CASE REPORT

Atypical craniocerebral eumycetoma: A case report and review of literature

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ABSTRACT

Craniocerebral eumycetomas are rare. They usually present with scalp swelling and discharging sinuses. Radiologically, they present as space-occupying lesions. We report a case of eumycetoma involving the left parietal cortex, bone, and subcutaneous tissue in a young male, farm laborer, who presented with seizures and blurring of vision. Imaging showed a dural based lesions enhancing moderately on contrast. To the best of our knowledge and belief, ours is the first published case in the English Literature where a eumycetoma has presented as a mass lesion without discharging sinuses. It is imperative to keep such atypical features of an infective etiology in mind because they may be one of differentials of "dural" based lesions where only a biopsy may suffice in the absence of significant mass effect to prove the diagnosis.

Key words: Cranium, eumycetoma, fungal granuloma, osteomyelitis

Introduction

Mycetoma, the most often affecting lower extremities, is a chronic, localized, slowly progressive, granulomatous subcutaneous infection characterized by tumefaction (swelling), exudates containing grains composed of the etiological agent of the infection, draining to the skin surface via interconnected sinus tracts, and deformity.[1,2] It is caused either by fungi (eumycotic mycetoma) or filamentous higher bacteria (actinomycotic mycetoma). More than 20 species of fungi and bacteria have been identified as etiologic agents. They gain entry into the skin usually by implantation following trauma. We herewith report a patient with eumycotic mycetoma affecting the scalp, skull bone, dura and underlying brain parenchyma presenting with many atypical features.

Case Report

A 26-year-old male farm laborer presented with chief complaints of 3–4 episodes of generalized tonic-clonic seizures, blurring of vision and headache of 5 months duration without any history of fever or any other constitutional symptoms. There were no neurological deficits on examination. Also, no scalp swelling or draining sinuses were evident. Computed tomography scan of the brain revealed a left parieto-occipital moderately enhancing, dural based lesion with significant mass effect. The overlying bone showed hyperostosis with intermittent punched out defects [Figure 1]. He was taken up for emergency surgical debulking and biopsy due to persistent mass effect. Intraoperatively, subcutaneous tissue was found to be densely adherent to the parietal bone. The dura was thickened and was an adherent to the bone that was punched out at various places [Figure 3]. The lesion was excised en bloc along with involved dura and bone. Postoperative period was uneventful. Subsequent magnetic resonance imaging (MRI) showed adequate excision along with a resolution of mass effect [Figure 2]. Histology showed dense collagen infiltrated by inflammatory infiltrate extending into bone and underlying brain parenchyma. There were multiple suppurating granulomas composed of neutrophils, lymphocytes, mononuclear cells and few multinucleated gaint cells. In the center of the abscess, there were granules measuring 2–4 mm with central pale color and Splender-Hopple phenomenon. The central pale area was composed of vesicles and septate hyphae, highlighted on Gomori methenamine silver stain and periodic acid-Schiff stain [Figure 4]. Tissue was submitted for culture, but it did not yield any growth. Based on the morphology, a diagnosis of pale grain mycetoma, probably *Pseudallescheria* spp. was made. The patient was started on anti-fungal therapy with itraconazole 200 mg twice a day. Patient was doing well at 3 months follow-up.

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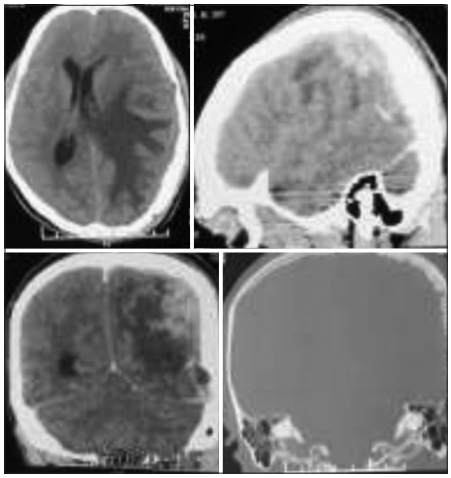

Figure 1: CT scan brain showing left parietal bone hyperostosis with subdural collection,focal cerebral oedema, mass effect ,midline shift and small hyper densities in parietal lobe

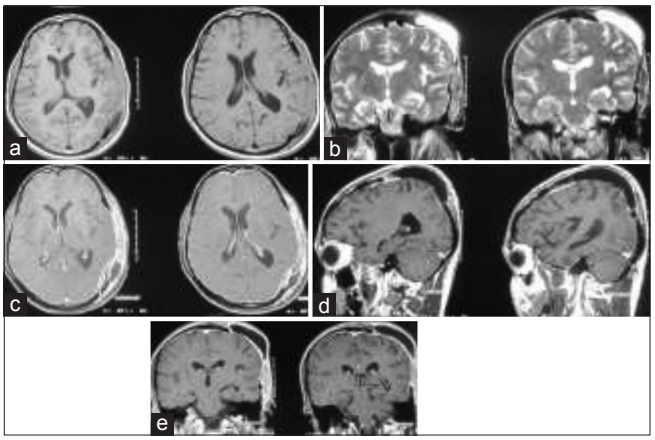

Figure 2: (a&b) MRI brain T1W and T2W axial section showing relieved mass effect with subcutaneous collection at operative site and no parenchymal lesion. (c,d and e) T1W post contrast of brain showing enhancement of subcutaneous collection at operative site

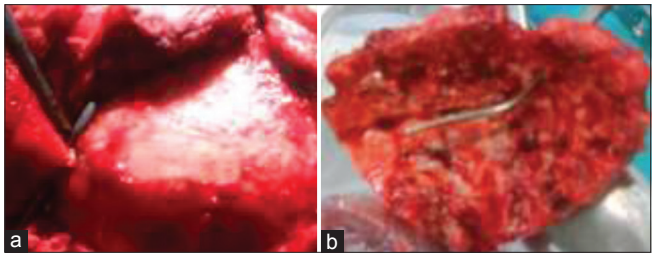

Figure 3: (a) Intraoperative picture showing subcutaneous tissue entering into brain through small whole in parietal bone (no external sinus to the skin). (b) removed parietal bone with bony destruction and punched out appearance

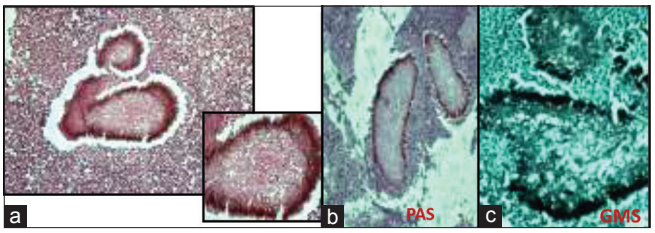

Figure 4: (a) photomicrograph showing granule with pale center and splender-hopple phenomenon amidst suppurative inflammation. Hematoxylin and eosin ×4 Inset showing eosinophilic material in the periphery of the granule with neutrophils clinging to it Hematoxylin and eosin ×40. (b) Photomicrograph showing filamentous hyphae in the center of the granule periodic acid schiff ×40. (c) Photomicrograph showing filamentous hyphae in the centre of the granule and vesicles in the periphery.Gomori s methenamine silver stain ×40

Discussion

The first description of mycetoma was attributed to Dr. John Gill, who reported "Madura foot" in a dispensary report of the Madras Medical Service of the British Army in India, in 1842.[3,4,5] In 1846, Colebrook from the same dispensary described further cases and introduced the term Madura foot for a distinct disease entity of that region. Vandyke Carter who studied the condition between 1860 and 1874, described the changes caused in the bony structures and soft tissues of the affected parts and then identified filaments of a fungus found in the grains within the sinus tracts of the diseased area.[6] Pinoy in 1913 recognized the possibility of classifying cases of mycetoma by grouping the causative organisms.[2] Usually the dorsal aspect of the foot is involved, but lesions of the hand, leg, torso, arm, and thigh have also been reported, lesions of scalp with involvement of cranial bones and brain is rare. When the scalp is involved, the infection usually starts in the back of the head or neck. The lesions are usually restricted to site of entry of organisms. There are a number of reports of cranial and intracranial mycetomas due to actinomycetes from India. *Pseudoallscheria boydii* and Madurella mycetomatis are the most common organisms causing eumycetoma. However, cranial eumycetomas are extremely uncommon and very rarely reported. Our patient had no such discharging sinuses. The surgery was intended to excise the lesion completely. However, only a biopsy may also suffice in the absence of any mass effect. Table 1 shows the summary of reported cases of eumycetomas till date. These lesions may also present without involvement of brain parenchyma. In 1950, Hickey reported on three patients of cranial eumycetoma without penetration of the dura.[7] One case involved the cranial vault quite extensively in a 20-year-old shepherd, and only a biopsy was done (not surgical excision). The other two cases, in a 30-year-old farmer and a 24-year-old shepherd, were sinocranial and involved the orbits and paranasal sinuses. In both cases, the lesion was surgically removed, necessitating enucleation of the eyeball in the farmer. In 1975, the first reported case of mycetoma involving the cerebral cortex in the English literature was reported by Natarajan *et al*.[3] Since then, the intra-parenchymal involvement has been reported once only in year 2008.[8] Ours is the third case in this series that had many differing salient "atypical" features. In our case, the diagnosis of eumycetoma was never suspected

Table 1: Review of literature

Case reports	Patient complaints	Past history	Imageology
Natarajan *et al.* 1975	25 years female farm laborer presented withScalp swelling of rightparietal region of twoand half years duration along with focal seizures and left hemiplegia History ofcarrying material over head was present	No history of previous surgery/trauma	Plain X-Ray-Intracranial space occupying lesion ; brain abscess at parietal region and bone and soft tissue involvement
Narayana *etal.* 2007	21-years female presented with history of pain in the left ear, serous discharge, impaired hearing for 6 months and right sided weakness along with headache and vomiting for 4 months	She underwent mastoidectomy on the left side 8 years before for chronic suppurative otitis media	CT scan of head revealed a solid cystic left cerebellopontine angle lesion causing severe brainstem compression and edema with hydrocephalus and erosion of petrous apex. MRI showed a multilobulated lesion in the left cerebellopontine Angle isointense on T1 weighted images and avid contrast Enhancement with no evidence of meningeal enhancement
Vamseemohan *et al.* 2008	18 years male farm laborer presented with left posterior parietal scalp swelling with discharging sinuses of one and half year duration with GTCS and doubtful history of thorn prick	Incision and drainage of left posterior parietal scalp swelling was done which resulted in discharging sinus	CT Scan-Intracranial space occupying lesion with punched out appearance of left parietal bone and thickened subcutaneous tissue
Munwwar ahmed 2011	35 years male presented with seizures 1 year, slowly increasing left parietal swelling 6 months, sudden onset of weakness of right upper limb and lower limb and inability to speak 6 days	History of trauma to left parietal scalp	MRI brain one year ago showed left parasagittal , homogenously enhancing, dural based mass lesion with surrounding oedema involving underlying cortex CT done at time of presentation showed intensely enhancing lesion with features of osteomyelitis in the adjacent parietal bone Repeat MRI after 1 year revealed iso- to hypointense mass and surrounding oedema in the left frontal region on T2W and FLAIR images
Present case	26 year male farm laborer presented with GTCS with no scalp swelling and discharging sinuses	No history of previous surgery/trauma	Dural based Left parieto-occipital enhancing lesion with hyperostosis and punched out appearance of the overlying bone

CT – Computed tomography; MRI – Magnetic resonance imaging; FLAIR – Fluid attenuated inversion recovery; GTCS – Generalised tonic clonic seizures

preoperatively or even intra-operatively due to the absence of purulent material, sulfur granules or a blackish pigmentation as it was a pale grain eumycetoma. The characteristic sign of pedal eumycetoma is dot in circle sign (T2-weighted hyperintense granuloma with central hypointense dots of fungal grains separated by hypointense fibrous walls). Also, a conglomeration of multiple hypointense dots of fungal grains against the background of inflammatory granulomas was also seen.[9] The grains were identified as hypointense dots on both T1-weighted and T2-weighted images, and their MRI signals were attributed to the magnetic-susceptibility of paramagnetic elements of grains. Radical surgical excision of the lesion is the mainstay of treatment as a response to medical therapy alone is not very effective and associated with late relapses.

Generalised tonic clonic seizures, Magnatic resonance imaging, Computed tomography.

Conclusions

Cerebral maduromycetoma is a rare entity. Only three cases including this one have been reported in the literature. Such atypical presentations of infective etiology should always be kept in mind while analyzing the differentials of a dural based lesion.

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