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Case Report

Isolated Spinal Neurocysticercosis: Unusual Ocular Presentation Mimicking Pseudotumor Cerebri

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Spinal intradural cysticercosis is a rare manifestation of neurocysticercosis. We report a unique patient who showed visual symptoms and normal imaging of the brain caused by isolated spinal neurocysticercosis. A 59-year-old male patient was admitted to the emergency unit with a history of severe headache and progressive blurred vision. Brain computed tomographic scanning and magnetic resonance imaging showed normal cerebral anatomy without hydrocephalus. The fundoscopic evaluation by an ophthalmologist showed bilateral papilledema. Perimetry studies revealed visual field defects in both eyes. With the diagnosis of pseudotumor cerebri, a lumbar tap was attempted; however, we could not drain the cerebrospinal fluid in spite of appropriate attempts. Lumbar magnetic resonance imaging revealed multilevel intraspinal lesions that were confirmed histologically to be neurocysticercosis. An intraoperative lumbar puncture revealed an increased opening pressure and cytochemical analysis showed elevated cerebrospinal fluid protein level. The headache resolved immediately after surgery. However, the visual symptoms remained and recovered only marginally despite antihelminthic medications after six months of operation.

Key Words: Spinal neurocysticercosis · Pseudotumor cerebri · Cerebrospinal spinal fluid.

INTRODUCTION

Neurocysticercosis (NCC), involvement of the central nervous system by Taenia solium, is one of the most common parasitic diseases. NCC of the central nervous system is classified as the parenchymal type, subarachnoid type, intraventricular type, or spinal type according to the involved location. For such infections, the frequency of the spinal type has been reported to be 0.7-5.0%^{4,6)}. However, isolated spinal NCC, in the absence of intracranial involvement, accounts for only 25% of spinal type NCC9,10). Spinal NCC may present with back pain, sensory deficits, motor weakness or hydrocephalus related symptoms¹²⁾. However, visual symptoms related spinal NCC are extremely rare. As far as the authors' knowledge, this is the first report that visual symptoms developed as a result of increased intracranial pressure caused by isolated spinal NCC in a male patient who had a high protein level in CSF analysis. We report a rare case of isolated spinal NCC that showed unusual visual symptoms.

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CASE REPORT

A 59-year-old man was admitted to the emergency unit with a history of severe headache and progressive blurred vision. Nausea usually had been present with the headaches. The symptoms began three months previously but were aggravated seven days before admission. The patient had been healthy without prior medical concerns before admission. On examination, the patient was alert and the vital signs were stable. There were no pathological reflexes detected and the motor strength was intact. The pupils were isocoria and reacted to light. A computed tomography scan and magnetic resonance imaging (MRI) of the brain showed no significant abnormalities; there were no intraparenchymal or intraventricular cysticercal lesions (Fig. 1). In addition, there were no subcutaneous nodules. The peripheral blood findings included a leukocyte count of 8700/mm3 (eosinophil 3.5%) and hemoglobin 12.8 mg/dL, without significant abnormalities. Fundoscopic evaluation by an ophthalmologist showed bilateral papilledema. Perimetry studies revealed bilateral symmetrical nasal and superior visual field defects, bilaterally. The patient was diagnosed with PTC and lumbar taps were attempted to confirm increased intracranial pressure; however, we could not obtain CSF in spite of appropriate attempts. Attempts at placing the needle for the LP at the L4-L5 and L3-L4 interspace resulted in little CSF outflow under pressure. A lumbar spine MRI was then performed. The MRI of the thoraco-

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Fig. 1. Computed tomographic scan reveals no intraparenchymal abnormalities.



Fig. 2. Sagittal precontrast and postcontrast T1-weighted images demonstrate diffuse multiple abnormal signals at conus medullaris, cauda equinea, and thecal sac.

lumbar spine revealed multiple cystic contrast enhancing lesions extending from T12-L1 and L5-S1. The conus medullaris appeared to be compressed and distorted by the mass (Fig. 2). Under general anesthesia, the patient underwent microsurgical removal. Laminectomy from T12 to L1 and L4 to L5 was performed. Several white encapsulated cysts containing clear fluid adhered to the arachnoid membraine but they were dissected without difficulty. About 1.5-2 cm sized four cysts were removed. The opening pressure, before incision of the dura, was 260 mm-H₂O; cytochemical analysis revealed four mononuclear cells/ mm³, 92 mg/dL protein, and 40 mg/dL glucose. During the surgery, mixed lesions of a solid and cystic nature were detected (Fig. 3). The pathological findings showed fibrosis, local calcification, and chronic inflammation on the walls of the remnants of the larva (Fig. 4). The patient was treated with antihelminthic medication (tablet Zentel® 400 mg, manufactured by Smith-Kline Beecham Pharmaceuticals) with oral dexamethasone for six weeks. The headache improved significantly after surgery. However, the vision, after six months of treatment, recovered only marginally to persistent papilledema and visual field de-

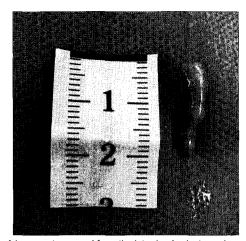


Fig. 3. A large cyst removed from the intradural spinal canal.

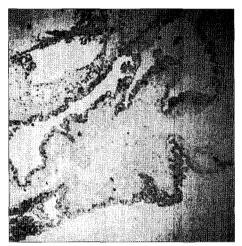


Fig. 4. Microscopic finding shows the anatomy of the cyst wall to be consistent with cysticercosis. Histological characteristics include microvilli, thin tegument and loose connective tissue (H&E $\times 100$).

fects. Six months after surgery, CSF analysis showed normal composition and opening pressure (180 mmH 20). We recommended a lumbo-peritoneal shunt, however, the patient refused due to the relieved headache symptoms.

DISCUSSION

Intraspinal involvement by cysticercosis is rare even in endemic areas; it has been reported in only 1 to 5% of patients with NCC and most such cases are associated with concomitant cerebral involvement. Isolated spinal involvement of cysticercosis, either intramedullary or extramedullary, is rare^{1,2,5)} Sotelo et al.¹¹⁾ reported that the frequency of the spinal type of neurocysticercosis was 0.77% among 753 cases of active neurocysticercosis. The spinal form may require more aggressive treatment due to the natural confines of the spinal canal. The location and size of the lesion, and the inflammatory response or arachnoid scarring generated by cyst breakdown are the factors involved in causing symptoms¹³⁾. It has been reported that the frequency of the intraspinal type of neurocysticercosis is significantly low-

er than cerebral parenchymal neurocysticercosis due to the small vessel diameter, low blood flow and the structure of the spinal cord making it harder to invade than the cerebrum³⁾. Spinal NCC usually presents with back pain, motor weakness, sensory deficits, or bladder involvement^{2,12)}. Pseudotumor cerebri (PTC) is a syndrome defined clinically by the features of elevated intracranial pressure, normal brain imaging, normal cerebrospinal fluid (CSF) and papilledema. To date, there has been only one case report of visual symptoms associated with increased intracranial pressure caused by isolated spinal NCC7). In that case, pseudotumor cerebri was present, confirmed by the normal composition of the CSF. However, in the present patient, the CSF analysis showed abnormally high protein level, which ruled out the possibility of PTC because normal CSF composition is main criteria of PTC. It is encountered most frequently in young, corpulent women. The mechanism underlying the visual symptoms in PTC and our patient are not fully understood; however, the symptoms might result from poor absorption of CSF by the meninges. The subsequent increase in extracerebral fluid volume might have resulted in elevated intracranial pressure. However, because of a slow and insidious progression, there is ample time for the ventricular system to compensate; this might explain why there was no dilation of the cerebral ventricles in this patient as there would be with PTC. Increased intracranial pressure causes stress on the peripheral aspects of the brain, including the cranial nerves. Stagnation of axoplasmic flow in the optic nerve (CN II) results in papilledema and transient visual obscurations; when the abducens nerve (CN VI) is involved, the result can be intermittent nerve palsy and diplopia. Obstruction of the CSF flow at the arachnoid villi or subarachnoid space is the usual mechanism causing PTC in patients with subarachnoid NCC. Therefore, the patient with intraspinal NCC can also develop symptoms mimicking PTC8). In this case, although the brain CT and MRI scans performed at the initial evaluation were normal, the CSF opening pressure revealed elevated intracranial pressure. There has been only one reported case of isolated spinal NCC associated with visual disturbances in an overweight woman¹²⁾. However, the CSF analysis showed a normal protein level confirming the diagnosis of PTC in that patient. In our case, the CSF analysis revealed abnormally high protein level and it could rule out PTC. High protein in the CSF, which increases the viscosity of CSF, can also lead to increase of the intracranial pressure and interference with axoplasma flow of the optic nerve fibers, resulting in papilledema¹⁴⁾.

CONCLUSION

Although spinal NCC is rare, it should be kept in mind as a possible cause of an unusual ocular presentation mimicking pseudotumor cerebri. Both increased intracranial pressure and high protein level in the CSF can be responsible for this rare ocular presentation by spinal NCC.

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