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Case Report

Intramedullary tuberculoma of the conus medullaris: case report and review of the literature

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Objective: To illustrate the dilemmas in the diagnosis and management of intramedullary tuberculomas of the spinal cord.
Methods: Case report of a 32 year-old man with tuberculous meningitis. The presence of unexplained urinary retention and progressive weakness in the legs led to the discovery of an additional tuberculoma of the conus medullaris.
Setting: Dicle University Diyarbakir, Turkey.
Results: The patient was on a 1-year course of isoniazid, pyrazinamide and rifampicin, and responded well to conservative treatment. Our patient's unique features were represented by the worsening of neurological symptoms while being treated with adequate anti-tuberculous medication.
Conclusion: We present a case of intramedullary tuberculoma of the conus medullaris to illustrate the dilemmas in the diagnosis and management of this curable disease, and review of the literature to date.
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Keywords: tuberculosis; conus tuberculoma; urinary retention; surgical therapy; magnetic resonance imaging (MRI)

Introduction

It is generally assumed that the incidence of neurotuberculosis is related to the prevalence of tuberculosis in the community. Tuberculosis is still a serious problem in our country. The incidence of tuberculosis is 43–46 in 100 000 inhabitants and it presents in a variety of clinical pictures.[1] Tuberculous meningitis is reported as the most common type of neurotuberculosis.[2] Tuberculoma of the spinal cord is extremely rare, being present in approximately two of every 1000 cases of central nervous system (CNS) tuberculosis.[3] The ratio of cranial to spinal tuberculous is approximately 1 : 42.[4] With the declining incidence of tuberculosis, it is to be expected that the neurological manifestations will become more rare. However, the disease is still endemic in developing countries. The potential for excellent recovery with adequate treatment is a stimulus for continuing awareness of the condition.

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We report a tuberculoma of the conus medullaris in a patient with tuberculous meningitis and aim to illustrate the dilemmas in the diagnosis and management of this potentially curable disease by reviewing the literature to date.

Case report

A 32 year-old man was transferred to our hospital from a peripheral hospital where he was admitted with 10 days history of fever, headache, lethargy, night sweats, and a persistent productive cough with haemoptysis. Physical examination showed low-grade fever, and diffuse rhonchi. The laboratory studies revealed haemoglobin (Hgb) of 10.7 g/dl, 10 000 mm^3 white blood cells (WBC), and 47 mm/h erythrocyte sedimentation rate (ESR). In the cerebro spinal fluid (CSF) analysis at initial admission, WBC was 420/mm^3 with 90% lymphocytes protein level was 76 mg/dl, glucose was 2.1 mmol/l and sputum was positive for acid fast bacilli. The chest X-ray demonstrated apical cavitations and diffuse interstitial infiltration. The patient was diagnosed as pulmonary tuberculous

infection, and therapy with rifampicin, isoniazid, and pyrazinamide was started. Because of our lack of laboratory facilities, in our case mycobacterium tuberculosis was not tested for sensitivity to anti-tuberculous agents.

His complaints resolved over 9 weeks and he was discharged on the same drug regime. Unfortunately, relapses occurred twice due to poor compliance with therapy.

Six months later, the patient was admitted to our department suffering from urinary incontinence, lower limb pain and progressive weakness in both legs and impotence. He was not able to stand and both plantar responses were flexor. Joint position and vibration sense were totally absent. Laboratory tests were Hgb 11.2 g/dl, ESR 60 mm/h, WBC 10 000 mm^3 (90% lymphocytes). CSF analysis was as follows: WBC 285 mm^3 with 85% lymphocytes, protein level 190 mg/dl, glucose 4.2 mmol/dl (serum glucose 7.3 mmol/dl), and negative for acid fast bacilli. Chest and thoracolumbar spinal X-rays were normal. Cranial CT was normal except for mild ventricular dilatation. Spinal MRI showed a diffuse nodular appearance in conus medullaris after intravenous gadolinium (Figures 1, 2 and 3).

The lesion was assumed to be intramedullary conus tuberculoma. The patient was treated with high dose steroids and triple drug chemotherapy. The patient responded well to conservative treatment and discharged at the end of 2 weeks. Two months after the second admission, the neurological status of the patient gradually deteriorated and presented with urinary incontinence, severe right lower limb and moderate left limb weakness. Sensory testing showed loss of pain, temperature and light touch in knees distally in a stocking fashion.

Our patient's unique features were represented by the worsening of his neurological symptoms while being treated with adequate anti-tuberculous medication. Further investigation showed a tuberculoma and surgery was offered.

Sixteen mg of dexamethasone was administered intravenously just before surgery. On operation, laminectomy, from T12 to L1 was performed and the dura was opened. The dura mater was tense, the conus was swollen with no evidence of an extra-

Figure 1 Gadolinium-enhanced MRI of the thoracolumbar spine, revealing a widening of the lower thoracic cord with an enhancing intramedullary lesion from T_{12} to L_1

Figure 2 Intramedullary diffuse enhanced lesion typical of tuberculoma of conus medullare in sagital T1 weighted image

medullary tumor. A midline myelotomy was performed at T_{12}-L_1 and a grayish mass was excised using an operating microscope. Intramedullary lesion was firm and adherent to neural tissue. Histopathological detection of the surgical specimen on the light microscopy revealed granulomatous tissue containing a number of Langhan's giant cells, inflammatory cells and caseating necrosis (Figure 4).

After the operation, the patient suffered from urinary retention, which resolved in 3 weeks. The patient's paraperesis slowly improved. His leg function gradually improved and he was able to stand and walk with a walker after 3 weeks postoperatively. His sexual dysfunction was not improved. The triple antituberculous treatment was continued for a year.

Discussion

Tuberculosis is a chronic bacterial infection characterized by the formation of granulomas in infected tissue and cell-mediated hypersensitivity. Tuberculosis is endemic in Turkey. The estimated yearly prevalence of tuberculosis is 3.58 per thousand in Turkey, and it is even higher in the general population in southeastern regions.[1] Recent statistic reports state the incidence of tuberculous infection at 11.1 per 100 000 population in Western countries and neuro-tuberculosis involves 0.5 to 2% of extra-pulmonary tuberculous infection in the general tuberculosis prevalence.[2,5] Spinal cord involvement is much less common than the brain at a ratio of approximately 1 : 42.[3] This ratio correlates with the spinal cord and brain weight ratio, and their blood supplementation ratio.[3–5] Six conus medullaris tuberculoma cases were reported in English literature up to date[2,5–9] (Table 1).

Intramedullary spinal tuberculoma is a rare condition and seems to be a disease of relatively young patients and probably originates from hematogenous seeding from an extraneural focus (usually pulmonary and renal), or as a part of miliary tuberculosis.[4]

The clinical presentation of intramedullary tuberculomas is that of a subacute spinal cord compression with appropriate motor and sensory findings depending on the level of the lesion.[10]

Spinal cord tuberculomas are divided into two types. The first type lesions comprise lesions generally 2 to 3 cm in diameter and have relatively thin membranes often containing granular calcified material. These lesions are generally attached to the dura, yet are relatively easily separated from the spinal cord. The second form of tuberculoma is one of diffuse occupation of the subdural masses with a relatively

Figure 3 Intramedullary diffuse non-homogen hyperintensed lesion typical of tuberculoma of conus medullare in T2 weighted axial view

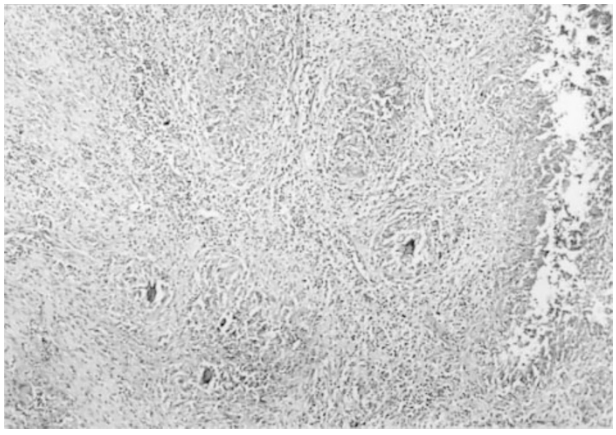

Figure 4 Photomicrograph showing small, caseating granulomas with giant cells, epithelioid histiocytes, and lymphocytes (hematoxylin and eosin, original magnification × 40)

Table 1 Reported cases of involvement of the conus medullaris in the literature

Author	*Age*	*Sex*	*Diagnostic method*	*Chemotherapy*	*Steroid therapy*	*Surgery*	*Clinic outcome*
Bradbury*[6]	*	*	*	*	*	*	*
Choksey[5]	31	F	Myelogra.	+	+	+	Moderate
Gupta[7]	*	*	MRI	+	*	–	*
Sie[8]	24	M	MRI	+	–	+	Good
Dehoux[2]	58	M	MRI	+	+	–	Good
Suzer[9]	20	M	MRI	+	+	–	Good
Our Case	32	M	MRI	+	+	+	Moderate

*not available; – was not performed

avascular greyish tuberculoma granulation tissue.[5] The present case is an example of the second form of spinal tuberculoma.

It is reported that patients with intradural extramedullary tuberculoma have presented with a gradual onset over weeks to months with progressive weakness of legs, sometimes associated with sphincter or sensory deficit.[4,5] A history of past or concurrent tuberculosis exposure to the disease was often obtainable, and patients had frequently suffered from tuberculous meningitis.[3,11,12] Intracerebral tuberculoma are known to occur quite frequently in association with tuberculous meningitis.[2,4,11,12]

Intramedullary tuberculoma, although a rare entity, must be considered in the differential diagnosis of subacute spinal cord compression, particularly in patients with evidence of extraneural tuberculous disease. Modern neurodiagnostic studies can aid in the localization of the tuberculous process to the extradural, intradural extramedullary, or intramedullary compartment.[7] Although myelography used to be the diagnostic procedure of choice in the evaluation of intramedullary tuberculosis, MRI is clearly superior in sensitivity and specificity. The advent of MRI may facilitate the diagnosis of an intramedullary mass consistent with tuberculoma.[7,13] The prognosis for neurologic improvement is good with appropriate therapy.[3,12,14] Medical therapy remains the mainstay of treatment, but when confronted with a progressing neurologic deficit, surgery may be indicated. In the articles reviewed,[3–5,8–10,15] surgery was performed in the majority of patients mainly to establish the diagnosis.

In conclusion, medical therapy remains the mainstay of treatment, but when confronted with a progressing neurological deficit, the neurosurgeon may need to remove the lesion. Exact surgical indications for spinal tuberculoma are not precisely determined, surgical decompression may play a role with the concomitant use of anti-tuberculous medication. In cases of conus medullaris tuberculoma which do not respond to treatment with antituberculous and steroid therapy, diagnostic and therapeutic surgical intervention should not be delayed. The application of modern neurodiagnostic studies, and microsurgical techniques associated with adequate medical treatment make this a potentially curable lesion.

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