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Case Report

Intramedullary tuberculoma of thoracic spine: A rare location of an endemic disease

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ABSTRACT

Tuberculosis (TB) is endemic in India, alone contributing for two thirds of total cases worldwide. Spinal tuberculosis accounts for 1-2% of all cases of tuberculosis. However, spinal intramedullary tuberculoma occurs in as few as 2 out of 1000 cases of tuberculosis involving central nervous system. This may present in form of meningitis, tubercular spondylitis, arachnoiditis or tuberculomas mostly. The most common site of involvement is found to be the thoracic region of spine affecting relatively younger age groups and often seen in association with extraspinal tuberculosis disease. The source of intramedullary tuberculosis is mostly hematogenous spread from a site of primary TB elsewhere in the body which is usually the lungs. Due to rarity of the disease misdiagnosis is frequent. Magnetic Resonance Imaging is the investigation of choice and histopathological examination and culture are confirmatory methods for diagnosis of tuberculosis. Microsurgical resection is better option for rapidly progressing intramedullary tuberculosis cases. Here we report a rare case of isolated spinal intramedullary tuberculoma of thoracic spinal region.

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1. Introduction

The involvement of nervous system as a site of extrapulmonary tuberculosis is quite common. This may present in form of meningitis, tubercular spondylitis, arachnoiditis or tuberculomas mostly. The occurrence of intracranial tuberculoma is common in contrast to isolated spinal intramedullary tuberculoma which is found rarely. The ratio of intracranial to intraspinal tuberculosis is approximately 20:1 to 42:1.¹⁻⁴ The incidence rate of spinal tuberculosis is found to be only 2 out of 100,000 cases of tuberculosis and 2 out of 1000 cases of tuberculosis involving central nervous system.^{5,6} Among all the cases of spinal tuberculosis reported in literature, isolated spinal

intramedullary tuberculomas (ISIMT) comprise 8%.⁷ The most common site of involvement is found to be the thoracic (T) region of spine affecting relatively younger age groups and often seen in association with extraspinal tuberculosis disease.³ Here we report a rare case of isolated spinal intramedullary tuberculoma of thoracic spinal region.

2. Case Report

A 27 year old male patient presented to our hospital, with low back pain, parasthesia and weakness in lower limbs with bladder and bowel incontinence for last 3 months. His presentation was progressive in nature. General examinations were within normal limits. On auscultation, chest was bilaterally clear with symmetrical vesicular sounds. Cardiovascular and per abdomen examination were also within normal limits. Examination of the spine,

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revealed gibbus at lower thoracic region. On neurological examination sensory loss to all modalities was present below T12 dermatomal level. Deep tendon reflexes and superficial reflexes were absent in both lower limbs.

Blood reports were normal. Chest X-ray was clear and. The patient was seronegative for human immunodeficiency virus (HIV). Magnetic Resonance Imaging (MRI) of thoracic region revealed a single intramedullary “target sign” lesion at T10 level which was hypointense on T1- weighted images and exhibited hyperintensity on T2-weighted images. On contrast study, the lesion showed signal enhancement at T10 vertebral level on T1-weighted imaging. (Figure 1). Additionally, a brain MRI was performed to rule out any associated intracranial tubercular lesion which was clear.

Based on the clinico-radiological findings, a diagnosis of isolated intramedullary tuberculoma at T-10 level was made and the patient was put on antitubercular therapy (ATT) with dexamethasone.



Fig. 1: MRI Thoracic spine is showing intramedullary ring enhancing lesion

The patient was followed up for improvement but after taking the treatment for one month, his condition was not improving and he persistently complained of increase in severity of pain and weakness. To manage the worsening clinical condition of the patient, microsurgical resection of the lesion was planned. The patient was operated in prone position and microsurgical lesional resection was performed and excision of intramedullary tuberculoma was performed (Figure 2). The operative procedure was uneventful. The excised specimen was sent for culture and histopathological examination. Tissue culture was also positive for mycobacterium tuberculosis. Histopathological examination revealed multiple necrotizing granulomas composed of epithelioid histiocytes exhibiting kidney shaped vesicular nuclei with pink nucleoli and abundant foamy cytoplasm. Granulomas also showed multiple Langhans giant cells and were surrounded by collar of lymphocytes and fibroblasts (Figure 3). The most likely diagnosis of tuberculosis was confirmed by PCR.



Fig. 2: Intramedullary tuberculoma at thoracic region

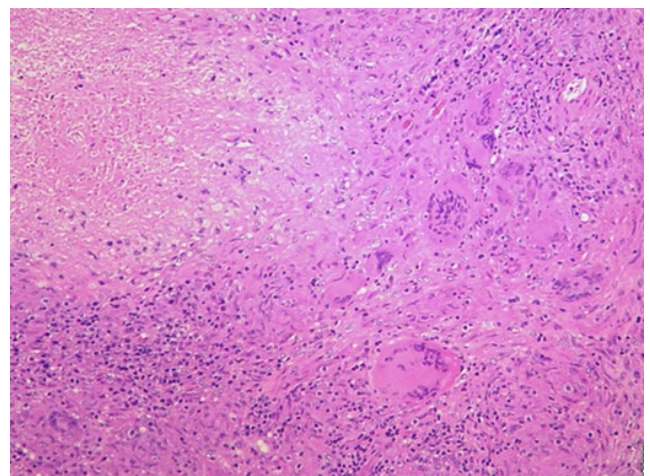


Fig. 3: Hematoxylin and eosin stained section showing necrotizing granuloma bordered by epithelioid cells and multiple langhan's giant cells

The patient was discharged after 18 days postoperatively. On a follow-up visit after 8 weeks, patient showed significant relief in the neurological symptoms and there was no improvement in the motor power of both lower limbs. Physiotherapy and ATT was continued postoperatively.

3. Discussion

Tuberculosis is caused by the aerobic bacteria *Mycobacterium tuberculosis*, usually involves high oxygen tension areas like the apex of lungs as the favored site of infection. While lung is the most common primary site, spine is the most common site for extrapulmonary involvement.

The occurrence of intracranial tuberculoma is common in contrast to isolated spinal intramedullary tuberculoma which is found rarely. The ratio of intracranial to intraspinal tuberculosis is approximately 20:1 to 42:1.¹⁻⁴ The incidence rate of spinal tuberculosis is found to be only 2 out of 100,000 cases of tuberculosis and 2 out of 1000 cases of tuberculosis involving central nervous system.^{5,6} Among all the cases of spinal tuberculosis reported in literature, isolated spinal intramedullary tuberculomas (ISIMT) comprise 8%.⁷

The source of ISIMT is mostly hematogenous spread from a site of primary TB elsewhere in the body which is usually the lungs.^{2,8} Incidence of isolated ISIMT is extremely rare even in a countries with endemic TB and contributes only 8% of all cases of spinal tuberculosis worldwide.⁷ Important risk factors in a case of suspected ISIMT are alcoholism, poor sanitation, low socioeconomic strata, history of exposure to a TB positive patient, HIV infection and immunosuppressant therapy. All these risk factors were absent in the present case. Although the clinical course and presentation of ISIMT is widely variable and specific to the vertebral level of spine affected, some common complaints that the patient presents with include fever, loss of appetite, lethargy, symptoms of sensory and motor impairment, back pain and impotence.⁸⁻¹⁰

The differential diagnosis include astrocytic glioma, ependymocytoma, hemangioblastoma, spinal cord metastasis and lymphomatous spread to the spinal cord. MRI is the imaging modality of choice in ISIMT. The MRI features of ISIMT can vary depending on the stage of tuberculoma formation (non caseating, caseating with a solid center, and caseating with a liquid center). In its earliest stage (non caseating), tuberculoma is characterized by severe inflammatory reaction with poor formation of the collagenous capsule. T1-weighted images show isosignal or slightly hyposignal intensity, and T2-weighted images show hypersignal intensity. The granuloma exhibits homogeneous enhancement after contrast administration on T1-weighted images. Later during the caseating stage, collagen becomes richer in the surrounding capsule, and

rim enhancement becomes visible on an MRI after contrast administration. A solid caseating tuberculoma appears iso- to hypointense on T1- weighted images and hypointense center with an iso- to hyperintense rim on T2-weighted images. When the solid center of the caseating tuberculoma liquefies, T1-weighted images show hyposignal intensity and T2-weighted images show hypersignal intensity at the center with a hypointense rim. However, these findings are not specific to tuberculoma.

Spinal deformities, like, gibbus and kyphosis are common in ISIMT depending on the level and extent of lesion. In intramedullary metastasis, posterior elements of the vertebral bodies are predominantly involved. Intervertebral disc and disc space are spared till late. Paraspinal soft tissue pus collection is uncommon. Lymphomatous metastasis commonly involves the cauda equina. In addition, multiple vertebral involvements are common. Fluorine-18-fluorodeoxyglucose positron emission tomography combined with computed tomography (FDG PET-CT) can help in differentiating spinal tuberculosis from spinal metastasis. FDG PET/CT demonstrates multiple FDG-avid lymph nodes and bone lesions in spinal cord metastasis. In spinal tuberculosis, para-discal FDG avid lesions and cold abscess are common

Major issue of concern regarding ISIMT is that there is no consensus with regard to the medical and surgical treatment in these cases. In practice, most researchers recommend antituberculous therapy, with or without corticosteroid, as the initial treatment when ISIMT is suspected. There are cases of ISIMT where microsurgical resection is beneficial as in our case. Microsurgical intervention should be considered in patients with rapidly deteriorating profound neurological deficits in spite of ATT.^{9,10}

4. Conclusion

Intramedullary tuberculoma is an incredibly rare diagnosis which poses a unique diagnostic dilemma to the clinician, as there lacks both a gold standard for diagnostic methodology and specific treatment guidelines for disease treatment and/or eradication. Although rare, this diagnosis should be included on the differential for any patient who presents with progressively worsening symptoms. Further studies analyzing what treatment regimens are most effective for ISIMT would be beneficial for improving patient outcomes. Early surgery has an important role in the treatment.

5. Source of Funding

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6. Conflict of Interest

The author declares that there is no conflict of interest.

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