

## Case Report

# A Case of Neurocysticercosis in Entire Spinal Level

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Cysticercosis is the most common parasitic infection affecting the central nervous system. Spinal neurocysticercosis (NCC) is very rare compared with intracranial NCC and requires more aggressive management because these lesions are poorly tolerated. The authors report a case of intradural extramedullary cysticercosis of the entire level of spine with review of the literature.

**KEY WORDS :** Cysticercosis · Spinal · Parasitic infection.

## INTRODUCTION

Neurocysticercosis (NCC), one of the most prevalent forms of parasitosis of the central nervous system (CNS), is caused by the larvae of the tapeworm *Taenia solium* in the nervous system. It typically involves the brain parenchyma, intracranial subarachnoid space or ventricular system, mainly in endemic regions. In these areas, the incidence of NCC accounts for up to 4% of the general population<sup>1)</sup>. The parasitic involvement of the spinal canal is a relatively rare event even in endemic areas with an estimated incidence of 1.5 to 3% among the total NCC<sup>2)</sup>. The authors report a 42-year-old woman who had complaints of progressive bilateral leg motor weakness, finally diagnosed with spinal NCC along the entire level of spine (C2-L2). The report includes comments on the surgical experience and a review of the literature in terms of treatment modality.

## CASE REPORT

A 42-year-old female with a history of hydrocephalus visited the institute complaining of unsteady gait, progressive lower-extremity weakness over several months. Muscle strength was Grade III/VI in lower extremities bilaterally with enhanced deep tendon reflexes. She had mental retar-

ation and nearly total deafness for a long time, probably from childhood. Non-enhanced computed tomography (CT) of the head performed one year before had revealed hydrocephalus that was not investigated further as the headache and vomiting had subsided. Magnetic resonance imaging (MRI) of the brain was normal. MRI of the spine revealed multiple intradural, extramedullary cystic structures displacing the spinal cord along the C2-L2 levels. The mass lesion was distributed throughout the entire level of the spine. (Fig. 1). The NCC was considered on neuroimaging and serological studies preoperatively. We operated the level of T3-T5 and L1-L3 where the spinal cord was severely compressed. The operation included laminectomy and intradural exploration, during which we found thickened wall of the sac adhered to the spinal cord, longitudinally (Fig. 2). Complete resection of the lesion was impossible because of its huge size. We performed a limited resection of the thickened longitudinal sac. Sharp excision of the sac was required for contained cyst removal, ultimately allowing free flow of multiple translucent cysts from the excised sac. On gross examination, the each cyst was a thin-walled and contained whitish fluid which is consistent with neurocysticercosis without scolices. The final diagnosis was made as a cysticercosis after histopathological examination (Fig. 3). The patient was given a course of albendazole with steroids for 8 weeks after surgery. Her neurological status gradually improved over several months. After 6-months he was able to walk independently. Repeated serial MR imaging revealed successful decompression of the spinal cord, and the patient experienced no recurrence of symptoms during the 12-month follow-up period (Fig. 4). The enzyme-linked immunosorbent assay

• Received : February 29, 2010 • Revised : September 30, 2010  
• Accepted : October 5, 2010  
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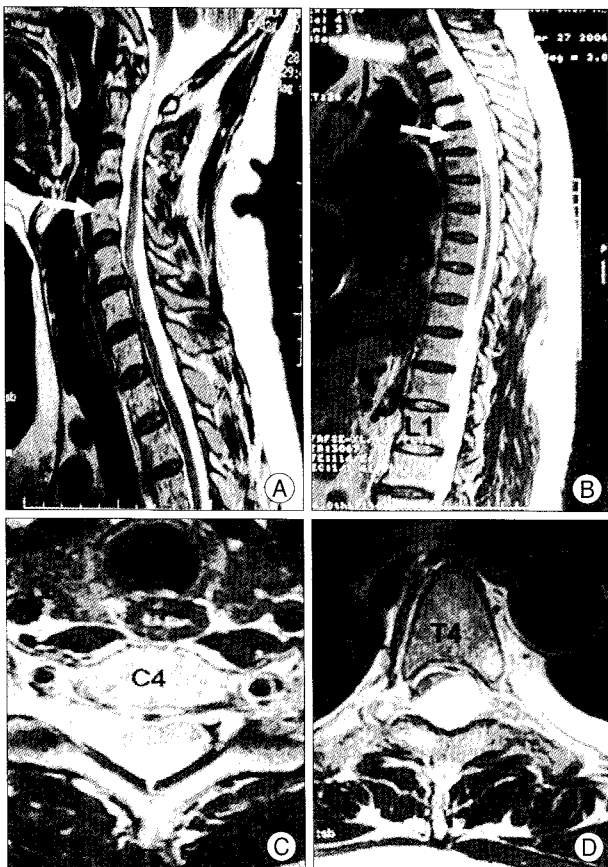


Fig. 1. Preoperative MRI of cervical and thoracic spine. Sagittal T2 weighted MR images revealing cystic mass lesion throughout cervical spine A : arrow indicates C4) and thoracolumbar spine B : arrow indicates T4). Axial T2-weighted MR images revealing a large posterior cyst and anterior displacement of the spinal cord at C4 and T4 level (C and D).

(ELISA) test showed no evidence of infection during follow-up period.

## DISCUSSION

Despite the high incidence of NCC in endemic areas, spinal NCC is very rare compared with intracranial NCC. There have been fewer than 200 cases of spinal NCC reported in the world literature<sup>2,11</sup>. The low incidence of spinal involvement is believed to be related to the fact that the blood flow to the brain is approximately 100-fold greater than to the spine<sup>7,9</sup>. Intradural spinal cysticercosis can be subdivided in leptomeningeal (subarachnoid) or intramedullary forms (parenchymal), and the former is more prevalent than the later. Leptomeningeal form occur in approximately 80% of cases which is assumed to result from larval migration through the ventricular system into the spinal subarachnoid space, where their movement could be assisted by gravity<sup>3</sup>. Queiroz et al.<sup>3,9</sup> reported that spinal distribution of cysticerci occurred as follows : 34% in the cervical; 44.5% in the thoracic; 15.5% in the lumbar; and 6% in the sacral region.



Fig. 2. Intraoperative photograph of the spinal cord demonstrating the presence of wide extended sac containing multiple cysticerci in the subarachnoid space at T2-4 level. The spinal cord (arrowheads) is severely displaced and flattened. The neurocysticercal sac (\*) occupying most of the spinal canal which compress the spinal cord.

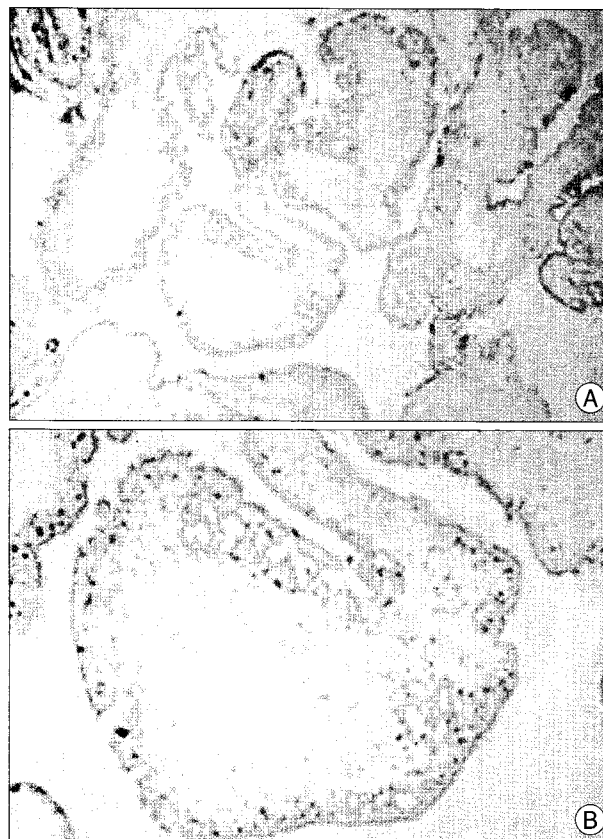


Fig. 3. Histologic finding. The degenerated scolex is covered by a wavy, somewhat refractile cuticle & a subcuticular pseudoepithelial layer (H & E, magnification (A :  $\times 40$ , B :  $\times 100$ )).

The disease may have a pleomorphic quality that is related to individual differences in the number and location of the lesions within the CNS<sup>4,12</sup>. In an older study, the authors reported that 30% also had intracerebral involvement in patients afflicted with spinal cysticercosis, whereas in a more recent study the authors noted 100% concurrent intracere-



**Fig. 4.** Postoperative sagittal T2-weighted magnetic resonance imaging at one year after surgery demonstrating improved compression of the spinal cord, but spinal cord thinning is remained at thoracic level.

bral involvement<sup>10</sup>. Some authors have reported that spinal involvement typically appears a few months or years after the development of hydrocephalus and/or episodes of cysticercosis-related meningitis<sup>5</sup>. Hydrocephalus may be explained by the presence of cysticerci within the ventricular system, arachnoiditis causing obstruction of the ventricles outlets (isolated ventricle), or subarachnoid natural CSF pathways. Spinal NCC-related symptoms are myelopathy and progressive weakness, induced by spinal cord and/or cauda equina compression. This case study has pleomorphic clinical entity in that the patient exhibited progressive spastic paraparesis due to spinal involvement with hydrocephalus and signs of dementia for more than 10 years.

In this case, laboratory test including the ELISA showed no evidence of ongoing intracranial infection and neuroimaging showed arrested hydrocephalus, assumed to be from a previous infection. Magnetic resonance imaging is the diagnostic study of choice for evaluating spinal NCC because it provides noninvasive multiplanar images of any potential intraspinal pathological entity. Immunodiagnostic tests of serum samples have been widely used to confirm the diagnosis of neurocysticercosis. The ELISA performed to test CSF samples is more accurate than that of serum; its sensitivity is 87% and

its specificity is 95%<sup>11</sup>. The treatment of spinal NCC includes a combination of specific cysticidal drugs, noncysticidal therapy and surgery, but there is a lack of definite guidelines due to the rarity of spinal involvement.

The efficacy of medical therapy with praziquantel or albendazole is well described in a number of cases by various authors with the resolution of neurological deficits<sup>12</sup>. Albendazole may be more effective than praziquantel<sup>6</sup>. Surgical treatment is indicated in cases of spinal NCC when patients experience severe and progressive neurological dysfunction regardless of whether medical therapy has been attempted. Excision of extramedullary lesions is often difficult because of the arachnoidal scarring secondary to cyst degeneration, but sharp dissection, gentle irrigation, and Valsalva maneuvers may assist in extirpating adherent cysts<sup>9</sup>. All cases of spinal neurocysticercosis, surgery does not provide an decisive cure without morbidity because of the various mechanisms responsible for neurological symptoms in spinal neurocysticercosis. However, in patients with acute onset of symptoms and in those where the diagnosis is doubtful, surgical excision is necessary to eliminate the compressive element. This is both to confirm the diagnosis and to provide maximal chances of recovery before any irreversible cord changes occurs.

In this case, due to the widespread nature of the cystic lesion, it was inevitable to do a limited excision rather than total resection. However, we did get an appropriate decompression of spinal cord without significant complications.

## CONCLUSION

Spinal neurocysticercosis should be considered in the differential diagnosis in high-risk populations with symptoms suggestive of a spinal mass lesion. Current therapy for leptomeningeal spinal cysticercosis is laminectomy and resection followed by medical management. In the case that total resection is not feasible due to entire spinal lesion such as this case, the limited resection with cyst removal and chemotherapy for the treatment of spinal neurocysticercosis may be an option.

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