

CASE REPORT WITH REVIEW OF LITERATURE

Intracranial en-plaque tuberculoma impersonating en-plaque meningioma: Case report and brief review of literature

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ABSTRACT

Background: Tuberculosis of the central nervous system continues to be a major health hazard in developing countries like India. There are various manifestations of central nervous system tuberculosis including meningitis and space occupying lesions. We present a case of tuberculoma en-plaque which is often initially confused with a meningioma on presentation. We also review the literature relevant to this unusual entity.

Methods: A search of PubMed, PubMed Central, the Cochrane Library, and MEDLINE were performed to identify all English language reports of intracranial en-plaque tuberculomas. The combinations of text strings "tuberculoma," "en*," and "plaque" were used to perform the query on PubMed. Only the studies reporting intracranial en-plaque tuberculomas were included.

Results: Literature review revealed six reports on en-plaque tuberculomas including seven patients. The mean age at presentation was 40.5 years, and no sex predilection was seen. Five of the six reported studies used anti-tuberculous therapy with or without surgical excision of the mass. Most of the patients in reported cases recovered fully.

Conclusion: It is imperative to consider tuberculoma as an important differential when encountering intracranial en-plaque masses as en-plaque tuberculomas can be effectively treated with a combination of anti-tuberculosis therapy and surgical resection, yet consequences of a missed or late diagnosis can prove fatal.

Key words: En-plaque tuberculoma, meningitis, space occupying lesions, tuberculoma

Introduction

Intracranial tuberculomas are usually secondary to a tuberculous focus elsewhere in the body, most commonly the lungs.^[1] However, the absence of extra-cranial evidence of the disease is not uncommon.^[2] Intracranial tuberculoma has been described both in the supratentorial and infratentorial compartments, but the incidence of intracranial tuberculomas has been on a decline.

An en-plaque tuberculoma is an uncommon variant of a tuberculoma, and few cases have been described in the literature. It resembles an en-plaque meningioma in the first instance, both radiologically and in gross appearance. The treatment options include medical therapy, surgical excision, or a combination of both. Here, the authors report a case of en-plaque tuberculoma and review the literature of this uncommon entity.

Case Report

A 73-year-old male presented to the outpatient Department with complaints of a headache, generalized tonic-clonic

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seizure episodes, left hemiparesis for 6 months, and vomiting for 2 months. His weakness had gradually progressed as he was walking with support at presentation, seizure frequency was one to two episodes per month, and the headache was localized to the right frontoparietal region. He had no visual complaints, history of trauma, fever, or a chronic cough. There was no past history of any major illnesses, and family history was unremarkable.

On examination, the patient was afebrile, conscious and oriented to person, place, and time. The higher mental functions were normal. No cranial nerve deficits were seen, and fundoscopic examination was normal. Left hemiparesis was apparent with power grade 3/5 in both upper and lower limbs with an associated increase in tone and exaggerated deep tendon reflexes. Babinski's sign was present on the left side. The neck was supple with no signs of meningism.

All routine investigations were normal including the chest X-ray. A magnetic resonance imaging (MRI) brain was done which showed an irregular, ill-defined hypointense lesion over right frontoparietal convexity on T1-weighted with intense enhancement on gadolinium contrast [Figure 1], hyperintense on T2-weighted. A presumptive diagnosis of an en-plaque meningioma was made, and surgery was planned accordingly. During surgery the resection of a firm, slightly yellowish, mass adherent to the dura, and infiltrating the brain was achieved [Figure 2]. Part of the adherent dura was excised with the mass [Figure 3]. The overlying bone was normal. The histopathological examination showed multiple tubercles

comprising necrotic areas, epithelioid cells, Langhans type of giant cells, and acute and chronic inflammatory cells [Figure 4]. Ziehl Neelsen staining showed the presence of acid-fast bacilli confirming the diagnosis of tuberculoma. The postoperative computed tomography (CT) scan showed a complete resection of the en-plaque mass [Figure 5].

The postoperative recovery was uneventful, and the patient was discharged on anti-tuberculosis therapy (ATT) and phenytoin. At 2 and 6 months follow-up, he had completely recovered from hemiparesis and was symptom-free.

Methods

A search of the existing medical literature was done in an attempt to include all reports of en-plaque tuberculomas, as well as manuscripts, detailing imaging and histopathological characteristics, management, and outcomes. A search of PubMed, PubMed Central, the Cochrane Library, and MEDLINE were performed to identify all English language reports on en-plaque intracranial tuberculomas. The combination of text strings "tuberculoma," "en*," and "plaque" separated by Boolean operators was used to perform the query on PubMed. Only studies reporting intracranial en-plaque tuberculomas were included.

Results

The review of the literature yields very few reports on the presentation of en-plaque tuberculomas. The initial literature search yielded 16 results; command/tuberculoma*[title]

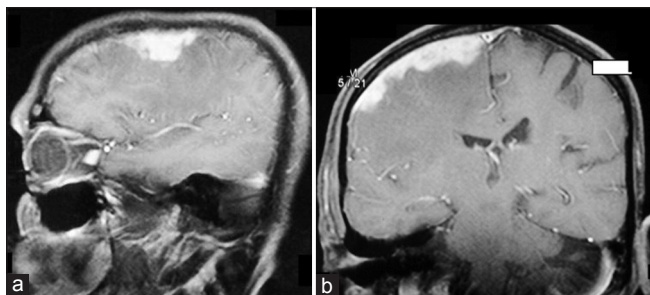


Figure 1: Location of en-plaque tuberculoma on T1W Gadolinium-enhanced magnetic resonance imaging. Sagittal view (a) and coronal view (b)

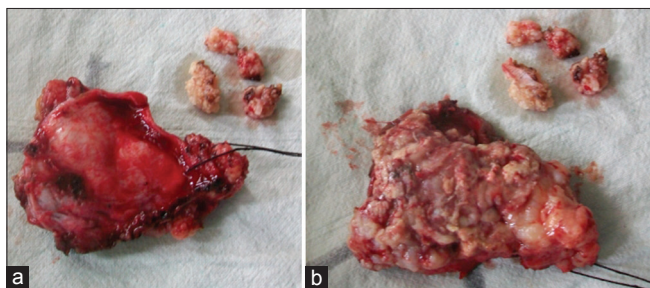


Figure 3: Gross appearance of the tuberculoma with adherent dura (a) and the parenchymal surface (b)

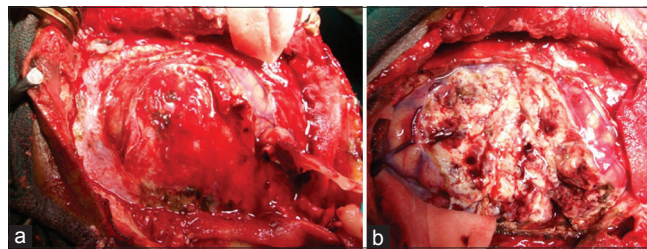


Figure 2: Intraoperative photograph. Dura adherent to the underlying tuberculoma (a). Brain after removal of the tuberculoma (b)

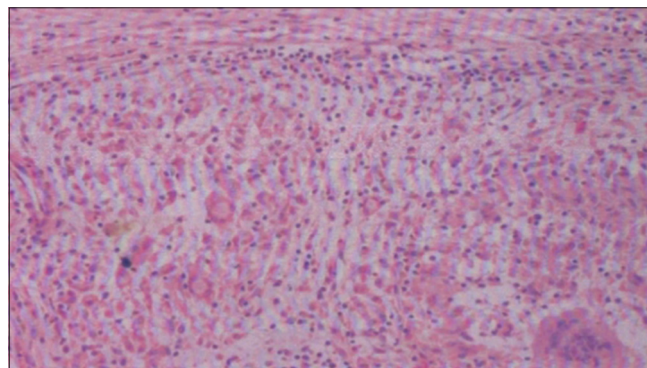


Figure 4: Histopathological microscopic picture showing Langhans giant cells, epithelioid cells, inflammatory cells, and necrotic areas

and (en* OR *plaque). A careful review of the titles, abstracts, and where needed, the manuscript, helped us to determine that 10 articles were not relevant to this review as they did not focus on intracranial en-plaque tuberculoma. This left six studies which fit out inclusion criteria.

Being such a rarely reported diagnosis, there is a lack of unifying characteristics that can aid in understanding of this disease process. The review of the literature yielded a sum of seven cases in six studies.^[2-7] The average age of presentation was 40.5 (25–73) years with no increased prevalence by sex. Relevant particulars of these reported cases are detailed in Table 1.

Discussion

Intracranial tuberculomas were very common in the past, reportedly up to 30% of all intracranial tumors.^[8] Their incidence have been on a decline ever since and presently are rarely seen in the developed world. Nevertheless, it is still fairly common in developing countries like India.

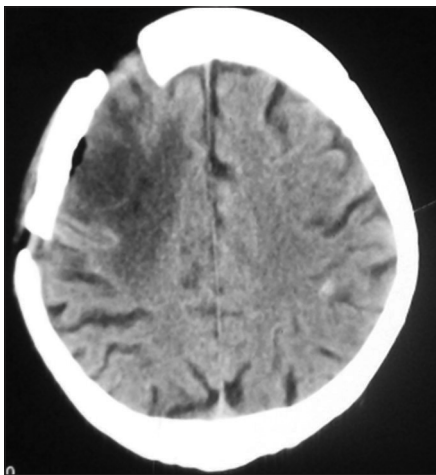


Figure 5: Postoperative computed tomography scan. Hypodense area showing total removal of en-plaque mass

Intracranial tuberculomas have been described both in the supratentorial and infratentorial compartments, the latter being more common in children.^[9] They usually result from a hematogenous spread of tubercle bacilli to the leptomeninges and brain parenchyma from tuberculous infection elsewhere in the body, commonly the lungs; however, the reported incidence of a concomitant extra-cranial tuberculous infection is only 30–50% of cases.^[8]

Tuberculoma en-plaque is a rare manifestation of intracranial tuberculoma wherein the mass is diffusely infiltrating the dura, often resembling a meningioma en-plaque. It was first reported as an autopsy finding in 1927 by Pardee and Knox^[10] who described it as, “a plaque like meningitic process without exudation, usually situated in the frontal and parietal regions.” They have been described along the cerebral convexity,^[2,4] tentorium cerebelli,^[5,6] dura over petrous bone,^[3] and posterior falx.^[7] They are usually seen in young adults, as opposed to our case who was 73-year-old. They usually present with a headache, vomiting, blurring of vision, focal seizures or weakness of the limbs, and neck tilt.^[4,5] On CT scan, it appears as a dural based, densely enhancing mass.^[3] MRI is the investigation of choice in which the lesion is hypointense on T1-weighted, and a hypointense lesion with surrounding hyperintense edema on T2-weighted and enhances well with Gadolinium contrast.^[2] Aggressive surgical resection has been the approach toward en-plaque tuberculomas, as was done in our case, probably due to the imprecise preoperative diagnosis. Since, the medical therapy is generally effective for intracranial tuberculomas; it is desirable to have a high index of suspicion for these cases.

Neurological manifestations of en-plaque tuberculomas largely depend upon location within the central neuraxis, however, common unifying symptoms include nausea, vomiting, headache, seizures, and vision changes.^[6,11-14] Typical management in cases reported include either medical management with ATT and steroids or surgical excision followed

Table 1: Review of literature on reported cases of intracranial en-plaque tuberculoma

| Study | Age (years)/sex | Presentation | Management | Imaging | Outcome |
|----------------------------------|-----------------|------------------------------------|-------------------------|--|--|
| *Welchman 1979 ^[3] | NA | NA | Surgical resection | Mass effect; poor to no peripheral definition; isodense to slightly hyperdense on CT without calcification | NA |
| Bauer et al. 1996 ^[2] | 34 male | Weakness, HA, numbness | ATT, surgical resection | Hypointense of T1 MRI, hyperintense on T2 MRI; mass effect | Full recovery |
| Ng et al. 1996 ^[4] | 35 female | HA, vomiting, syncope | ATT, surgical resection | Densely enhancing mass with mass effect and calcifications | Moderate recovery with right-hand weakness |
| Singh et al. 1999 ^[7] | 62 male | HA, vomiting, seizure, stiff neck | ATT + steroids | NA | Full recovery |
| Dubey et al. 2002 ^[5] | 14 female | HA, visual blurring | ATT, surgical resection | NA | Full recovery |
| Alkan et al. 2003 ^[6] | 25 female | HA, hearing loss, neck pain, fever | ATT + steroids | Hypointense on T1 MRI; hypointense centrally and hyperintense peripherally on T2 MRI | Full recovery |
| Kumar 2013 (present report) | 73 male | Seizure, HA, hemiparesis | ATT, surgical resection | Hypointense on T1 MRI; Hyperintense on T2 MRI | Full recovery |

*Report of two cases. ATT – Anti-tuberculous therapy; CT – Computed tomography; HA – Headache; MRI – Magnetic resonance imaging; NA – Not available

by ATT. Of the seven reported cases reviewed, two underwent only medical management, two had surgical excision only, and three had a combination of surgical intervention and medical management, as was the clinical course in our patient [Table 1]. There was a low complication rate reported from surgical resection of en-plaque tuberculomas; of the cases reviewed, all experienced a full recovery with the one exception of mild hand weakness.

Previous reports described CT imaging characteristics of tuberculoma en-plaque to be initially ambiguous from a meningioma, noting poorly defined density and contrast enhancement varied from moderate to intense.^[3,4] Preoperative differentiation of tuberculoma en-plaque from a meningioma may allow a trial with medical management with ATT and steroids. Singh *et al.* described the use of polymerase chain reaction assay and DNA amplification to diagnose en-plaque tuberculomas.^[7] Preoperative diagnosis of tuberculoma en-plaque is otherwise challenging, especially in the absence of extra-cranial tuberculous affection. Most cases reported were confirmed postoperatively (including our case) by histopathological evaluation of the excised mass and demonstration of granulomas, caseating necrosis, and Langhans type giant cells consistent with tuberculosis.

Conclusion

The authors present a case report of an en-plaque tuberculoma, as well as provide, to our knowledge, the only compiled analysis of all reported cases with aims to establish unifying presentations of the disease, management strategies used and outcome reports. Though a rare diagnosis, areas in which tuberculosis is endemic should consider en-plaque tuberculoma among the differential of presenting en-plaque mass lesions. En-plaque tuberculoma can be effectively treated with a combination of ATT and

surgical resection, yet consequences of a missed or late diagnosis may prove fatal.

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Conflicts of interest

There are no conflicts of interest.

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