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21 cm long intra-medullary tuberculoma of dorsal spinal cord. A rare lesion and its management

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ABSTRACT

Intramedullary tuberculoma (IMT) is a rare form of spinal tuberculosis. The incidence of this disease is 01-02/100,000 patients (1). Occurs by hematogenous dissemination from focus elsewhere in the body. Magnetic resonance imaging (MRI) is helpful for diagnosing IMT at an early stage and it is also very useful in follow-up. CSF PCR studies are confirmatory. Histopathology is the Gold Standard. AKT drug therapy is the mainstay of treatment for IMT, with surgery indicated in select patients. Here, we describe a female patient with long-segment dorsal intramedullary tuberculoma with compressive myelopathy treated with surgery and put on AKT who improved clinically.

Keywords
intramedullary,
tuberculoma,
21cm

INTRODUCTION

Tuberculosis can theoretically affect any tissue of the human body, but in practicality, there are different predilections and incidences for different tissues. The spectrum of Spinal TB includes – Tubercular Spondylodiscitis – potts disease (most common), TB myelitis (next common) and intra-spinal TB (rare)(2). Intraspinal TB further can be TB arachnoiditis, meningitis and Intra-Medullary Tuberculoma (IMT)(2). IMT is extremely rare entity with incidence. Spinal IMTs are extremely rare, seen in only 1- 2 of 100,000 cases of TB (1) and 2 of 1000 cases of CNS TB (2). The first report of IMT was given by Albercrombie in 1828. (3) In 1960, Lin et al. reviewed literature of IMT and accounted for 104 cases, of which majority were diagnosed post-mortem (3). The first magnetic resonance imaging (MRI) documented description of tuberculoma was given by Rhoton et al. in 1988(4). Clinical presentation is of a compressive myelopathy but with MRI showing Intramedullary lesion, can be misdiagnosed for Intra-Medullary tumour. Presentation of Intra-Medullary cord syndrome is unusual compared to common spinal TB presentations. Here we report such a case with such unusual presentation, where sound knowledge of MRI interpretation, corelation with past history of Koch elsewhere in body, helped to diagnose TB and we discuss the treatments given and clinical outcome.

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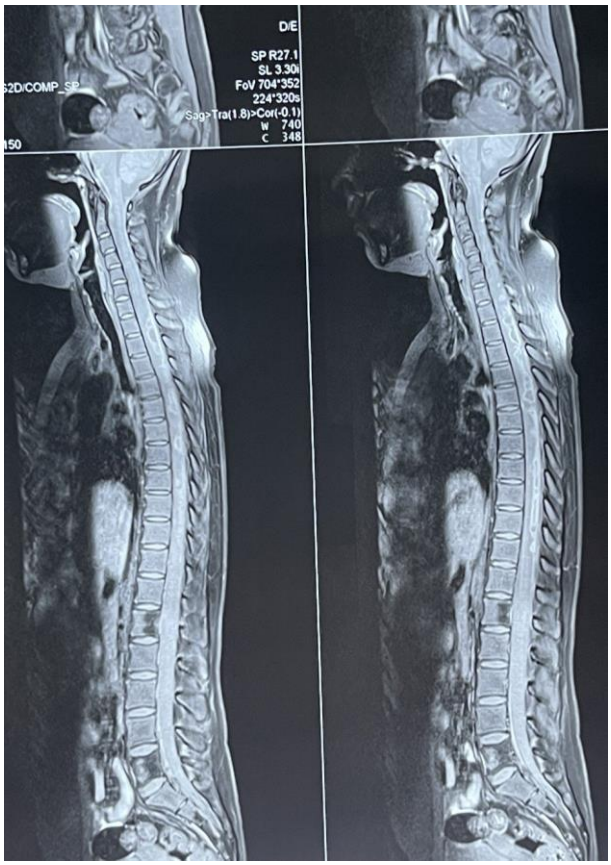
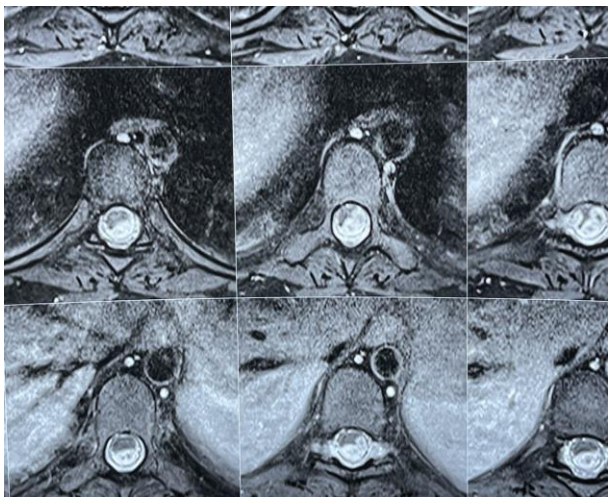
CASE REPORT

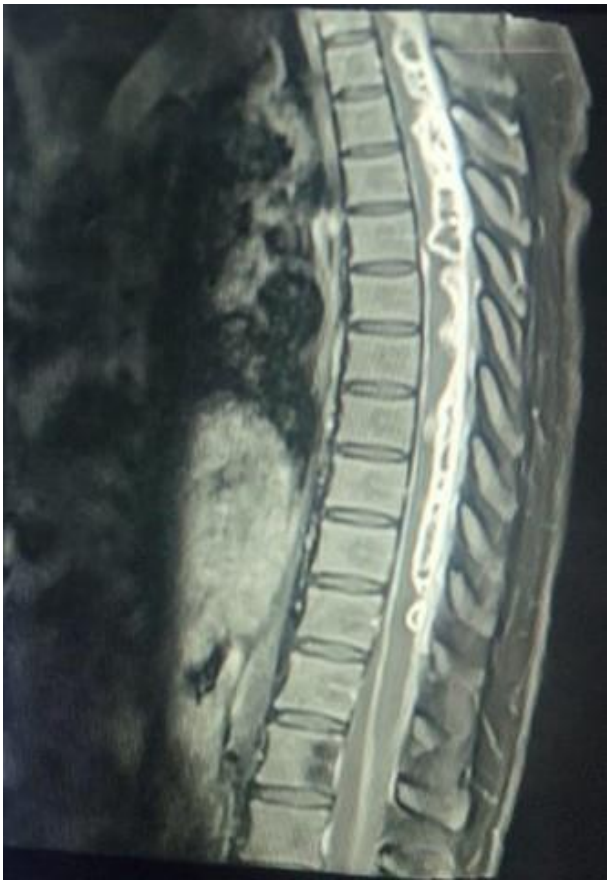
A 17-year-old female, a student, presented with complaints of back pain which was sudden insidious onset ,was described as persistent dull ache and was radiation to both lower limbs and which was gradually increasing intensity over 1 month until presentation ; this was a/w slow progressive weakness over both lower limbs and pt was rendered bed ridden at presentation ; a/w bladder and bowel incontinence for which she was foley catheterised. She gives a past history of CNS TB with communicating Hydrocephalus for which she was right mpvp shunted at private hospital 06 years back and she had completed AKT - during which she developed Ethambutol induced ocular toxicity. She had no other comorbidities, no other operative history. On examination, her higher mental functions and cranial nerves were intact. Her muscles on lower limbs started to show atrophy.

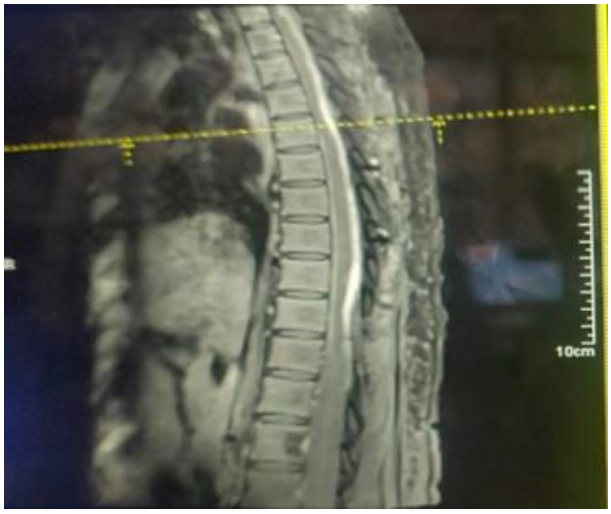
Tone was increased on both lower limbs. Muscle power was grade 05 in all muscle groups of both upper limbs while it was grade 02 over all muscles of the lower limbs. Reflexes were normal over all 4 limbs. Plantars were mute. She had sensory impairment below D6 corresponding to vertebrae level and altered sensation with hypoesthesia. Clinical provisional diagnosis was made of myelopathy and could be due to TB spondylodiscitis (i/v/o past history) and patient was investigated further. Routine blood investigations were done and analysed. Erythrocyte Sedimentation Rate was done and was found to be elevated. 'Triple H' was found to be normal and Plain chest radiograph was also found to be normal. Contrast enhanced MRI was done – this showed a long segment patchy intradural, intramedullary lesion with T1 iso-intense and T2 hypo-intense extending from c7 to d10 level with maximum thickness at d6 level. It was peripheral rim enhancing on contrast. CSF showed elevated protein, increased cellularity with 100% lymphocytic predominance. There were also small tuberculomas in the basal brain but with no mass effect/ hydrocephalus. These findings lead us to shift diagnosis to that of Intra-Medullary Tuberculoma and hence consulted pulmonologists for starting AKT(i/v/o past history of AKT and ethambutol toxicity) and decision was taken to put her on Bedaquilline based AKT. Simultaneously, we also opted for surgery for spine as she was having frank myelopathy with neurodeficits, aim was to achieve excision of the lesion and decompress where necessary. Intraoperatively we found a intr-dural extrapial long segment tuberculoma from c7 to d10 levels – this was excised totally and cord was free hence laminae placed back. Histopathology was suggestive of chronic granulomatous inflammation suggestive tuberculosis and

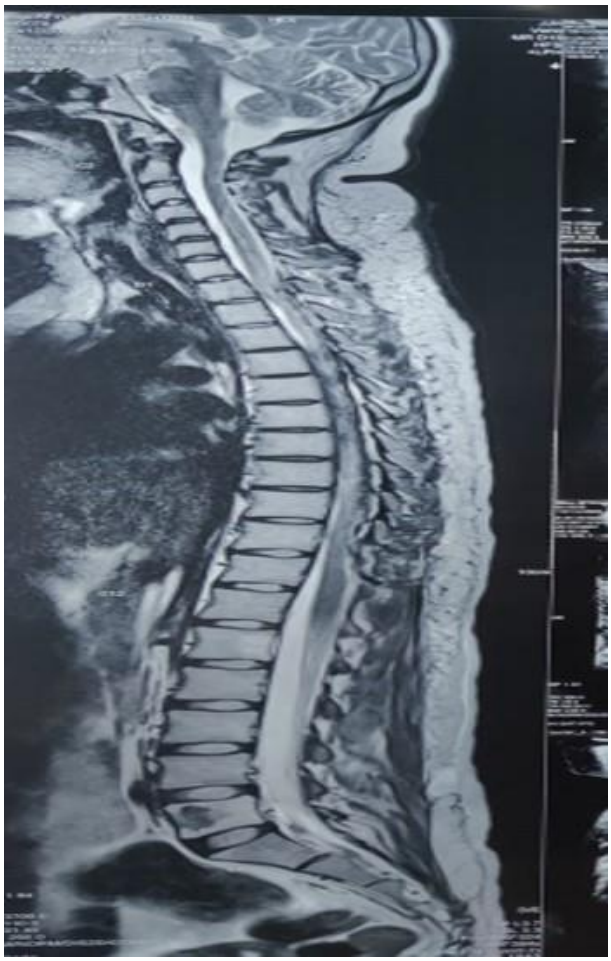




Figures 1, 2, 3. Pre op MRI Cervico dorsal spine with contrast showing intradural intramedullary contrast enhancing lesion.







Figures 4, 5. Post operative MRI with contrast s/o complete excision of lesion.

GeneXpert on the tissue sample was positive for M.Tb and with Rif resistance detected. This confirmed our diagnosis of Spinal Intra-medullary tuberculoma. On POD 02, MRI showed significant radiological resolution and on POD 10, patient was discharged with Power on both lower limbs grade 03- a clinical improvement. Patient kept on follow up with plan to repeat MRI at 03 months.

Discussion

CNS TB accounts for approximately 0.5-2% of all cases of TB, carries a high mortality and neurological morbidity,(1) . Spinal TB is very rare and the common forms of spinal TB are tuberculous spondylitis, TB myelitis, and intraspinal TB. Intraspinal TB could be spinal meningitis, arachnoiditis, IMT, and abscess. IMT is very uncommon and till now roughly 150 cases have been reported(6). The thoracic segment is the most common site of IMTB and hematogenous spread is usually the main etiology followed by CSF seeding – this is a/w longer segmental involvement (7). Most cases of intramedullary tuberculomas are subacute and present with progressive symptoms suggestive of a compressive myelopathy.

Our patient had the picture of such subacute cord compression with past history of TB in brain hence corroborating with CSF seeding theory and also consistent with this is finding of a long segment

involvement. Immunoosuppresive states in general are known to favour disseminated Kochs hence was also done and found negative in our patient. Raised ESR is again favourable for diagnosis of Koch. Advent of MRI has made diagnosis of IMT more accurate and earlier. In the early phase, the tuberculoma is characterized by severe inflammatory reaction which causes severe edema. At this stage, the gel capsule is not well formed. During this stage, the enhancement after contrast examination is uniform. T1WI and T2WI both show equal signal intensity(early phase). As the gel content in the tuberculoma increases, the peripheral edema begins to disappear(intermediate phase). As a result, T1WI shows isointense lesions while T2WI shows low or isointense lesions. Contrast MRI shows central hypointensity with rim enhancement. With the development of caseation(late phase), T2WI shows a typical "target sign," which means that it exhibits a range from the low signal target to the high signal rim and also from the center of the low signal rim to the peripheral parts. The caseous substance appears hyperintense at the center, which gives the characteristic target sign. The low signal rim in the external region is composed of collagen fibers produced by fibroblasts.

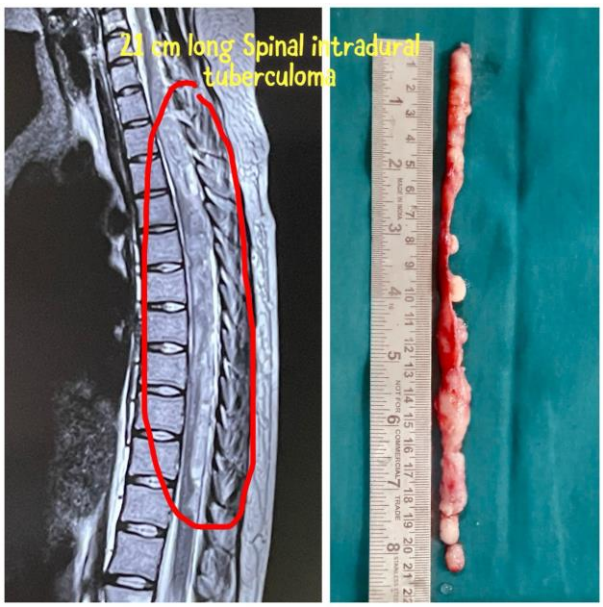


Figure 6. 21 cm long specimen of tuberculoma.

The target sign is a valuable indicator that helps differentiate spinal tuberculoma from other intramedullary lesions. Rim enhancement and presence of sharp margins also differentiates IMT from intramedullary tumours.(8)(9)(10)(11) . In our patient, there was t2 hypointensity with peripheral rim enhancement and central hypointensity on contrast. This corresponds to the intermediate phase. Further CSF study was done which showed elevated protein and increased cellular count with 100% lymphocytes. This is a well-accepted classical picture for CNS Koch. Hence at this juncture, we decided to start AKT immediately. Now this girl had already received AKT and already got Ethambutol toxicity. So we consulted pulmonologists and ophthalmologists and started her on BDQ based AKT with ophthalmological safe drugs. Indications for surgery in IMT.

1. Gross neurological deficits
2. Worsening of neurological status during Rx
3. Paradoxical enlargement of lesion during Rx

Our patient had neurodeficits and myeopathy which had to be hence operated. Surgical resection was performed through posterior approach from c7 to d10 Level. Midline durotomy was performed and showed a thick mass of 11 cm extrapial tuberculoma. The patient underwent gross total excision of the mass and there was no need as such of bony decompression. The tissue was sent for HPE which revealed a granulomatous lesion with a central area of caseation in keeping with a tuberculoma. Histopathological diagnosis is also confirmatory for TB. We also further confirmed with GeneXpert. Reason for GeneXpert was twofold, we wanted to find out why in a immunocompetent patient there is reccurence and dissemination of TB apart from confirming diagnosis.

GeneXpert showed us Mtb and confirmed resistance to Rif – hence making our calculated assumption correct. This child had initially itself CNS TB in the brain with communicating hydrocephalus which was incompletely evaluated at the time (could be due to treatment done at periphery and was odne in 2016 when GeneXpert may not have been in vogue) and was shunted and received line one AKT but which was already a resistant strain of Mtb. This then over period of time formed small tuberculomas in basal brain and disseminated to long segment of cord by CSF and gave rise IMT. Hence aptly managed with BDQ AKT and optimum surgery.

At POD 2 ther was significant radiological resolution. Clinical improvement was evident at Day 10 with marginal increase in power in both lower

limbs. Patient to be kept in follow up and MRI and clinical assessment repeated at 3 months. This case is being reported for its sheer rarity and to highlight diagnostic features and importance of surgery along with medical treatment to achieve good clinical results.

Conclusion

Rare entity of IMT is to be kept in mind in endemic nations like India when patient has intramedullary cord symptoms in picture of history of Kochs disease. Although medical management is to be instituted at earliest, this case report emphasised role of surgery in this disease. Look for drug resistance / immunosuppressive states in such disease and treat those properly which in turns will only help eradicating this entity. This case could provide some evidence-based data, thus contributing to the future research studies and clinical practice.

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