Case report

Spinal adhesive arachnoiditis following surgery treatment for an intramedullary tuberculoma of the conus medularis: A case report

ABSTRACT

Aims: Describe a rare case of an intramedullary tuberculoma (IMT) complicated with chronic adhesive arachnoiditis (CAA).
Presentation of case: A 19-year-old- female presented with complaints of weight loss, progressive weakness of both lower limbs and urinary retention. Neurological examination corroborated the motor dysfunction of the lower limbs, associated with sensory disturbances. Magnetic resonance imaging (MRI) detected a ring enhancing intramedullary mass expanding the conus medullaris. A provisional diagnosis of neoplastic origin lesion was given, and the patient had complete resection of the mass. Histopathology was consistent with tuberculoma. 8 months after beginning postsurgical antituberculous therapy the neurological symptoms worsened and follow-up MRI showed a distorted spinal cord with arachnoid cysts and septations, findings corresponding to CAA. Despite a new surgical intervention, the patient remained with lasting disability.
Discussion: IMT is an uncommon presentation of spine tuberculosis. The clinical manifestations mimic those of any space occupying lesion (SOL) and are dependent of the location in the spine, thus a differential diagnosis with neoplastic, inflammatory and other granulomatous lesions is required. MRI is the optimal method for evaluating and diagnosing IMT. Pharmacological and surgical interventions aim to decompress the spinal cord and recover neurological functions. However, unexpected complications as CAA can occur impeding a satisfactory recovery.
Conclusion: The diagnosis of IMT is challenging due to its low frequency and imaging features. Therefore, this report emphasizes the importance of recognition the clinical and radiological presentation of IMT to allow an early diagnosis, ease the selection of the therapeutic interventions and avoid complications.

Keywords: Tuberculosis, intramedullary tuberculoma, magnetic resonance imaging, chronic adhesive arachnoiditis.

1. INTRODUCTION

Tuberculosis (TB) is a multifaceted disease secondary to the infection by the bacillus *Mycobacterium tuberculosis* [1]. Central nervous system tuberculosis (CNS) TB stands out among other extrapulmonary variants of TB on account of its significant morbidity and devastating complications. Tuberculous meningitis, tuberculoma, miliary TB, tuberculous abscess, tuberculous encephalopathy and spinal TB constitute the spectrum of CNS TB [2]. In the spine arachnoiditis, spondylitis and myelitis represent the vast majority of cases, however there are rare presentations as the tuberculoma [2,3]. To the date about 175 cases of intramedullary tuberculoma (IMT) have been reported in the literature [4]. Given its low

frequency, the prompt recognition of its clinical-radiological features, in which Magnetic resonance imaging (MRI) play an important role, is of vital importance for the selection of the therapeutic interventions and avoid complications [5].

In this report we summarize an unusual case of IMT affecting the conus medullaris which progressed to chronic adhesive arachnoiditis (CAA), despite medical treatment.

2. CASE PRESENTATION

A 19-year-old female was admitted complaining of weight loss, sensory disturbances of the left lower limb and progressive weakness of both lower limbs for the past 6 months. 1 week before her admission persistent back pain and a feeling of incomplete voiding of urine added. On examination, cognitive functions and cranial nerves were normal. The muscle strength in the right lower extremity was 4/5 and in the left lower extremity was 3/5. There was hypoesthesia in areas below the T8 dermatome, her reflexes were brisk and a rest tremor was found in the right lower extremity. The patient tested negative to HIV. Other blood parameters and plain X-Rays of the chest and dorsolumbar spine were all normal. MRI of the brain and whole spine revealed an intramedullary mass at T12-L1 level, causing a fusiform enlargement of the conus medullaris (Fig. 1). The oval-shaped mass measured 1.4 x 1 x 0.9 cm, had sharp margins and showed a central isointense signal to the cord with a hyperintense halo in T1 weighted images (WI) and heterogenous intensity, mainly hypointense in T2 WI, surrounded by cord edema spanning from T6 to L1. On post-gadolinium T1 sequence the mass exhibited an avid ring-enhancement pattern. Since this mass behaved as a space occupying lesion (SOL) a provisional diagnosis of glioma or ependymoma was given and resection surgery was offered to the patient.

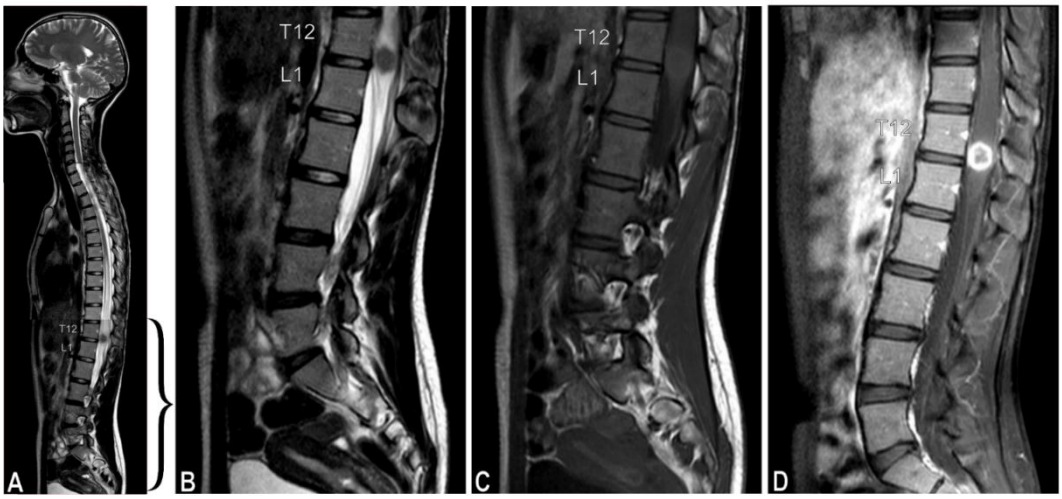

Fig. 1. (A) Neuroaxis MRI and (B) sagittal T2 weighted image of the thoraco-lumbar spine showing a hypointense intramedullary lesion at T12-L1 level associated with cord edema spanning from T6-L1. (C) On sagittal T1 weighted image a fusiform dilation of the conus medullaris is seen. (D) Gadolinium-enhanced T1 weighted image revealing a ring enhancing pattern of the lesion.

The excision of the lesion was done through posterior approach via laminectomy from T11 to L2 level. Midline durotomy exposed a whitish, avascular, multilobulated, soft tumor, reaching the pial surface and infiltrating the dorsal nerve roots, thereby the tumor was dissected

employing microsurgical technique. Histopathological examination showed granulomatous chronic inflammatory changes characterized by granulomas composed of central caseating necrosis encircled by peripheral multinucleated Langhan's-type giant cells, epithelioid cells and lymphocytes. Ziehl-Neelsen staining identified *M. tuberculosis* (Fig. 2). These findings were consistent with IMT and accordingly, the patient started antituberculous pharmacotherapy (APh). However, 8 months after surgery and APh, the weakness of both lower limbs and sensory disturbances worsened, leading to the confinement of the patient to bed. She was readmitted and a follow-up MRI at this point showed a distorted and tethered spinal cord secondary to cord atrophy alternating with cord edema, loculated arachnoid cysts with mass effect on the cord and arachnoid septations, extending from T6 to L1 levels. Beneath L2 level the nerve roots were clump forming a soft tissue mass. After gadolinium a pial and dural enhancement was observed (Fig. 3-Fig. 4). The previous findings corresponded to CAA and a new surgical intervention with cyst drainage and mycrolisis of adhesions was performed. Despite these procedures the strength of the lower extremities was not recovered.

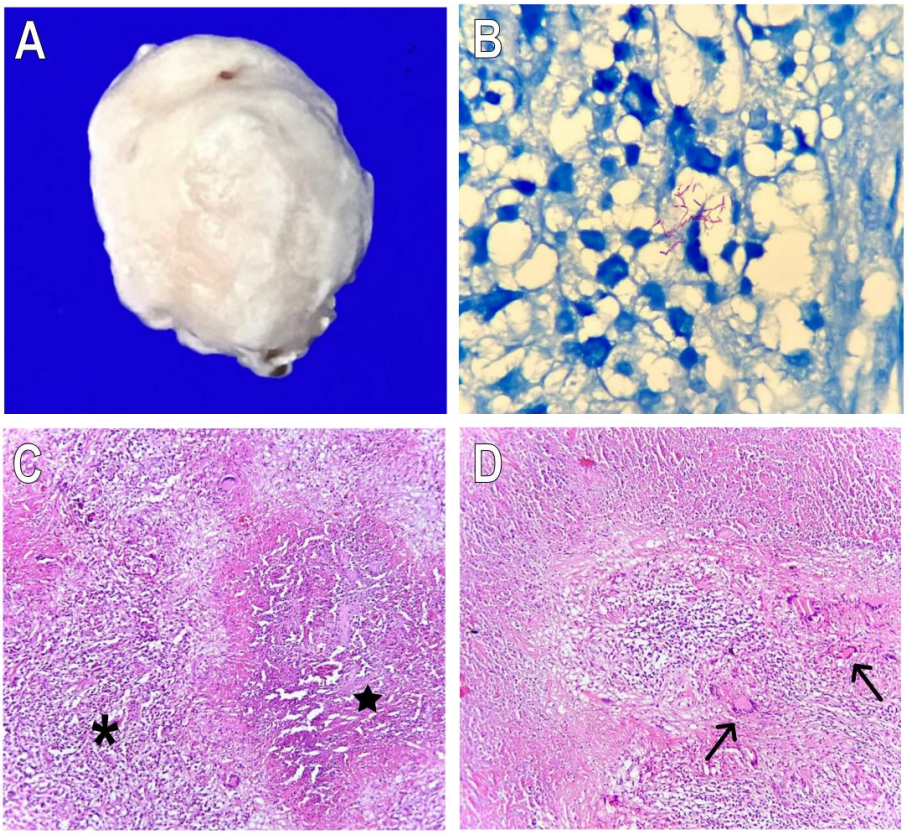

Fig. 2. (A) Photograph of the pathologic specimen obtained via laminectomy showing a whitish and rounded soft tumor. (B) Photomicrograph highlighting acid-alcoholic resistant bacilli at Ziehl Neelsen stain (100 x). (C-D) Photomicrographs of hematoxylin and eosin stain showing a granulomatous lesions with central caseating necrosis (star) surrounded by epithelioid cells, lymphocytes (asterisk) and peripheral giant Langhan´s giant cells (arrows).

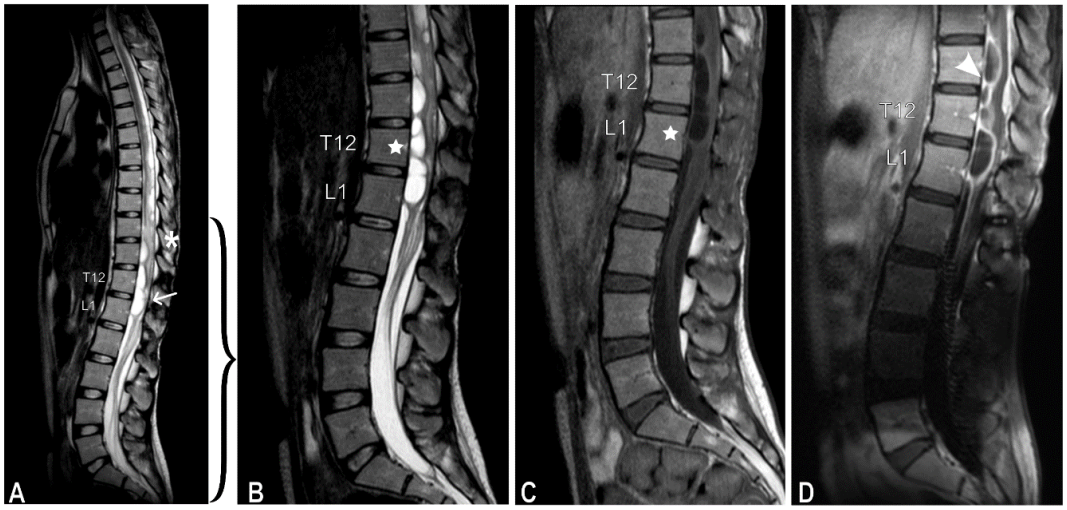

Fig. 3. Follow-up MRI. (A) Sagittal T2 weighted image of the thoraco-lumbar spine showing a distorted cord, with edema, and alternating segments of tethered cord (asterisk) and atrophy (arrow) extending from T6 to L1. (B-C) Sagittal T2 and T1 weighted images focused on low thoracic and lumbar spine exhibiting loculated arachnoid cyst with mass effect on the cord, delimited by arachnoid septations (star). (D) Gadolinium-enhanced T1 weighted image revealing pial and dural enhancement (arrow head).

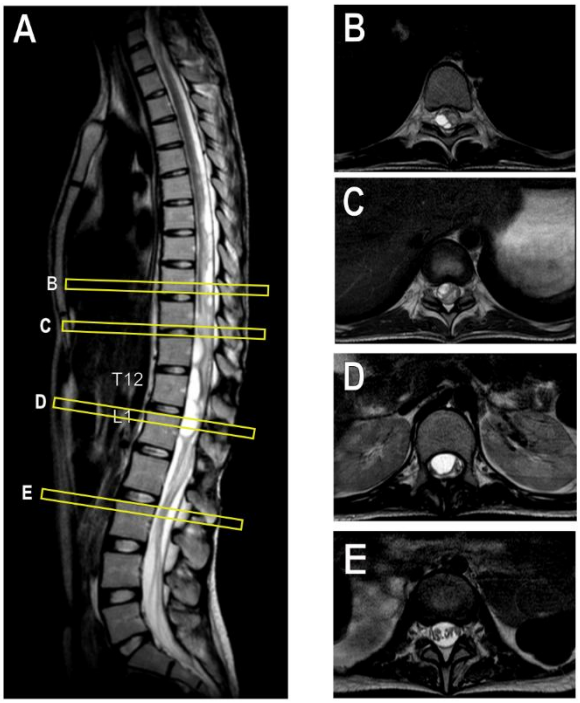

Fig. 4. Follow-up MRI. (A) Sagittal T2 weighted image of the thoraco-lumbar spine showing axial sections at different levels (yellow rectangles). (B-C) Axial T2 weighted images showing loculated arachnoid cyst with a tethered cord, (D) cord atrophy and (E) cauda equina nerve fibers forming a soft tissue mass at both margins of the thecal sac

3. DISCUSSION

IMT was first described by Abercrombie in 1828, occurs in 2 of 1000 cases of CNS TB and the intracranial tuberculoma to IMT ratio is approximately 42:1 [4, 7]. As in our case, the greatest incidence of IMT is found in young people (mean age: 29.7 years) and that has been attributed to reactivation of a previous TB infection by immunodeficiency disorders, although our patient has neither history of TB nor evidence of immunosuppression [5, 8]. Clinical presentation differs according the location of the lesion, but motor weakness (100%) and sensory involvement (50%) are frequently described [5]. The bilateral lower limbs weakness associated to back pain, paresthesia and urinary disfunction allowed us to suspect a conus medullaris syndrome, thus stablish the location of the disease in the spine [9]. Miyamoto et al [10] reviewed 15 cases of IMT and found that the cervical and thoracic cord were the preferred segments affected with 47% of the cases each and the remaining 6 % corresponded to IMT of the conus. It has been proposed that these frequencies are determined by the distribution of the blood flow in the spinal cord, with thoracic segment receiving 45% of the entire supply of the cord [11].

Clinical presentations of IMT are indistinct from any other SOL, hence MRI is the optimal method to characterize intramedullary lesions [5]. IMT depicts a varying appearance according to each stage of the granulomatous lesion formation. In the early phase a non-caseating tuberculoma corresponds to a solid lesion. Subsequently, a central caseating zone of necrosis surrounded by external collagenous tissue capsule shapes a solid caseating tuberculoma, which behaves isointense at T1WI and isointense to hypointense at T2WI. At contrast-enhanced imaging a ring enhancement with hypointense center that gradually become hyperintense, as the caseation progress, gives the appearance of the target sign. The imaging findings and histological examination of our patient was consistent with this latter phase. A tuberculoma with central liquefaction is formed in the last stage and it may be indistinguishable from an infectious abscess [5,8]. This variability in the MRI appearances may mimic neoplastic, inflammatory, demyelinating, vascular and other granulomatous lesions. In addition to the unusual imaging findings, the absence of past or concurrent TB infection in a young adult with no comorbidities led to doubt in diagnosis [5,12]. Surgical treatment was indicated due to the suspicion of a neoplastic SOL, the size of the lesion and the rapid onset of the neurological dysfunction. Once diagnosis of IMT was confirmed we initiated APh, expecting an enhance in its effectiveness, after removal of the necrotic content and debulking of the tuberculoma [7,10].

TB is most common cause of infectious spinal arachnoiditis [13]. CAA is a devastating form of persistent pia-arachnoid inflammation that favors the formation of fibrinous exudate in the surface of the nerve roots and the spinal cord, which in association with fibroblast proliferation and collagen deposition lead to intrathecal scars and dural adhesions affecting cerebrospinal fluid (CSF) flow [14]. To the extent of our knowledge this is the first report that relates CAA to IMT. The uncommon relation between CAA and IMT can be explained by the tuberculoma structure itself, which is surrounded a collagenous capsule and nervous tissue, features that difficult the extension to the meningeal layers. Etiology of CAA is heterogeneous, with trauma, previous spinal surgery, infections and epidural anesthetics among the main causes. Therefore, we believe that the surgical intervention synergized the TB infection to develop CAA. MRI is the gold standard for the diagnosis of CAA, having 92% sensitivity and 100% specificity [14,15]. In the follow-up MRI of our case, we found the most frequent findings linked to CAA affecting the thoraco-lumbar spine, such as cord atrophy and tethering, cord edema, loculated arachnoid cyst, arachnoid septations and pial-dural enhancement [15]. The cauda equina nerve fibers distribution was described forming a soft tissue mass at both margins of the thecal sac, thus corresponding to the most advanced stage of the disease or group 3 proposed by Delamarter et al [16]. Despite the vast

descriptions of the imaging features of CAA, many studies suggest that MRI and clinical findings are not related, then clinicians should take decisions based on clinical data [17]. The progressive behavior of the motor and sensory symptoms in our patient prompted the election for surgical intervention to release the cicatricial adhesions and recover CSF flow. However, as many other severe cases, our case was refractory to surgery, remaining with lasting disability.

4. CONCLUSION

In this report, we presented a unique case of two uncommon clinical-radiological presentations of CNS TB. As patients with IMT may show only unspecific signs of an SOL, MRI is essential for discerning the etiology among more frequent intramedullary lesions. The recognition of the imaging characteristics allows to reach an early diagnosis, easing the selection of the therapeutic interventions that may yield a better result and avoid complications. CAA is an unexpected complication after surgical removal of IMT with a complex and not entirely understood pathophysiology. Therefore, future studies are required that ameliorate the diagnosis and management of this devastating disease.

CONSENT

All authors declare that written informed consent was obtained from the patient for publication of this case report and accompanying images.

ETHICAL APPROVAL

As per international standards or university standards written ethical approval has been collected and preserved by the author (s).

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