

# Maxillary Sinus Aspergillosis in an Immunocompetent Adult Female

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## ABSTRACT

**Introduction:** Aspergillosis is the second most frequent opportunistic fungal infection of the paranasal sinuses which has seen a substantial increase in the incidence in recent years. Definitive diagnosis is based on radiological, histological examination and fungal culture.

**Case Report:** We present here a report of a chronic case of a fungal rhinosinusitis involving maxillary sinus in an immunocompetent adult female. The pathogen is identified as *Aspergillus flavus* complex. The case was successfully treated with surgical debridement and voriconazole.

**Conclusion:** Early diagnosis, surgical debridement and selection of appropriate antifungal agent is important for successful treatment in both immunocompromised as well as immunocompetent cases.

**Keywords:** Aspergillosis, Paranasal Sinuses, Immunocompetent

## INTRODUCTION

Aspergillosis is a fungal infection of the nasal cavity and paranasal sinuses in maxillofacial region<sup>5</sup>. Paranasal sinuses are second most frequent site of fungal infection after lungs. Approximately, 6-9% of rhinosinusitis cases have fungal etiology<sup>1</sup>. Aspergillosis is the second most frequent opportunistic fungal infection of the paranasal sinuses. *A. fumigatus*, *A. niger* and *A. flavus* are the species that most frequently affect humans<sup>2</sup>. The maxillary sinus is the most common sinus to be affected<sup>1</sup>. In recent years there has been an increase in the incidence of fungal rhinosinusitis due to diffusion of different pathogenic organisms and to the higher prevalence of diseases such as diabetes and chronic treatments with corticosteroids and immunosuppressants<sup>2</sup>.

Fungal rhinosinusitis is classified into an invasive and a non-invasive form. The non-invasive form are allergic sinusitis and aspergilloma. Invasive aspergillus infections can be either limited [chronic or indolent] or fulminant [acute], with a rapid malignant course advancing relentlessly to the destruction of the nasal cavity, the sinuses and the adjacent structures such as the orbit and the brain within a few days<sup>1</sup>. Clinico-radiological findings can be misleading as the lesions are locally destructive and mimic a neoplasm. A biopsy is necessary to establish the diagnosis.

Aspergillosis is mainly transmitted by inhalation of spores, but in the maxillary sinus it can also come through an orsinusal iatrogenic communication secondary to a dental procedure, such as a root canal treatment or a dental extraction<sup>2</sup>. Aspergillus rhinosinusitis may present with nasal polyps, anterior or posterior nasal discharge, nasal obstruction, epistaxis, headache, anosmia, proptosis and

snoring<sup>3</sup>. It should be suspected in a patient with recurrent or refractory sinusitis, not responding to antibiotic therapy<sup>4</sup>. Our study describes a case of maxillary sinus aspergillosis with symptoms of recurrent sinusitis in an immunocompetent patient.

## CASE REPORT

A 50-year-old female with a history of swelling in the right maxillary region since one year presented with pain and increase in the size of swelling since five months. Patient gives a significant past history of nasal polyp in 2000, right maxillary mass in 2013, nasal bleeding in 2017 and maxillary abscess with external drainage in May, 2020 for which the patient underwent repeated surgery. Histopathology revealed granulomatous inflammation with foreign body giant cell reaction with no evidence of malignancy. Patient also gives history of treatment with fluconazole for one year. Patient is a known diabetic since four years. At presentation now, there is no history of nasal obstruction, nasal discharge, fever, loss of vision or any systemic symptoms.

On examination, a firm to hard swelling over the right maxillary region, protruding into upper gingivobuccal sulcus was seen. CT scan revealed large irregular soft tissue lesion in right maxillary sinus with resorption, destruction and extension into the skin and integument - suggestive of granulomatous or neoplastic etiology (Fig 1 & 2). Biopsy of the lesion was performed which showed abundant septate hyphae (Fig 3), with inflammatory cells and multinucleated giant cells with no evidence of malignancy. On Potassium hydroxide (KOH) mount it showed hyaline septate hyphae (Fig 4). On culture in Sabouraud's dextrose agar, rapidly growing, velvety, yellowish green colonies are seen (Fig 5). On microscopy, *Aspergillus flavus* complex identified as filamentous structures with septate hyphae branching at an acute (45°) angle with conidiophores bearing globose vesicles was confirmed (Fig 6). Phialides produced directly from entire surface of vesicle giving rise to short chains of

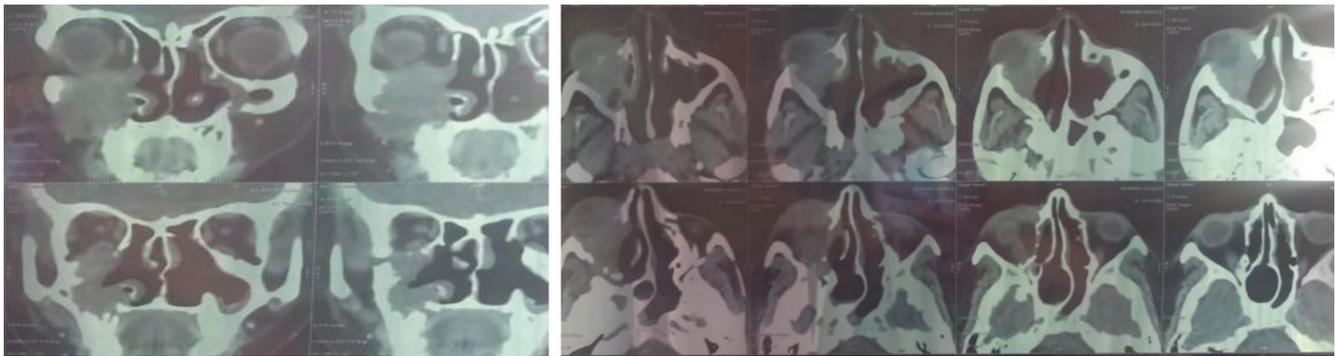
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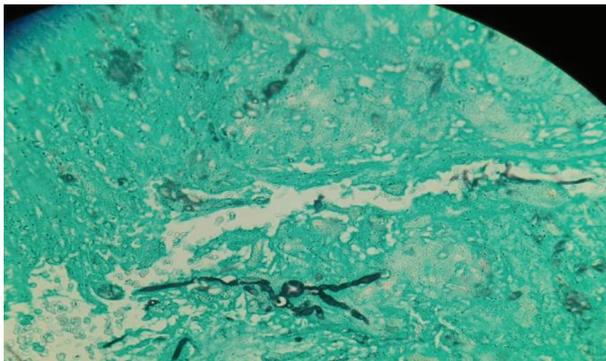
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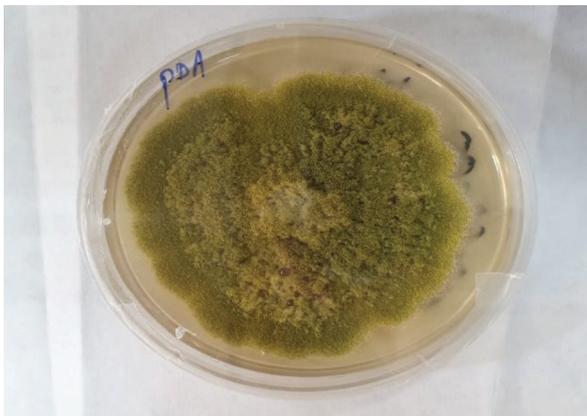
**Figure-1 & 2:** CECT PNS images depicting lesion and extension into right maxillary sinus



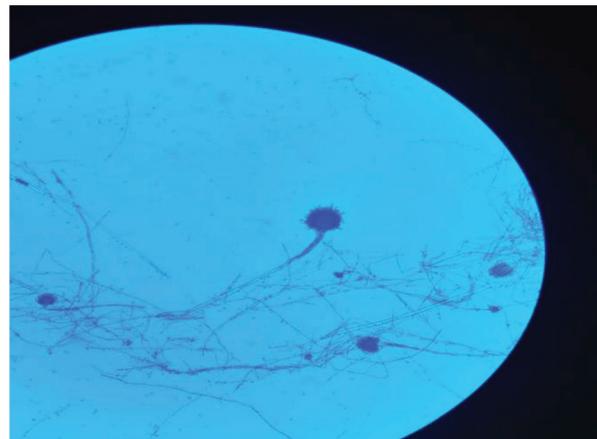
**Figure-3:** Fungal hyphae in histopathological section of excised mass stained with GMS stain



**Figure-4:** KOH mount with septate hyphae with conidiophore and hyaline conidia



**Figure-5:** *Aspergillus flavus* complex growth on culture media



**Figure-6:** Microscopic appearance of *A. flavus* complex in LPCB mount

hyaline spherical conidia was also noted.

Patient was operated for right maxillary sinus mass excision and started on voriconazole. On follow up of patient after 6 weeks, pain has subsided and there is regression in size of the swelling.

**DISCUSSION**

Fungi has been increasingly recognized as important pathogens in severe acute and chronic sinusitis in immunocompromised hosts. They have been detected in more than 90% of nasal lavages in immunocompetent patients with rhinosinusitis<sup>7</sup>. Fungal infections of the paranasal sinuses need to be diagnosed early in order to avoid significant mortality and morbidity<sup>1</sup>. Factors that predispose to fungal infections in sinuses include polyps and stagnant secretions, other factors like neutropenia, inappropriate use

of antibiotics, immunosuppressive drugs, corticosteroids, uncontrolled diabetes mellitus, HIV infection, trauma, burns, and radiation therapy<sup>1</sup>. In the paranasal sinuses, headache, nasal congestion, fever, and pain in the face and around the eye are common presenting features<sup>1</sup>.

Invasive aspergillosis arising from the nose and paranasal sinuses can cause an intra-orbital and intra-cranial extension mainly along the skull base and larger vessels. Intracranial and intra-orbital extension is associated with decreased survival rate and increased surgical morbidity<sup>1</sup>.

Both MRI and CT scan can help to establish a diagnosis of invasive fungal sinusitis. Clinico-radiological findings can sometimes be misleading as the lesions are locally destructive and might mimic a neoplasm. Biopsy is necessary

to establish the diagnosis<sup>1</sup>. Hyphae are typical and specific for each fungus; *Aspergillus* spp. shows septate hyphae that branch at 45° angles. Fungal cultures on Sabouraud's dextrose agar is needed to confirm the diagnosis.

As invasive maxillary sinus aspergillosis is rare in immunocompetent patients, early precise recognition is necessary in order to achieve optimal treatment results. Delayed diagnosis and treatment may lead to a poor therapeutic outcome<sup>1</sup>.

The comprehensive management of paranasal sinus fungal infection includes early diagnosis, histological typing, appropriate antifungal therapy, and surgery<sup>6</sup>. The prognosis is directly related to the severity of the underlying disease, the extent of the disease when treatment begins, and the aggressiveness of the treatment<sup>6</sup>. For the treatment of invasive aspergillosis of the sinuses, it is recommended to continue oral antifungal therapy for at least 4-6 months to prevent recurrence of the disease<sup>8</sup>. Regular post-operative follow-up is recommended in all the cases with CT scan and nasal endoscopy every 3-4 months. Early diagnosis of recurrent disease requires prolonged systemic antifungal chemotherapy<sup>1</sup>.

## CONCLUSION

Invasive maxillary sinus aspergillosis should be considered in the differential diagnosis of maxillary sinusitis that does not respond to standard conservative therapy with antibiotics even in immunocompetent patients. Early diagnosis and therapeutic intervention, with selection of an appropriate antifungal agent and surgical debridement, is the key to successful treatment of invasive aspergillosis.

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