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Case Report

Isolated Tuberculoma Mimicking Foramen Magnum Meningioma in the Absence of Primary Tuberculosis: A Case Report

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Central nervous system tuberculosis is a devastating complication of systemic tuberculosis. Intradural extramedullary (IDEM) tuberculoma at the foramen magnum is rare, and mimics en plaque meningioma. We report the case of a 53-year-old woman who presented with dysesthesia of the tongue and lower cranial nerve (CN) palsy, with onset 4 months prior to admission. The neurologic examination revealed left upper-limb weakness and hypoesthesia on the sole and dorsum of the left foot. Other physical examinations revealed no features of tubercular infection. Laboratory investigations likewise showed no signs of infection or inflammation. Magnetic resonance imaging of the brain showed an IDEM mass originating from the left intradural surface at the foramen magnum extending to the C2 segment and compressing the brainstem and upper cervical cord. The mass was isointense/hypointense on T1- and T2-weighted images and homogeneously-enhanced on postcontrast images. The lesion also exhibited the dural-tail sign and was preoperatively diagnosed as en plaque meningioma. The patient underwent surgery via the left transcondylar fossa approach with partial laminectomy of the atlas. Intraoperatively, the mass exhibited a dural origin and encased the vertebral artery and lower CNs, with strong adhesions. While the histopathological study of the mass was strongly suggestive of tuberculoma with multifocal granulomas, caseous necrosis, and Langerhans giant cells, extensive diagnostic studies failed to detect *Mycobacterium tuberculosis* itself. Although the patient had recurrence with multisystem involvement, she responded well to antitubercular treatment. IDEM tuberculoma of the foramen magnum may present as en plaque meningioma. Histopathology is required for a definitive diagnosis. Prompt surgical resection and decompression with adequate antitubercular treatment yield better neurological outcomes.

Keywords: Isolated central nervous system tuberculoma, Intradural extramedullary tuberculoma, Foramen magnum, Far-lateral approach

INTRODUCTION

Tuberculosis (TB) is still an important pathology in the developing countries and recently in the developed countries as well. Extrapulmonary complications of TB is seen in 15%–21% of patients infected with tubercle bacilli.[1] Central nervous system (CNS) involvement as extrapulmonary complication is seen in approximately 10% of TB patients with the most common presentation being tuberculous meningitis (TM).[2,3] Tuberculomas, a less common presentation, are well defined focal masses resulting from *Mycobacterium tuberculosis* infection and mostly occur as a paradoxical response to antitubercular treatment (ATT). Based on their location, CNS tuberculoma may be extradural, intradural extramedullary (IDEM) or intramedullary.

IDEM tuberculoma is a rare presentation and is notorious as it mimics en-plaque meningioma. IDEM tuberculoma mostly occurs secondary to TM, however a few cases have been reported in absence of primary tubercular disease.[2,4,5] To the authors' knowledge, this is the fourth case of isolated IDEM tuberculoma of the spinal cord in absence of primary TB. This rare case is also featured with its unique imaging findings mimicking foramen magnum meningioma.

CASE REPORT

A 53-year-old lady presented with the complaints of dysesthesia of the tongue and numbness over the left side of the face for 4 months prior to presentation. Neurologic examination revealed slight weakness of her left upper limb and hypoesthesia over the medial aspect of left sole and foot. Deep tendon reflexes were normal. Other physical and systemic examinations revealed no abnormalities. Laboratory investigations were also within normal limits and there we no signs of infection or inflammation. The patient's CRP was tested multiple times and was within the range of 0.04–0.33 mg/L and did not have a single value. Magnetic resonance imaging (MRI) of brain revealed a mass extending from the foramen magnum to the craniovertebral (CV) junction and compressing the brain stem and upper cervical cord. The mass was hypo- to iso-intense on T1-(TIWI) and T2-weighted images (T2WI) and was homogeneously enhanced on postcontrast images with dural-tail sign (Fig. 1A-C). Cerebral angiography showed faint tumor staining from the muscle branches and pial vessels. Based on these imaging findings, the mass was diagnosed as en-plaque meningioma of the foramen magnum and was opted for surgical resection.

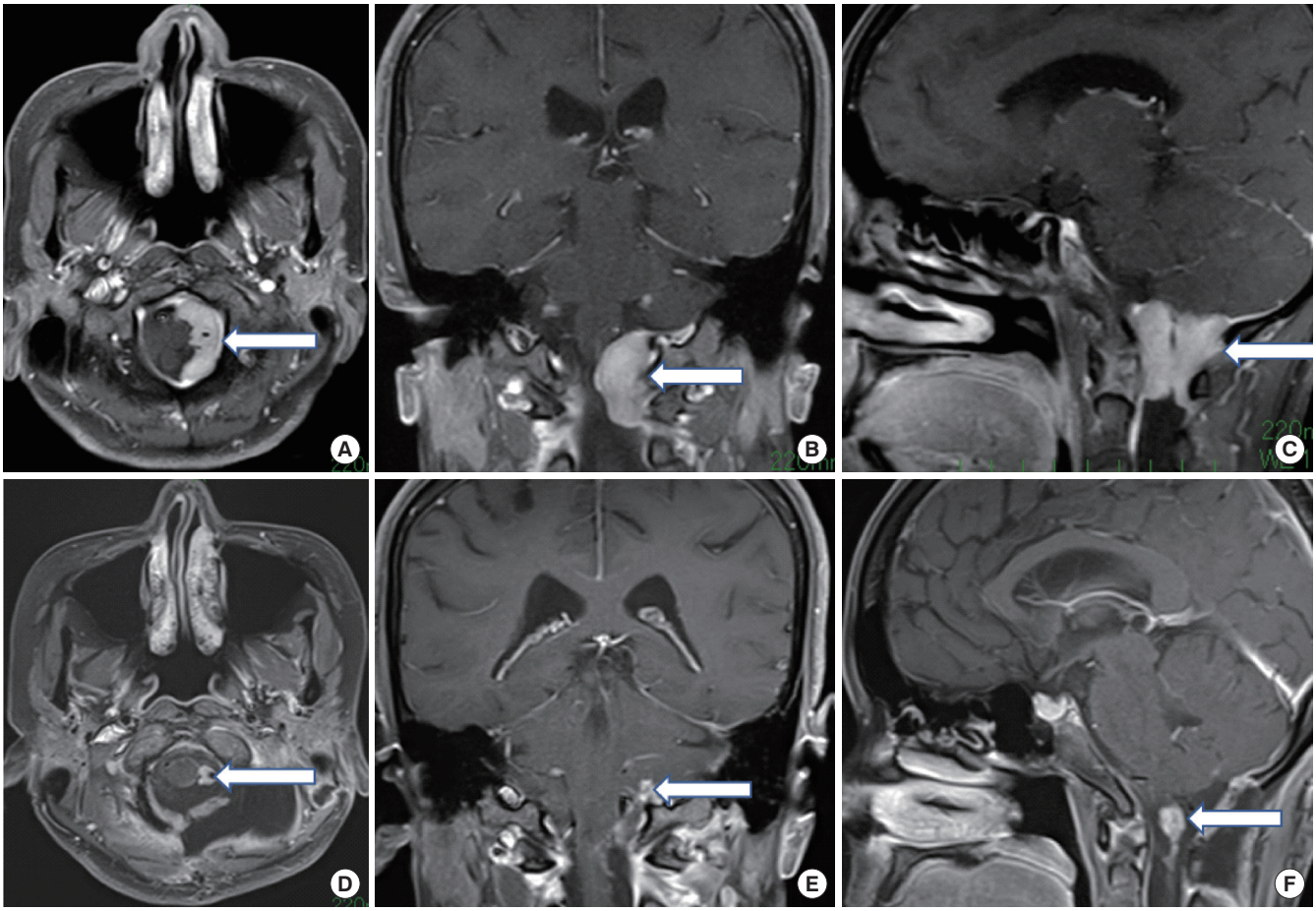

Fig. 1. (A-C) Preoperative magnetic resonance (MR) images of patient with isolated intradural extramedullary tuberculoma showing homogeneously enhanced mass and dural-tail sign and significantly compressing the upper cervical cord (arrows). (D-F) Postoperative MR images of the patient showing residual mass (arrows) strongly adhered to the vertebral artery and posterior spinal arteries.

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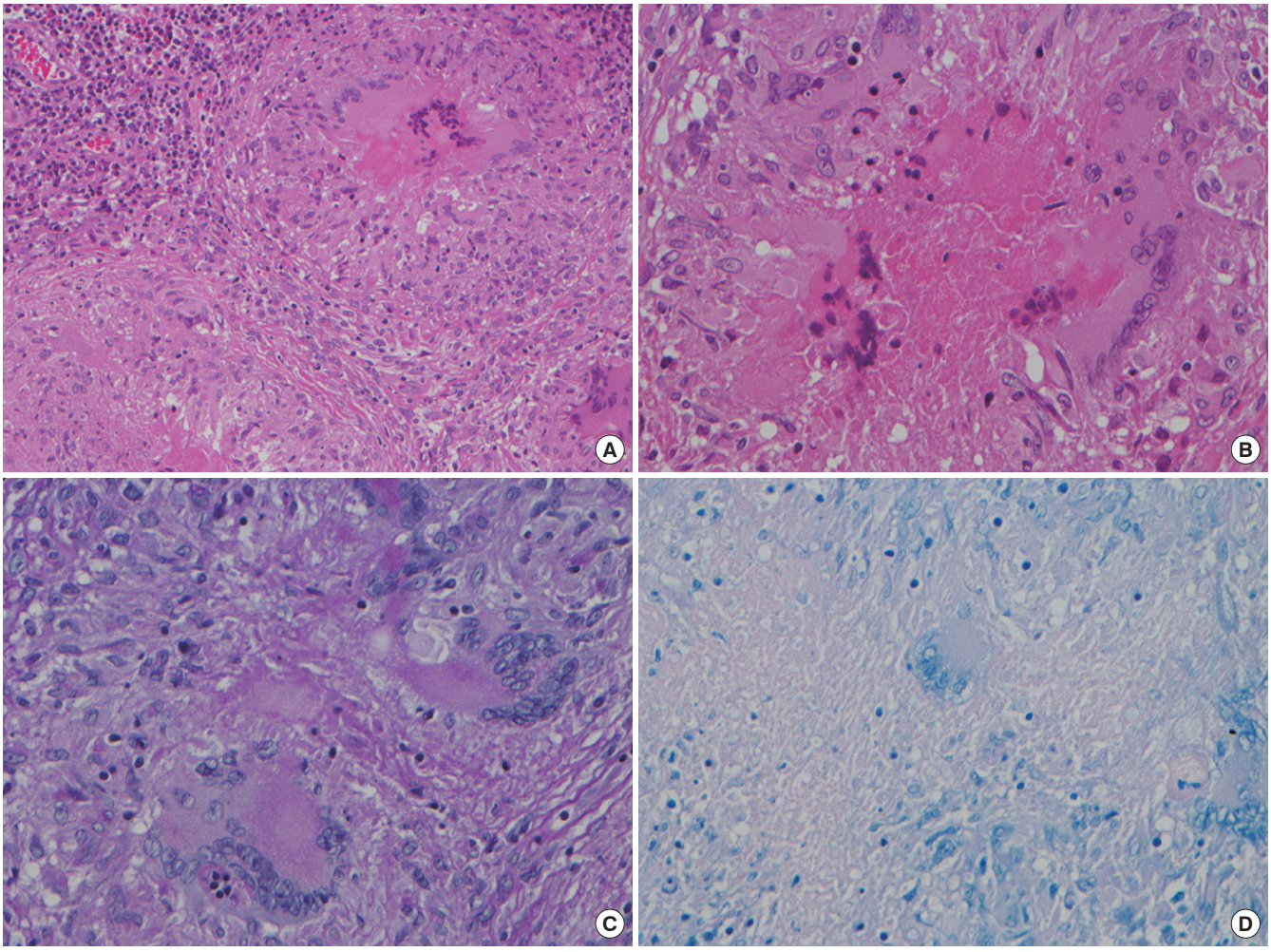

Fig. 2. Histopathological study showing chronic granulomatous inflammation with multifocal granulomas with caseous necrosis, Langerhans giant cells and epithelioid cells suggestive of tuberculoma (Hematoxylin and Eosin stain: A, ×200; B, ×400). The mass was negative for Periodic acid–Schiff stain (C) and Ziehl–Neelsen stain (D).

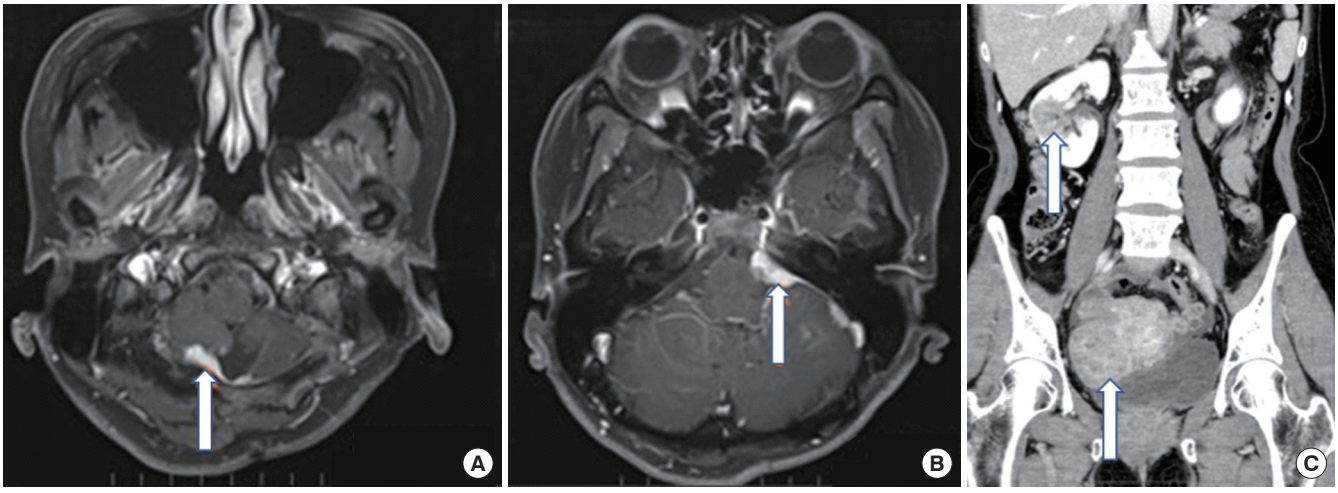

Fig. 3. Magnetic resonance images during observation period showing dissemination of the disease with multisystem involvement, dural dissemination (A, B) (arrows), right renal and tubo-ovarian mass (C) (arrow).

The surgery was performed via the left transcondylar fossa approach with partial laminectomy of the atlas. The firm mass with strong dural attachment encased the left vertebral artery (VA), lower cranial nerves, C2 rootlets, and the posterior spinal arteries. The surgery achieved decompression of the brainstem and upper cervical cord. A portion of the mass around the VA was intentionally left because of its strong adhesion to the arterial wall (Fig. 1D-F). Postoperatively, there was remarkable improvement in the neurological symptoms with no additional neurological deficits.

Histopathological examination revealed the mass to be a chronic granulomatous inflammation with multifocal granulomas of various sizes with caseous necrosis, Langerhans giant cells and epithelioid cells which are strongly suggestive of tuberculoma (Fig. 2). However, cultures did not grow *M. tuberculosis* and the stains were negative for acid-fast-bacilli and fungi. Polymerase chain reaction was also negative for tubercle bacilli complex. Computed tomography scan of the whole body revealed no focal lesions other than residual mass in the CV junction. In the absence of direct evidence of presence of *M. tuberculosis*, the patient was kept under observation without specific antitubercular treatment. The patient demonstrated recurrence of the disease with extensive dural involvement and right tubo-ovarian and renal mass 10 months after the surgery (Fig. 3). Following this, even though acid-fast bacilli were not isolated, based on the typical histopathological findings of a tuberculous granuloma, the initial mass was finally diagnosed as IDEM tuberculoma and rest as tubercular dissemination. She is now under treatment with antitubercular drugs and is showing good response to the treatment with gradual decrease in the size of extracranial masses.

This study was approved by the ethics committee of Hiroshima University (appproval number: 259).

DISCUSSION

1. IDEM Tuberculoma of the Spinal Cord

Extrapulmonary complications of TB are commonly seen in TB endemic regions and in immuno-compromised patients. CNS involvement by TB can be either intracranial or spinal. TM is the most common presentation of intracranial tuberculous infection and tuberculoma involving the cerebrospinal axis presents as a very rare complication.[2,3,6] Based on location of tuberculoma, it can be extradural, IDEM and intramedullary. Intradural tuberculoma is estimated to be approximately 2%–5% of CNS tuberculoma while IDEM location is even rarer.[2,3,7,8] Roca[7] reported a review of 22 cases of IDEM tuberculoma of the spinal cord from 1981 to 2004. According to the review, IDEM tuberculoma occurred predominantly in adolescents and young adults with median age of 27 years (interquartile range, 19–37 years). There was no apparent sex predilection (male:female = 1.2:1). Human immunodeficiency virus-infection with immunodeficiency was reported in a minority of patients (3 of 22) with no immunodeficiency reported in other patients. All but single case of IDEM tuberculoma occurred weeks after TM despite of antecedent ATT. This paradoxical formation of the tuberculoma may represent the reaction of the host immunity to the circulating antigens released from bacilli killed by ATT.

Although multiple sites were involved in IDEM tuberculoma, the most frequent site was thoracic spine (~95%) followed by lower cervical spine and least at lumbar region. Similar to the Pott's spine, this predilection may be a result of hematogenous spread of primary or postprimary pulmonary infection. To the author's knowledge, this case represents the first reported case of IDEM tuberculoma at foramen magnum mimicking an en-plaque meningioma.

Table 1. Summary of isolated tuberculoma of the spinal cord in the absence of primary tuberculosis

Study	Age (yr)/sex	Location/level	Immunocompromised	MRI features
Konar et al. (2011)[4]	40/M	L1–L4 spine	No	T1WI: isointense T2WI: heterointense Post-Gd: homogenous enhancement
Mirzai (2005)[2]	40/M	C6–T1 levels	No	T2WI: iso-/hypointense Post-Gd: marked enhancement
Compton and Dorsch (1984)[5]	45/M	C3–T1 levels	No	N/A (myelography)
Present case (2018)	53/F	Foramen Magnum, CV junction, C1 level	No	T1WI: isointense T2WI: isointense Gd-T1: homogeneous enhancement

MRI, magnetic resonance imaging; TIWI, T1-weighted image; T2WI, T2-weighted image; Gd, gadolinium; CV, craniovertebral.

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2. Isolated Tuberculoma of the Spinal Cord in Absence of Primary TB

Only 4 cases (including the present case) of isolated tuberculoma of the spinal cord in absence of the primary tubercular disease have been reported (Table 1). The exact mechanism of these rare instances is not clear but may have occurred as a host response to subinfectious dose of *M. tuberculosis* circulating in the body fluids. Apart from absent constitutional symptoms and normal acute phase reactants, IDEM tuberculoma presents with myelopathy manifested as various combination of sensory, motor and bowel-bladder symptoms. Given the fact that clinical presentation of IDEM tuberculoma is nonspecific and the general symptoms of tubercular infection might also be absent, we need high index of suspicion for the exact diagnosis.

The imaging features of IDEM tuberculoma are also nonspecific and as seen in our case, may mimic en-plaque meningioma. In MRI as well, the signal intensities were similar to that of meningioma with iso-/hypointense on T1WI and iso-/hyperintense on T2WI with homogeneous enhancement on postcontrast images. En-plaque meningioma usually derives its blood supply from the pial vessels and the same findings were seen in IDEM tuberculoma as well as in our case. The imaging features signifying en-plaque meningioma and intraoperative findings of strong dural attachment in our case suggestive of meningioma made the definitive diagnosis nearly impossible without histopathological examination. Studies have suggested that intraoperative diagnosis of tuberculoma with frozen section is sometimes difficult and it may be difficult to distinguish from other intracranial pathologies.[9,10] Bauer et al.[11] also have stressed on the necessity of biopsy and histopathology for the definitive diagnosis of tuberculoma. These findings suggest that diagnostic work-up for IDEM tuberculoma should be done in cases with features of meningioma with compressive myelopathy even in absence of constitutional symptoms of tubercular infection. IDEM tuberculoma must also be included as one of the differentials during the diagnostic work-up of en-plaque meningioma.

Treatment guidelines for IDEM tuberculoma is not well established.[7] Surgical removal must include decompression and excision as much as possible preventing the additional neurological morbidity followed by intensive treatment with antitubercular drugs as the biopsied of partially excised lesion may exhibit persistent lesion even after full course of ATT.[12] Intensive treatment with antitubercular drugs for an adequate duration (6–9 months) is sufficient for intramedullary tuberculoma but may need to be prolonged to 1 year or more depending on the radiological state of the lesion.[2,3,5,13] The compressive symptoms in IDEM tuberculoma warrants surgical excision in addition to intensive ATT to ensure better neurological outcome.

CONCLUSION

IDEM tuberculoma, though extremely rare at foramen magnum, may mimic en-plaque meningioma and should be considered as one of the differentials even in absence of primary tubercular disease in an immuno-competent patient. Radiological and clinical features may be inconclusive and required histopathology for the definitive diagnosis. Prompt surgical resection and decompression ensures better neurological outcome in addition to intensive and adequate ATT.

CONFLICT OF INTEREST

The authors have nothing to disclose.

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