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A case series on isolated maxillary sinus involvement in mucormycosis

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ABSTRACT

Mucormycosis is a potentially fatal fungal infection that can affect various parts of the body, including the sinuses. Isolated maxillary sinus involvement usually indicates an early diagnosis of Mucormycosis. The aim of the study was to present a case series on the clinical presentation and management outcomes in patients with isolated maxillary sinus involvement in Mucormycosis to emphasize on the early diagnosis and management of the condition. A retrospective study of 10 patients who presented to the RGGGH ENT department with isolated maxillary sinus involvement in Mucormycosis that was confirmed by diagnostic nasal endoscopy, CT scan and fungal KOH swab and who were managed by endoscopic sinus surgery. In our study, out of 10 cases, two patients had unilateral symptoms and eight patients had bilateral symptoms. Most common presentation was nasal obstruction, nasal discharge, and facial pain. Complete disease clearance was achieved in all cases by endoscopic sinus surgery with debridement. Diabetes was the most common underlying comorbidity, present in all 10 patients. All patients were treated with a combination of antifungal therapy and surgical debridement. All patients showed improvement on follow-up, with no recurrence or mortality reported during the follow-up period. Mucormycosis with isolated maxillary sinus involvement is a timely presentation of mucormycosis but potentially life-threatening due to the nature of the disease to spread rapidly. Prompt diagnosis and management are crucial for improving outcomes in these patients. A combination of antifungal therapy and surgical debridement appears to be an effective treatment approach in these patients.

Key words: Mucormycosis; Isolated maxillary sinus; Endoscopic sinus surgery

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INTRODUCTION

Mucormycosis is a rare but potentially fatal fungal infection caused by fungi of the order *Mucorales*.[1] Immunocompromised people, such as those with uncontrolled diabetes, hematological malignancies, solid organ transplants, and prolonged corticosteroid therapy, are the population most commonly affected.[2,3] The illness usually affects the brain, lungs, and sinuses.

Isolated maxillary sinus involvement is an early presentation of mucormycosis.[4] It is often associated with uncontrolled diabetes and is characterized by the invasion of the fungal hyphae into the vascular structures, leading to tissue necrosis, thrombosis, and infarction.[5] During the post-COVID era, there was a significant rise in the number of cases of Mucormycosis that were probably attributed to the inappropriate steroid usage.[6] The clinical presentation of isolated maxillary sinus mucormycosis can be non-specific, with symptoms such as nasal congestion, nasal discharge, and facial pain. The diagnosis of isolated maxillary sinus mucormycosis requires a high index of suspicion, as the symptoms can mimic those of other sinonasal diseases.[7]

The maxillary sinus is the largest of the paranasal sinuses (PNSs) and because of its proximity to important structures like the orbit and the skull base, if the infection is not identified and treated immediately, it can cause significant consequences.[8] Hence, this study aims to establish the importance of early diagnosis and prompt management

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which are crucial to improve patient outcomes in Mucormycosis.

MATERIALS AND METHODS

In this retrospective study, we collected the case details of 10 patients, who were admitted in ENT ward, RGGGH during the period of July 2021–August 2021. All the patients presented with clinical symptoms suggestive of Mucormycosis. Most of the patients presented with complaints of facial pain, nasal obstruction, and nasal discharge. All patients were previously diagnosed diabetics with nine of them having had COVID-19 infection in the past. All patients did CT PNS, which showed heterogeneous intensities within PNSs, which is unilateral in two patients and bilateral in eight patients. All patients were started on intravenous liposomal amphotericin B, underwent endoscopic sinus surgery and continued liposomal amphotericin B postoperatively for 3 weeks. We followed all patients postoperatively at fortnightly intervals for the first 3 months and once in 2 months thereafter. None of the patients had recurrence or required revision surgery.

RESULTS

Our study focused on 10 patients (Table 1) with mucormycosis who presented with disease of the maxillary sinus alone. Of these, six were male and four were female (Figure 1), with a mean age range of 50–60 years. Bilateral maxillary sinus involvement was observed in eight cases, while the remaining two had unilateral maxillary sinus disease (Figure 2). All patients had a known history of diabetes. Moreover, nine out of 10 patients had a prior COVID-19 infection (Figure 3), with seven of them receiving oral or intravenous corticosteroids (Figure 4). We performed diagnostic nasal endoscopy, which revealed a characteristic finding of necrosis with secretions in the middle meatus. Common CT PNS findings included mucosal thickening in the maxillary sinus, which was heterogeneous, with some cases showing decreased density due to tissue necrosis. In all patients, a KOH swab from the middle meatus showed the presence of broad aseptate fungal hyphae. Treatment of patients involved the administration of intravenous liposomal amphotericin B and endoscopic sinus surgery with debridement. Post-operative histopathology showed the presence of broad aseptate fungal elements with right-angled branching, angioinvasion, and bony invasion. Patients were monitored through diagnostic nasal endoscopy at fortnightly intervals for the first 3 months, and then once in 2 months.

Our results showed that all 10 patients had complete disease clearance without recurrence.

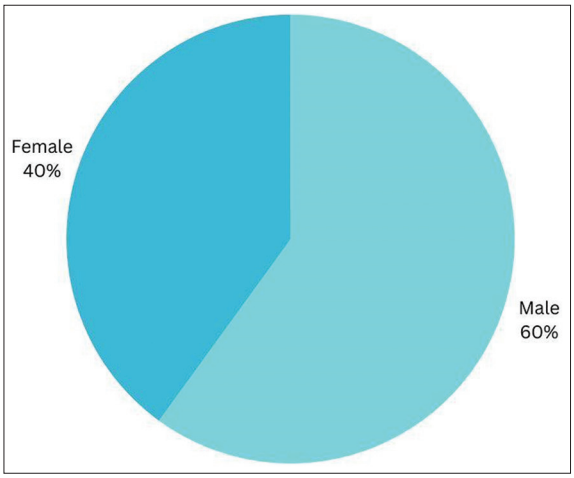

Figure 1: Sex distribution

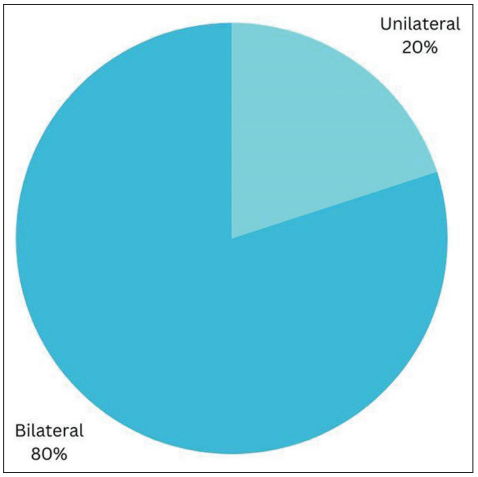

Figure 2: Laterality of maxillary sinus involvement

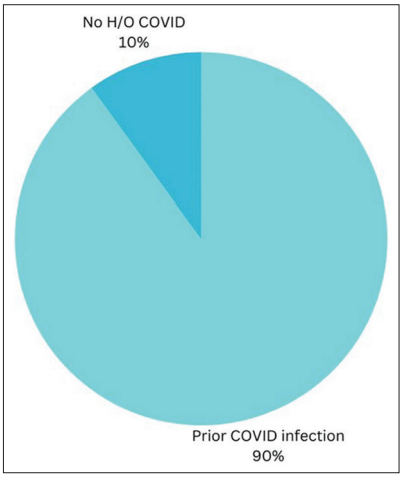

Figure 3: Post-COVID infection

The following chart represents sex distribution in patients with isolated maxillary sinus mucormycosis.

The following chart represents laterality of maxillary sinus involvement.

The following figure shows the of patients who had a history of COVID infection in the past.

The following figure shows the number of patients who received corticosteroids.

DISCUSSION

Isolated maxillary sinus involvement in mucormycosis is an early presentation of invasive mucormycosis.[9,10] Patients usually have an associated immune compromised state such as diabetes mellitus, hematological malignancies, and post-transplant patients;[11,12] moreover, after the COVID pandemic, there was an enormous rise in the number of cases of this otherwise rare disease.[13,14] Since isolated involvement of maxillary sinus is an early stage in the course of the disease the patients usually present with mild non-specific symptoms such as nasal discharge, nasal obstruction, and facial pain which usually leads to clinical suspicion of rhinosinusitis.[15] Thus, performing diagnostic nasal endoscopy in these patients is of utmost importance as it characteristically shows areas of necrosis and secretions in the middle meatus (Figure 5).[16-18]

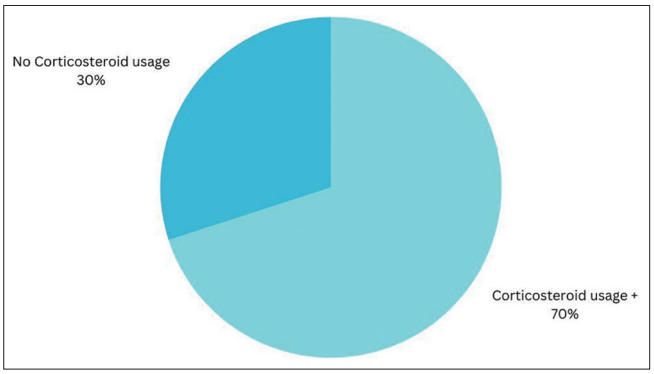

Figure 4: Corticosteroid usage

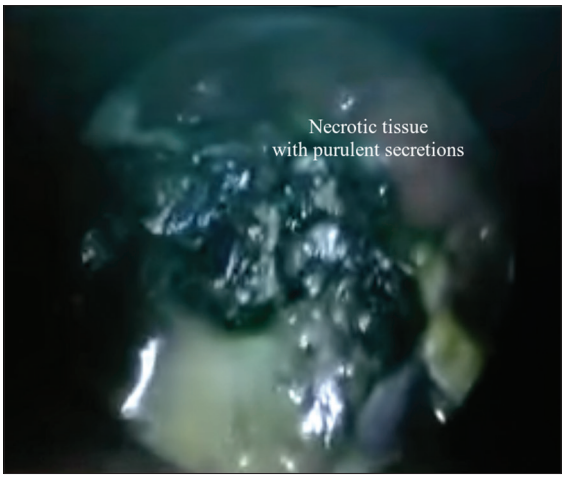

Figure 5: Diagnostic nasal endoscopy showing areas of necrosis and secretions in the middle meatus

KOH swab from the middle meatus serves as a quick aid in diagnosis[19,20] showing broad aseptate fungal hyphae with the right angle branching (Figure 6) diagnostic of mucor.[21]

CT PNS reveals maxillary sinus mucosal thickening which is heterogeneous,[22] with some cases displaying decreased density as a result of tissue necrosis (Figure 7).[23] The disease may be unilateral or bilateral involving both maxillary sinuses (Figures 8-10).

The cornerstone of management involves surgical debridement through endoscopic sinus surgery till fresh

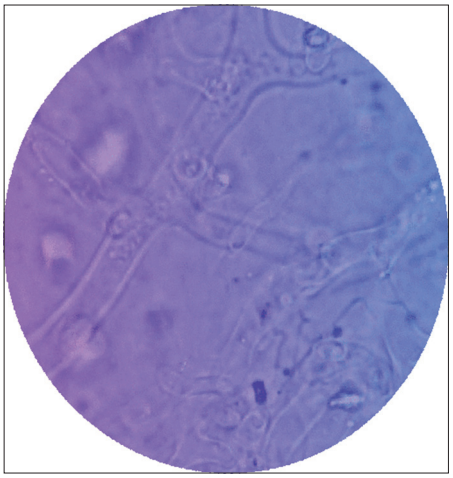

Figure 6: KOH mount from middle meatal secretions showing broad aseptate hyphae with right angled branching

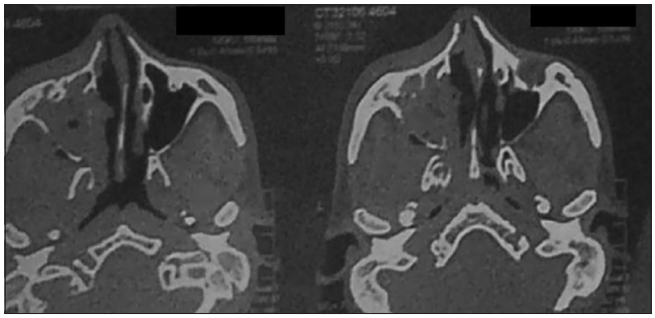

Figure 7: CT PNS showing disease in left maxillary sinus with bony erosion

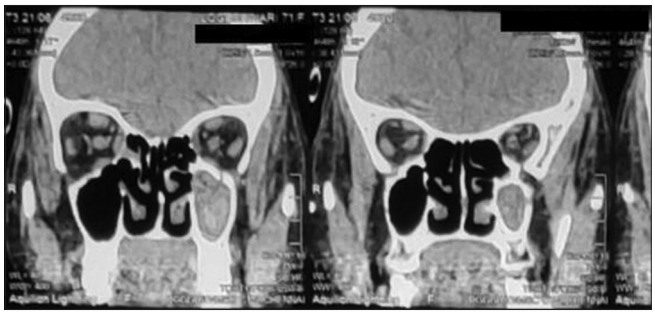

Figure 8: CT PNS image showing heterogeneous opacities in the left maxillary sinus

Table 1: Mucormycosis with isolated maxillary sinus involvement – case details

S. No	Age (years)	Sex	Symptoms	Sinus involvement and Laterality on CT	Comorbidities	H/O COVID/ steroid course	Pre Op DNE	Pre op Fungal KOH	Management	Post Op HPE	Post op DNE
1	52	F	Nasal obstruction Nasal discharge	Maxillary Bilateral	Diabetes mellitus Hypertension	+/+	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
2	56	M	Nasal obstruction Facial pain	Maxillary Bilateral	Diabetes mellitus	-/-	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
3	46	F	Nasal obstruction	Maxillary Bilateral	Diabetes mellitus Hypothyroidism	+/-	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
4	50	F	Nasal obstruction Facial pain	Maxillary Bilateral	Diabetes mellitus Hypertension	+/+	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
5	60	M	Nasal obstruction Facial pain	Maxillary Bilateral	Diabetes mellitus CAD	+/+	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
6	62	M	Nasal obstruction Nasal discharge	Maxillary Unilateral	Diabetes mellitus	+/+	Left middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
7	57	M	Nasal obstruction Facial pain Nasal discharge	Maxillary Bilateral	Diabetes mellitus Hypertension CAD	+/-	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
8	60	M	Nasal obstruction Facial pain	Maxillary Bilateral	Diabetes mellitus Hypertension	+/+	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease

(Contd...)

Table 1: (Continued)

S. No	Age (years)	Sex	Symptoms	Sinus involvement and Laterality on CT	Comorbidities	H/O COVID/ steroid course	Pre Op DNE	Pre op Fungal KOH	Management	Post Op HPE	Post op DNE
9	58	F	Nasal discarge Facial pain	Maxillary Unilateral	Diabetes mellitus Hypertension	+/+	Left middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease
10	55	M	Nasal obstruction Facial pain	Maxillary Bilateral	Diabetes mellitus	+/+	Bilateral middle meatus necrotic secretions	Broad aseptate Fungal elements +	ESS with debridement	Mucormycosis with e/o angioinvasion and bony invasion	Postoperative changes+No fungal debris/residual disease

Figure 9: CT PNS image showing bilateral maxillary sinus involvement

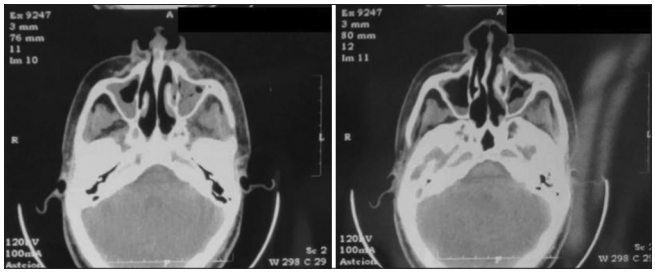

Figure 10: CT PNS image showing bilateral maxillary sinus involvement

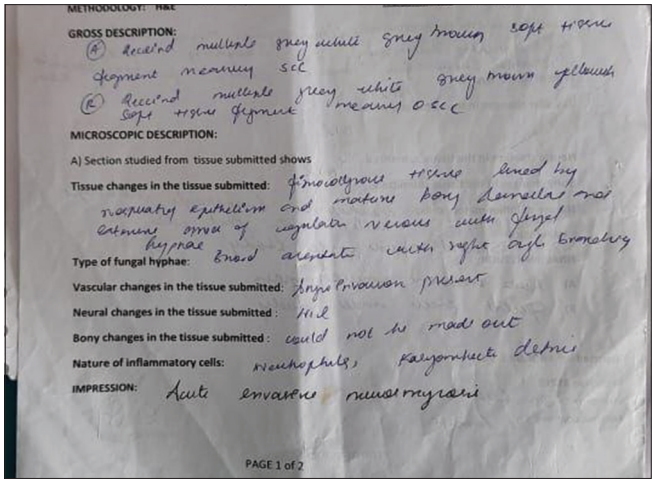

Figure 11: Histopathology report showing acute invasive fungal sinusistis

bleeding occurs which in turn provides good response to antifungal medications such as liposomal amphotericin B. Associated immunocompromising conditions such as diabetes mellitus must be managed appropriately.[24] Diagnosis can be further confirmed by histopathology which shows the characteristic invasive nature of the fungi- angioinvasion and bony invasion (Figures 11 and 12).

Since maxillary sinus involvement without other sinus disease is an early stage in the disease, thorough and

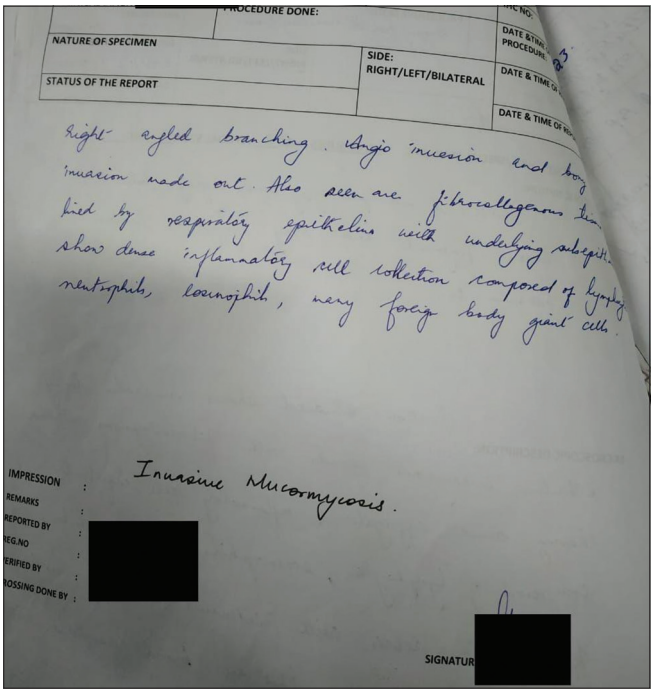

Figure 12: Histopathology report showing invasive mucormycosis

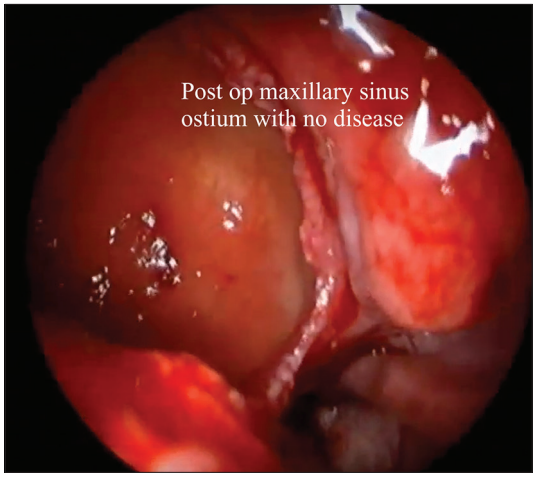

Figure 13: Post-operative diagnostic nasal endoscopy showing maxillary sinus ostium

extensive debridement of the sinuses promptly provides a good outcome in the patients with fewer chances of recurrence of disease.[25] Regular follow-up of patients via nasal endoscopy is essential to detect recurrence (Figure 13).

CONCLUSION

Early detection of mucormycosis by strong suspicion of the condition especially in immune compromised individuals with expeditious and extensive surgical debridement by endoscopic sinus surgery, appropriate antifungal therapy (Liposomal Amphotericin B), and strict glycemic control provides good patient outcome preventing mortality from this potentially fatal condition.

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