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Case Report

Orbital proptosis in a young immunocompetent female patient

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Introduction

Invasive fungal infections of the nasal and paranasal sinuses are emerging health care problems and are increasingly being reported in healthy immunocompetent persons. Fungal rhinosinusitis (FRS) broadly encompass a wide spectrum of immune and pathological responses that include invasive, chronic, granulomatous and allergic disease. A wide range of fungal species are involved but Aspergillus species is the most common etiological agent. Granulomatous invasive FRS is one of the variants particularly prevalent in Sudan, India, Pakistan and Saudi Arabia. Herein we report a case of chronic granulomatous invasive fungal sinusitis in an immunocompetent adult female.[1–3]

Case report

A 28 year young female patient immunocompetent and non-diabetic from urban background presented with right sided nasal obstruction, mucous discharge and gradual development of proptosis and telecanthus of 3 months duration. There was no history of nasal bleed or visual defect. Anterior rhinoscopy revealed a multilobulated polypoid mass in the right nasal cavity not bleeding to touch. Detailed examination of head and neck and laboratory parameters did not reveal any abnormality. Hematological and biochemical parameters were within normal limits. A plain radiograph followed by a CT scan of the paranasal sinuses was reported as follows: A polypoidal mass lesion epicentered in the right ethmoidal sinus extending into the right nasal cavity and the right orbit causing destruction of the bony septae of the ethmoidal sinus and medial wall of the orbit resulting in deviation of the bony nasal septum to the left, but no intracranial extension of the lesion was noted (Fig. 1). A partial functional endoscopic sinus surgery (FESS) was performed and a polypoidal mass with large quantity of blackish yellow debris was removed and sent for histopathological examination (HPE) that revealed a chronic granulomatos lesion with multinucleated giant cells.

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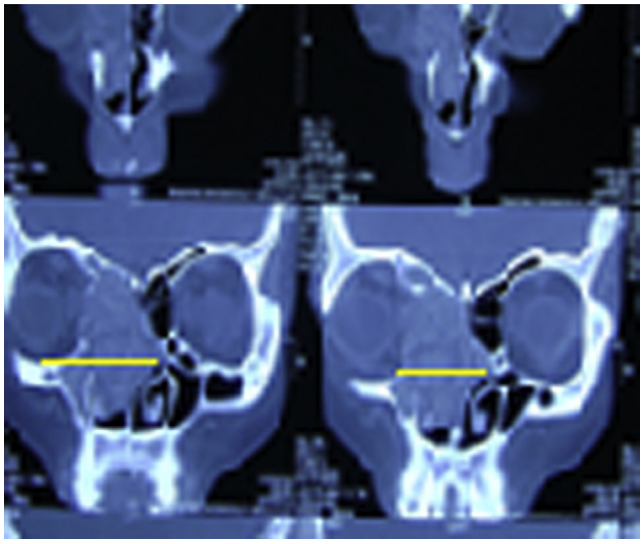

Fig. 1 – CT scan right paranasal sinus: A polypoidal mass lesion epicentered in the right ethmoidal sinus extending into the right nasal cavity and the right orbit causing destruction of the bony septae of the ethmoidal sinus and medial wall of the orbit resulting in deviation of the bony nasal septum to the left. But, no intracranial extension of the lesion was noted.

Acid fast bacilli (AFB), malignant cells and fungal elements were not seen. Based on the HPE findings, a diagnosis of tuberculosis was made and the patient was started on antitubercular treatment (ATT). She, however, reported back to hospital after 6 weeks of ATT without any regression in proptosis or telecanthus. Recurrence of the nasal mass was confirmed on anterior rhinoscopy and a repeat CT scan. FESS was carried out again with complete removal of the mass along with all its extensions. Appropriate media were inoculated for bacterial isolation. A 20% KOH mount and culture on plain Sabouraud's dextrose agar (SDA) and SDA with gentamicin and cycloheximide at 25 °C and 37 °C respectively was done for isolation of fungal pathogens.

No significant bacterial growth was seen after 48 h of incubation in 10% CO_2 and no fungal elements were seen on direct KOH mount. Tissue sections showed noncaseating granulomatous lesions with foreign body type of multinucleated giant cells and infiltrating lymphomononuclear cells, predominantly plasma cells (Fig. 2). Gomori's methenamine silver (GMS) stained tissue sections revealed hyaline septate hyphae with dichotomous branching (Fig. 2). Fungal elements within the giant cells were not visualized on GMS stain. Neither could we demonstrate any ''negative image'' of the fungal hyphae inside the giant cells on H&E staining. There was no evidence of vasculitis, vascular proliferation or perivascular fibrosis in the lesion. The fungal culture on plain SD agar yielded velvety yellow to green colonies with a golden-brown tan on the reverse within a week. Microscopic examination of the colony on plain SD agar (Fig. 3) by tease mount in lacto-phenol cotton blue stain showed conidiophores with biseriate phialides covering the entire vesicle (Fig. 3) and the isolate was identified as *Aspergillus flavus*. Based

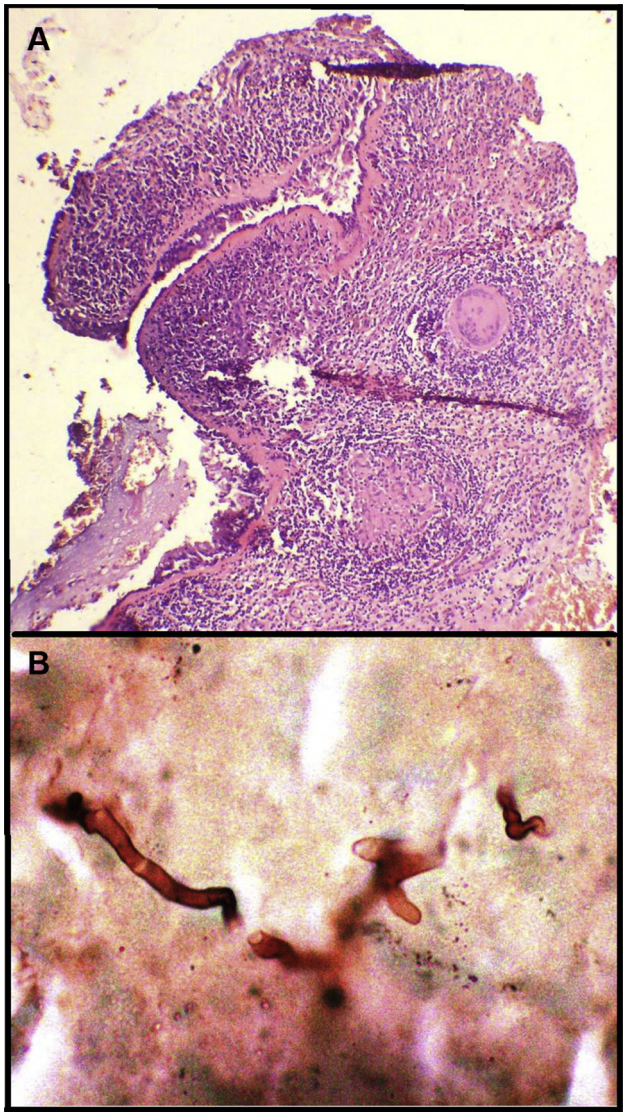

Fig. 2 – A: Photomicrograph of the paranasal mass (H&E stain; 400×) showing noncaseating granulomatous lesions with foreign body type of multinucleated giant cells and infiltrating lymphomononuclear cells. B: Gomori's methenamine silver (GMS; 400×) stain showing hyaline septate hyphae with dichotomous branching.

on the overall clinical, radiological and laboratory findings, the case was diagnosed as ''Granulomatous invasive fungal sinusitis caused by *A. flavus*''.

Oral Itraconazole 100 mg bid with a short course of steroid therapy was given during the post-operative period. The response to therapy was excellent and within 15 days there was complete regression of the proptosis without recurrences on follow up at 3 months.

Discussion

Granulomatous invasive FRS is typically characterized by a clinical course of 12 weeks or more and associated with an