

Case Report

Multiple Tuberculoma Involving the Brain and Spinal Cord in a Patient with Miliary Pulmonary Tuberculosis

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Although tuberculosis of the central nervous system is well known, the incidence of intramedullary tuberculomas is low and a combination of intramedullary with intracranial tuberculomas is extremely rare. We report a case of disseminated tuberculoma involving brain and spine with miliary pulmonary tuberculosis in a 66-year-old woman initially presenting with fever, general weakness, back pain and motor weakness of both lower extremities. Despite medical therapy, she developed progressive motor weakness of both lower extremities with muscle strength 1/5 in both lower extremities. Urgent surgical intervention was followed and her muscle power and motor functions were improved gradually. The anti-tuberculous drugs were continued and the follow-up magnetic resonance imaging (MRI) of brain and spine showed that the lesions had become smaller or disappeared.

KEY WORDS : Tuberculoma · Spine · Brain · Magnetic resonance imaging.

INTRODUCTION

Although tuberculosis remains a major health problem in developing countries, tuberculoma involving the central nervous system is still uncommon compared with the involvement of other systems²³. Intramedullary tuberculomas are rare and constitute only 0.2 to 0.5% of all central nervous system (CNS) tuberculomas²¹. Among patients with spinal tuberculosis, 55% present with vertebral body involvement, 39% with intraspinal granulomatous lesions without bone involvement, and only 7% with intramedullary lesions¹³. The combination of intramedullary and intracranial tuberculomas is extremely rare and only five cases have been reported in the literature^{6,13,19,21,23}.

Most cases of intramedullary tuberculomas are subacute with progressive spinal cord compression symptoms. Given the rarity of spinal intramedullary tuberculoma, there is no standardized treatment protocol for this condition. We report a rare case of disseminated tuberculomas involving the spinal cord and brain, and discuss the role of surgery in the treatment of these rare lesions.

CASE REPORT

A 66-year-old woman was admitted with fever, general weakness, back pain and motor weakness of both lower extremities for two weeks. Examination at this time revealed spastic paraparesis with muscle strength of 3/5 in both lower extremities, impaired sensations below the L1 level, and exaggerated deep tendon reflexes. Plain radiographs showed narrowing of the L1-L2 space with irregularity and osteolysis of the endplates. The MRI of lumbar spine revealed hyperintense lesions in marrow and disc of L1-L2 in T2-weighted images with spinal cord compression, and a hyperintense lesion expanding the cord from T10 to T11 in T2-weighted images and the lesion showed prominent ring enhancement with contrast medium (Fig. 1). Also, the MRI of cervical spine showed small enhancing nodules in C2-C3 level (Fig. 2). The patient showed negative for HIV, but her chest computerized Tomography (CT) revealed randomly distributed miliary nodules in both lung fields, which was highly suggestive of miliary pulmonary tuberculosis. The tuberculin test was positive and the MRI of the brain revealed multiple enhancing nodules in deep white matter and subcortical area (Fig. 3). She was treated with rifampicin 600 mg, isoniazid 300 mg, pyridoxamine 1500 mg, ethambutol 1200 mg and pyridoxine 25 mg. Despite medical therapy, she developed progressive motor weakness of both lower

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extremities with decrease of anal sphincter tone and aggravated urinary incontinence. The neurologic examination revealed spastic paraplegia with muscle strength 1/5 in both lower extremities, with impaired sensations below L1 level and exaggerated reflexes. As showing progressive neurologic deficit in spite of adequate medical management, emergent operation was planned. Total laminectomy of L1-L2 and subtotal laminectomy of T10-T11 was done, and myelotomies were followed at T10 and T11 using the operating microscope. On examination, the intramedullary lesions were firm and well-circumscribed with a readily definable plane. The gross pathologies of the three fragments of fibrous tissue were 0.2 cm, 0.3 cm, 0.5 cm diameter encapsulated, gray, firm, rubber-like masses. The histologic examination of the tumor specimen revealed diffuse chronic inflammatory cell infiltration with granulation tissue and focal abscess formation (Fig. 4). Results of a polymerase chain reaction for mycobacterial DNA sequences (TB-TPCR) test applied to the specimens were positive (Fig. 5).

Under the diagnosis of multiple central nervous system (CNS) tuberculomas (spinal cord and brain) with miliary pulmonary tuberculosis, the anti-tuberculous drugs were continued. The patient has been well after surgery, with no new neurologic worsening. At a 5-month follow-up visit after surgery, the muscle power in the lower limbs had improved to grade 4/5, but bladder dysfunction persisted. The follow-up MRI of brain and spine showed the lesions had become smaller or disappeared (Fig. 6).

DISCUSSION

Spinal intramedullary tuberculomas are rare lesions even in areas where tuberculosis is endemic³⁾. Intramedullary tuberculomas are rare and constitute only 0.2 to 5% of all CNS

tuberculomas²⁰⁾. Arseni and Samtica found only five intramedullary tuberculomas in their series of 210 cases of CNS tuberculosis²⁾. Concurrent intramedullary and intracranial tuberculomas are very rare and only 5 cases has been

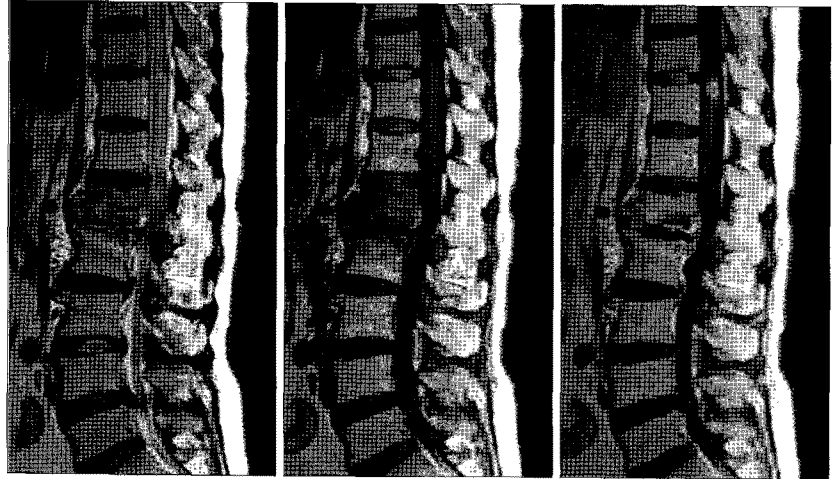


Fig. 1. The magnetic resonance image of thoracolumbar spine revealed hyperintense lesions in marrow and disc of L1-L2 in T2-weighted images with spinal cord compression, and an hyperintense lesion expanding the cord from T10 to T11 in T2-weighted images with prominent ring enhancement with contrast medium.

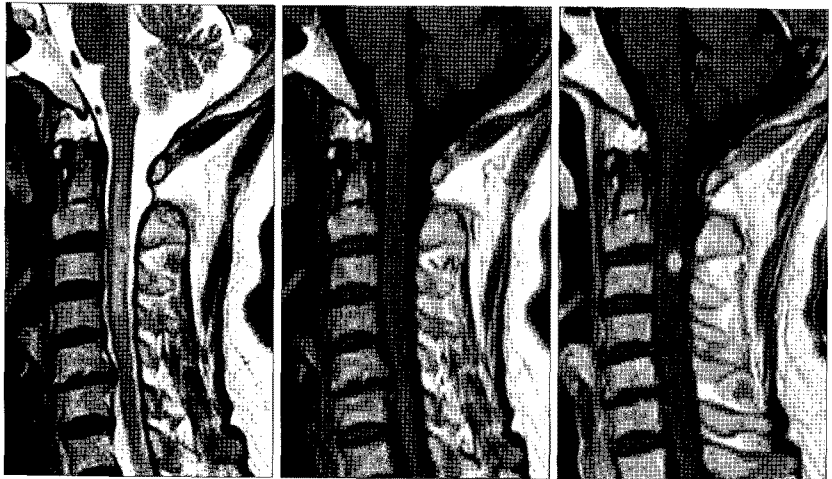


Fig. 2. The magnetic resonance image of cervical spine showing a small enhancing nodule in C2-C3 level.

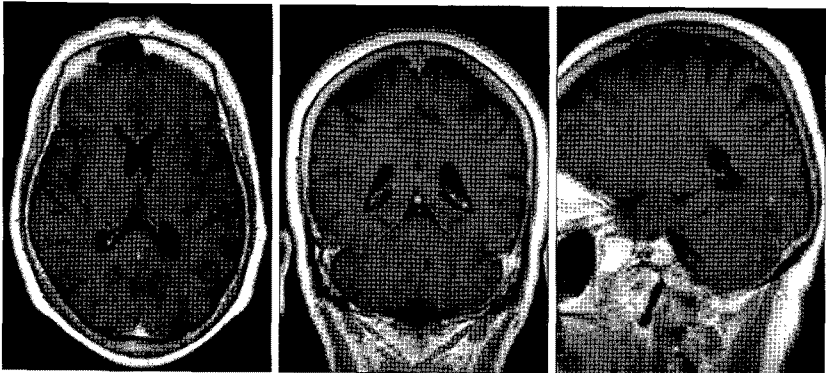


Fig. 3. The magnetic resonance image of the brain showing multiple enhancing nodules in deep white matter and subcortical area.

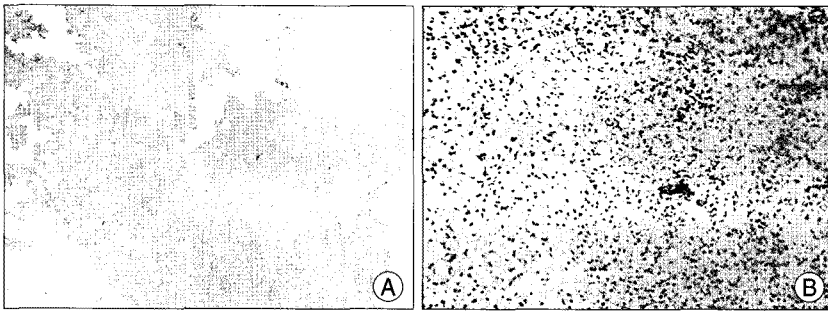


Fig. 4. The histologic examination of the tumor specimen revealing diffuse chronic inflammatory cell infiltration with granulation tissue and focal abscess formation (hematoxylin-eosin, original magnification, A : $\times 40$, B : $\times 200$).

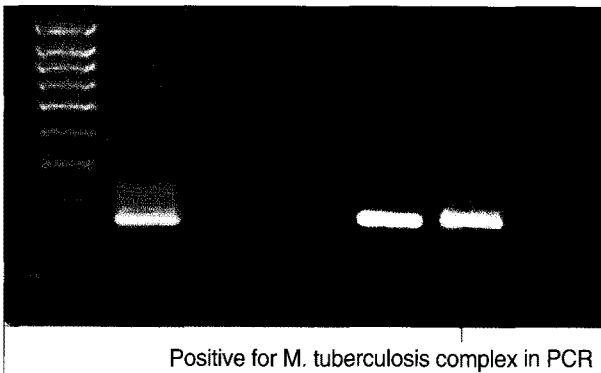


Fig. 5. Results of a polymerase chain reaction for mycobacterial DNA sequences (TB-PCR) test applied to the specimens are shown to be positive.

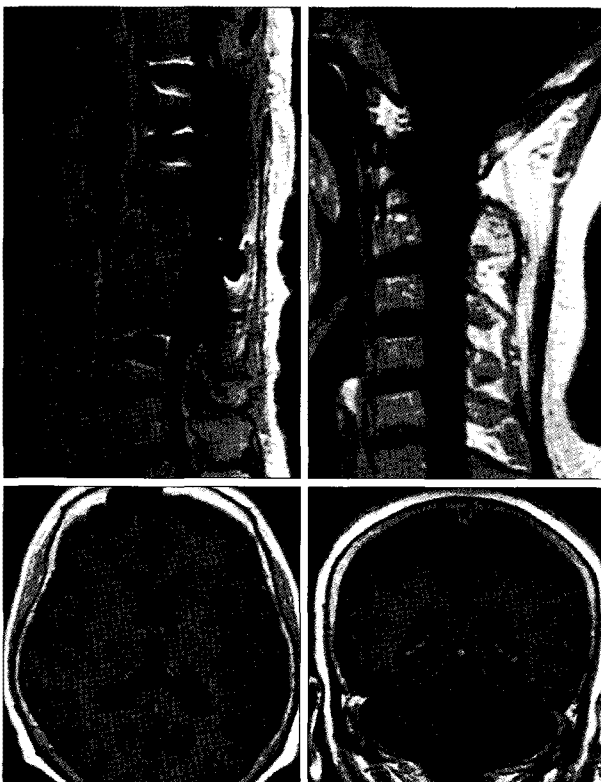


Fig. 6. The follow-up magnetic resonance image of spine and brain demonstrating the lesions had become smaller or disappeared.

reported so far^{6,13,19,21,23}. Thacker and Puri²¹ reported a 6-year-old girl who presented with progressive paraparesis in whom imaging revealed intramedullary tuberculoma with incidentally discovered multiple intracranial tuberculoma. Yen et al.²³ re-ported a 67-year-old man with known pulmonary tuberculosis who developed symptoms of spinal cord compression. Imaging revealed an intramedullary tuberculoma along

with multiple intracranial tuberculomas. In both cases, the intracranial lesions were incidentally discovered, as in our patient. However, in the cases reported by Huang et al.⁶ Shen et al.¹⁹ and Muthukumar et al.¹³, the patients were symptomatic for both intracranial and intramedullary lesions. Most of the reported intramedullary spinal tuberculomas are solitary. However, multiple intramedullary spinal tuberculomas have been increasingly reported after the use of MRI was introduced^{7,8,10,11,15,16,19}. We suggest that MRI of the brain should be performed in the case of multiple intramedullary spinal tuberculomas because of the possible presence of early asymptomatic intracranial tuberculomas.

The imaging modality of choice for diagnosis of these lesions is MRI. The MRI has revolutionized the imaging of tuberculomas and the diagnosis can be made with reasonable certainty, avoiding the need for an invasive procedure. Intracranial tuberculomas have been described as low-intensity lesions with or without central hyperintensity (because of varying amount of caseous necrosis) on T2-weighted images and as hypo- to isointense lesions on T1-weighted images. The varying appearance of intracranial tuberculoma is attributed to the evolving nature of the granulomatous lesion. During the stage of chronic granulomatous inflammation, there is homogenous enhancement due to breakdown of the blood-cord barrier. Subsequently, collagen gets deposited along the capsule of the lesion, and the contents get transformed into caseous material. During this stage, the lesion presents with ring enhancement⁹. Intense peripheral enhancement may be explained by prominent vascularity seen on microscopy.

The differential diagnosis of intramedullary tuberculomas includes granulomas such as tuberculomas and cysticercal granulomas, and neoplastic lesions such as astrocytoma, metastasis or lymphoma. In this case, the clinical picture and the size of the lesion combined with the classical ring enhancement and surrounding edema was thought to be typical of a tuberculous granuloma. The resolution of the pathological changes in the brain as well as the spinal cord

as seen on the MRI after the institution of anti-tuberculous treatment confirmed our diagnosis.

The ideal treatment of intramedullary tuberculoma remains controversial. Both surgical and medical treatment have yielded good results in different series^{1,3-5,9,12,17,22,23}. Many authors have recommended medical treatment of intramedullary tuberculomas with good results^{4,5,14,18}. The purpose of surgical intervention is to decompress the cord when progressing neurologic defects occur and also makes it possible to examine the tissue pathologically. The important feature of our patient is progressive weakness of both lower extremities, with decrease of anal tone and aggravated urinary incontinence during effective medical treatment. In our case, we excised the intramedullary spinal tuberculomas and administered antituberculous treatment after surgery. The patient's neurologic symptoms improved, and the MRI follow-up showed complete disappearance of the intracranial and intramedullary tuberculomas. Our experience, although limited, has shown that neurological improvement is likely to occur when surgical intervention is undertaken before irreversible cord damage takes place. We believe that, even if the imaging features of intramedullary tuberculoma are characteristic and even if there is evidence of tuberculosis elsewhere in the body, the decision to undertake surgery should be based on the neurological status of the patient and not on the imaging features alone. Patients who present with profound neurological deficits should undergo early surgical decompression even if the treating physician is reasonably sure about the diagnosis of intramedullary tuberculoma. Undue delay entailed by prolonged medical therapy might lead to irreversible cord damage, and the patient might not improve neurologically even if the lesion disappears radiologically. Muthukumar et al.¹³ reported 2 patients who presented with intramedullary tuberculomas. In his report, one patient who had paraplegia preoperatively did not improve after late surgery. If early surgery had been undertaken in his patient, the chances of neurological improvement would have been better. We believe that surgical intervention should be considered for cases showing progressive deficits in spite of adequate medical management. Prospective studies are required to clarify the role of surgery and medical therapy in the treatment of these rare lesions.

CONCLUSION

We report a rare case of disseminated tuberculomas involving the spinal cord and brain. Medical treatment is generally advocated as the initial therapy for intramedullary

tuberculoma, and surgery is reserved for medical failure. However, this case suggests that urgent surgical intervention should be considered for cases showing progressive neurologic deficits.

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