CASE REPORT



A rare case of intramedullary tuberculoma: Complete resolution after medical treatment and role of magnetic resonance imaging in diagnosis and follow-up

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ABSTRACT

Intramedullary tuberculoma (IMT) is a rare form of spinal tuberculosis. IMT has an incidence of 2 cases per 2000 cases of central nervous system tuberculosis. Magnetic resonance imaging (MRI) can be helpful for diagnosing IMT at an early stage and it is also very useful in follow-up. Three stages of IMT have been described in MRI based on the evolution of the lesion. Medical therapy is the mainstay of treatment for IMT though there are surgical options for select patients. Here, we describe a patient with dorsal IMT who improved clinically as well as radiologically with antituberculous treatment and steroids.

Key words: Complete resolution, intramedullary tuberculoma, magnetic resonance imaging, medical treatment

Introduction

Tuberculosis (TB) of the central nervous system (CNS) is rare, with an incidence of 0.5-2% in patients with systemic TB.^[1] Spinal TB presents commonly as tuberculous spondylitis or arachnoiditis. Intramedullary spinal tuberculomas (IMTs) are rare, even in geographic areas where TB is endemic. The first report of IMT was given by Albercrombie in 1828.^[1] In 1960, Lin et al. reviewed literature of IMT and accounted for 104 cases, of which majority were diagnosed post-mortem.^[1] In 1999, Ratliff et al. reviewed a total of 148 cases of IMT in literature.^[2] In India, Dastur reported 74 cases of paraplegia secondary to TB without evidence of Pott's spine.^[3] In this series, IMT accounted only for 8% of cases. Spinal IMTs are extremely rare, seen in only 2 of 100,000 cases of TB and 2 of 1000 cases of CNS TB. IMT is very uncommon and till now roughly 150 cases have been reported. From India, Sharma and Gupta have reported 10 and 8 cases of IMT, respectively, and they also discussed

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the treatment aspects of IMT.^[4,5] The first magnetic resonance imaging (MRI) documented description of tuberculoma was given by Rhoton *et al.* in 1988.^[6] Based on the evolution of IMT, three stages of IMT were described in MRI.^[7] Treatment for IMT could be medical, surgical, or both. Here, we report a rare case of IMT, discuss the evolution of lesions on serial MRIs, and document complete neurological recovery and radiological resolution following medical therapy.

Case Report

A 23-year-old male, a software engineer by profession, presented with complaints of weakness of both lower limbs of 5 days duration, which initially started in left lower limb and subsequently involved the right lower limb. He also complained of loss of dexterity involving both upper limbs since 2 days. Along with this complaint, he also has a band-like feeling in the upper abdomen, urinary hesitancy, and a feeling of incomplete voiding of urine. On examination, his higher mental functions and cranial nerves were normal. All his reflexes were brisk and there was ill-sustained clonus on the right side. His upper limb power proximally was 5, while distally it was 4. His lower limb power was 3 and it followed a pyramidal distribution of weakness. He had sensory impairment below T7 corresponding to vertebral level D5 and altered sensation below C6. General physical examination and other system examinations were normal. A provisional diagnosis of cervical myelopathy was made and patient was investigated. His complete blood count, renal profile, liver function tests, human immunodeficiency virus (HIV), Venereal Disease Research Laboratory test, and hepatitis B surface antigen were negative. His chest X-ray



showed right apical TB. Erythrocyte sedimentation rate was high and Mantoux was reactive. MRI whole cord revealed an iso- to hypointense lesion at D5 level and a swollen cord on T1-weighted imaging (T1WI). The lesion was hypointense on T2-weighted imaging (T2WI), with gross edema appearing hyperintense, extending up to the cervical and lower dorsal level. Contrast-enhanced MRI showed a ring-enhancing lesion at D5 with sharp margins. The oval-shaped lesion measured $1.44 \text{ cm} \times 0.91 \text{ cm}$. Cerebrospinal fluid (CSF) analysis showed increased cells with lymphocyte predominance and elevated protein. CSF polymerase chain reaction for mycobacterium TB was positive. A diagnosis of IMT and pulmonary TB was made and patient was started on antituberculous treatment (ATT) with steroids. His upper limb weakness could be attributed to the surrounding inflammatory edema which had extended till cervical and lower dorsal level. Over a period of 2 weeks, the patient improved very well and was able to walk without support. His upper limb weakness improved. Over a period of 3 months, his band-like feeling at upper dorsal level gradually improved and his lower limb power fully improved except for mild hesitancy while passing urine.

MRI was repeated at 2nd and 6th months after starting ATT. Steroids were given for duration of 2 months. His second MRI showed significant decrease in the size of the lesion and his third MRI showed almost total disappearance of the lesion except for an insignificant edema at D5 level.

Discussion and Conclusion

Out of the estimated global annual incidence of 9.4 million TB cases, 1.98 million cases were found to have occurred in India, of which 0.87 million were infectious cases, thus catering to a fifth of the global burden of TB.^[8] TB is one of the earliest opportunistic diseases to develop amongst persons infected with HIV. HIV infection is the most powerful risk factor for the progression of TB infection to TB disease. World Health Organization estimated the prevalence of 6.7% of HIV (5.5-7.9%) in TB patients in India in 2008.^[8] CNS TB accounts for approximately 0.5-2% of all cases of TB, carries a high mortality and neurological morbidity, and disproportionately afflicts children and HIV-infected individuals. The exact burden of CNS TB in India is unknown. Spinal TB is very rare and the common forms of spinal TB are tuberculous spondylitis, TB myelitis, and intraspinal TB. Intraspinal TB could be spinal meningitis, arachnoiditis, IMT, and abscess. IMT is very uncommon and till now roughly 150 cases have been reported. Tuberculomas develop following hematogenous dissemination of bacilli from an infection elsewhere in the body, usually lung. Our patient had right apical active TB [Figure 1]. Advent of MRI has made diagnosis of IMT more accurate and earlier. In the early phase, the tuberculoma is characterized by severe inflammatory reaction which causes severe edema. At this stage, the gel capsule is not well formed. During this stage, the enhancement after contrast examination is uniform.



Figure 1: T1 contrast coronal image showing the ring-enhancing lesion and its dimensions, and the arrows at the right side showing apical tuberculosis

T1WI and T2WI both show equal signal intensity. As the gel content in the tuberculoma increases, the peripheral edema begins to disappear. As a result, T1WI shows isointense lesions while T2WI shows low or isointense lesions. Contrast MRI shows central hypointensity with rim enhancement. With the development of caseation, T2WI shows a typical "target sign," which means that it exhibits a range from the low signal target to the high signal rim and also from the center of the low signal rim to the peripheral parts. The caseous substance appears hyperintense at the center, which gives the characteristic target sign. The low signal rim in the external region is composed of collagen fibers produced by fibroblasts. The target sign is a valuable indicator that helps differentiate spinal tuberculoma from other intramedullary lesions. Rim enhancement and presence of sharp margins also differentiates IMT from intramedullary tumors.^[4,7,9,10] In our case, the imaging done at the time of presentation corresponds to the early stage with uniform enhancement and gross edema [Figures 2 and 3]. MRI taken 2 months later corresponds to the intermediate stage with decrease in size of lesion and the gross disappearance of edema [Figures 4 and 5]. MRI taken 6 months later shows complete resolution of the lesion [Figures 6 and 7]. The characteristic target sign was not seen, but sharp margins, ring enhancement, presence of right apical TB, raised protein, CSF polymerase chain reaction (PCR) for mycobacterium TB positivity, and response to ATT confirmed the diagnosis of IMT. Treatment options for IMT could be medical, surgical, or both. Gross neurological deficits, worsening of neurological status despite treatment, and paradoxical enlargement of lesion during treatment are indications of surgery. Otherwise, the mainstay of treatment is medical.[11]

Our patient was put on isoniazid (INH) 300 mg/day, rifampicin (RF) 450 mg/day, pyrazinamide 1500 mg/day, and ethambutol 800 mg/day daily for 2 months, followed by INH and RF for 4 months. Pyridoxine at 40 mg/day was given for



Figure 2: T2-weighted sagittal image showing the lesion at D5 level, indicated by central arrow, and the extent of cord edema is delineated by the top and lower arrow heads



Figure 3: T1 contrast sagittal image showing the ring-enhancing lesion at D5 level



Figure 4: T2 sagittal image taken 2 months later showing decrease in the size (arrow marked) of the lesion as well as gross reduction of cord edema



Figure 6: T2-weighted image taken 6 months later showing complete disappearance of the lesion

all 6 months. Prednisone at 1 mg/kg body weight was given for 1 month and then subsequently tapered over a period



Figure 5: T1 contrast image showing decrease in size of the lesion when compared to the images taken 2 months ago



Figure 7: T1 contrast image showing no abnormal enhancement in the cord, indicating complete resolution of the lesion

of 1 month. The ring-enhancing lesion decreased in size at 2 months and completely resolved after completion of ATT. His

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right apical lung lesions also had regressed. This case of IMT is being presented for its rarity and to highlight the radiological features of IMT and its complete clearance with medical therapy.

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