Spinal adhesive arachnoiditis following surgical treatment for an intramedullary tuberculoma of the conus medullaris: A case reportfrom western Mexico

ABSTRACT

Objectives: Describe a rare case of intramedullary tuberculoma (IMT) complicated by chronic adhesive arachnoiditis (CAA).
Presentation of case: A 19-year-old woman 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 that expanded the conus medullaris. A provisional diagnosis of neoplastic origin lesion was made and the patient received complete resection of the mass. Histopathology was consistent with tuberculoma. 8 months after beginning postoperativeantituberculostherapy, neurological symptoms worsened and follow-up MRI showed a distorted spinal cord with arachnoid cysts and septations, findingsthat corresponded 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 onthe location in the spine, thus a differential diagnosis with neoplastic, inflammatory, and other granulomatous lesions is required. Being IMT an encapsulated lesion,it is unexpected thatthe inflammatory responseextends to pia-arachnoid layers conducting to complications, such asCAA. The association between IMT and CAA must be recognized, since it undermines the outcomes of therapeutical interventions.
Conclusion: The diagnosis of IMT complicated by CAA is challenging due to its low frequency. Therefore, this report emphasizes the importance of recognizing the clinical and radiological featuresof this association to allow an early diagnosis, andfacilitate the selection of therapeutic interventions.

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

1. INTRODUCTION

Tuberculosis (TB) is a multifaceted disease secondary toinfection by the bacillus *Mycobacterium tuberculosis* [1]. Central Nervous System Tuberculosis (CNS) TB is a distinct disease among other extrapulmonary variants of TB due to its significant morbidity and devastating complications[2]. Tuberculous meningitis, tuberculoma, miliary TB, tuberculous abscess, tuberculous encephalopathy, and spinal TB constitute the spectrum of CNS TB [3].In the spine, arachnoiditis, spondylitis, and myelitis represent the vast majority of cases;however, there are rare presentations such as tuberculoma [3,4].

1.1 Intramedullary tuberculoma

Tuberculomas are well defined focal masses that result from TB infection. In the spine tuberculomas may be extradural, intradural extramedullary or intramedullary in location [3,4].To the date about 175 cases of intramedullary tuberculoma (IMT) have been reported in the literature [5,18,19,20,21]. Given its low frequency,prompt recognition of its clinical and radiological features, in which magnetic resonance imaging (MRI) plays an important role, is of vital importance for the selection of therapeutic interventions and avoid complications [6].

1.2 Chronic adhesive arachnoiditis

TB is themost common cause of infectious spinal arachnoiditis [7]. Chronic adhesive arachnoiditis (CAA) is a devastating form of persistent pia-arachnoid inflammation that favors the formation of fibrinous exudateson the surface of nerve roots and spinal cord, which, in association with fibroblast proliferation and collagen deposition, lead to intrathecal scars and dural adhesions that affect cerebrospinal fluid (CSF) flow. This scar forming disease has a highly variable clinical presentation and may progress to an incapacitating condition [8].

In this report, we summarize an unusual case of IMT affecting the conus medullaris that progressed to CAA, despite medical treatment.

2. CASE PRESENTATION

A 19-year-oldmestizowomanfrom western Mexico with no family history of tuberculosis or immunosuppressionwas 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 admission, persistent back pain and a feeling of incomplete urine voiding added. On examination, cognitive functions and cranial nerves were normal. Muscle strength in the right lower extremity was 4/5 and in the left lower extremity was 3/5. Hypoesthesiawas foundin areas below the T8 dermatome, her reflexes were brisk and a rest tremor was found in the right lower extremity.Laboratory tests were hemoglobin of 10 g/dL, white blood cells of 6,700/mcL, platelets of 397,000 cells/mcL, serum creatinine of 0.7 mg/dL, blood urea nitrogen of 23.1 mg/dL, and glucose of 70 mg/dL.Hypochromic, microcytic anemia was treated with oral iron supplementation.The patient tested negative for HIV. Plain X-rays of the chest and dorsolumbar spine were normal. Magnetic resonance imaging of the brain and entire spine revealed an intramedullary mass at theT12-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 the T1 weighted images (WI) and heterogeneous intensity, mainly hypointense in T2 WI, surrounded by cord edema spanning from T6 to L1. On the 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.

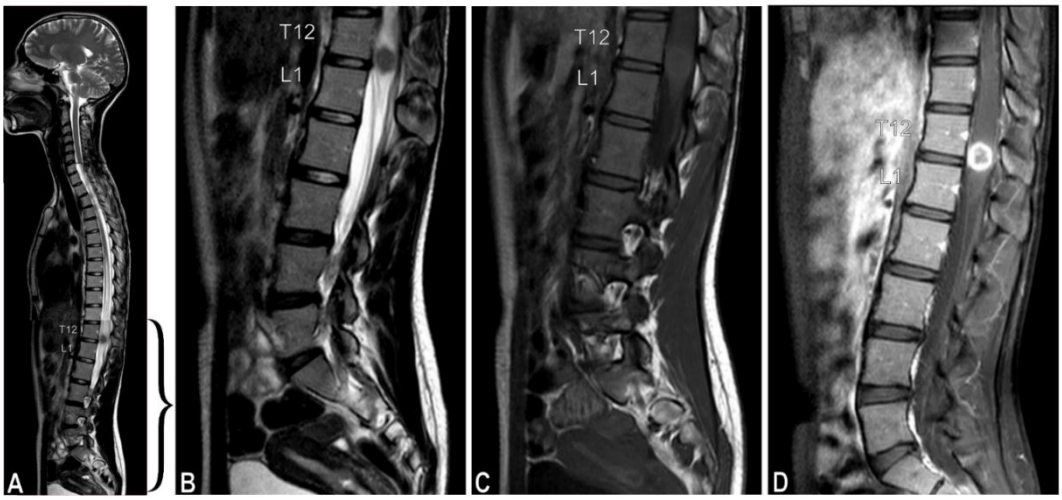

Fig. 1. (A) Neuroaxis magnetic resonance imaging and (B) sagittal T2 weighted image of the thoracolumbar spine showing a hypointense intramedullary lesion at theT12-L1 level associated with cord edema spanning from T6-L1. (C) In the sagittal T1 weighted image a fusiform dilation of the conus medullaris is seen. (D) Gadolinium-enhanced T1 weighted image that reveals a ring enhancing pattern of the lesion.

The excision of the lesion was made by posterior approach through laminectomy from the level T11 to L2.The midline durotomy exposed a soft,whitish, avascular, multilobulated, soft tumor, reaching the pial surface and infiltrating the dorsal nerve roots, thereby dissectedthe tumor usingmicrosurgical technique. Histopathological examination showed chronic granulomatous inflammatory changes characterized by granulomas composed of central caseating necrosis surrounded 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 therefore the patient started antituberculous pharmacotherapy (APh). However, 8 months after surgery and APh, the weakness of both lower extremities 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. At the bottom level of L2, the nerve roots were clumpsthat formeda soft tissue mass. After gadolinium, pial and dural enhancement was observed (Figs.3-Fig. 4). The previous findings corresponded to CAA and a new surgical intervention with cyst drainage and adhesionmicrolysis was performed. Despite these procedures, the strength of the lower extremities was not recovered.

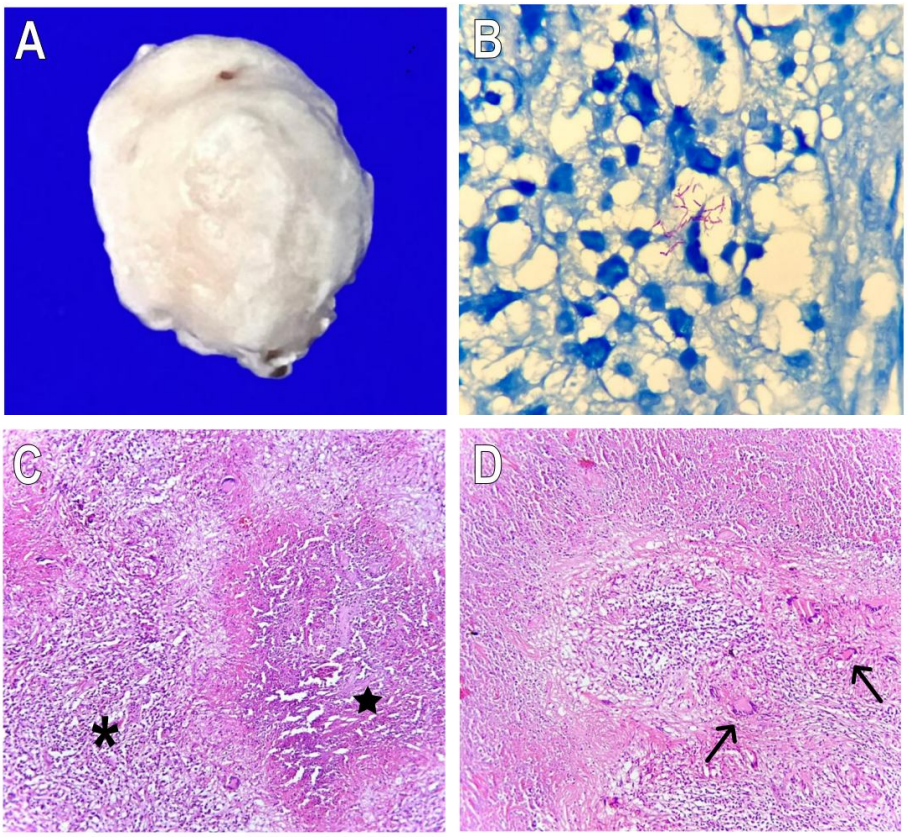

Fig. 2. (A) Photograph of the pathologic specimen obtained by laminectomy showing a whitish and rounded soft tumor. (B) Photomicrograph highlighting acid-alcoholic resistant bacilli at Ziehl-Neelsen stain (100 x). (C-D) Photomicrographs of hematoxylin and eosin stainingshowinggranulomatous lesions with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

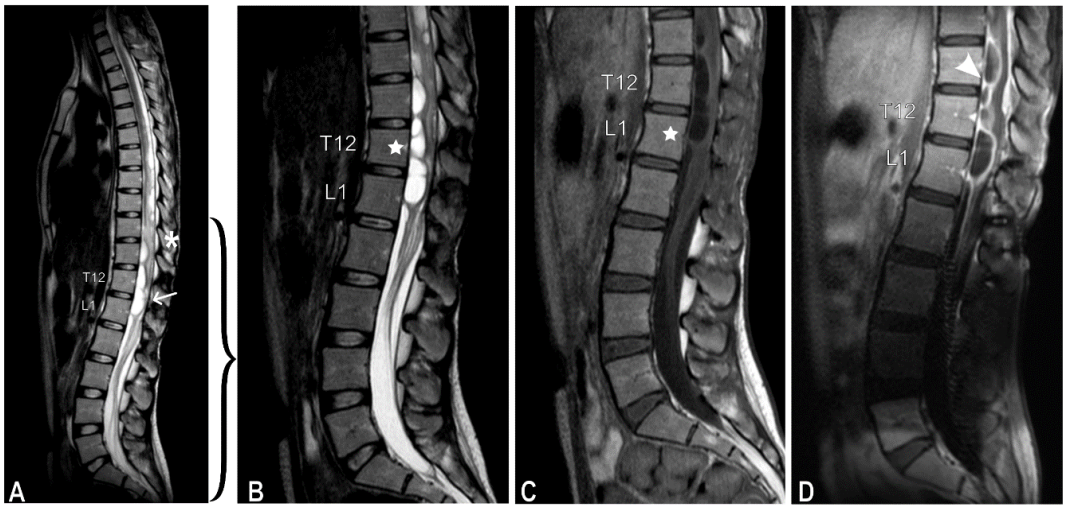

Fig. 3. Follow-up magnetic resonance imaging. (A) Weighted sagittal T2 image of the thoracolumbar spine showing a distorted cord, with edema, and alternating segments of

thetethered cord (asterisk) and atrophy (arrow) extending from T6 to L1. (B-C) Sagittal images weighted T2 and T1 focused on thelow thoracic and lumbar spine exhibiting a loculated arachnoid cyst with amass effect on the cord, delimited by arachnoid septations (star). (D) T1 weighted gadolinium-enhanced image revealing pial and dural enhancement (arrow head).

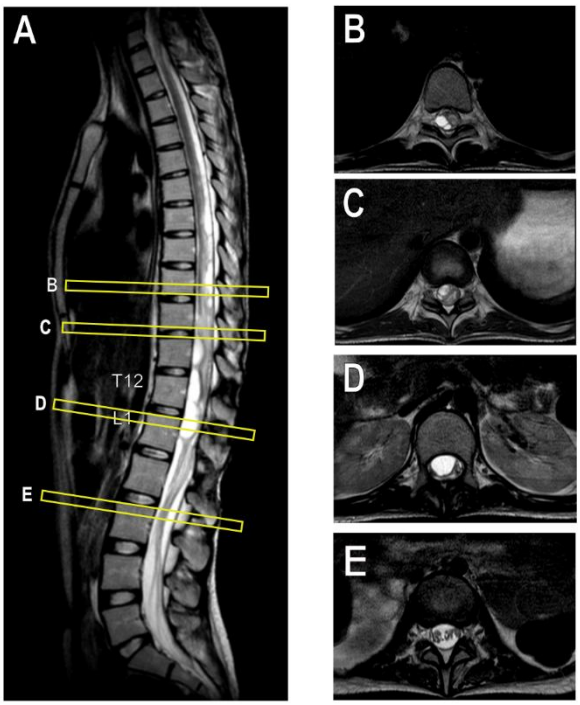

Fig. 4. Follow-up magnetic resonance imaging. (A) Weighted sagittal T2 image of the thoracolumbar spine showing axial sections at different levels (yellow rectangles). (B-C) Axial T2 weighted images showing loculated arachnoid cyst with a tethered cord, (D) cord atrophy, and (E) cauda equina nerve fibers 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 outof 1000 cases of TB of CNS and the intracranial tuberculoma to IMT ratio is approximately 42:1[5, 9]. As in our case, thehighest incidence of IMT is found in young people (mean age: 29.7 years) and has been attributed to thereactivation of a previous TB infection by immunodeficiency disorders, although our patient has neither a history of TB nor evidence of immunosuppression [6, 10].The clinical presentation differs according to the location of the injury, but motor weakness (100%) and sensory involvement (50%) are frequently described [6]. The bilateral weakness of the lower extremities associated withback pain, paresthesia and urinary dysfunction allowed us to suspect a conus medullaris syndrome, thus establishing the location of the disease in the spine [11]. Miyamoto et al.[12] 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. These frequencies havebeen proposed to be determined by the distribution of blood flow in the spinal cord, with thethoracic segment receiving 45% of the entire supply of the cord [13].

The clinical presentations of IMT are indistinct from any other SOL;therefore, MRI is the optimal method to characterize intramedullary lesions [6].The IMT shows a varying appearance according to each stage of the formation ofgranulomatous lesion. In the early phase, a noncaseating tuberculoma corresponds to a solid lesion. Subsequently, a central necrosiscaseating zone surrounded by anexternal collagenous tissue capsule forms 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 becomes hyperintense, as the caseation progresses, gives the appearance of the target sign. The imaging findings and histological examination of our patient were consistent with this last phase. In the last stage, a tuberculoma with central liquefaction is formed and may be indistinguishable from an infectious abscess [6,10]. This variability in the appearance of the magnetic resonance may mimic neoplastic, inflammatory, demyelinating, vascular, and other granulomatous lesions. In addition to unusual imaging findings, the absence of previous or concurrent TB infection in a young adult without comorbidities led to doubt in diagnosis [6,14]. Surgical treatment was indicated due to suspicion of a neoplastic SOL, the size of the lesion, and the rapid onset of neurological dysfunction. Once IMT diagnosis was confirmed, we initiated APh, expecting anincrease in its effectiveness, after removal of necrotic content and debulking of the tuberculoma [9,12].

To the best of our knowledge, this is the first report that relates CAA to IMT. The uncommon relationship between CAA and IMT can be explained by the tuberculoma structure itself, which is surrounded bya collagenous capsule and nervous tissue, characteristics that makeextension to the meningeal layersdifficult.The 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 TB infection to develop CAA[7].Magnetic resonance imaging is the gold standard for the diagnosis of CAA, having 92% sensitivity and 100% specificity [8,15]. In the follow-up MRI of our case, we found the most frequent findings related to CAA affecting the thoracolumbar spine, such as cord atrophy and tethering, cord edema, loculated arachnoid cyst, arachnoid septations, and pial-dural enhancement [15]. The distribution ofcauda equina nerve fibers was describedasforming 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;therefore, clinicians should make decisions based on clinical data [17]. The progressive behavior of motor and sensory symptoms in our patient prompted the choice of surgical intervention to release cicatricial adhesions and recover CSF flow. However, as many other severe cases, our case was refractory to surgery and remainedwith lasting disability.

4. CONCLUSION

In this report, we presented a unique case of two uncommon clinical and radiological presentations of CNS TB. As patients with IMT may show only unspecific signs of an SOL, MRI is essential to discern the etiology among themore frequent intramedullary lesions.Recognition of imaging characteristics allows one reach an early diagnosis, easing the selection of 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 to improve the diagnosis and management of this devastating disease.

CONSENT

All authors declare that written informed consent was obtained from the patient for thepublication of this case report and theaccompanying 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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