fistula over the right side of face with conjunctival congestion since five days. There was history of intermittent fever since seven days. There was no history of blurring of vision, swelling of eye lid or any other complaint. There was no prior history of diabetes mellitus, hypertension or any other chronic medical illness. There was no history of any malignancy or immunosuppressive therapy in the past. On examination pulse was 88 per minute, regular, blood pressure was 120/74 mm hg in right arm supine postion, spo2 was 94 percent on room air and respiratory rate was 24 breaths per minute. On local examination there was a fistula present on the aspect of right maxillary area with pus discharge and there was a white coloured lesion observed on the palate of the patient. On systemic examination bilateral air entry was equal however there were bronchial breath sounds present in the inframammary area with bronchophony present. Heart sounds were normal, abdomen was soft non tender and conscious and oriented. Nasopharyngeal Swab was sent for COVID-19 which turned out to be positive. HRCT thorax was done which showed bilateral ground glass opacity with a CT Severity score of 14/25 and CORAD 6 (Figure 1). MRI paranasal sinus was done suggestive of sinusitis along with maxillary sinus fistula (figure 2). Swab was sent for culture from the fistula which showed growth of Rhizpopus species (Figure 3). Lab investigations are mentioned in table 1. Patient was advised surgical debridement in view of extensive spread of mucormycosis with oral and cutaneous lesions (Figure 4 and 5) but the patient and her relatives were not willing for the same and opted for conservative approach. Patient was started on liposomal amphotericin B, remdesavir and other supportive measures. Patient's HbA1c was found to be raised along with raised Fasting Blood Sugar and Post meal blood sugar she was thus started on Insulin in view of uncontrolled sugar levels.

Figure 1 Showing bilateral ground glass opacity in HRCT Thorax

Figure 2 MRI Paranasal Sinus showing features of maxillary sinusitis along with maxillary sinus fistula

Table 1 Showing Lab investigations of the patient

CBC	Hb-10.7gm/dl, MCV:82fl, Platelet count 110000/dl, WBC Count 3980/dl
LFT	Total Protein-5.5gm/dl, Albumin-2.8gm/dl, Globulin-2.7gm/dl, aspartate aminotransferase 33 units/l , alanine aminotransferase 28 units/l, AlkanlinePhophatase 110 IU/l, Total Bilirubin :1.4mg/dl,
KFT	Creatinine:0.9mg/dl, Urea46mg/dl, Sodium-139 mmol/l, Potassium – 3.6 mmol/l
CRP	21.9mg/dl
D-Dimer	0.99
Serum Ferritin	750ng/ml
HbA1c	7.4

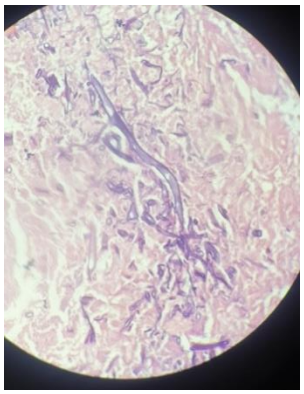

Figure 3 Showing growth of Rhizopus species with septate hyphae branching at right angles

Figure 4 Showing oral lesions of the patient

Figure 5 Showing Pus discharge coming out of maxillary sinus fistula along with conjunctival congestion

3. DISCUSSION

Mucormycosis is known to be precipitated in Diabetes Mellitus and immunocompromised states. In Diabetes Mellitus there is hyperglycemia which leads to hypeglycation of iron sequesting protein as a result of which there is release of free iron in blood. There is ultimately upregulation of GRP 78 receptor and increased expression of COTH protein which enables the fungus to bind and cause invasion resulting in mucormycosis. Diabetes also leads to defective phagocytic function of immune cells thus resulting in fungal invasion (Figure 6). In complications of Diabetes like diabetic ketoacidosis there is upregulation of iron sequesting protein again resulting in increase of free iron in blood and ultimately synergising fungal binding and invasion (Mekonnen et al., 2021).

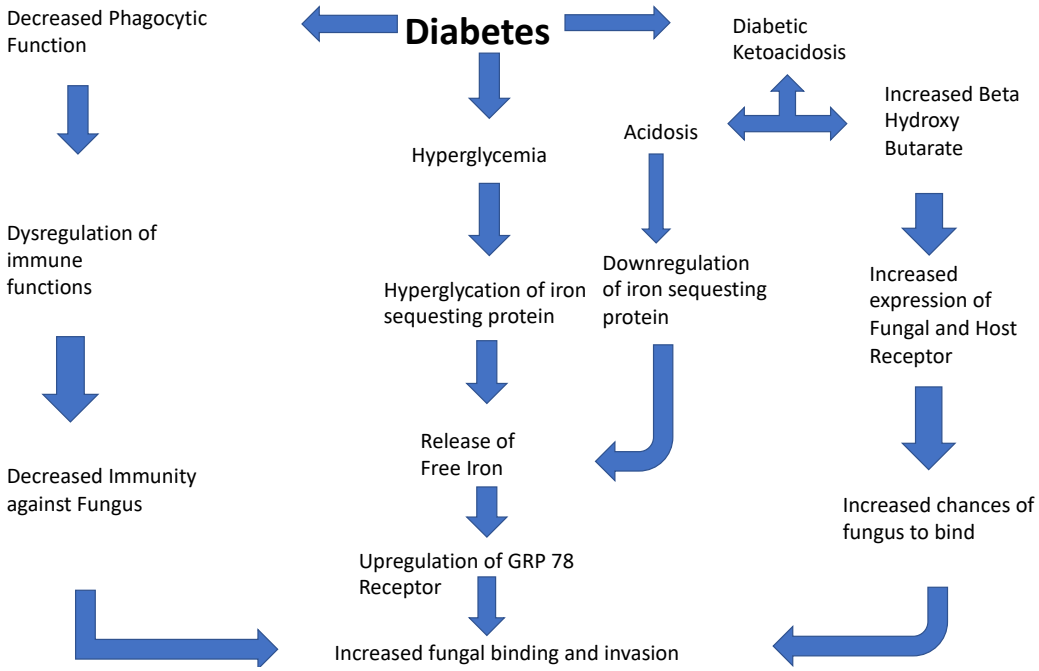

Figure 6 Showing Pathophysiology of Mucormycosis in Fungal Infection

Cutaneous Mucormycosis is a manifestation of mucormycosis where either there is an external implantation of the fungus or hematogenous spread of the fungus to the dermis. External implantation can be by the means of exposure to infection, penetrating injuries, insulin syringes or surgical dressings. Hematogenous spread can be from a focus of fungal infection. This cutaneous spread of mucormycosis manifests as involvmenent of the muscle, fascia and bone. Ultimately it requires surgical debridement.

Histopathological examination is the most sensitive for detection of mucormycosis. There is thick wallen, ribbon like hyphae that branch at right angles. Culture may also be positive in some cases. MRI Brain or paranasal sinus is an extremely useful modality which provides a clear idea about the extent of the disease. Treatment approach starts with early initiation of treatment, management of the predisposing factors and surgical debridement. Liposomal amphotericin B remains the drug of choice in mucormycosis.

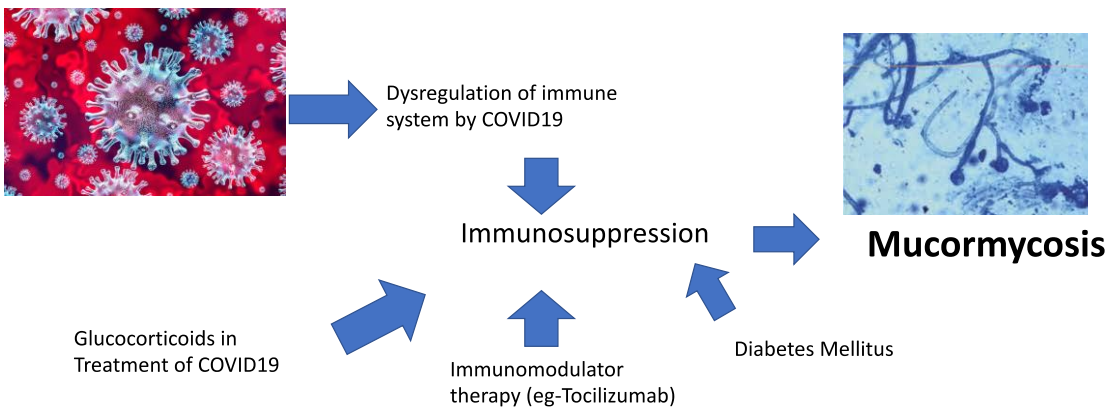

Figure 7 Showing pathophysiology of Mucormycosis in COVID-19

In cases of COVID-19 there is predisposition to mucormycosis as there is dysregulation of the immune system and use of glucocorticoids and immunomodulaters in treatment for COVID 19 further increases the risk of contracting mucormycosis. Another major factor is Diabetes which is commonly seen in COVID-19 due to increased predisposition to COVID-19 in diabetic patients and increased expression of ACE 2 on beta cells of pancreas (Figure 7).

In our case, the patient presented with a discharging fistula from the maxillary sinus over the right side of face which turned out is a cutaneous manifestation of Mucormycosis. Patient tested positive for COVID-19 with Ground glass opacity on HRCT Thorax predominantly in the lower lobes of the lung which is a common finding in COVID-19 (Jain et al., 2020). Patient was not known diabetic previously but had raised blood glucose levels and HbA1c and was started on Insulin for the same. Mucormycosis was managed with Liposomal Amphoterecin B since the patient and relatives were not willing for surgical intervention and Liposomal amphotericin B remains the drug of choice for conservative management of mucormycosis (Mehta & Pandet, 2020). Thus we highlight that through increased vigilance cutaneous manifestation of mucormycosis can be diagnosed correctly and early initiation of treatment prevents morbidity and mortality in such individuals.

4. CONCLUSION

Hence, we conclude that cutaneous manifestation of mucormycosis as a primary presentation in COVID-19 is rare but shouldn't be ignored as it may result in serious complication and increased mortality which can be prevented by early diagnosis and initiation of treatment. We also advice the physicians treating COVID-19 to use glucocorticoids and immunomodulator therapy judiciously in order to prevent unwanted complications which can be mitigated. Also, a regular check of blood sugar levels in COVID-19 patients in recommended as diabetic patients are more prone to present with mucormycosis in the future.

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Conflict of interest

The Authors have no conflicts of interest that are directly relevant to the content of this clinic-pathological case

Financial Resources

There are no financial resources to fund this study

Informed Consent

Informed Consent was obtained from the patient.

Author's Contribution

All the authors contributed equally to the case report.

Data and materials availability

All data associated with this study are present in the paper.

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