A Hossini, W Ammouri, K Quenum, N Fatemi, N Raouzi, A Saïdi, M Jiddane, F Bellakhdar

Citation

Abstract
Intradural extramedullary tuberculoma of the spinal cord is a very rare presentation of non osseous spinal tuberculosis. We reported on, a case of a 22 year-old female who was treated for pulmonary tuberculosis since five months. The patient developed spastic paraplegia, which progressed rapidly, 20 days before admission. MRI of spine revealed an intradural extramedullary lesion at the T3-T4 and T7-T12 levels. After surgery, the histopathology of the lesion confirmed the diagnosis of tuberculosis.

INTRODUCTION
In endemic countries, tuberculomas of the spinal cord without disco vertebral lesions are rare, contrary to the spondylodiscitis tuberculosis. However, intradural extramedullary tuberculoma is a very rare presentation of non osseous spinal tuberculosis.

Here, we reported a case of intradural extramedullary tuberculoma at T3-T4 and T7-T12 levels revealed by spastic paraplegia, which progressed rapidly, in a patient who was treated since five months for pulmonary tuberculosis.

CASE REPORT
A 22-year old Moroccan woman was admitted in our department with progressive paraplegia and urinary incontinence. The patient had a history of miliary tuberculosis disease and was actively treated since 5 months (initially four drug therapy that included, streptomycin, isoniazid, rifampin, and pyrazinamide during two months and then isoniazid and rifampin). The patient had a perfect adherence and tolerance to therapy, becoming afebrile after two weeks.

At the fifth months of the treatment, the patient was suffering for dorsal and lumbar pain, weight loss, anorexia, constipation, urinary incontinence, improved of her general condition and a lower extremity weakness 20 days before her admission.

Physical examination revealed a body temperature at 37°, normal consciousness, and spastic paraplegia with hypoesthesia below T4 dermatome. Deep tendon reflexes of lower extremities were lively and the Babinski reflex was bilaterally positive. Laboratory findings were as follows: erythrocyte sedimentation rate 115 mm/h, C –reactive protein 48 mg/dl, white blood cell count 13400/mm³, and lymphocytes 1200/mm³. HIV testing was negative. The examination of lumbar cerebrospinal fluid was normal and there was no tubercle bacillus detected.

Chest and spine radiography were normal. MRI of spine revealed two intradural extramedullary lesions at the T3-T4 and T7-T12 levels. The lesions are isointense in the T1 and T2-weighted image with an annular and multilocular enhancement after Gadolinium injection (figures 1 and 2). The diagnosis of intradural extramedullary tuberculoma was suspected on the base of the history, of pulmonary tuberculosis and the results of the imaging.

Figure 1
Figure 1: Sagittal MRI on T1 (a), T2 (b) and T1 post contrast (c) weighted images, shows two posteriors intradural extramedullary lesions on T3-T4 and T7-T12 levels. The lesions are isointense to the marrow on T1 and T2 weighted images, with an intensif peripheral Gadolinium enhancement.

Figure 2
Figure 2: Axial post contrast MRI on T1 weighted image show a posterior intradural extramedullary lesions which driving back in front of the marrow. The lesions contain many compartment of pus.

Laminectomy at T3-T4 and T7-T12 vertebrae levels was performed. At the opening of the dura mater we observed a yellow-greyish colored intradural extramedullary process. This process had a tough consistency and was avascular without plan of cleavage. The lesions were gently removed from the dura and a partially excision was made. Histologic examination confirmed the diagnosis of tuberculoma. Also, theses intradural granulomas contained tubercle bacillus at direct exam and in the culture confirming the diagnosis of tuberculosis. After the operation, the patient continued the anti-tuberculous medication (isoniazid 300 mg, rifampin 600 mg, pyrazinamide 1500 mg, ethambutol 1200 mg). During a follow-up of 12 months, the patient experienced a partial improvement in motor strength.

DISCUSSION

Neuraxis is affected uncommonly by tuberculosis as compared to other organ systems. Intradural extramedullary tuberculoma is a very rare presentation of non osseous spinal tuberculosis and the first case was described by Bucy and Oberhill in 1950. Dastur’s review, 64% of tuberculomas are extradural, 8% are intramedullary and only 1% are intradural extramedullary. The rest of the lesions involve the arachnoid without dural involvement. This lesion has been described in a predominantly young population of both genders. An advanced HIV infection was present in a minority of the cases reported in the literature, with no other immunodeficiencies.

Parsons and Pallis have classified intradural extramedullary tuberculoma in two groups: the first group includes rounded or oval and tough lesions, measuring 2 to 3 cm in diameter. Theses lesions are limited, with or without arachnoiditis and tied to dura mater with a net plane of cleavage to the spinal allowing a mass excision; the second group (the case of our patient) includes diffuses lesions with greyish granulomas mass. Theses group of lesion are avascular and are accompanied by arachnoiditis. Complete excision is impossible because theses lesions have a cylindrical disposition around the spinal and a diffuse vertical extension.

Intramedullary tuberculomas are usually associated with pulmonary infection and the dissemination is done by blood way. In our case, the patient had not tuberculous meningitis. Whereas, in the literature, intradural extramedullary tuberculoma occurred usually weeks after tuberculosis meningitis, while patients were taking effective antituberculous medications. These suggest that, despite an adequate response to antituberculous treatment, an ongoing inflammatory process takes place in the arachnoid, which leads to the development of a tuberculoma. This complication might be a modality of paradoxical reaction to antituberculous medication. Such reaction consists of an exacerbation in symptoms, signs, and laboratory or radiographic manifestations of tuberculosis, probably related to an immune response to antigens released as bacilli are killed by effective chemotherapy with produce of excessive tubercular protein. This protein stimulates the antigen-reactive lymphocytes in the tuberculoma to proliferate, resulting in the chain of events that is responsible for expansion of tuberculoma or visualizations of previously unapparent tuberculomas.

The lesion of intradural extramedullary tuberculoma is often single, dual localisation (like our case) is rarely reported.
Patients with intradural extramedullary tuberculoma presented with progressive spinal compression over weeks to months, with paraparesis, hypoesthesia with a sensory level, and bladder dysfunction, rarely patients present an acute myelitis. 

History of past or concurrent tuberculosis in patients with general symptoms (fever, asthenia, night sweat and the alteration of the general condition) and the presence of a second tuberculous localisation can help for the diagnosis of intradural extramedullary tuberculoma. But the absence of these conditions doesn’t rule out the possibility of tuberculoma.

Histologic examination of intradural extramedullary tuberculoma revealed epithelioid granuloma with caseous necrosis. Within the granuloma there is little or no mycobacterium tuberculosis. 

MRI is nowadays the diagnostic method of choice. It should be done to rule out any intradural or the more common vertebral involvement by tuberculosis. Typically a subdural longitudinal collection is observed at any level of the spine, which posteriorly pushes the spinal cord, and strongly enhances after gadolinium injection. The presence of annular lesion at MRI points to an infectious origin. Also, MRI can diagnosis the association with arachnoiditis, guides the surgery to decompression of the spinal cord and is necessary to control the evolution of the collection under antituberculous medications.

Treatment must be rapid and effective to prevent neurological sequelae. Early surgery is necessary to the decompression of the spinal cord and to obtain the histologic or bacteriologic confirmation of tuberculosis. Contrary to limited extramedullary tuberculoma, complete excision of extended tuberculoma is impossible, because it was closely adhered to the cord, leaving no clear plane of cleavage with it.

Antituberculous medications should be administered after surgery to prevent tuberculous meningitis. The use of corticosteroids is also reasonable and generally recommended. Such medication has been used in paradoxical reactions to antituberculous medication, to reduce interstitial oedema and perilesional granulomatous vasculitis.

Despite an early management of patient with intradural extramedullary tuberculoma, the prognosis still unfavourable. In a serie of 22 cases reported by Roca et al, 77% of patients remained with persistent walking difficulty and the prognosis was not significantly influenced by age, gender, time elapsed since beginning of symptoms to diagnosis of intradural extramedullary tuberculoma, surgical treatment, intensive antituberculous treatment, use of corticosteroids and the presence of plane of cleavage (p>0,05).

CONCLUSION

Tuberculosis of central nervous system is common in endemic countries and is dominated by pott’s disease. However, intradural extramedullary tuberculoma is infrequent, it should be suspected in any patient with symptoms of back pain and focal neurological deficits with a history of tuberculosis, and early imaging by MRI should be done to diagnosis of the collection and to control there evolution after surgery and antituberculous medications.

References


Author Information
Adil Hossini
Neurosurgery department, University Ibn Sina hospital

Wafa Ammouri
Internal medicine department, University Ibn Sina hospital

Kisito Quenum
Neurosurgery department, University Ibn Sina hospital

Nizar Fatemi
Neurosurgery department, University Ibn Sina hospital

Nabil Raouzi
Neurosurgery department, University Ibn Sina hospital

Amar Saïdi
Centre of anatomy pathology

Mohamed Jiddane
Neuroradiology department, University Ibn Sina hospital

Fouad Bellakhdar
Neurosurgery department, University Ibn Sina hospital