NON-OSSEOUS INTRADURAL TUBERCULOMA OF THE THORACIC SPINE WITH COMPRESSIVE MYELOPATHY

Shyam Duvuru¹, Vivek Sanker², Naureen Syed³, Prakash Gupta⁴, Sanjana Rajurkar⁵, and Tirth Dave⁶

¹Apollo Speciality Hospitals Madurai
²Noorul Islam Institute of Medical Science and Research Foundation Medicity
³UTMDACC
⁴Virgen Milagrosa University Foundation
⁵Datta Meghe Institute of Higher Education & Research
⁶Bukovinian State Medical University

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INTRODUCTION:
Tuberculosis remains a significant global health burden, with central nervous system (CNS) involvement being a rare but serious manifestation. While pulmonary tuberculosis is the most common form, extrapulmonary manifestations can affect various organs and systems. The CNS involvement in developing countries constitutes nearly 10% of all tuberculosis patients [1]. Tuberculomas with compressive myelopathies without bone involvement are an even rarer occurrence. In the differential diagnosis of extensive spinal cord injuries, particularly in young patients with a history of pulmonary tuberculosis or tuberculous meningitis, it is important to consider the possibility of an intradural extramedullary tuberculoma [2].

Here we will be discussing a case of intradural extramedullary tuberculosis of the thoracic spine, with progressive neurological dysfunction. Surgery aims to decompress the spinal cord and remove the tuberculoma, thereby relieving the pressure on the neural tissues and preventing further neurological deterioration [3-5]. The specific surgical technique employed depends on the location and extent of the tuberculoma.

Following surgery, anti-tubercular therapy is initiated to target the underlying tuberculosis infection. This typically involves a combination of multiple anti-tuberculosis medications, such as isoniazid, rifampicin, ethambutol, and pyrazinamide [2]. The duration of anti-tubercular therapy may vary but generally lasts several months to ensure complete eradication of the infection. Continual postoperative monitoring assumes paramount importance in assessing neurological recovery and treatment response. Concurrently, physiotherapy and rehabilitation assume significance in facilitating the functional restoration and enhancing the overall quality of life [1].

This report aims to discuss the diagnostic challenges, treatment strategy, and clinical outcomes, highlighting the importance of a multidisciplinary approach in managing this uncommon condition.

CASE REPORT:
A 29-year-old man, who had been under treatment for disseminated tuberculosis for seven months, was admitted with progressive weakness of both lower limbs leading to difficulty in walking. There was no history of fever, cough, palpitations, or breathlessness. Physical examination revealed a palpable spleen but no lymphadenopathy. He had spastic paraparesis with involvement of right side more than the left. There was severe girdle pain at T5 level. The patient underwent a computed tomography (CT) chest, which
revealed multiple patchy resolving ground glass opacities and interlobular septal thickening in bilateral lower lobes, predominantly superior and posterior basal segments, suggesting resolving tuberculosis. Also, mild centrilobular emphysema in bilateral upper lobes with sub-centimeter right paratracheal and left paraaortic lymph nodes, the largest measuring 7 mm, were present. Other findings included splenomegaly measuring 17.6 cm and calcified hepatic granulomas (segment VII).

Magnetic Resonance Imaging (MRI) thoracic spine revealed a single 2.1 *1.7 * 1.45 cm intradural/juxta medullary and extramedullary mass appearing as well-defined T1-weighted low signal/ T2-weighted FLAIR bright signal with central necrosis and marginal enhancement after gadolinium (Gd) suggestive of immature tuberculoma at T5 level with surrounding edema (Figure 1: A, B, & C).

Figure 1: A, B, & C: The MRI images of the thoracic spine. A) Coronal T1-weighted MRI image shows a ring-like enhancing intradural lesion after gadolinium. B) Sagittal T1-weighted MRI image shows a 2.1 X1.7 X 1.45 cm intradural/juxta medullary and extramedullary mass with central necrosis and marginal enhancement after gadolinium suggestive of immature tuberculoma at T5 level. C) Axial T2-weighted image.

No evidence of vertebral TB, spinal tract infiltration, or engulfment was noted. A diagnosis of D5 Intradural extra medullary space-occupying lesion (SOL) with spastic paresis, cord compression, compressive myelopathy, disseminated tuberculosis, and post-tuberculous medication-induced hepatic granuloma was made. D4-D6 laminectomy and microsurgical excision under IONM was planned to remove the lesion. After pre-anesthetic assessment and consent, the patient underwent the procedure with the head held in a horseshoe, subperiosteal muscle dissection made, and laminae exposed. Durotomy was performed under a microscope and the lesion was visualized. A grayish-white, firm lesion with a central necrotic area was exposed (Figure 2).
Figure 2. The capsule and necrotic component is seen which is densely adherent to spinal cord.

The pus was aspirated, followed by internal bulking using ultrasonic surgical aspirator (Figure 3, 4, 5). The thin rim of the capsule was left behind but was coagulated well. Homeostasis was achieved, and the dura was closed using a 5-0 prolene continuous suture. The patient tolerated the procedure well. After surgery, he was treated with antibiotics, anti-diabetics, steroids, anti-tubercular drugs, anti-inflammatory, gastro protectives, antiemetics, and other supportive measures. Physiotherapy measures were instituted. The neurologist was involved in his paraparesis care, and an endocrinologist for glycemic control. He gradually improved symptomatically, able to walk with minimal support.

Figure 3. Intraoperative picture showing a densely adherent lesion with neovascularization.
Figure 4. The capsule is incised and pus is delivered off the lesion

Figure 5. Post-excision status with decompression of the spinal cord

DISCUSSION:

Tuberculosis (TB) remains a common health issue in developing countries even though TB has not been seen widely as before [6]. Osseous TB involvement may account for up to 35% of patients with extrapulmonary TB, and vertebral osteomyelitis infections (Pott’s disease) account for 25–60% of all osseous infections caused by TB [7-9]. Approximately 10% of patients with extrapulmonary tuberculosis (TB) experience central nervous system (CNS) involvement, typically resulting from the bacteremic stage of the disease. During this stage, tuberculous lesions known as Rich foci may develop in the meninges or within the brain or spinal cord tissue [10,11]. After several months or even years, these foci can rupture into the cerebrospinal fluid, leading to meningitis, or they can enlarge and form tuberculomas within the brain or spinal cord parenchyma.

The Mycobacterium bacilli can also spread hematogenously to various parts of the body, including the brain, due to minor bacteremia. The pathogenesis of central nervous system tuberculosis involves the development of small foci of tuberculosis, known as Rich foci, in the brain, spinal cord or meninges [10]. Earlier studies by Rich and McCordock showed that TBM requires direct inoculation of the bacilli through a meningeal focus in the central nervous system. However, later studies showed that disseminated tuberculosis plays...
a crucial role in the development of tuberculosis [12]. Tumor necrosis factor alpha (TNF-\(\alpha\)) is a critical cytokine in \textit{M. tuberculosis} neuropathogenesis. Although it helps in granuloma formation and control of mycobacterial infections, local production of TNF-\(\alpha\) in the central nervous system can alter the permeability of the blood-brain barrier and promote the progression of TBM [13].

\textit{M. tuberculosis} has a unique ability to enter and multiply in macrophages. Human microglia are selectively infected by \textit{M. tuberculosis} and the CD14 receptor facilitates engulfment of unopsonized bacilli by microglia. Microglia infected with \textit{M. tuberculosis} produce a variety of cytokines and chemokines, including TNF-\(\alpha\), IL-6, IL-1\(\beta\), CCL2, CCL5, and CXCL10. Microglia play a central role in the neuropathogenesis of tuberculosis, and their infection can lead to immunosuppressive effects, especially in more virulent strains [14].

Tuberculous meningitis is the most common form of central nervous system (CNS) tuberculosis (TB) compared to tuberculomas. Similarly, among various forms of spinal TB involvement such as Pott disease, non-osseous spinal TB, tuberculous spinal meningitis, and tuberculous arachnoiditis, non-osseous spinal tuberculosis is infrequent. In a review conducted by Dastur, 74 cases of spinal tuberculomas were analyzed, revealing that 65% were located extra-durally, 8% were intramedullary, 5% were intradural extramedullary, and 20% were arachnoidal [15-17]. Non-osseous spinal tuberculomas typically originate from a primary pulmonary focus, spreading hematogenously or through direct extension from hilar lymph nodes [18]. In our case, a rare and unexpected involvement occurred in the form of a non-osseous intradural extramedullary (IDEM) tuberculoma of the spinal cord, resulting in paraparesis despite the patient receiving antitubercular therapy (ATT). To our knowledge, this is the first reported case of a tuberculoma at the juxta-medullary location in the thoracic region.

The clinical presentation of the case aligns with the findings reported in the literature, which often show a higher prevalence among males [19]. The most common presentation in our case was spastic paraplegia, post-ATT liver failure, and respiratory problems associated with tuberculosis and COVID-19. Nevertheless, tuberculomas are slow growing with areas of necrosis that are encapsulated, avascular, and infrequently calcification. Our patient exhibited involvement of the thoracic spinal cord, which aligns with the reported incidence of up to 70% of cases in the literature. The thoracic spine is the most frequently affected site in Pott’s syndrome [20]. The radiological imaging findings of tuberculomas can vary depending on the stage of the lesion. On MRI, tuberculomas may present with non-caseating granulomas or caseating granulomas characterized by a solid or liquid center [21]. The tuberculous lesion typically appears isointense on T1W images, isointense to hypointense on T2W images, and exhibits ring enhancement with a hypointense center on gadolinium-enhanced MR scans. As the lesion undergoes caseation, the center becomes bright and gives rise to a target sign, as observed in our case [22-24].

A combination of surgical intervention and medical treatment has shown excellent outcomes. While some authors argue that anti-tuberculous therapy alone is sufficient when a paradoxical reaction develops into a tuberculoma [25,26], a literature review on intradural extramedullary spinal tuberculoma confirms the limited effectiveness of medical therapy alone and highlights the need for surgical intervention [27-33]. The reason for the poor response to chemotherapy in this condition remains unclear. Therefore, we believe that surgery is warranted when intradural extramedullary spinal tuberculoma arises as a paradoxical response to therapy.

The list of similar published cases is mentioned in Table 1:

<table>
<thead>
<tr>
<th>Case Report</th>
<th>Case age/Sex</th>
<th>Symptoms</th>
<th>Radiographic Findings</th>
<th>Treatment Plan</th>
<th>Outcome</th>
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<tr>
<td>Name et al., Year [Ref]</td>
<td>Age/Condition</td>
<td>Symptoms</td>
<td>Diagnosis</td>
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<tr>
<td>Kim et al, 2000 [34]</td>
<td>49/F</td>
<td>Seizure, facial palsy, left upper extremity weakness, paraparesis, hypesthesia below the T6 dermatome, urine incontinence</td>
<td>IDEM mass between the T1 and T2 spinal levels</td>
<td>Surgery and antituberculous drugs</td>
<td>Paraparesis has improved, and urine function is normal</td>
</tr>
<tr>
<td>Muthukumar et al, 2006 [35]</td>
<td>14/F</td>
<td>Tonic-clonic seizure</td>
<td>Lesion in the spinal cord</td>
<td>T7-T12 Laminectomy</td>
<td>Power gradually increased during a three-month period</td>
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<td></td>
<td>45/F</td>
<td>Numbness, sphincter problems, and lower extremity inability</td>
<td>T1-weighted scans show lesion spreading the cord, whereas T2-weighted images show hypointensity with a central region of hyperintensity.</td>
<td>T2-T5 Laminectomy</td>
<td>The patient’s neurological condition had not improved</td>
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<td>Hristea et al, 2008 [36]</td>
<td>20/M</td>
<td>Lower-limb weakness and disorientation</td>
<td>Obliterated thoracic and lumbar subarachnoid space</td>
<td>IV dexamethasone and intrathecal methylprednisolone</td>
<td>The patient was able to walk without assistance, had modest sensory loss, moderate spasticity, and no sphincter problems</td>
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<td>40/M</td>
<td>Right-leg paresthesia, walking difficulties, and bladder incontinence</td>
<td>Multiple intramedullary lesions at D4, D5—6, D8, D10, and D11</td>
<td>IV dexamethasone and intrathecal methylprednisolone</td>
<td>The patient was able to walk on his own</td>
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<tr>
<td></td>
<td>29/M</td>
<td>Sensory loss, paresthesia, gradual lower limb weakness, and walking difficulties</td>
<td>At T8-9, a solitary intramedullary mass suggests tuberculoma</td>
<td>IV dexamethasone and intrathecal methylprednisolone</td>
<td>The patient was able to walk on his own</td>
</tr>
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Table 1: List of case reports having similar presentation

CONCLUSIONS:

Intradural extramedullary (IDEM) tuberculoma is a rare manifestation of central nervous system (CNS) tuberculosis, often arising as a result of paradoxical reactions to antitubercular medication. When patients on antitubercular therapy develop new-onset neurological deficits indicative of compressive myelopathy, IDEM tuberculoma should be considered as one of the potential causes. Early and accurate diagnosis, along with timely initiation of appropriate medical or surgical interventions, is crucial to prevent irreversible dysfunction. Excision plays a pivotal role in the treatment of this uncommon condition.

CONFLICTS OF INTEREST:

None declared.

AUTHOR CONTRIBUTION:

All the authors contributed equally in drafting, editing, revising and finalizing the case report.
ETHICAL APPROVAL:
The ethical approval was not required for the case report as per the country’s guidelines.

CONSENT:
Written informed consent was obtained from the patient to publish this report.

DATA AVAILABILITY STATEMENT:
The data that support the findings of this article are available from the corresponding author upon reasonable request.

REFERENCES: