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INVASIVE ASPERGILLOSIS PRESENTING AS SCALP OSTEOMYELITIS: A RARE CASE REPORT

Puneet Agarwal[1], Uma Shankar Agarwal[2], Surendra Kumar Thalor[1], Ram Singh Meena[2], Saroj Purohit[2]
[1]Assistant Professor, Department of Dermatology, SMS Medical College & Hospital, Jaipur
[2]Professor, Department of Dermatology, SMS Medical College & Hospital, Jaipur
Corresponding Author:
Dr. Uma Shankar Agarwal
397, Shree Gopal Nagar, Gopalpura Bypass, Jaipur, • Email: dr.usag@gmail.com

Abstract

Aspergillus is a mold whose spores are commonly found in air. It primarily causes infection in immunocompromised individuals. We report a rare case of osteomyelitis due to Aspergillus niger in an immunocompetent patient.

Key words: Fungal Osteomyelitis, Invasive Aspergillosis

Introduction

Aspergillus is a mold whose spores are commonly found in air. It primarily causes infection in immunocompromised individuals.[1] Three types of aspergillosis are seen: invasive aspergillosis, chronic (and saprophytic) forms of aspergillosis; and allergic forms of aspergillosis. Invasive aspergillosis (IA) usually involves the sinopulmonary tract, with the lung being the most common site of infection, while osteomyelitis due to Aspergillus species is rare.[2] We report a rare case of osteomyelitis due to Aspergillus niger in an immunocompetent patient.

Case Report

A 45 year old male, manual laborer by occupation presented to the OPD with complaint of sinuses over scalp for four years associated with bilateral hearing loss and loss of vision from right eye. According to his wife, he had headache predominantly on right side four years back which was followed by redness and swelling of right eye after 15 days. He took some treatment and when the swelling resolved there was corneal opacity and loss of vision. Over the next 5-6 months the pain persisted. Then he had difficulty in hearing from left ear followed by right leading to complete hearing loss in both ears. For the next one year there was no complaint except pain in the right frontal area. Thereafter an ulcerated nodule developed over occipital area with pus discharge. It was followed by formation of multiple nodules over occipito- frontal area of scalp and the mastoid area over the next 6-7 months. The nodules eventually became non-healing ulcers. Later on, the patient had discharge of pus from both ears and lateral margin of the left eye. These sinuses persisted for next one and a half years with on and off pus discharge. There was a history of weight loss of 7- 8 kg over the course of the illness. There was no history of any trauma, surgical intervention, cough, fever, night sweats and ear, nose or oral cavity infection prior to onset of symptoms. He had an MRI (brain and orbit) done in November 2013 suggesting ill defined diffuse lesion in the orbital fat in right retrobulbar region surrounding the extraocular muscles and the optic nerve and bilateral mastoiditis.

On examination there were multiple draining sinuses present over scalp with necrotic edges (Fig. 1-3). There was a greenish waxy discharge. The discharge was also coming through auditory meatus. In the right eye corneal opacity was seen. A sinus was also present over left cheek with discharge of clear fluid on mastication and talking. The patient was pale and had bilateral mobile, slightly tender posterior cervical lymph nodes.

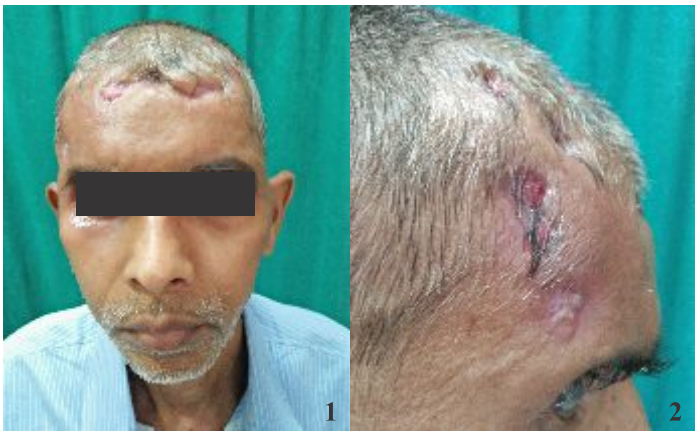

Figure 1 & 2 : 1: Multiple draining sinuses over frontal and temporal area of scalp with corneal opacity in right eye.
2: Multiple draining sinuses over frontal and temporal area of scalp

A preliminary diagnosis of scrofuloderma, actinomycetoma and subcutaneous mycosis were kept and all routine investigations of the patient were sent along with Mantoux test (MT), sputum for AFB, skin biopsy for histopathology and tissue culture. The pus was also sent for culture and KOH examination. Cartridge based nucleic acid amplification test (CBNAAT) was done from pus for tuberculosis. A contrast-enhanced magnetic resonance imaging (CEMRI) of brain and orbit was advised to assess the extent of the lesion. Patient was found to be severely anemic with

Haemoglobin 4.3g/dL and total red blood cell count 2.11*106/µL. His MT and CBNAAT were negative, chest X ray did not reveal any lesion and the KOH mount was negative for fungal hyphae. Histopathology showed granulomatous inflammatory lesion but no organism was seen on Periodic acid Schiff (PAS) and Ziehl-Nielson (ZN) stain. His CEMRI (brain) suggested multiple bilateral frontal and occipital cutaneous-subcutaneous lesions involving underlying bones with no gross intra cranial extension. There was soft tissue mucosal thickening in bilateral ethmoid air cells, frontal and sphenoid sinuses, suggestive of sinusitis and also there was evidence of bilateral mastoiditis. His CEMRI (orbit) suggested enopthalmos of right eye ball with loss of normal right retrobulbar fat. Skull X-ray showed osteolytic changes with destruction of outer and inner table of skull involving frontal and occipital bones. The pus culture grew Aspergillus niger. An ENT opinion was sought for ear discharge and hearing loss. His ear examination showed bilateral subtotal tympanic membrane perforation with polypoidal mucosa and was advised contrast-enhanced CT scan of temporal bone and audiometry. CECT (temporal bone) suggested multiple lytic lesions in frontal, left sided sphenoid, bilateral petrous and occipital bones along with sphenoid and maxillary sinusitis. A mass was seen causing encasement of petrous part of bilateral internal carotid arteries also causing destruction of bone forming inner ear. Also there was thickening of mucosa of middle ear cavity and epitympanum. The audiometry suggested bilateral hearing loss. Since skin biopsy was inconclusive and no conclusion could be reached about etiology on culture, a biopsy was planned from the mass seen in CECT (temporal bone). Histopathology revealed many bony trabeculae with focal presence of mucosal lining. Intertrabecular spaces showed mixed inflammatory infiltrate with presence of giant cells. On PAS stain, at one focus a single fungal colony was seen with few branching, septate hyphae, branching at acute angle [Fig. 4,5]. Due to presence of fungal hyphae in histopathology, Aspergillus niger was considered to be the causative organism as it was grown on culture.

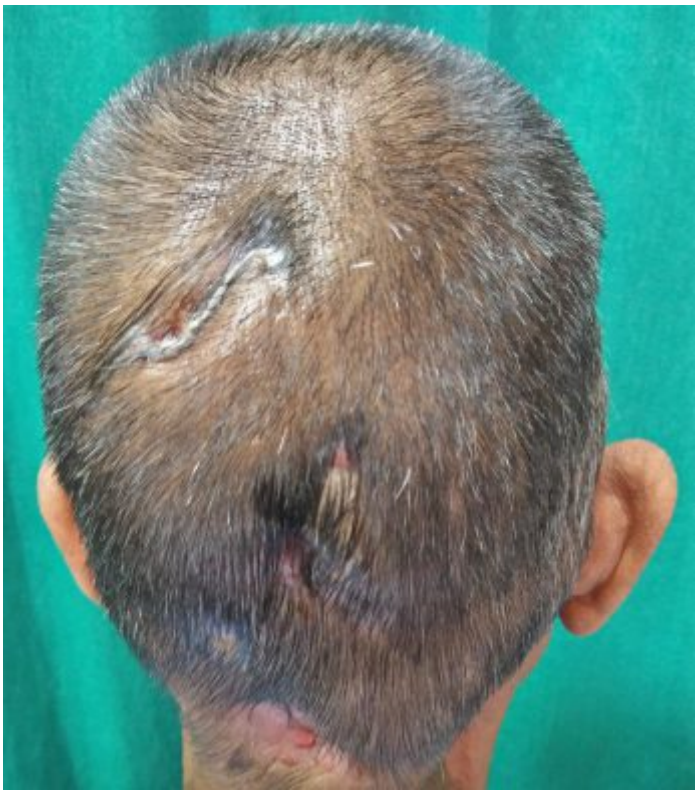

Figure 3 : Multiple draining sinuses over occipital area of scalp.

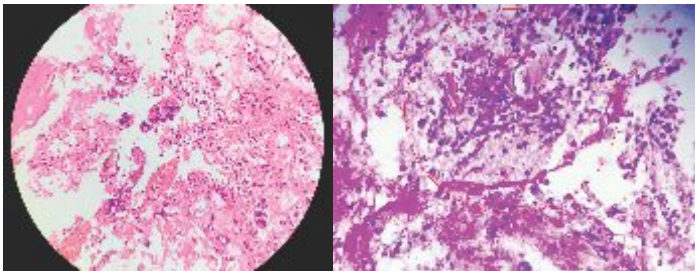

Figure 4 & 5: 4: On histopathology Intertrabecular spaces showed mixed inflammatory infiltrate with presence of giant cells (H&E,10X). **5:** On PAS stain, at one focus a single fungal colony was seen with few branching, septate hyphae, branching at acute angle. [In circle] (PAS, 40X).

The patient was started on liposomal amphotrericin B (1mg/kg/day) intravenously along with itraconazole 200 mg BD. The patient responded dramatically to the treatment with significant reduction in pus discharge after a week. The sinuses began to heal and granulation tissue was seen to grow in the ulcers. The discharge from the ear also reduced significantly.

Discussion

Aspergillus species are ubiquitous saprophytic organisms. More than 300 species are known, but only a few of them cause opportunistic infections.[2] Osteomyelitis due to Aspergillus is rare. It is caused by: [3,4]

(1) Contiguous spread of infection, like from sinus infection affecting cranium or pulmonary infection affecting ribs or vertebrae

(2) Hematogenous spread from a primary focus

(3) Trauma or maybe iatrogenic

The incidence of Aspergillus affecting the bone among all cases of invasive aspergillosis (IA) is estimated to be 3%.[2] Amongst the infective Aspergillus species the most common isolates from osteomyelitis lesions are Aspergillus fumigates followed by Aspergillus flavus and Aspergillus nidulans. Less frequently isolated species included Aspergillus terreus, Aspergillus niger, Aspergillus versicolor and Aspergillus flaviparus.[5]

Clinically IA manifests with pain and tenderness followed by sinus tract formation with purulent discharge (green waxy pus).[6] According to Infectious Diseases Society of America (IDSA), diagnosis of Aspergillosis requires histopathological documentation of infection and a positive microbiological culture from a normally sterile site. Other methods are PCR and detection of Galactomannan and (1-3)-ß-D-Glucan in serum and bronchoalveolar lavage. The IDSA recommended treatment for Aspergillus osteomyelitis is surgical intervention, where feasible, combined with voriconazole. Other useful antifungals

are liposomal amphotericin B, isavuconazole, caspofungin, micafungin, posaconazole and itraconazole. Therapy should be continued for a minimum of 8 weeks, frequently requiring longer courses (> 6 months).[7]

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