Spinal intramedullary tuberculoma

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Abstract: Spinal intramedullary tuberculosis is a rare disease entity. The authors report two healthy adult patients presented with progressive paraparesis due to histologically proven intramedullary tuberculomas. The spinal magnetic resonance imaging (MRI) scans revealed intensely enhancing intramedullary lesion. Surgery was performed through laminectomy and complete resection was achieved. Both patients improved after surgery with normal motor power and follow-up MRI showed complete excision. *(p85-88)*

Introduction

Tuberculosis remains an important pathological entity in developing countries.

There are approximately 30 million active cases in the world, 10 million new cases occur annually, and tuberculosis probably causes 6% of all deaths worldwide.[17] The incidence of tuberculosis in North America and Europe had been in decline since 1986, when a reversal in this trend was thought to occur secondary to the spread of the acquired immunodeficiency syndrome and an increase in the number of immunocompromised patients.[2] Central nervous system (CNS) tuberculosis is a rare entity, affecting 0.5-2% of patients with systemic tuberculosis.[13] The spinal cord is much less commonly involved than the brain at a ratio of approximately 1:20-1:42.[4,9,12,16,17] This correlates well with the average ratio of mean spinal cord to brain weight of 1:47, suggesting that the location of these lesions is determined by the relative distribution of neural tissue, although other factors may be involved.[17]

Spinal cord disease from tuberculosis is most frequently due to Pott's disease. Mathai and Chandy reported that 42% of their spinal operations for non-traumatic paraplegias were for compression resulting from tuberculous spondylitis (Pott's disease).[17] Dastur reviewed 74 cases of tuberculous paraplegia without evidence of Pott's disease and discovered that extradural granulomas occurred at a rate of 64%, arachnoidal lesions without dural involvement occurred at a rate of 20%, intramedullary lesions occurred at a rate of 8%, and subdural/extramedullary lesions occurred at a rate of 1%.[3] Spinal intramedullary tuberculomas are extremely rare, seen in only 2 of 100,000 cases of tuberculosis and 2 of 1,000 cases of tuberculous CNS disease. Approximately 150 cases have been reported in the world literature to date, mainly in third world countries.[17-18]

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Case Report

Case 1: A 16-year-old male patient, presented with progressive spastic paraparesis and urinary incontinence of 3 months duration. No past history of TB. On examination there was spastic paraparesis grade 2-3 power with sensory level at L1 and severe spasm of back muscles. We could not detect any tuberculous focus in his body. Magnetic resonance imaging (MRI) of the dorsolumbar spine with contrast showed lower dorsal spinal cord and conus medullaris expansion by intramedullary mass lesion, exhibiting mixed low and slightly bright signals on T1-weighted image (Fig. 1), bright signals of the upper and lower part and low centre signals on T2-weighted image (Fig. 2) with intense enhancement after Gd-DTPA injection of the lower part of the lesion (Fig. 3). Surgery was performed in the prone position. The procedure involved laminectomies from T10 through L1, followed by a midline myelotomy. The lesion was well-circumscribed and was excised completely using the microscope. Histopathological examination revealed a granulomatous lesion that contained Langerhans' giant cells, inflammatory cells, and evidence of caseating necrosis. The patient was prescribed a 12 months course of anti-tuberculous drugs. Patient improved slowly and on follow-up after 6 months he had grade 5 motor power in both lower limbs but still had urine incontinence. Follow-up MRI (Fig. 4) with contrast 9 months later showed no residual granuloma.

Case 2: 45-year-old female presented to KAUH with back pain for 6 months and progressive lower extremity

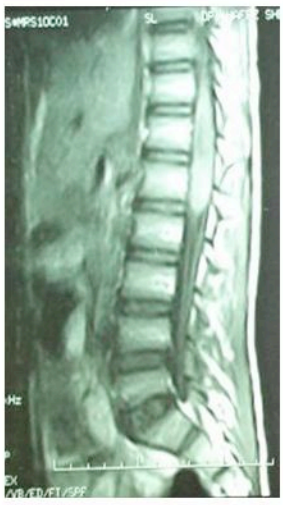
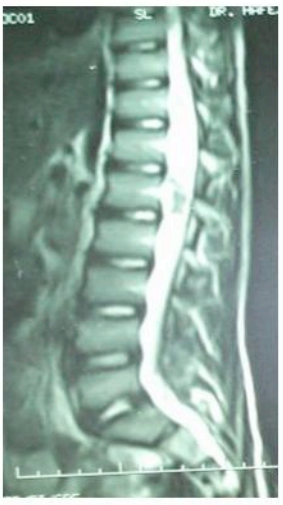
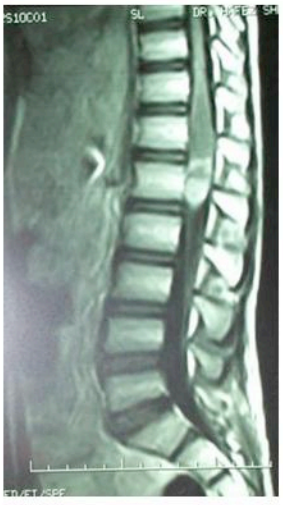
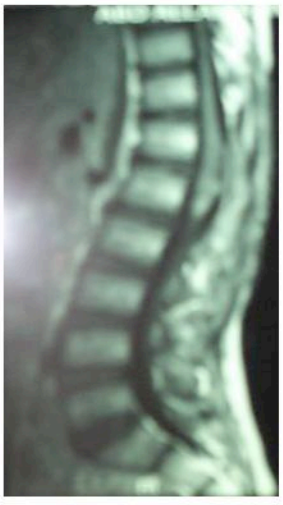

Figure 1 - T1-WI MRI scan demonstrating mixed low and bright signals of intramedullary lesion of lower dorsal spinal cord and conus meullaris. **Figure 2 -** T2-WI MRI scan demonstrating central hypointensity with surrounding bright signals of the lesion. **Figure 3 -** Enhanced T1-WI MRI demonstrating intense enhancement of the lower part of the lesion. **Figure 4 -** Postoperative enhanced T1-WI MRI after one year of follow up showed no residual tuberculoma

weakness which manifested in a fall down the stairs 2 months later. After a 2 week period, the patient began to have episodic urinary incontinence with further progression of lower extremity weakness to the point of inability to ambulate. General examination was within normal and there was no back swelling or tenderness. Neurological examination revealed grade 3 paraparesis with a sensory level at the 9th thoracic dermatome and her rectal tone was diminished. Routine laboratory investigations, including CBC and ESR, and her chest x-ray were within normal. A MRI scan of her entire spine revealed a swollen cord with diffuse oedema (Fig. 5). T1-weighted MRI scan revealed 20 x 25 mm intramedullary lesion at T12/L1 level brightly enhancing after administration of contrast (Fig. 6). Surveillance MRI of the entire neuraxis and brain did not reveal additional lesions. Based on the MRI findings, she was provisionally diagnosed as having primary CNS lymphoma. The patient underwent 2 level laminectomy and complete microsurgical resection was achieved trough midline myelotomy. There were no changes on the pia of the spinal cord apart from focal swelling. A relatively vascular and firm intramedullary lesion was encountered with no clear cleavage from the spinal cord tissue Histological analysis showed non-caseating granulomas surrounded by reactive gliosis and increased scattered histiocytes. Acid fast bacilli (AFB) stain demonstrated presence of numerous AFB in multiple specimens. The patient had a smooth postoperative

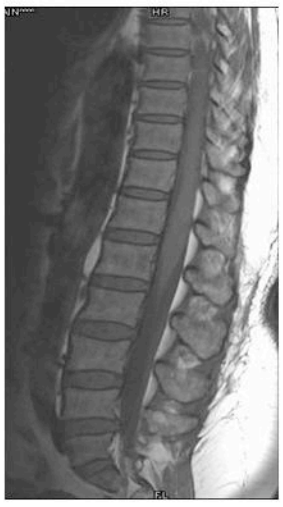
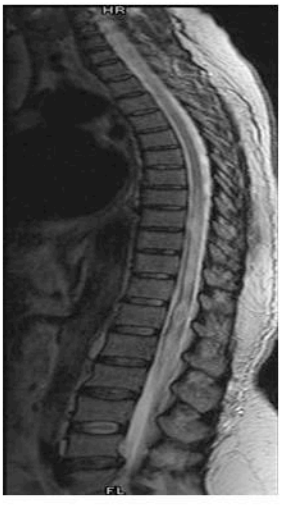

Figure 5 - (**a**) T1-WI and (**b**) T2-WI MRI demonstrating diffuse cord swelling and oedema with isointense intramedullary lesion

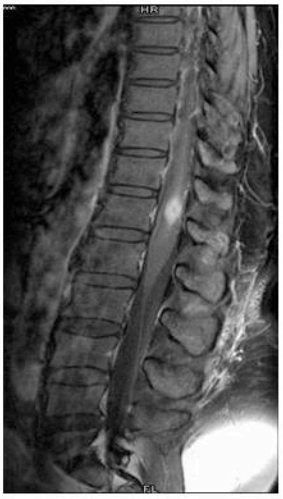

Figure 6 - Enhanced T1-WI MRI demonstrating intense homogenous enhancement of the lesion

recovery with marked improvement of motor and bladder function. She was prescribed 9 months course of antituberculous therapy. Postoperative MRI follow-up confirmed complete resolution of the lesion (Fig. 7). At 3 year follow-up, she had complete neurological recovery and with no recurrent lesion on MRI scans.

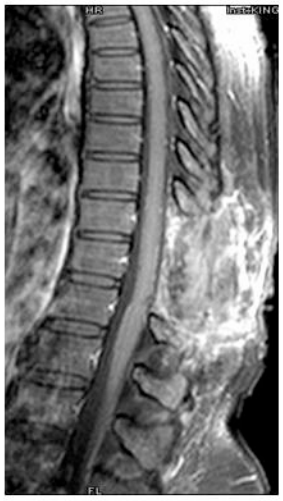

Figure 7 - One year postoperative follow up enhanced T1-WI MRI demonstrating no residual tuberculoma

Discussion

Tuberculosis is a serious disease, that continues to occur worldwide, particularly in developing regions. When the CNS is affected, morbidity and mortality are high.[4]

Spinal intramedullary tuberculomas were first described by Serra in 1840.[16] Lin, et al reviewed the literature of spinal intramedullary tuberculomas in 1960 and found reports of 104 cases, 88 of which were found on post-mortem.[16] In a more recent study, Ratliff found only 148 cases of spinal intramedullary tuberculomas mentioned in the literature.[14] Sharma, et al reported 10 cases of spinal intramedullary tuberculoma.[18]

The various types of spinal intramedullary tuberculous lesions include tuberculoma, spinal cord oedema, and cavitation.[9] Widespread use of MRI has allowed more accurate and frequent detection of intraspinal tuberculoma.[5,8,15,18]

Most of the reported cases of spinal intramedullary tuberculomas are associated with foci of tuberculosis elsewhere in the body.[16] They are caused by the blood borne infection of the tubercle bacilli, secondary to tubercular infection elsewhere, commonly from the lung.[9,11] Hanci, et al mentioned that history of previous tuberculosis or tuberculous contact is usually present and some of the cases of spinal intramedullary tuberculous abscess have been reported with spinal dysraphism and as a complication of diagnostic spinal puncture.[9]

Rao reported that although spinal intramedullary tuberculomas occur at any age and in either sex, the greatest incidence is found in young nulliparous women, attributed to reactivation of previous tuberculosis by pregnancy.[15]

Location of these lesions are distributed evenly in the cervical, dorsal and lumbar portions of the spinal cord.[16] However, 72% of those in the review of MacDonell, et al were located in the thoracic spinal cord.[12]

Spinal intramedullary tuberculomas are usually associated with intracranial involvement.[16] In our cases no intracranial tuberculomas were detected. Though multiple tuberculomas are seen in 10-33% of cases of intracranial tuberculomas, multiple lesions of spinal cord are very uncommon and sparsely reported in the literature.[16] Gupta, et al reported multiple intramedullary lesions with skip areas in seven of their patients.[6] Lin, et al has also reported a case of multiple areas of spinal involvement by tuberculomas in a patient on treatment for tuberculous meningitis.[11] Clinically, patients with spinal intramedullary tuberculomas present with signs and symptoms depending on the location of tuberculoma in the spinal cord.[12] Earlier, the role of imaging in spinal intramedullary tuberculoma was restricted to plain films and myelography, which were not much assistance, being able to show only cord swelling.[17] However, this has undergone radical change with the advent of MRI. The role of MRI in the diseases of spine and cord is well documented, as it helps in delineation and better characterisation of these lesions.[6-8,18]

Rhoton, et al published the first MRI documented description of spinal intramedullary tuberculoma, in 1988, and authors of subsequent studies were better able to characterise these lesions as the technology became more advanced.[7,16] In serial MRI scans of all types of tuberculous spinal cord lesions, the initial images reveal a homogeneously enhanced site that is suggestive of myelitis or early stage tuberculoma, and this irregularity gradually transforms into a ring-shaped lesion with a hypointense centre.[8,16]

Spinal intramedullary tuberculomas have been described as low intensity lesions with or without central hyperintensity (because of varying amount of caseous necrosis) on T2-weighted image and as hypo to isointense lesions on T1-weighted imaging.[8,16] Other series describe iso to hyperintensity on T1-weighed imaging, at the site of the tuberculoma with peripheral hypointensity due to oedema/myelomalacia.[7] The peculiar hypointensity or isointensity seen on T2-weighted imaging in the tuberculomas may reflect restricted mobile protons within high protein content in organized caseation, cellular and collagenous layers, the presence of heterogeneously distributed free radicals produced

by macrophages during active phagocytosis and/or highly immobile saturated fatty acids.[10] Loose liquified caseation in the centre may produce central hyperintensity on T2-weighted imaging.[7] Intense peripheral enhancement may be explained by prominent vascularity seen on microscopy, as the MR findings in our case.[7]

Mainstay of treatment is medical, unless otherwise indicated, surgery can be differed or avoided.[18] Gupta, et al reported 10 cases of intraspinal tuberculosis in which follow-up MRI scans confirmed response to medical treatment.[5] Rao also reported four patients with intramedullary tuberculoma who were medically managed with success.[15] Similar result was reported by Sharma, et al in a series of 10 cases of spinal intramedullary tuberculoma.[18] In contrast, Citow and Ammirati reported one case of spinal intramedullary tuberculoma that progressed despite aggressive antituberculosis treatment and necessitated surgical intervention.[1] Although medical therapy has proven effective, the intramedullary type of intraspinal tuberculoma is a firm and well-circumscribed lesion, and these features allow surgical removal without undue trauma to surrounding functional cord.[17] In addition, it is possible that enlargement of the lesion and progression of caseating necrosis will produce more neurological deficits while the patient is undergoing medical treatment.[1,3] This finding is echoed by the intraoperative appearance of the mass: no clear boundary or plane was observed in cases described by Gupta, et al.[15] This is in contrast to the well-circumscribed, firm masses described by Rhoton, et al and Lin, et al.[11,16]

Rao mentioned that patients harbouring any lesion suggestive of spinal intramedullary tuberculosis are treated medically and undergo regular follow-up examination.[15] Surgical intervention is considered only in patients with: 1) gross neurological deficits; 2) poor response to medical treatment; 3) deterioration in neurological status during treatment; and 4) any paradoxical enlargement of the lesion observed on follow-up MR imaging.[15]

Conclusion

Spinal intramedullary tuberculoma is a rare condition, needing proper evaluation and imaging. The treatment, by and large, is drug therapy. In patients with rapid deterioration mass should be removed without delay to eliminate any mass effect as quickly as possible. An extended course of antituberculous treatment should be prescribed immediately after the lesion is removed. Multidrug resistance (MDR) to TB must also be considered in those not responding to 3 or 4 first-line drugs.

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