

# Utility of Aspiration Cytology in Clinically Unexpected Cases of Mixed Infection of *Aspergillus* and *Mucor* in Oculo-Rhino-Cerebral Mycosis: A Case Series

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## ABSTRACT

**Introduction:** Fungal rhinosinusitis comprise a spectrum of disease processes varying clinically, histologically as well as biologically. It can be broadly divided into two categories based on histopathological findings into invasive and non invasive depending on invasion of the mucosal layer. Invasive fungi can be effectively treated if fungus is identified rapidly. Both mucormycosis and aspergillosis are life threatening fungal infection usually occurring in immunocompromised or in patients with uncontrolled diabetes mellitus. Here we present two different cases of mixed fungal infection of mucor and aspergillus.

**Case report:** 27 year non diabetic male presented with swelling in left lower lid and 35 year non diabetic male with swelling on right side of cheek are the two cases reviewed here. Contrast-enhanced computed tomography Paranasal sinuses in these cases suggestive of either malignancy or invasive fungal disease. Aspiration cytology performed from both cases revealed inflammatory infiltrate along with abscesses and hyphae of fungus. Endoscopically removed tissue sent for histopathological examination showed numerous giant cells and presence of broad aseptate as well as septate hyphae with acute angle branching within giant cell consistent with mucormycosis and aspergillosis.

**Conclusion:** Patient responded well to therapy in both of the cases. Here we emphasized the importance of early detection and aggressive treatment in the management of the mixed fungal infection which is more fatal as compared to single fungal infection.

**Keywords:** Aspergillosis, Mucormycosis, Rhinosinusitis

## INTRODUCTION

Mycotic infections of the paranasal sinuses are on the rise globally. They are common in north India, and are now being recognized in other parts of India as well.<sup>1,2</sup> The spectrum of fungal diseases of the paranasal sinuses ranges from allergic sinusitis to invasive disease (fungal rhinosinusitis). Opportunistic fungal infection can be life threatening in the presence of immunosuppression or uncontrolled diabetes mellitus due to their invasive potential. Mucormycosis of oculo rhino cerebral is an uncommon rapidly spreading invasive fungal infection and is life threatening with mortality rate nearing 50%. Its coexistence with aspergillus and candida makes it more uncommon.<sup>3</sup>

Early diagnosis is important as the lesions may be rapidly progressive and destructive and may even be fatal. These lesions can mimic malignancy clinically and on radiology.

Fine needle aspiration (FNA) is a safe and simple technique widely used for rapid diagnosis of head and neck lesions. It can be used for diagnosis of fungal etiology in rhino-sinusoidal inflammation. In literature there are only few case reports which describe diagnosis of rhino-sinusoidal mycoses on cytology.<sup>4-6</sup> We present two such cases diagnosed on FNA.

## CASE REPORT

A 27 yr old immunocompetent male reported with history of swelling in left lower lid since 9 month along with watery discharge from the eye. Patient was sent for contrast enhanced computed tomography paranasal sinuses which revealed mass involving left ethmoid, maxillary sinus eroding lamina papyracea of left orbit suggestive of either malignancy or inverted papilloma. Similarly a 35 year old male of rhinocerebral aspergillosis presented with swelling on right side of cheek for 20 days. No complain of fever, ear discharge, nasal bleeding. Contrast enhanced computed tomography paranasal sinuses showed lytic destructive expansile soft tissue mass in right maxillary sinus with extension into right orbit and right infratemporal fossa suspicious of Neoplastic etiology.

Patients were presumed to be immunocompetent as they had not received chemotherapy, steroids, or other immunosuppressive agents. None of the patients had a diagnosis of cancer, chronic liver disease, end-stage renal failure, diabetes mellitus, HIV infection, or congenital immunodeficiency. The clinical and radiological suspicion of malignancy in both cases.

Both the patient underwent fine needle aspiration cytology. Smear prepared and stained with May Grunwald Giemsa and Hematoxylin & Eosin stains. Smears examined were cellular and showed inflammatory infiltrate along with abscesses,

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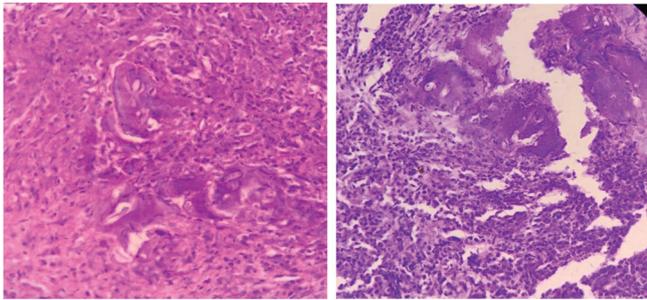
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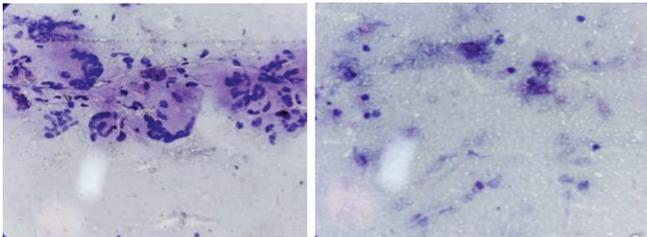
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**Figure-1 & 2:** Smear showed inflammatory infiltrate along with abscesses, necrotic debris and hyphae of fungus. (H & E 400x)



**Figure-3 & 4:** Sections show numerous giant cells and ill-formed Granuloma with the presence of broad aseptate as well as Septate hyphae with acute angle branching within giant cells and inflammatory infiltrate Consistent with mucormycosis and aspergillosis. (H & E 400x)

necrotic debris and hyphae of fungus. ( Fig. 1 and 2)

Based on FNA diagnosis, the patients were treated with endoscopically guided surgical debridement and thereafter the tissue sent for histopathological examination. Histopathological sections confirmed the diagnosis as showed numerous giant cells and ill formed granuloma with presence of broad aseptate as well as septate hyphae with acute angle branching within giant cell and inflammatory infiltrate consistent with diagnosis of mucormycosis and aspergillosis. (Fig. 3 and 4)

## DISCUSSION

Fungal sinonasal sinusitis constitutes 6-9% of all rhinosinusitis. *Aspergillus* and *Mucor* are the most common non invasive and invasive fungal infection respectively of head and neck region. Both can pose diagnostic and therapeutic challenge in view of mimics as malignancy.<sup>7</sup>

Here in this case series we basically focussed on the usefulness of aspiration cytology in early diagnosis of fungal infection.

Mucormycosis is an opportunistic suppurative infection which spreads by direct as well as hematogenous dissemination and is associated with vascular invasion resulting in thrombosis, embolism, and infarction.<sup>8</sup> The predisposing factors for mucormycosis are uncontrolled diabetes mellitus (particularly in patients having ketoacidosis), underlying malignancies, renal failure, organ transplant, long-term immunosuppressive therapy, and cirrhosis.<sup>9</sup> However, the existence of acidosis in large tissue damage by some trauma with some immunodepression may be the explanation for the development of this type of infection in previously healthy individual.

Aspergillosis is a well-known fungal infection most commonly caused by *A. fumigatus*. If diagnosis and therapeutic interventions are delayed it may result in massive tissue destruction and, eventually death.

The current study assesses the role of pre-operative FNA for this purpose. FNA provided a reliable generic diagnosis of inflammation and sparse to moderate inflammatory infiltrate and foreign body giant cells with or without with necrosis were seen in both cases. A specific diagnosis can be made when fungal profiles are identified. Demonstration of an inflammatory reaction determines that the organism represents true infection and not contamination.

Sharma D et al also presented a case comparable with our case of invasive maxillary lesions in immunocompetent patients, clinically suspected of malignancy however fine needle aspiration cytology showed fungal hyphae, morphologically suggestive of *Aspergillus*, which was later confirmed on histopathology.<sup>4</sup>

Singh N et al presented a case report to highlight the clinical comparable with or case utility of a simple FNA procedure as the initial diagnostic modality in cases of fungal sinusitis, which can masquerade clinically as a neoplastic lesion.<sup>6</sup>

Deepa Pandey et al reported a case of mixed opportunistic infection with *Mucor*, *Aspergillus* and *Candida* in oculo-rhino-cerebral mycosis in immunocompromised and diabetic patient. No such history was found in our patients.<sup>10</sup>

Singhal N et al assess the utility of Fine needle aspiration cytology for early diagnosis of invasive fungal rhinosinusitis where aspiration cytology was performed from the maxillary/ethmoid sinus in patients with a destructive mass lesion in the maxilla mimicking clinically and radiologically as malignancy cytology findings were suggestive of fungal etiology which was later confirmed histopathologically.<sup>11</sup>

## CONCLUSION

Fungal rhinosinusitis can closely mimic malignancy, clinically. FNA is a reliable, simple and quick technique for its diagnosis. The search for fungal profiles should be more aggressive in smears rich in inflammatory and foreign body giant cells, or in cases with a strong clinical or radiological suspicion. Thus, preoperative FNA diagnosis obviates the need for a diagnostic biopsy, allows rapid diagnosis as cultures take time and helps to plan proper treatment to suit individual patients.

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