Case Report

Spinal cysticercosis: A report of two cases with review of literature

ABSTRACT

Neurocysticercosis is the most common parasitic infection of the central nervous system worldwide. However, Cysticercosis affecting the spine is considered extremely rare. We report two cases of spinal cysticercosis with review of literature.

Key words: Cysticercosis; intramedullary; spine.

Introduction

Cysticercosis is the most common parasitic central nervous system (CNS) infection worldwide.[1] The vast majority of neurocysticercosis are usually found at meningo-basal (30%), parenchymal (20%), intraventricular (17%), intraspinal (1%), or mixed locations (32%).[2] Intraspinal cases are extremely rare; incidence of spinal cysticercosis varies from 0.7% to 5.85%.[3] Spinal cysticercosis commonly affects the subarachnoid space compared to the spinal cord substance.[4] A purely intramedullary location as reported in this paper is quite exceptional. We report two cases of spinal cysticercosis, diagnosed only after histopathological examination, one of which was intramedullary in its location.

Case Reports

Case 1

A 60-year-old male presented with a 3-month history of a gradually progressive weakness of both the lower limbs with associated bowel and bladder dysfunction for 20 days. Neurological examination disclosed spastic paraparesis with decreased motor power in both the lower limbs.

Magnetic resonance imaging (MRI) of the brain was normal. MRI of the dorsolumbar spine was performed on a 3 Tesla MRI scanner. Multiplanar conventional T1-weighted image (T1WI), T2-weighted image (T2WI), and postcontrast T1WI were acquired. A well-defined intramedullary cystic lesion was seen in the conus at T11 vertebral level causing focal swelling of the cord, homogeneously hypointense on T1WI, and hyperintense on T2WI with slight peripheral edema. The subarachnoid space from T11 to T12 was narrow due to the marked expansion of the spinal cord [Figure 1a and b]. A differential diagnosis of granulomatous lesion and cysticercosis was made.

The lesion was approached through a laminectomy from T11 to T12 exposing a swollen spinal cord with tense duramater at T11. A midline myelotomy was performed revealing a well-circumscribed white cystic lesion. The cyst was

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dissected free from the surrounding spinal cord parenchyma without much difficulty and was removed totally.

Histopathology showed the typical parasitic cyst with multiple layers [Figure 2a and b]. This case was reported as spinal cysticercosis.

Postoperatively, the patient was treated with albendazole. After 2 weeks, the patient improved enough to ambulate with a walking aid. The postoperative course was uneventful and the patient was discharged. Regular follow-up (3 years) is being done, and there are no signs of recurrence till date.

Case 2

A 25-year-old female presented with a 5-year history of a gradually progressive weakness of both the lower limbs with associated pain and difficulty in walking for 1 month. Neurological examination disclosed decreased motor power in both the lower limbs.

MRI of the brain was normal. MRI of the dorsolumbar spine was performed on a 3 Tesla MRI scanner. Multiplanar conventional T1WI and T2WI were acquired. A large, oval-shaped cystic lesion was seen involving the D12 to L2 vertebral level intraspinally intradural extramedullary indenting the spinal cord, homogeneously hypointense on T1WI [Figure 3a and b]. Arachnoid cyst, epidermoid cyst, and cystic meningioma were considered in the differential diagnosis.

The patient underwent a laminectomy from T12 to L2. The dura was tense at L1. On opening the dura, a 30 mm long, smooth spinal cord enlargement was seen. A midline myelotomy was performed revealing a well-circumscribed white cystic lesion. The cyst was dissected free from the surrounding spinal cord parenchyma without much difficulty and was removed totally [Figure 4].

Histopathology showed the typical parasitic cyst with multiple layers [Figure 2c and d] along with the sucker of the larval form. This case too was reported as spinal cysticercosis.

Postoperatively, the patient was treated with albendazole. The postoperative course was uneventful and the patient was discharged. Follow-up (1 year) is being done showing a recovery in the motor power of her lower limbs.

Discussion

Neurocysticercosis continues to be a major public health problem with a worldwide distribution, showing a higher prevalence in the developing regions of the world.[5] However, the prevalence is a variable depending mainly on sociocultural and economic factors. In addition, immigration from endemic to nonendemic areas is also one of the major factors influencing the prevalence.

CNS cysticercosis affects men and women equally. The peak incidence is between the third and fourth decades of life. It typically involves the brain parenchyma, intracranial

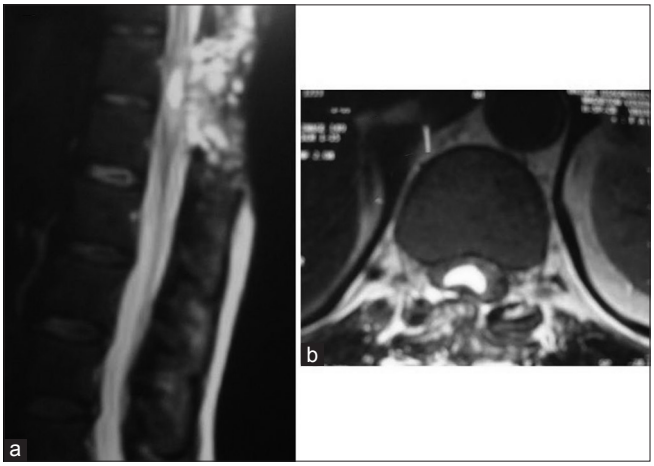

Figure 1: (a and b) Magnetic resonance imaging (axial T2 weighted images) at the level of conus medullaris reveals an intradural extramedullary cystic lesion compressing/displacing the conus and cauda equina rightward

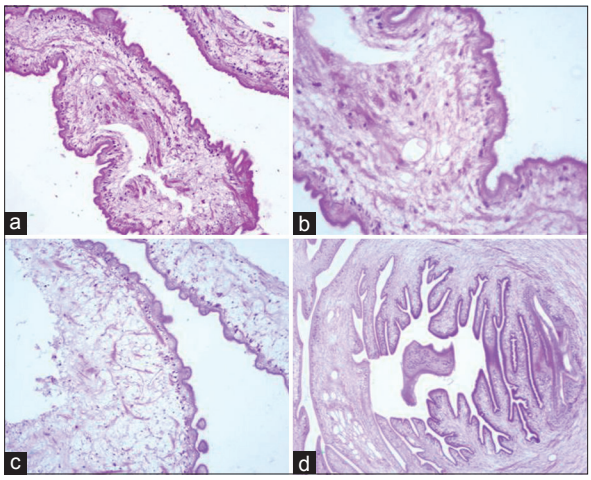

Figure 2: (a and b) Histopathology showing all three layers (cuticular, cellular, and reticular) of neurocysticercosis (Hand E, ×200). (c and d) Histopathology showing the three layers (cuticular, cellular, and reticular) of neurocysticercosis and the sucker (H and E, ×200)

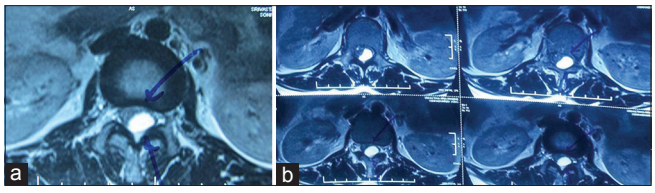

Figure 3: (a and b) Magnetic resonance imaging (sagittal and axial T2 weighted images) at the level of conus showing a thin-walled intramedullary cystic mass expanding the conus with perifocal edema. No obvious solid components identified

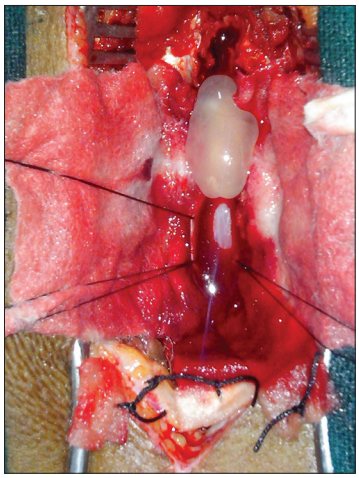

Figure 4: Microphotograph showing the intraoperative cyst

subarachnoid space, or ventricles. Spinal cysticercosis is rare; it may be leptomeningeal, intramedullary, or epidural. Among these, leptomeningeal is the most common, intramedullary is rare, and epidural is extremely rare.[2] Most cases of spinal cysticercosis are usually associated with cerebral cysticercosis. Isolated spinal cysticercosis either intramedullary or extramedullary is extremely rare.[6] In our cases, there was no evidence of either cerebral or systemic cysticercosis.

Clinical manifestations of spinal cysticercosis depend on the number and topography of lesions, individual's immune response to the parasite, and the presence or absence of previous infestations. Clinically, the most common manifestation of spinal neurocysticercosis is root pain and progressive weakness in contrast to the parenchymal neurocysticercosis that manifests with epileptic seizures; subarachnoid neurocysticercosis that manifests with headache while intraventricular neurocysticercosis manifests as subacute or intermittent syndrome of intracranial hypertension. Neurological damage in spinal intramedullary cysticercosis is attributed to the following factors: (i) mechanical compression caused by the cyst, (ii) due to the cord edema as a result of inflammation caused by degenerating larva remnants, and (iii) gliosis.[7]

Sotelo and Carpio had defined three clinical stages of neurocysticercosis, namely, active, transitional, and inactive neurocysticercosis. Escobar has also defined four pathological stages of neurocysticercosis. They are vesicular, colloidal vesicular, granular nodular, and nodular calcified. Vesicular is active form, colloidal vesicular and granular nodular represent transitional stages while nodular calcified stage is an inactive stage of neurocysticercosis.[8] Clinical suspicion of spinal cysticercosis is difficult, especially when there is neither previous history of any parasitic infestation nor associated cerebral neurocysticercosis. In patients with clinical suspicion of neurocysticercosis, cerebrospinal fluid (CSF) examination should be done as it provides a reliable evidence of inflammation; further immunoblot test may be performed to confirm the diagnosis of neurocysticercosis.[9] CSF examination was not done in our cases; otherwise, it could have provided a provisional diagnosis. Apart from CSF studies, MRI is one of the most useful diagnostic tools providing useful information in the evaluation of spinal neurocysticercosis patients. MRI, in addition to the diagnosis also provides precise information about the disease activity and its location, carrying important therapeutic implications.[10] Mathuriya *et al*. had described MRI findings for the different stages of intramedullary cysticercosis; the pathognomonic diagnostic feature is the presence of cyst with an eccentric mural nodule representing the scolex showing a hypointense rim with hyperintense core on T2W1 and hypointense or isointense lesion on T1W1.[11] However, these are not specific and the differentials include: Arachnoid cyst, ependymal cyst, neurenteric cyst, sarcoidosis, ependymoma, and infections including abscess.[12] In cases of spinal cysticercosis, the entire neuraxis should be evaluated to detect any additional lesion. In contrast to the existing literature, both our cases had isolated spinal cysticercosis.

A single therapeutic approach is not justifiable in spinal cysticercosis considering the pleomorphic nature of the disease. The decisive factors for the treatment of spinal cysticercosis are activity of the disease and location of parasites. Two mainstays of therapy are medical and surgical intervention. Surgical intervention should be considered in patients with acute onset of symptoms and in those where the diagnosis is in doubt as it not only provides decompression but also confirms the diagnosis after the histopathological examination and provides maximum chances of recovery avoiding the irreversible cord changes.[13] Results of surgical intervention are variable; Mohanty *et al*. in a case series of spinal intramedullary cysticercosis reported drastic recovery in 7/8 (87.5%) cases.[14] However, Sharma *et al*. reported improvement only in 60% of cases.[15] Both our cases had surgical removal of the cysts facilitating decompression and resolution of the cord edema, preventing any further damage, showing reasonably good recovery with minimal morbidity. Recent literature shows that according to American Society for Microbiology Current Consensus Guidelines for treatment of neurocysticercosis, treatment of intramedullary/extramedullary spinal cysticercosis should be surgical.[16]

Conclusion

Spinal intramedullary cysticercosis remains a diagnostic challenge. It should be considered in the differential diagnosis of spinal intramedullary/extramedullary lesions. Surgical treatment is recommended in patients with extensive lesions showing progressive neurological deficits. Medical treatment may be recommended in patients with early neurological symptoms under surveillance as surgical intervention may be required in case of neurological deterioration or failure of medical therapy.

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Conflicts of interest
There are no conflicts of interest.

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