



Case Report

Spinal intramedullary tuberculoma in a 3-year-old girl

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ABSTRACT

Background: Spinal intramedullary tuberculoma (IMT) is a rare manifestation of extrapulmonary tuberculosis (TB). Presentation of TB in the pediatric age group is a significant contributor to mortality.

Case Description: A young vaccinated girl presented to the neurosurgery department with difficulty walking and urinary incontinence. A magnetic resonance imaging performed outside the hospital showed a hyperintense intramedullary lesion extending from T6 to T9. The patient underwent T6–T9 laminoplasty with intramedullary lesion decompression under neuromonitoring. The dense adherence of the lesion to the cord and nerve roots permitted only debulking. Histopathological examination confirmed the diagnosis of tuberculoma. The patient was started on antitubercular treatment and was then subsequently discharged. After 8 months, the patient was reviewed and showed improvement in her symptoms and complete resolution of the lesion on imaging. The patient has now developed hydrocephalus on the latest computed tomography imaging, which may be due to tubercular meningitis or arachnoiditis.

Conclusion: Complete resolution of spinal IMT is possible with a combined treatment approach.

Keywords: Antitubercular treatment, Pediatric spine, Spinal intramedullary lesion, Tuberculoma

BACKGROUND

Tuberculosis (TB) is one of the top 10 causes of under-5 mortality worldwide.^[2] Further alarmingly described by Ramos *et al.*,^[15] from 1998 to 2015, 1282 pediatric cases of TB were diagnosed, with 45.5% being under 5 years of age. In India, a subcontinent where TB is endemic, pulmonary, and extrapulmonary manifestations of the infection is commonplace. A rare subtype of extrapulmonary TB includes spinal intramedullary tuberculoma (IMT). Different schools of thought exist when it comes to management. Of the recorded cases, here is one case where a combined approach was taken to treat the patient.

CASE DESCRIPTION

A 3-year-old girl was brought by her parents to the neurosurgery department with complaints of difficulty walking for 4 months. The patient had a backward bending posture while walking. She swayed to one side and dragged her right lower limb. She also had difficulty wearing her slippers. She was not able to bend forward or sit for a long duration of time. Ten months ago, she had a history of headaches that lasted for a week. The patient also had urinary incontinence for

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4 months. The patient had one episode of seizures post which she developed lower limb weakness. There was no history of trauma to the spine and no history of coming in contact with a patient who had TB. The patient was vaccinated according to the World Health Organization schedule. The patient had a history of pneumonia at the age of 1 month, following which she was admitted to the intensive care unit for 1 week.

On examination, vitals were stable. Neurological examination showed that the tone of both lower limbs was increased. Plantar reflexes were extensor bilaterally. A magnetic resonance imaging (MRI) from an outside clinic showed a hyperintense intramedullary lesion, on T2W imaging, from T6 to T9 [Figure 1]. On sagittal T1W imaging, with contrast, there was a fusiform dilation of the cord at the same levels with no enhancement. A similar finding was present on the axial cuts [Figure 2]. The scan also showed hydrocephalus. However, on examination, there were no signs of raised intracranial pressure (ICP). The child underwent a T6 to T9 laminoplasty with intramedullary lesion decompression under neuromonitoring. Intraoperatively, the lesion was yellowish, fibrous, and densely adherent to underlying cord and nerve roots. Hence, only debulking of the lesion was performed. Postoperatively, mild deterioration of motor power in both lower limbs was noted. Histopathological examination of the lesion showed features suggestive of tuberculous granulomatous inflammation [Figure 3].

The diagnosis was then established. A pulmonology consultation for initiation of antitubercular treatment (ATT) was taken. The child was started on ATT and was subsequently discharged after being symptomatically better. The patient was reviewed 8 months after her initial visit while still on ATT, and she showed improvement in her gait abnormalities and is walking with support. The review scans [Figure 4] showed complete resolution of the lower lesion but now showed evidence of chronic arachnoid adhesions at the T3 level. A CT brain performed at the same time showed evidence of hydrocephalus, most likely noncommunicating

in nature [Figure 5]. This may be due to tubercular meningitis or arachnoiditis. The patient was advised to continue her ATT and was discharged.

DISCUSSION

Spinal TB is a common site of extrapulmonary TB, accounting for approximately 50% of skeletal TB.^[4] Spinal involvement accounts for 1–2% of all cases of TB.^[3] Although spinal TB is a frequent finding, what is infrequent is the occurrence of its manifestation as an IMT as it only forms approximately 8% of all cases of spinal TB.^[9] Further adding to its rarity is its presentation in the under-5 population. A thorough literature search showed that our report is one of the few documented cases below 5 years.^[1,8,10,13,17] Including our case, only four patients have undergone an operation. Symptoms vary based on the level of spinal involvement. With cervical spine TB, symptoms suggestive of the spinal cord or root compression will be present, whereas, with thoracic or lumbar involvement, lower extremity weakness leading to possible paraplegia may be the presentation.^[4] Age and immunocompetency seem to play a role in the pathogenesis as spinal TB is more common in the younger age groups.^[4] As



Figure 1: (a) T2W sagittal image showing a hyperintense intramedullary lesion from T6 to T9. (b) T2W axial image showing the same hyperintense lesion.

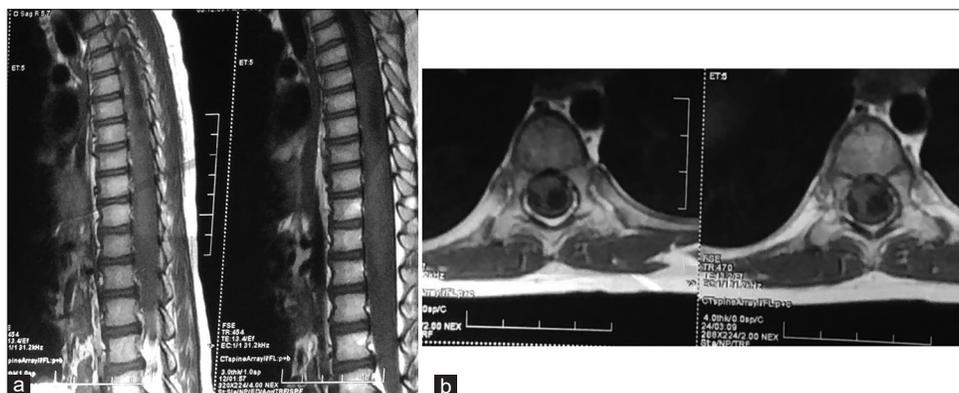


Figure 2: (a) T1W sagittal images with contrast showing fusiform dilation of the spinal cord from T6 to T9 with no enhancement. (b) T1W axial image showing the same finding.

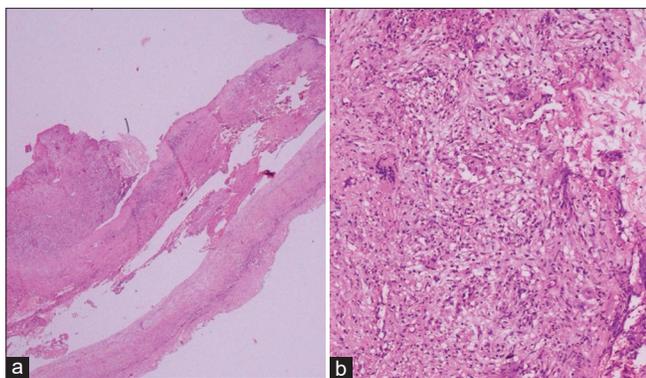


Figure 3: (a) Low-power view ($\times 20$) of thickened and hyalinized connective tissue (meninges) strips with infiltration by large nodular foci of dense inflammation. The slide was stained with hematoxylin and eosin. (b) A closer view ($\times 200$) revealed coalescing loose epithelioid cell granulomas and scattered Langhan's multinucleated giant cells on a background of lymphocytes and plasma cells. The slide was stained with hematoxylin and eosin.

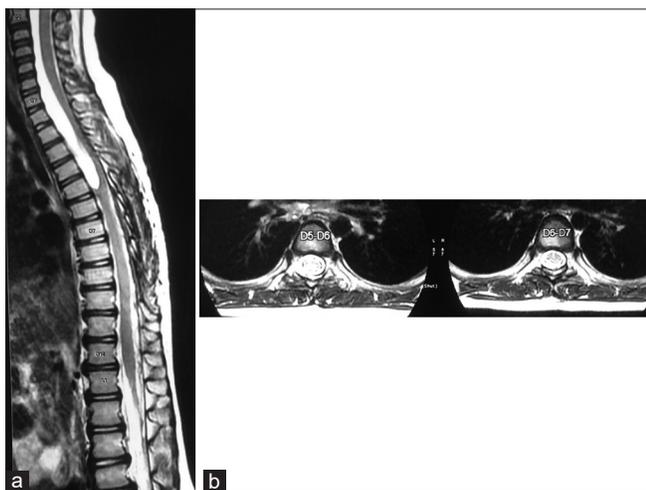


Figure 4: (a) T2W sagittal image taken at the 8-month review with the patient on antitubercular treatment. There is a complete resolution of the lesion. There are chronic arachnoid adhesions present at the T3 level. (b) T2W axial image showing complete resolution of the lesion.

indicated in the present case and previous pediatric reports, the influence of the Bacille Calmette–Guerin vaccine in the prevention of spinal IMT is questionable.^[5,17]

MRI is the imaging modality of choice for spinal IMT.^[11] Radiological findings of the spine may vary depending on the phase of illness, translating to differing levels of intensities seen on T1W and T2W sequencing. However, a characteristic finding is the “target sign” seen on T2W sequencing on the MRI.^[11] Identification of this sign will help rule out other differentials. Our patient had MRI spine findings showing a hyperintense lesion on T2W sequencing from T6 to T9 and a nonenhancing lesion on T1W sequencing in the same levels.

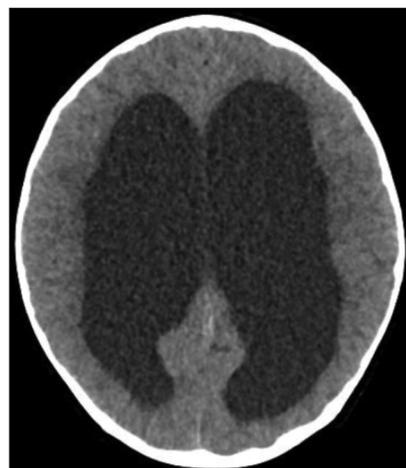


Figure 5: Computed tomography brain taken at the 8-month review showing features suggestive of noncommunicating hydrocephalus. There is dilation of the fourth ventricle, aqueduct, third ventricle, and bilateral lateral ventricles with transependymal cerebrospinal fluid seepage. There is no dilation of the foramina of Luschka or Magendie. We believe that this was caused as a result of tubercular meningitis or arachnoiditis.

MRI brain also showed hydrocephalus. Of the previous case reports, there have been variations in presentation, imaging findings, management, and outcomes of the chosen intervention for the patients [Table 1]. We initially considered the diagnosis to be intramedullary astrocytoma based on the MRI findings as it is a common entity in this age group. Consideration was also given to ependymoma. The intraoperative findings and the histopathological examination made tuberculoma a more likely diagnosis.

Kulkarni *et al.*^[10] suggested that surgical interventions be performed if the patient is unresponsive to chemotherapy, the diagnosis is uncertain, or if there is rapid neurological deterioration. As our patient had signs of compressive myelopathy without raised ICP, decompression of the lesion was planned, and a biopsy was simultaneously taken. We explained to the family that in case of any raised ICP in the postoperative period, measures might have to be taken to deal with the same appropriately. We never considered management with only ATT for this patient as we initially thought the diagnosis to be a tumor instead of a tuberculoma. Therefore, we did not choose to manage this patient with only ATT and saw the need for operative intervention. Medical management is more preferred than surgical management to avoid complications such as postoperative meningitis.^[12] The existing literature suggests operated patients of spinal IMT need to be put on ATT (with or without steroids) postoperatively. Most of the previous reports have started ATT empirically or based on clinical evidence of TB. Our case was unique as there were no clinical signs and symptoms of TB in any other part of the body except the spinal cord.

Table 1: Compilation of cases under the age of 5 years.

Study	Age/sex	Symptoms	CNS imaging	Treatment	Outcome
Present report	3-year-old female	Difficulty walking, headache, urinary incontinence, seizures	MRI spine – Hyperintense intramedullary lesion from T6 to T9 noted on T2W imaging and a fusiform dilation of the cord, at the same levels, with no enhancement noted on T1W imaging with contrast MRI brain – Hydrocephalus	Surgical- 1. T6-T9 laminoplasty with intramedullary lesion decompression under neuromonitoring 2. ATT	Postoperatively mild deterioration of motor power in both lower limbs was noted. At the 8-month review, the patient had improvement in her gait abnormalities and complete radiologic resolution of the lesion. MRI imaging performed at the time of review showed chronic arachnoid adhesions at the T3 level. A CT brain performed at the same time also showed hydrocephalus.
Chagla et al. ^[11]	9-month-old female	Spastic tetraparesis	MRI brain – Hypointense well-circumscribed lesion extending from C2 to C7	Surgical- 1. C2-C7 laminoplasty with subtotal excision and lax duroplasty 2. ATT	Improvement in muscular strength was noted in the immediate postoperative period. At the 3-year follow-up, imaging showed complete resolution of the lesion.
Pike et al. ^[13]	23-month-old male	Seizure, neck mass	CT spine – Tuberculous spondylitis involving the T10-T11 vertebrae with a large paravertebral mass measuring 5 cm in diameter MRI spine – Contrast T1W images showed a 1 cm rim-enhancing lesion with central nonenhancing necrosis occupying the right side of the cord at C5–C7	Surgical- 1. Anterior spinal debridement and vascularized rib placement with posterior spinal instrumentation and fusion 2. ATT	At 3 months postoperatively, the patient had an adequate spinal alignment with a normal neurologic examination. The spinal alignment was normal at 6 months postoperatively.
Sujatha et al. ^[17]	1-year-old male	Fever, vomiting, lethargy, right lower limb weakness	MRI brain – Multiple ring-enhancing lesions over the left parietal lobe, left occipital lobe, and left cerebellar hemisphere MRI spine – Two ring-enhancing lesions at the level of T1 and T12	ATT	The patient improved on follow-up. One year later, a repeat MRI scan showed complete resolution of the tuberculomas.
Kulkarni et al. ^[10]	4-year-old male	Fever, headache, vomiting	MRI brain – Multiple ring-enhancing lesions in the cerebrum, cerebellum, brainstem, and intramedullary region of cervical cord at the C3/C4 level	ATT	The patient had a reasonable improvement.

(Contd...)

Table 1: (Continued)

Study	Age/sex	Symptoms	CNS imaging	Treatment	Outcome
Khalid <i>et al.</i> ^[8]	4-year-old male	Fever, acute flaccid paralysis	MRI spine – Holocord expansion with altered signal intensity with peripheral enhancement from T6 to conus medullaris MRI brain – Significant communicating hydrocephalus	Surgical- 1. Partial laminectomy 2. ATT	Uneventful postoperative recovery period. Improvement of the symptoms was noted.

MRI: Magnetic resonance imaging, ATT: Antitubercular treatment, CT: Computed tomography

In addition, our diagnosis was confirmed by biopsy and subsequent histopathological examination.

We are aware that other granulomatous conditions share some similar microscopic findings as TB, such as sarcoidosis.^[6,16] An article from 2001 estimated the prevalence of pediatric sarcoidosis in India to be 2%.^[7] The prevalence of pediatric TB in India is difficult to quantify due to inconsistent reporting and coordination between the private and public sectors. The Revised National TB Control Programme (RNTCP) provisions manage the cases under the public sector. A district in India estimated the reporting inequalities to be as much as 17% in the private sector and 8% under the RNTCP.^[18] We did not deem subsequent culture with TB-specific media to be necessary since TB is endemic to India and was most likely the diagnosis in this case since there was a microscopic pattern of granulomatous inflammation.

Four of the previously mentioned cases of IMT have been recorded in India.^[1,8,10,17] Of these, only Khalid *et al.*^[8] identified acid-fast bacilli (AFB) using culture studies and then initiated treatment. Although Chagla *et al.*^[1] performed histopathological analysis, they have not mentioned anything regarding confirmation of TB by culture. Their patient was initiated on ATT. Kulkarni *et al.*^[10] performed magnetic resonance spectroscopy which provided biochemical clues suggesting TB without performing culture. The case by Sujatha *et al.*^[17] had cerebrospinal fluid (CSF) analysis findings suggestive of tubercular meningitis. However, they could not isolate AFB from the CSF and the other TB tests they performed were also negative. The latter two groups also started their patients on ATT without confirmation by culture. We believe that medical practitioners in India are justified in empirically starting ATT based on the endemic nature of the disease, clinical suspicion, and preliminary identification of granulomatous inflammation. Patient-reported outcomes should guide the decision to continue ATT usage. In this case, we made the correct decision as the patient's review MRI showed no remnants of the lesion at the initial site.

Continuation of the ATT will be required to address the arachnoid adhesions. Further follow-up will be required to ensure the resolution of this new finding and prevent possible post-tubercular kyphosis.^[14]

CONCLUSION

IMTs are a rare spinal manifestation of TB, especially in the under-5 category. Tuberculomas are more likely to occur in the brain rather than the spine. The treatment depends on the presentation of the patient, radiologic findings, and the surgeon's decision. We present this case to further the accounts made of this infrequent diagnosis and show that complete resolution of IMT is possible with a combined treatment approach.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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