

CASE REPORT

A Rare Case of Isolated Cervical Intramedullary Cysticercosis: A Surgical Dilemma

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ABSTRACT

Cysticercosis is the most common parasitic infection of the cerebral parenchyma and spinal cord involvement is rare. In spinal cord, it affects more commonly thoracic spinal cord while cervical involvement is rare. We report the case of a 30-year-old female patient with isolated cervical intramedullary cysticercosis. Patient presented with pain in neck and right upper limb. Her magnetic resonance imaging (MRI) cervical spine revealed dumbbell-shaped intramedullary lesion at C3 to C4 level.

Patient underwent surgery with complete excision of cervical intramedullary lesion. Histopathological report confirmed cysticercosis. Postoperatively, patient had complete relief in her clinical symptoms. In this report, we also discuss the principles of diagnosis and treatment of intramedullary cysticercosis in combination with literature review. Spinal neurocysticercosis should be considered as a differential diagnosis of spinal mass lesion in patients residing in endemic area, such as India. Both surgical therapy and medical therapy have a role in the management of spinal cysticercosis.

Keywords: Cysticercosis, Intramedullary, Spinal cord.

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INTRODUCTION

Cysticercosis is most common parasitic disease of central nervous system. It is caused by the larval stage of *Taenia*

solium. In endemic regions, the incidence of neurocysticercosis approaches 4% of general population. Spinal neurocysticercosis either intramedullary or extramedullary without intracranial neurocysticercosis is considered to be rare.

CASE REPORT

A 30-year-old female presented with neck pain for 6 months and pain in right upper limb for 5 months. Neck pain was dull aching type, deep-seated, and aggravating in supine position. Neurological examination revealed hypertonia in lower limbs. Power was Medical Research Council grade IV in elbow and wrist flexion as well as extension. Right finger grip was weak. There was hyperreflexia in biceps, triceps, supinator, and knee. Plantar response was extensor. Superficial abdominal reflexes were absent.

Her MRI cervical spine revealed dumbbell-shaped lesion at C3 to C4 level, which was hypointense on T1-weighted image (T1WI) and hyperintense on T2-weighted image (T2WI). There was peripheral ring enhancement on contrast administration (Fig. 1).

The patient underwent surgery due to progressive deterioration of power and severe pain. The lesion approached through the dorsal root entry zone and gross total resection of lesion done. C2, C3, C4, and C5 laminoplasty was performed prior to closure of wound in layers. The lesion was firm, well encapsulated with cystic component. It was avascular and yellowish in color (Fig. 2). In the immediate postoperative period, patient developed weakness in overhead abduction on right side. It gradually improved in 15 days. Cervical pain and pain in right upper limb improved postoperatively.

Final histopathology report was suggestive of fragment of cysticercus with foamy macrophages and lymphocytic infiltrate around it (Fig. 3). Patient was started on tab albendazole 600 mg OD (15 mg/kg/day). The MRI brain ruled out intracranial neurocysticercosis.

At 1-year follow-up, patient was completely asymptomatic without any neurological deficit and follow-up MRI showed no evidence of cysticercosis.

DISCUSSION

Encystment of larva of *T. solium*, i.e., pork tapeworm, is called as cysticercosis.¹⁻⁴ Paranoli first described human

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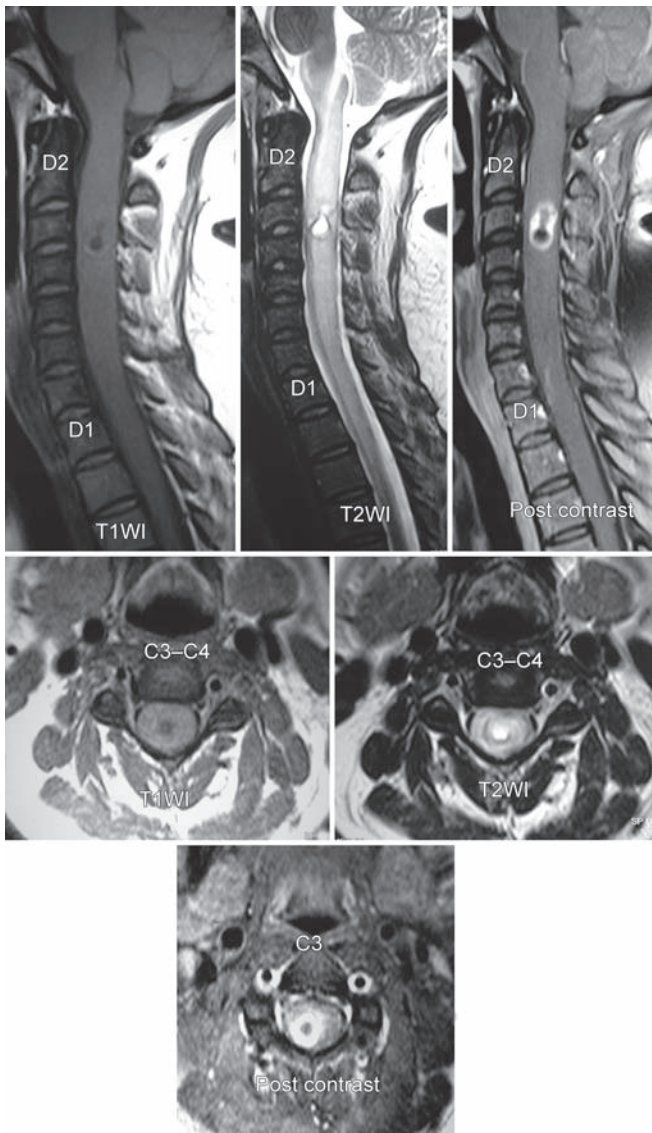


Fig. 1: The MRI cervical spine revealing dumbbell-shaped lesion at C3 to C4 level with peripheral ring enhancement on contrast administration

cysticercosis in 1,550 and the causative agent *T. solium* was recognized by Leuckart and Kuchenmeister, much later in the 19th century.⁵ Intracranial encystment is referred as neurocysticercosis. Cysticercosis is widely endemic in Brazil, Peru, Mexico, Korea, and India. It should be differentiated from teniasis, i.e., carriage of the adult tapeworm in the intestine. There are three stages to the life cycle: larva, embryo (or oncospheres), and adult (Fig. 4). *Taenia solium* can infect humans in two different ways: as adult worm or as the larva. Human intestinal tapeworm infection, i.e., teniasis, results from eating undercooked infested (measly) pork while the disease cysticercosis occurs when animals or humans become an intermediate host for the larval stage by ingesting viable eggs produced by proglottids. The most common route of ingestion is fecal oral route. Many larvae die naturally within 5 to 7 years.

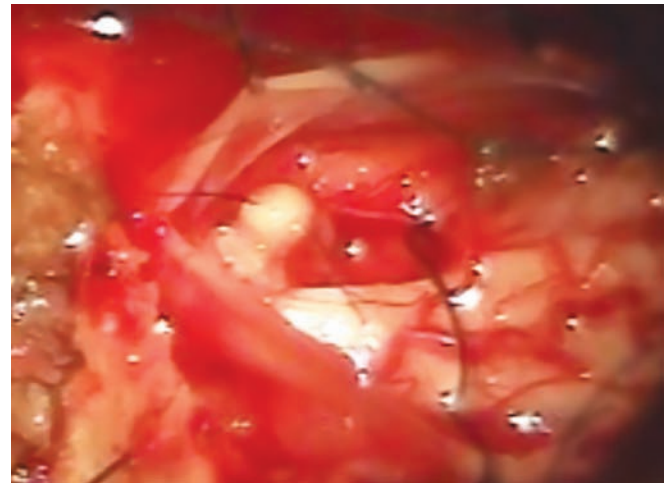


Fig. 2: Cervical myelotomy with delivery of firm, well-encapsulated cysticercus

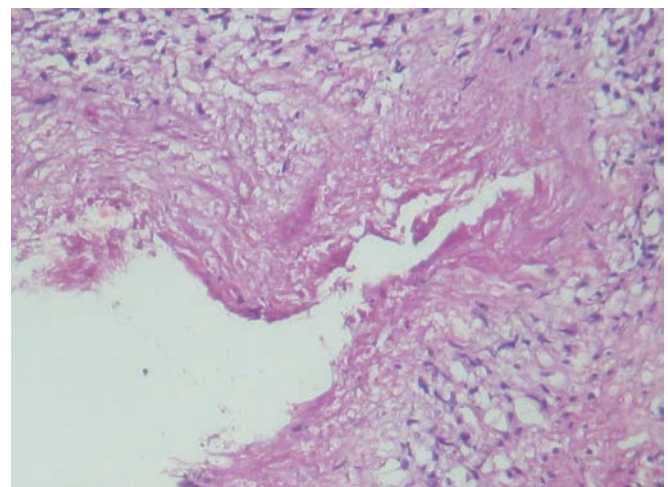


Fig. 3: Histopathological examination suggestive of fragment of cysticercus with foamy macrophages and lymphocytic infiltrate

Though cysticercosis is the most common parasitic infection of central nervous system, spinal cord involvement is less common. Spinal neurocysticercosis occurs in patients with intracranial neurocysticercosis in approximately 75% of the cases and isolated cases of spinal neurocysticercosis either intramedullary or extramedullary are considered to be rare.^{5,6} Spinal cysticercosis has been identified in the vertebral, extradural, intradural, but extramedullary and intramedullary regions. Intramedullary involvement in cysticercosis is seldom observed accounting for fewer than 20% of the intradural spinal cases.¹ Migration of the cysticercosis through the ventriculo-ependymal pathway and hematogenous dissemination has been identified to be the possible pathogenetic mechanisms.^{3,7} The higher incidence of spinal neurocysticercosis in the thoracic region may be related to the high blood flow in the thoracic segment of the spinal cord.

Symptoms produced by spinal cysticercosis depend on their location, size, and associated inflammation. Most

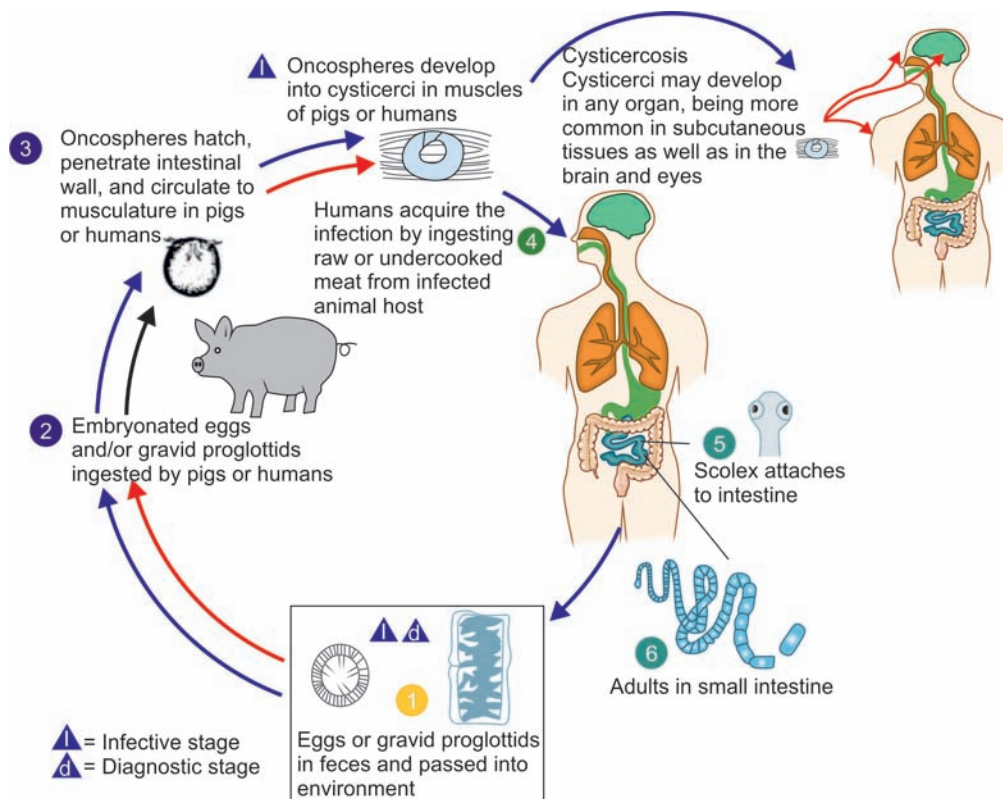


Fig. 4: Life cycle of *T. solium*

symptoms are produced due to mass effect produced by enlarging cyst. Symptoms can be radiculopathy or paresthesia. Corticospinal tract involvement leads to weakness ranging from monoparesis, paraparesis to quadriplegia. Bowel or bladder involvement can occur with intramedullary cysticercosis.

In the absence of previous history of neurocysticercosis or subcutaneous nodules, it may be difficult to clinically suspect intramedullary cysticercosis. High eosinophil count and calcification of soft tissues in the plain radiogram may be suggestive, but such findings are rare.⁸ Cerebrospinal fluid (CSF) and serum enzyme-linked immunoelectrotransfer blot assay against glycoprotein antigens may be supportive in suspected cases.⁹

Magnetic resonance imaging of spinal intramedullary lesions typically demonstrates cystic areas within the parenchyma and cyst fluid similar to that of CSF on both T1WI and T2WI.^{6,10} A subtle hypointensity may appear at the rim of the cyst on T2-weighted sequences. Irregular areas of peripheral enhancement after intravenously administered gadolinium have also been observed. Once diagnosis of cysticercosis is made, entire neuraxis should be evaluated to rule out brain parenchymal involvement.

When a patient does not come from an endemic area or spinal cord symptoms are the first manifestations of the disease, the diagnosis may only become apparent at the time of surgery.

It is of greater importance to determine a provisional diagnosis without or prior to surgery because of the high surgery-related morbidity, associated mortality rate of the disease, the potential earlier detection with neuroimaging evaluation, and the increasing effectiveness of medical management. Medical management is often adjunct to surgical management. Corticosteroids should be used in all patients. It may temporarily relieve symptoms and may help to decrease edema that tends to occur initially during treatment initially with anthelmintic drugs. The steroids should be tapered after anthelmintic is discontinued.

In patients presenting with acute or progressive neurological state, and in those where the diagnosis is missed or is in doubt, surgical excision is the choice of treatment as histopathology not only confirms the diagnosis, but early surgery also provides recovery before any irreversible cord damage takes place.¹⁰

Intramedullary spinal cysticercus cysts tend to be located superficially within the parenchyma (i.e., <3 mm deep). In most cases, the myelotomy is performed in the midline between the dorsal columns. Eccentric lesions may be approached through the dorsal root entry zone. After performing a superficial myelotomy, a cleavage plane can usually be developed with ease, thereby separating the cyst from the adjacent, albeit reactive cord parenchyma. Intraoperative ultrasound can help in localization of cyst.

Sharma et al¹¹ hypothesized that sensory and urological improvements likely were a result of decreased mass effect following decompressive surgery, but the refractoriness of motor deficits might have been a result of parenchymal gliosis from toxic parasitic metabolites. In our case, there was a complete improvement of sensory and motor symptoms in follow-up period of 1 year.

CONCLUSION

Spinal neurocysticercosis should be considered as a differential diagnosis of spinal mass lesion in patients residing in endemic areas, such as Brazil, Peru, Mexico, Korea, and India. In patients with spinal form, clinical and imaging features may be similar to intramedullary or extramedullary neoplastic mass lesions. Both surgical therapy and medical therapy have a role in the management of spinal cysticercosis.

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