

CASE REPORT

Rhinocerebral Mucormycosis Presenting as Oroantral Fistula

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ABSTRACT

Mucormycosis refers to a severe infection with fungi of order Mucorales, seen in diabetic and immunocompromised patients. Rhino-orbito-cerebral mucormycosis is known to exist in two forms, the well-known acute form and the less well-recognized chronic form. The most common presenting features of the chronic form are ophthalmologic, including ptosis, proptosis, visual loss and ophthalmoplegia. Here, we report a case of chronic rhinocerebral mucormycosis (RCM) presented with nonhealing oroantral fistula in a diabetic patient without any orbital symptoms. We are reporting this case for its unusual presentation.

Keywords: Rhinocerebral mucormycosis, Mucormycosis, Chronic rhinocerebral mucormycosis, Oroantral fistula.

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INTRODUCTION

Mucormycosis generally presents as an acute fulminant infection in diabetics or in patients with immunocompromised disease.¹ Chronic presentations of rhinocerebral mucormycosis (RCM) have also been described. In the chronic infection, the disease course is indolent and slowly progressive, often occurring over weeks to months. The most common presenting features of chronic rhinocerebral mucormycosis (CRM) are ophthalmologic and include ptosis, proptosis, visual loss and ophthalmoplegia. CRM occurs predominantly in patients with uncontrolled diabetes and ketoacidosis. The incidence of internal carotid artery and cavernous sinus thrombosis is higher in CRM patients than in those with the acute disease, although the overall survival rate for CRM patients is 83%.¹

Here, we report a case of chronic RCM presented with nonhealing oroantral fistula in diabetic patient without any orbital symptoms. We are reporting it for its unusual presentation.

CASE REPORT

A 75-year-old male patient was referred by the dental department for nonhealing oroantral fistula. He was a known diabetic and was on regular treatment. There was H/O right upper molar excision, 2 months back, following which he

developed oroantral fistula with salty discharge. Now he came to dental OPD for excision of other caried teeth so that the dentures can be fitted. The dentist referred the patient to ENT for the treatment of oroantral fistula.

On examination, patient's oral hygiene was bad. There was a fistula seen in place of right upper third molar with some white hard piece protruding out of it. Other ENT examination was normal, except tenderness over right maxillary sinus. We prescribed antibiotics, antihistaminic and anti-inflammatory drugs for 15 days as chronic maxillary sinusitis is the most common cause for nonhealing oroantral fistula. In the meantime, investigations were done. X-ray PNS showed haziness over right maxillary sinus. Thinking that unless the remaining part of tooth (white protruding part) is removed, the fistula will not heal, patient was again referred back to dental OPD. When dental surgeon tried to remove it the whole inferior part of maxilla came out as seen in Figure 1, with a very large resulting fistula of 2 × 2 cm size (Fig. 2). Surprisingly there was no bleeding. The specimen was sent for HPR keeping in mind possibility of malignancy or osteomyelitis or mucormycosis.

HPR reported as mucormycosis. Computed tomographic (CT) PNS confirmed the same findings with no extension to nose and orbit. Patient was admitted and intravenous amphotericin B was started. We also gave local amphotericin B douche. No further surgical treatment was necessary as partial maxillectomy was already done. Repeated endoscopic debridement were done till there were no necrotic tissue. The advantage we got in this case was that

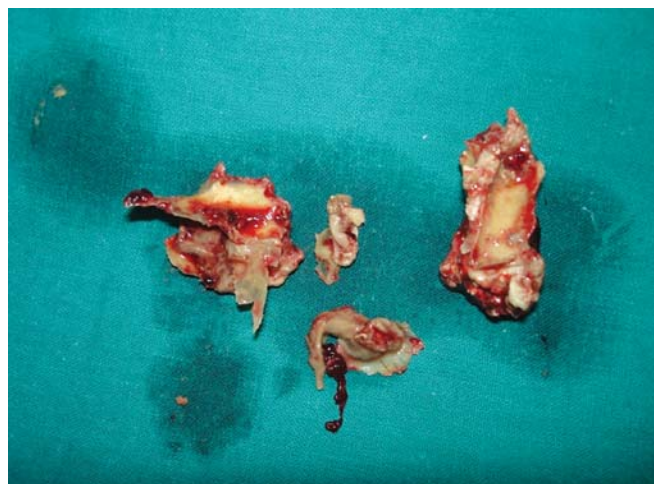


Fig. 1: Pieces of maxilla removed



Fig. 2: Oroantral fistula

the maxillary sinus could be visualized directly through the fistula. Patient recovered well but did not followed up later for correction of oroantral fistula.

DISCUSSION

Mucormycosis refers to a rare and severe infection with fungi of order Mucorales. *Rhizopus* species is the most common organism. The disease begins in the nose, extends to paranasal sinuses and quickly spread to brain and orbit, producing life-threatening orbitocerebral manifestations. The fungus directly invades blood vessels and propagates by arterial thrombosis causing ischemic necrosis of the surrounding tissue. Spread of disease is directly proportional to the time delay in diagnosis except in rare case of indolent variety of disease.² Rhino-orbito-cerebral mucormycosis is known to exist in two forms, the well-known acute form and the less well-recognized chronic form or indolent variety.

Though the disease is described in the literature as chronic form, the indolent variety will be a better term to use, because in chronic form the disease slowly progresses but in indolent form it is more or less static. In our case also, the disease was affecting right maxillary sinus only and did not spread to adjacent areas like orbit and nose. The patient developed fistula after the tooth excision.

Our case was also unique, as the most common presenting features of CRM are ophthalmologic and include ptosis, proptosis, visual loss and ophthalmoplegia.³ But, these features were absent in this case.

The most common oral sign of mucormycosis is ulceration of the palate, which results from necrosis due to invasion of a palatal vessel.⁴ Extension from the sinuses into the mouth causes painful, black necrotic ulcerations in the hard palate.⁵ The lesion is characteristically large and deep, causing denudation of the underlying bone. Ulcers

from mucormycosis have also been reported on the gingiva, lip and alveolar ridge.⁶

Mucormycosis presenting as nonhealing oroantral fistula is not reported till now. This case merits consideration as the presentation was unusual for indolent and for acute as well.

Osteomyelitis and bony involvement from sinonasal mucormycosis is usually absent despite deep extension of disease and when present, occurs last in the course of the disease.^{7,8} The late occurrence of bony involvement is explained by the angioinvasive nature of the fungus and characteristically deep extension of infection through perivascular channels that precedes frank bony destruction.^{7,8}

But in our case, bone involvement occurred quite early, before any soft tissue involvement.

Differential diagnosis of a lesion presenting as palatal perforation should also include tertiary syphilis,⁹ leprosy, tantrum oris, mechanical trauma, intranasal cocaine abuse,¹⁰ malignancies, especially nasal T-cell lymphomas, Wegener's granulomatosis and midline nonhealing granuloma.¹¹ But, evidence of diabetes or immunosuppression in a patient presenting with necrotic lesions of the nasal cavity and palate strongly favors the diagnosis of a deep fungal infection.

Plain radiographs of sinuses and orbits may demonstrate sinus mucosal thickening, with or without air-fluid levels, but this is not specific. CT scan with contrast or MRI may demonstrate erosion or destruction of bone or sinuses and help delineate the extent of the disease.⁷

CONCLUSION

Indolent RCM is rare and is seldom survived by immunosuppressed patients. It shows very slow progression over a period of months^{4,6} and is more or less static. Multimodal therapy with surgical debridement and antifungal chemotherapy is required for an optimal outcome along with correction of diabetes or immunosuppression. The overall survival rate for CRM patients is 83%.¹

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