Case Report

Cranial Mycetoma: A Rare Case Report with Review of Literature

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Abstract

Mycetoma is a chronic granulomatous infection caused by fungi or bacteria, known as eumycetoma and actinomycetoma, respectively. Mycetoma commonly affects young males belonging to low socioeconomic strata, usually barefooted agricultural workers. It mainly affects lower and upper limbs presenting as a painless swelling with discharging sinus. Rarely, is it encountered in the intracranial location. The diagnosis relies on the clinical presentation and identification of the etiological agents within the tissue, by histology and special stains. It is important to specify the fungal or bacterial etiology, because the treatment of each is completely different. The management of such infections is challenging and should involve early diagnosis, the use of antibacterials or antifungals, and surgical removal of the lesion. To the best of our knowledge, only seven cases of intracranial mycetoma have been reported. The present case highlights the rarity of this lesion, thereby contributing to the existing literature and presenting its diagnostic implications.

Keywords: Actinomycetoma, eumycetoma, intracranial, mycetoma, temporal

Introduction

Mycetoma is a chronic, localized, slowly progressive, granulomatous infection.[1] Based on its etiology, mycetoma is referred to as eumycetoma when the infection is caused by filamentous fungi and actinomycetoma when the infection is due to actinomycetes.[2] Mycetoma commonly affects young adults in the age range of 20–40 years belonging to low socioeconomic strata; usually barefooted workers in the rural areas engaged in farming and rearing sheep.[3] It shows a male predominance with a sex ratio of 3 : 1. Repeated local minor trauma or penetrating injury provides a portal of entry for the organism. It mainly affects lower and upper limbs, although it may involve any part of the body.[4] However, lesions of the scalp with involvement of cranial bones and brain are rare. It is important to specify the fungal or bacterial etiology, because treatment modalities differ. Managing such infections are still challenging and treatment should involve early diagnosis, the use of antibacterials or antifungals and surgical removal.

We report a case of cranial mycetoma in the brain parenchyma presenting as an abscess in a young male.

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Case Report

An 18-year-old immunocompetent male presented with complaints of headache, fever, and an episode of convulsions. There was no neurological deficit on examination. Routine urine, stool, and blood analysis were normal. All laboratory parameters were within the normal limits. Furthermore, no scalp swelling or draining sinus was evident. NCCT head revealed an ill marginated hypodense lesion in the temporal lobe with contiguous peri-lesional odema [Figure 1]. Based on clinical and preoperative radiological findings, a provisional diagnosis of brain abscess was made. The lesion was excised and sent for the histopathological examination. Grossly multiple gray black tissue fragments were received, cut sections of which showed black granules. The postoperative period was uneventful.

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Histopathological examination showed predominantly areas of acute and chronic inflammation. At places, colonies of compact aggregates of thin filamentous structures surrounded by dense inflammatory infiltrate were seen [Figure 2]. The center of the abscess revealed granules with central pale color and Splender – Hopple phenomenon. Morphological diagnosis of mycetoma was made, and special stains were performed. Mycetoma colonies showed positivity for Periodic acid Schiff [Figure 3a] and Gomori-methenamine silver [Figure 3b]. Grams stain was done to differentiate between actinomycetes and eumycetes, and it was negative. Tissue was not sent for culture. The final diagnosis of eumycetoma in the temporal region was rendered.

Discussion

"Madura foot" was first described by Dr. John Gill in 1842 in a dispensary report of the Madras Medical Service of the British Army in India.[5] Pinoy in 1913 recognized the possibility of classifying cases of mycetoma by grouping the causative organisms.[6] It is endemic in many tropical and subtropical countries such as India, Pakistan, and parts of Africa.[7]

Young adult males in the age range of 20–40 years are more frequently affected. Farmers, workers, and students are affected most, but no occupation is exempted. In general, the exposed areas of the body that include hands and feet are more frequently involved with mycetoma. The lesions are usually restricted to the site of entry of organisms for few weeks leading to swelling and nodule formation, which after few months results in discharging sinuses, having the presence of grains, which are pathognomic.[4] Histologically, the organisms of actinomycetoma display Splendore – Hoeppli reaction which is a central matted appearance surrounded by a peripheral deposition of eosinophilic infiltrate.[8]

The demonstration of the organisms by Gram staining helps in species identification. The organisms found in actinomycetoma are 1 μ thin, branching filaments which are Gram-positive and acid-fast, whereas the granules of eumycetoma are Gram-negative, septate hyphae, 4–5 μ thick.[9] Although culture is the gold standard for the diagnosis of eumycetoma, it takes a long time with false-positive results due to contamination of sample.[10]

Craniocerebral involvement is a very uncommon phenomenon, and only seven cases are reported in the medical literature till date, excluding the index case [Table 1]. Five patients were male and two were female. The average age among all the reported cases was 24.3 years. Our patient was an 18-year-old immunocompetent male. Unlike others, our patient had no typical history of swelling or discharging sinuses. Furthermore, there was no calvarial or dural involvement. The lesion was exclusively within the temporal lobe.

In 1950, Hickey *et al*. reported three patients of cranial eumycetoma without penetration of the dura.[11] In 1975, the first reported case of mycetoma involving the cerebral cortex

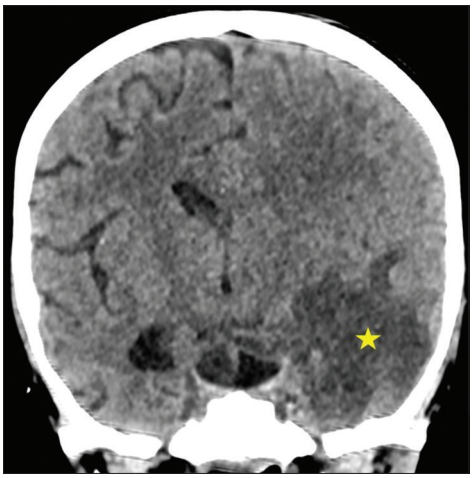

Figure 1: Coronal noncontrast computed tomography scan shows an ill-marginated hypodense lesion (yellow star) in the left temporal lobe with contiguous edema

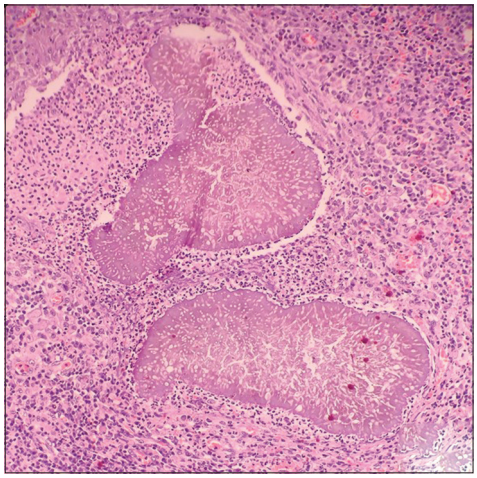

Figure 2: Photomicrograph showing granule with pale center and Splender-Hopple phenomenon amidst suppurative inflammation (H and E, ×200)

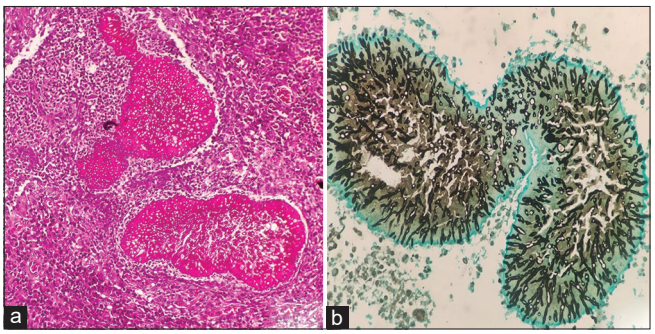

Figure 3: (a) Filamentous hyphae in the center of the granule periodic acid Schiff (PAS, ×200). (b) Filamentous hyphae in the center of the granule in the periphery-GMS stain (GMS, ×400)

in the English literature was reported by Natarajan *et al*.[5] The patient had a pyogenic brain abscess at the site of the cranial mycetoma. In 2007, a case of cerebellopontine angle eumycetoma in a young female was reported.[12] In 2008,

Table 1: Review of literature of intracranial mycetoma

Authors/ year	Age/ sex	Clinical features	History of trauma	Imaging	Culture	Management
Natarajan *et al*., 1975	25/ male	Focal seizure, left hemiplegia, discharging scalp sinus	Yes	X-ray: Right parietal bone defect with loculated air. Carotid angio: Left side shift of right ACA	*M. mycetomatis*	Surgery-twice. Chloromycetin, streptomycin penicillin
SaiKiran *et al*., 2007	21/ female	Left ear discharge with hearing loss and right hemiparesis	No	CT: Left CPA solid-cystic lesion with erosion of petrous. MRI-T1 hypointense with contrast enhancement	*P. boydii*	Surgery, antifungal
Beeram V *et al*., 2008	18/ male	Discharging scalp sinus and generalized seizure	No	CECT: Parietal punched-out bony lesion with enhancing intra and extradural mass	*M. mycetomatis*	Surgery, antifungal
Ahmed *et al*., 2011[14]	31/ male	Scalp mass, seizure, and right hemiparesis	Yes	CECT: Dural-based enhancing lesion with osteomyelitis of skull bone. MRI: Left parasagittal enhancing dural-based mass. Dot-in-circle sign in T2W	*M. grisea*	Surgery, antifungal
Goel *et al*., 2012[15]	17/ female	Scalp mass, seizure, and discharging sinus	Yes	X-ray: Right parietal lytic skull lesions. CECT: Hyperdense, enhancing extra-axial mass with scalloping and erosion of overlying bone	*M. mycetomatis*	Surgery, antifungal
Rao *et al*., 2015	26/ male	Generalized seizure, blurring of vision, and no scalp lesion	No	CECT: Left parietooccipital enhancing dural-based lesion with bone hyperostosis along with punched-out lesion	*P. boydii*	Surgery, antifungal
Behera *et al*., 2018[16]	32/ male	Generalized seizure and left-sided weakness	Yes	X-ray of the skull bone revealed multiple radiolucent areas, MRI of the brain revealed an extra-axial lesion	NA	Surgery, antifungal
Present case	18/ male	Headache, fever, and convulsions	No	NCCT of the head revealed an ill-marginated hypodense lesion in the temporal lobe with contiguous perilesional edema	NA	Surgery, antifungal

M. mycetomatis: Madurella mycetomatis, P. boydii: Pseudallescheria boydii, M. grisea: Madurella grisea, NA: Not available, ACA: Anterior cerebral artery, CT: Computed tomography, CPA: Cerebellopontine angle, MRI: Magnetic resonance imaging, CECT: Contrast-enhanced computed tomography, NCCT: Noncontrast computed tomography

Beeram *et al*. described a case of maduromycetoma involving the left parietal cortex, bone, and subcutaneous tissue in an 18-year-old young male farm laborer who presented with left parietal scalp swelling that had progressed into a relentlessly discharging sinus.[13] In 2010, Maheshwari *et al*. reported a case of Madurella infection of the paranasal sinuses that extended into the intracranial compartment. Rao *et al*. in 2015 reported a patient with eumycotic mycetoma affecting the scalp, skull bone, Dura and underlying brain parenchyma presenting with atypical features.[6]

Early diagnosis remains critical for treatment, reducing the associated morbidity with this condition. Surgical excision of the lesion is the mainstay of treatment as response to medical therapy alone is not very effective and associated with late relapses. It is also important to initiate the correct treatment with antibacterials or antifungals based on the species for optimal response. Our patient was started on anti-fungal therapy and is doing well at 6 months' follow-up.

Conclusion

Cerebral mycetoma is a rare entity. Only seven cases have been reported in the literature. Because of the chronic nature of disease, it is often diagnosed at an advanced stage. Hence, there is a need for a correct and prompt diagnosis after meticulous clinical examination and histopathology, along with special stains for species identification. It is important to distinguish between fungal or bacterial etiology as the treatment is different for each entity. Early diagnosis followed by medical therapy combined with surgical debridement is the treatment of choice, although long-term follow-up is necessary.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given his consent for his images and other clinical information to be reported in the journal. The patient understands that his name and initials will not be published and due efforts will be made to conceal his identity, but anonymity cannot be guaranteed.

Research quality and ethics statement

The authors followed applicable EQUATOR Network (http://www.equator-network.org/) guidelines, notably the CARE guideline, during the conduct of this report.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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