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Case Report

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INVASIVE FRONTAL BONE SINUS ASPERGILLOSIS – A CASE REPORT

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ABSTRACT

Aspergillus species are ubiquitous fungi present in environment with inhalation of spores(conidia) being a common event. However, inhalation of the spores causing fungal sinus infection with invasion of adjacent bony structures or disseminated invasive infection is a rare entity, seen only in immune suppressed individuals. Here, we report a case of a 24 years male, known case of Generalised tonic-clonic seizure on regular anti-epileptics, otherwise immunocompetent without any comorbidity who presented to us with recurrent swelling with discharge over left frontal region of forehead – at site of previously operated myofibroblastic tumour. Biopsy specimen was taken after surgical debridement of the swelling showed presence of fungal hyphae suggestive of Aspergillosis. The patient was treated with Amphotericin B followed by long term voriconazole therapy.

KEYWORDS- Aspergillosis, frontal sinus, fungal hyphae, voriconazole.

BACKGROUND

Aspergillus is a ubiquitous saprophytic Ascomycete fungus which grows by budding or branching. The spores of this fungus are introduced by inhalational route.[1] Aspergillus sinusitis can occur in normal hosts but is self-limiting . Invasive form seen only in immune –suppressed individuals like those with HIV-AIDS infection, long standing uncontrolled diabetics, persons on chronic steroid therapy, cancer chemotherapy patients and also reported in patients suffering from auto-immune diseases.[2] In our case, frontal sinus aspergillosis with invasion to adjacent frontal bone was noted in an otherwise healthy male post-operatively. So, our aim of reporting this case is to highlight that one should keep high index of suspicion for aspergillosis in case of paranasal sinusitis so that timely initiation of anti-fungals can avoid significant morbidity and mortality.

CASE

A 24years old male, known case of Generalised tonic clonic seizure(GTCS) on anti-epileptic medication since childhood, follow up case of operated left frontal lobe space occupying lesion(SOL) and frontal sinus SOL, presented to us with recuurent swelling left forehead with periorbital swelling along with discharge from incision site of operation.

He gave a past history of left sided headache for few months along with vertigo and dizziness for the same duration. He was investigated for same and his MRI revealed large expansile SOL at left frontal sinus cavity with invasion upto dural margin of left basi-frontal lobe. Surgical excision was done of the left frontal mass & sinus and a titanium mesh was placed over frontal sinus region with subgaleal drain which was later removed. Specimen from the left frontal sinus and mass were processed for histo-pathological examination. Microscopic examination of specimen sections showed features suggestive of inflammatory myofibroblastic tumour – benign in nature.

3years later of the surgery, he again developed swelling over left frontal region of forehead –which was again reoperated. Sections of incised parts were processed for H/P examination.

The specimen sections Hematoxylin and eosin stains showed dense fibrotic tissue and area of inflammatory infiltrate with degenerated material- dense sclerotic lesion with focal chronic inflammation. Special stain with GMS stain (Grocott-Gomori Methenamine silver stain) showed presence of fungal hyphae – septate and branched. It was diagnosed to be Apergillosis infection of left frontal bone and sinus and patient was referred to our institute.

When patient presented to us, the swelling was seen over left frontal region – size of about 5x4 cm, tender, firm in nature with an incision mark noted from which there was discharging pus. Rest general survey and systemic examination did not reveal any abnormality.

His routine investigations revealed- Total WBC count-6,300/mm3, neutrophil 44%, eosinophil-4%; Hb-11.6%,platelet-2.9lakhs/mm3. Liver function tests and renal profile, serum electrolytes were within normal limits. Chest X-ray and USG abdomen were normal. Discharge from the swelling was sent for microbiological examination. Gram stain and fungal stain and culture were negative. TB bactec culture and acid fast stains were negative. The paraffin block samples from resected frontal mass were restained and thoroughly examined which showed acute angled branched, septate hyphae suggestive of Aspergillus species. His serology – HbsAg, anti HCV and HIV serology were non-reactive.

Patient was treated with Injection Amphotericin B 1mg/kg for 2weeks, followed by tablet Voriconazole 200mg twice daily. His discharge resolved with regression of the swelling after 3 months. Then the patient was asked to come up for follow up at regular intervals.

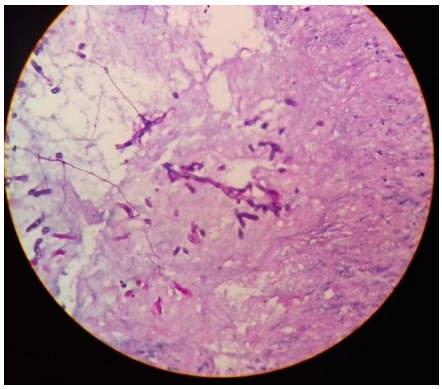

Fig. 1: Hematoxylin and eosin stain from excised section of left frontal sinus and bone mass showing dense fibrotic tissue, area of inflammatory infiltrate with degenerated material and admixed fungal hyphae.

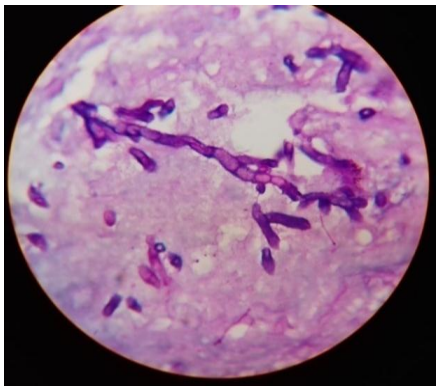

Fig. 2: H & E stain showing septate, acute angled, branched fungal hyphae.

DISCUSSION

Aspergillosis affecting paranasal sinus with invasive form invading adjacent bone in an otherwise immune-competent individual is rare entity reported in literature. Route of infection is via respiratory route, common reported species involved are Aspergillus flavus and A. fumigatus. Invasive aspergillus sinusitis is characterised by spread of fungus from paranasal sinus airspace to adjacent structures. Hypoxic environment caused by blockage of sinus osteum, secretion of tissue invading substances by fungus coupled with inappropriate host immune mechanism are responsible for invasive form of sinusitis seen in immunocompromised individuals.[2]our patient was on anti-epileptic drug levetiracetam and had frontal bone tumour which may be a risk factor for his developing invasive fungal infection. Following his excision of myofibroblastic tumour, a titanium plate mesh was placed – a foreign body which might have acted as a nidus for infection. [3]. Demonstration of fungus in tissues is essential to confirm invasive nature of the disease,[2] as was done in our case by H&E and GMS stains. Treatment consists of surgical debridement along with antifungal therapy.[4] Voriconazole is a newer triazole antifungal agent which inhibits fungal cell membrane synthesis by inhibiting ergosterol synthesis. It is found to be superior to amphotericin B and also other azoles.[5] In our case, we treated him with 2weeks of Amphotericin B followed by Tablet Voriconazole. Invasive frontal bone and sinus aspergillosis in an otherwise healthy male is a rare entity and thus deserves to be reported.

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