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CASE REPORT

Intramedullary craniovertebral junction tuberculoma: An uncommon location of a common disease

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Summary Central nervous system involvement is observed in no more than 10% of patients with systemic tuberculosis. Although CNS tuberculosis is not rare in endemic countries, such as India, intramedullary tuberculosis is not commonly reported. In this study, we report a case of a 40-year-old female who presented with a six-year history of insidious onset, gradually progressive, asymmetric quadriparesis. She was diagnosed with intramedullary tuberculoma at the craniovertebral junction and showed significant clinico-radiological improvement with medical management alone. To the best of our knowledge, this report describes the first case of intramedullary tuberculoma at the craniovertebral junction to be reported. With the increased availability of MRI in developing countries, it is now possible for clinicians to diagnose this condition without performing a biopsy. It is important for the clinicians in developed countries to be highly suspicious of intramedullary tuberculoma, as there has been a resurgence of CNS tuberculosis due to the emergence of the HIV pandemic.
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Introduction

Central nervous system (CNS) involvement is observed in no more than 10% of patients with systemic tuberculosis [1]. Although CNS tuberculosis is not rare in endemic countries, such as India, intramedullary tuberculoma is not commonly reported [2,3]. It is important for clinicians to diagnose this treatable condition as early as possible so that irreversible complications can be prevented. Here, we report a case of a 40-year-old

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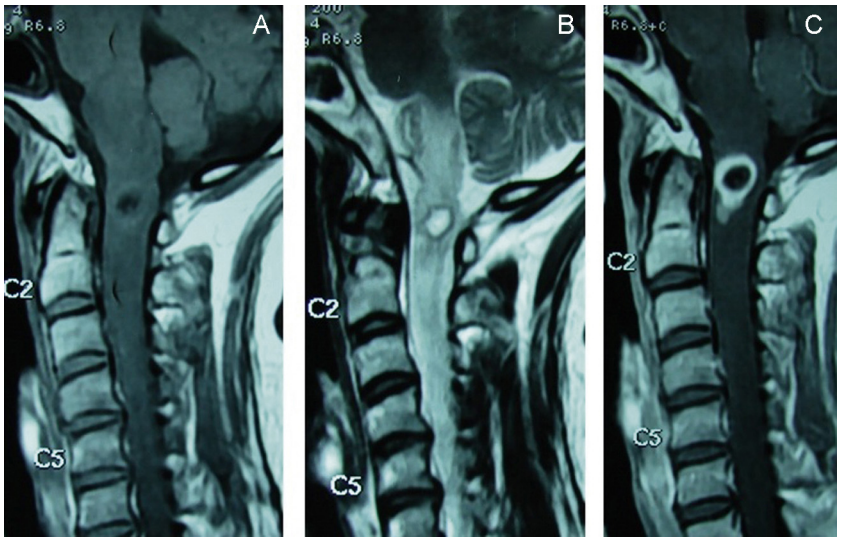

Figure 1 (A) MRI of cervical spine T1W sagittal image showing hypointense lesion with hyperintense rim at craniovertebral junction. (B) MRI of cervical spine T2W sagittal image showing hyperintense lesion with hypointense rim at craniovertebral junction. (C) MRI of cervical spine T1W+ contrast sagittal image showing thick ring enhancing lesion at craniovertebral junction.

female who presented with insidious onset, gradually progressive, asymmetric quadriparesis. She was diagnosed with intramedullary tuberculoma at the craniovertebral (CV) junction and showed significant clinico-radiological improvement with medical management alone. To the best of our knowledge, this report describes the first case of intramedullary tuberculoma at the craniovertebral junction to be reported.

Case report

A 40-year-old vegetarian female with no known chronic illnesses presented with insidious onset, gradually progressive (right more than left) quadriparesis of six years in duration followed by numbness in both upper and lower limbs for four years prior to her admission to our clinic. She had a history of significant weight loss over six months but no history of fever, cough, headache, vomiting or any swellings in the body. The patient's general physical examination was normal. Her cardiovascular, respiratory, and gastrointestinal system examinations were normal. On neurological examination, the patient was found to be conscious and well oriented with no cranial nerve deficit. A motor examination revealed spasticity in both upper and lower limbs (right more than left) with a medical research council (MRC) grade of 4−/5 in her right upper and lower limbs and a MRC grade of 4/5 in her left upper and lower limbs. She showed brisk reflexes in both upper and lower limbs and a bilateral extensor plantar response. Sensory examination revealed pan-sensory loss up to the C2 spinal level. Her neck movements were restricted and painful. Thus, a clinical diagnosis of compressive cervical myelopathy was made.

Contrast enhanced magnetic resonance imaging (MRI) of the patient's cervical spine revealed an intramedullary ring enhancing lesion at the C2 vertebral level with perilesional edema (Fig. 1A—C). A contrast enhanced MRI of her brain was also performed to rule out any intracranial lesions, and the results of the MRI were normal (Fig. 2). Blood investigations revealed a raised erythrocyte sedimentation rate (ESR) of 80 mm/h and normal liver and renal function tests. The patient's chest X-ray was normal. The enzyme-linked immunosorbant assay (ELISA) for human immunodeficiency virus

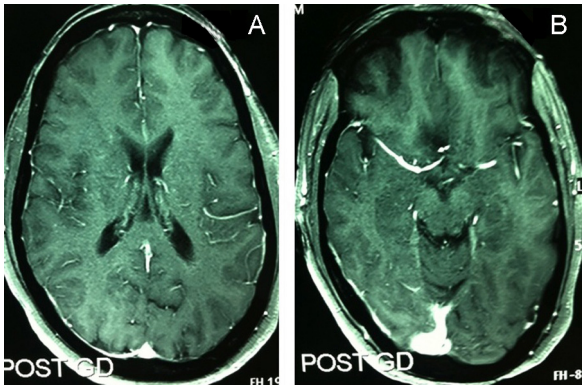

Figure 2 (A and B) T1+ contrast axial images of brain showing no intracranial tuberculoma.

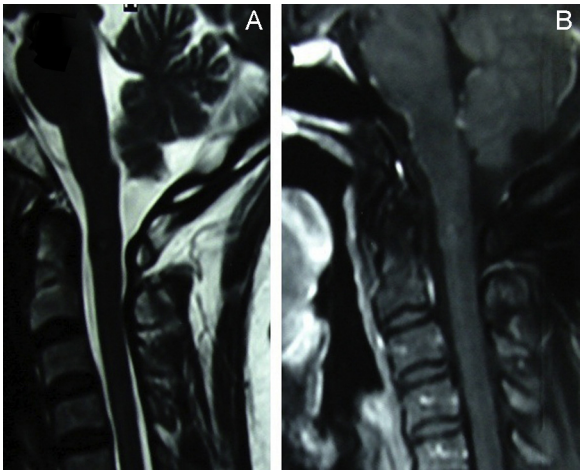

Figure 3 (A and B) MRI of cervical spine T2W and T1W+ contrast sagittal images (done four months after start of ATT) showing significant resolution of the lesion.

(HIV) was negative. CSF examination showed pleocytosis with 240 cells (80% lymphocytes), raised proteins (125 mg%), and low sugar (25 mg%), along with a positive polymerase chain reaction (PCR) for Mycobacterium tuberculosis (MTB). The India ink preparation of the patient's CSF to test for cryptococcal antigen was negative. Cysticercal antibody tests of the patient's serum and CSF by ELISA were negative. Thus, a final diagnosis of intramedullary tuberculoma of the cervical spine was made, and the patient was prescribed a five drug antitubercular therapy, including isoniazid (300 mg/day), rifampicin (450 mg/day), ethambutol (800 mg/day), pyrazinamide (1500 mg/day), and injectable streptomycin (0.75 g/day intramuscular), for three months followed by isoniazid and rifampicin for a total of 18 months. A short course of an injectable steroid followed by an oral steroid was also given to the patient in a tapering dose over the first two months of treatment. She started showing improvement in her motor symptoms after 20—25 days of antitubercular treatment (ATT) and steroids. At the patient's four month follow up visit, her power improved to a MRC grade of 4+/5 in both of her upper and lower limbs, and her sensory symptoms were also resolved. Her neck movements were painless with no restriction. After four months of treatment, her CSF examination was completely normal, and an MRI of her cervical spine showed a significant reduction in the size of the lesion and edema (Fig. 3A and B).

Discussion

Although tuberculosis (TB) can affect any part of the body, spinal cord involvement is rarely encountered compared to brain involvement [4,5]. Central nervous system tuberculosis has always been considered a great health hazard in endemic countries, such as India, with high rates of mortality and morbidity [6].

Pott's spine is the most common presentation of TB spine, whereas non-osseous spinal cord involvement usually occurs due to tuberculoma. Tuberculoma most commonly occurs at the dorsal spine and is usually extradural in location, with intramedullary tuberculoma comprising only 8% of all spinal tuberculomas [7]. In our patient, the tuberculoma was intramedullary at the CV junction, which, to the best of our knowledge, has never been previously reported.

The radiological presentation of tuberculoma depends on the stage of the disease [8]. In the early phase, there is a significant inflammatory reaction, and the tuberculoma is observed as a hypointense lesion on T1W and T2W images with homogenous contrast enhancement. In the later stage, as the capsule becomes richer in collagen, the tuberculoma appears isointense on T1W and T2W images with ring enhancement. With the development of caseation, the center of the tuberculoma appears bright, and the target sign is observable [9].

In the past, surgical treatment was considered the corner stone of the management of intramedullary tuberculoma, but recent case reports and a case series by Gupta et al. showed good results with medical management alone. Thus, in any case for which MRI is suggestive of intramedullary spinal tuberculoma, medical management should be considered as the first option, and surgical treatment should be reserved for limited indications, such as gross neurological deficits, poor response to medical treatment, deterioration in neurological status during medical treatment and paradoxical enlargement of the lesion observed on a follow-up MRI [10].

Because our patient presented with quadriparesis, a neurosurgical opinion was sought, but in view of the location of the lesion and the patient's slowly progressive and long-standing symptoms, medical management was chosen as the course of treatment. The surgical treatment was kept as a second line of management in case the patient had a poor response to medical management or had rapid deterioration, and the patient was advised close follow-up. Interestingly, the patient started showing improvement as early as 20—25 days after starting ATT and a tapering dose of steroids, and after four months of treatment, she had significant clinical improvement with normal CSF and a reduction in the size of her lesion and edema viewed by MRI.

CV junction space occupying lesions (SOLs) are different from SOLs of other sites in two respects. First, there can be a difference in the clinical presentation of CV SOLs compared to other SOLs, because the CV junction contains several important neurovascular structures, such as the medulla, vertebrobasilar system, lower cranial nerves (IX, X, XI and XII) and sympathetic trunk. Damage to these structures can lead to dysphagia, nasal regurgitation, hoarseness of the voice, Horner's syndrome, etc. which do not occur in patients with spinal SOLs at other sites. Secondly, CV junction tumors are surgically challenging. Their location combined with their close anatomical relation to critical vascular and neural structures makes radical tumor resection difficult [11,12].

Cysticercal granuloma is an important differential diagnosis of a ring enhancing lesion. In our case, it was ruled out based on the presence of significant edema on MRI, a positive PCR result for MTB in the CSF, and a negative cysticercal antibody test result in the blood and CSF. The patient's good clinico-radiological response to ATT further confirmed our diagnosis. Other possible differential diagnoses of intramedullary spinal SOLs include ependymoma, astrocytoma, and metastasis. Ependymoma and astrocytoma are the two most common intramedullary tumors. The cervical spine is the most common location of ependymoma, whereas it is the second most common location for astrocytoma after the dorsal spine. Clinically, it is not possible to differentiate between these two types of intramedullary SOLs, but they can be easily differentiated from tuberculoma on contrast enhanced MRI. On contrast enhanced T1 image, both ependymoma and astrocytoma show homogenous or heterogeneous enhancement, whereas tuberculoma shows ring enhancement. Intramedullary metastasis may present as a ring enhancing lesion on MRI, but, in this case, the patient's long history of symptoms of six years virtually ruled out the possibility of intramedullary metastasis [13]. The patient's good clinico-radiological response to ATT further confirmed our diagnosis.

Conclusions

Intramedullary tuberculoma has rarely been reported. With the increased availability of MRI, it is now possible for clinicians to diagnose intramedullary tuberculoma without performing a biopsy. Our case also highlights that conservative management should be the preferred treatment method for patients with intramedullary tuberculoma, and surgery should be reserved for cases with any neurological or radiological deterioration.

Conflict of interest

Funding: No funding sources.
Competing interests: None declared.
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