

Case report

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Neurocysticercosis is a common parasitic infection of the central nervous system. Spinal involvement is described in only 0.25–5.8 % of patients and pure spinal neurocysticercosis without cystic lesions in the brain is extremely rare.¹ We present a rare case of cervical intramedullary neurocysticercosis, mimicking spinal intramedullary ependymoma, which was successfully treated surgically and only diagnosed histopathologically after total resection.

Case Report

A 37-year-old, right-handed female patient presented to the Neurosurgical Out-Patient Clinic with progressive weakness of all four limbs for one and half months without hyperreflexia or sphincter dysfunction. There was no history of trauma, fever or coagulopathy. The

An unusual case of Cervical intramedullary neurocysticercosis mimicking ependymoma

Neurocysticercosis is a common parasitic infection of the central nervous system, caused by larvae of *Taenia solium*. Cysticercosis of spine is less common compared to that of brain itself. Anatomically, spinal cysticercosis can be classified as extraspinal (vertebral) or intraspinal (epidural, subdural, arachnoid, or intramedullary), where intramedullary type is extremely rare. Isolated spinal intramedullary neurocysticercosis without brain involvement is even more rare. Preoperative diagnosis of cysticercosis within the cord may be difficult; and the clinical and radiographic features may mimic that of more common intramedullary tumor. Here, we present a rare case of cervical intramedullary neurocysticercosis in an adult, presenting with progressive limb weakness, mimicking spinal ependymoma, clinically and radiographically and only diagnosed histopathologically after surgery.

Key Words: Spinal intramedullary neurocysticercosis, ependymoma, in toto resection

patient had no history of parasitic infection in the past. On neurological examination, the patient had asymmetrical quadriplegia, with motor power of 3/5 in right upper and lower limbs and 4/5 in the left side. However, her sensory examination (including pinprick, touch, temperature, position and vibration) was intact; and the deep tendon reflexes were normal throughout. There was no long tract signs i.e. Hoffmann's and Babinski signs.

Magnetic Resonance Imaging (MRI) of the cervical spine revealed approximately 22 x 12 mm sized enhancing lesion at C3/C4 cord level with widening of the cord. Furthermore, a subcentimetric cystic lesion was noted at upper end of the lesion with hemosiderin cap and adjacent cord edema (**Figure 1A and 1B**). Radiographically, it was consistent with the diagnosis of intramedullary ependymoma with proximal localized syrinx.

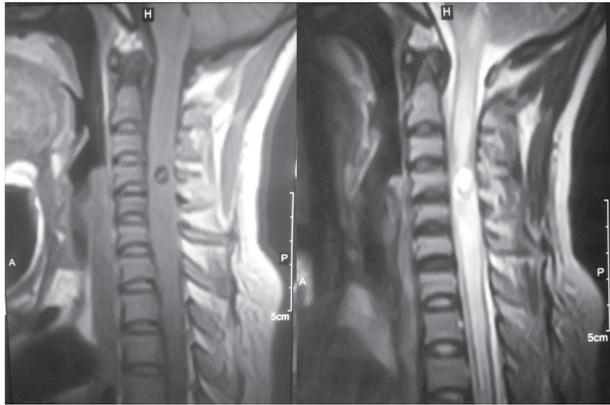


Figure 1: Centrally located enhancing well circumscribed lesion at C3-C4 level of spinal cord with its widening, and adjacent cord edema. Fig 1A: Iso intense in T1 weighted image. Fig 1B: Hyper intense in T2 weighted image with high signal intensity sub-centimetric cystic lesion.

With preoperative diagnosis of ependymoma, she underwent cervical laminoplasty C3-C5 and excision of lesion. Under general anesthesia, posterior midline incision extending from spinous processes of C2 to C6 was made in a standard fashion. Laminectomy of C3 to C5 was achieved using NSK high speed neuroelectric-drill with footplate. The dura was opened longitudinally. The cord was opened near the midline, avoiding the pial vessels. The pia mater was tacked up to the dural edges using 6-0 prolene suture for better exposure. The lesion was then completely excised after meticulously dissecting it off the surrounding normal cord tissue. After copious irrigation and meticulous hemostasis, the pia mater was closed interruptedly; and then, the dura was closed in continuous water-tight fashion. Laminoplasty was done by fixing titanium mini-cranial plates and screws on either sides of the three lamina. The incision was finally closed in layers and skin with staples. Intraoperative finding was 3.0 x 1.5 cm sized tumor which was minimally vascular with cystic swelling attached superiorly to it. Macroscopically, the lesion was grayish- whitish in color with firm to soft in consistency and had a well-delineated plane between the lesion and normal cord, consistent with ependymoma on gross examination (Figure 2 and 3).

Postoperative course was uneventful. There was no obvious neurological deterioration after surgery. Postoperatively, the patient underwent vigorous limb physiotherapy and gait training. The staples were removed on POD 11; and patient was discharged on 12th day. The neurological status remained the same at the time of discharge. The patient is on regular follow up with physiotherapy. At three-month follow-up, the patient was

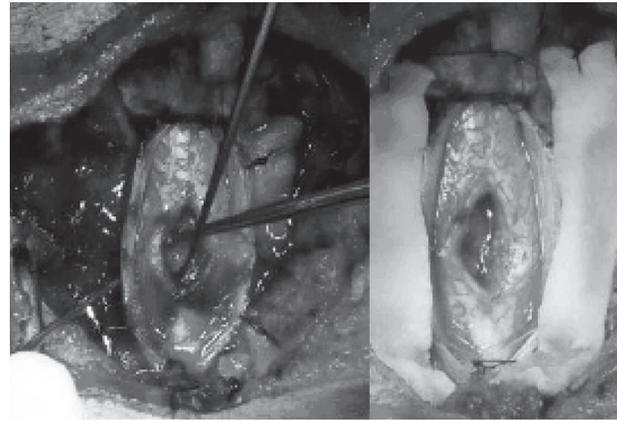


Figure 2: Intraoperative pictures showing the well-encapsulated lesion within the cord as it is being dissected off the normal cord (2A) and decompressed cord with tumor bed after complete excision (2B)

able to walk independently without support, presenting with only minimal weakness on the right leg (4+/5) and motor strength on rest of the limbs was 5/5. Patient was not given oral Albendazole as there was no evidence of any systemic parasitic infection.

Histological examination revealed cyst wall with pallisading epithelium histiocytes and chronic inflammatory cells along with parasite, suggestive of neurocysticercosis (Figure 4).

Discussion

Cysticercosis of central nervous system caused by larval infestation of *Taeniasolium* characteristically involves the brain parenchyma, intracranial subarachnoid space, or ventricular system.² Spinal involvement is usually associated with concomitant cerebral involvement. Isolated involvement of spine is extremely rare with predominantly extramedullary.³ Intramedullary cysticercosis often presents in the patients between 20 to 45 years old, ranging from 5 to 45 years old.⁴ The common clinical manifestations included pain, paraparesis, spasticity, bowel and bladder incontinence, and sexual dysfunction.⁵ In our case, the patient presented with progressive asymmetrical quadriparesis without spasticity or sphincter involvement. It is more endemic in countries like Latin America, Mexico and other Southeast Asian countries. The cause of this infection is known to be due to ingestion of cysticercal eggs in food contaminated by human or porcine feces via fecal-oral route.⁶

In regards to the distribution within the cord, cysticercus is commonly located in the thoracic level,

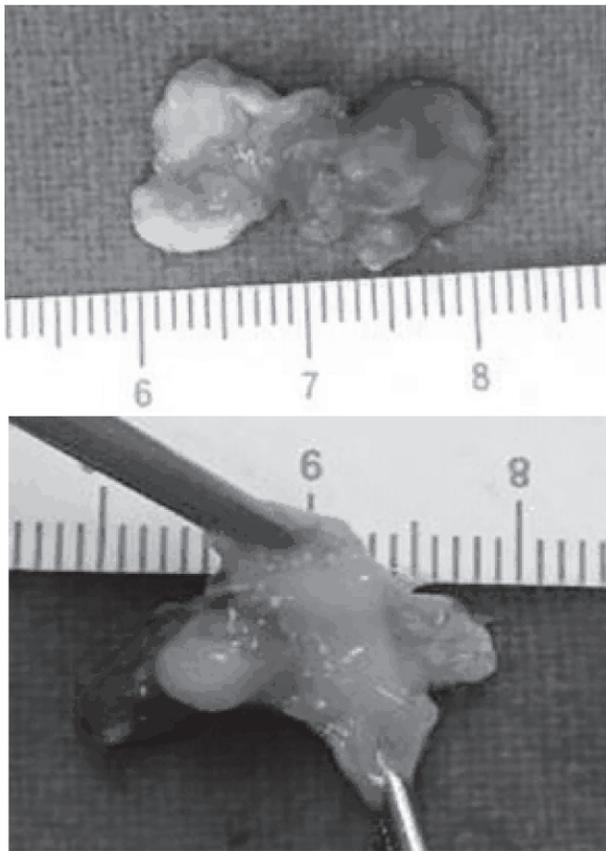


Figure 3: Gross specimen showing grey-white tissue

other parts of the cord.⁷ However, it is also thought that intramedullary cysticercus could migrate to the spinal cord via the ventriculo-ependymal pathway. It is difficult to clinically suspect spinal cysticercosis in the absence of previous history. Some findings like, high eosinophil count and calcification of soft tissues in the plain radiograms can be helpful. The vesicular stage of neurocysticercosis appear as a well defined hypointense cyst with hyperintense scolex inside on the T1- weighted image.⁸ The thickened cyst capsule in the colloidal stage is hyperintense on T1-weighted and hypointense on T2-weighted images. The absolute criteria to make diagnosis of neurocysticercosis are (1) histological demonstration of the parasite from biopsy of the brain or spinal cord, (2) cystic lesion showing scolex on CT/MRI, and (3) direct visualization of subretinal parasites on fundoscopic examination. Any one of the above criteria is sufficient for the diagnosis.⁹

The medical management of intramedullary spinal cysticercosis should be considered in patients with no neurological deficit and in cases diagnosed by cerebrospinal fluid assay. However, in patients presenting with acute or progressive neurological deterioration, and in those cases where the diagnosis is in dilemma, surgical removal if possible should be the treatment of choice.

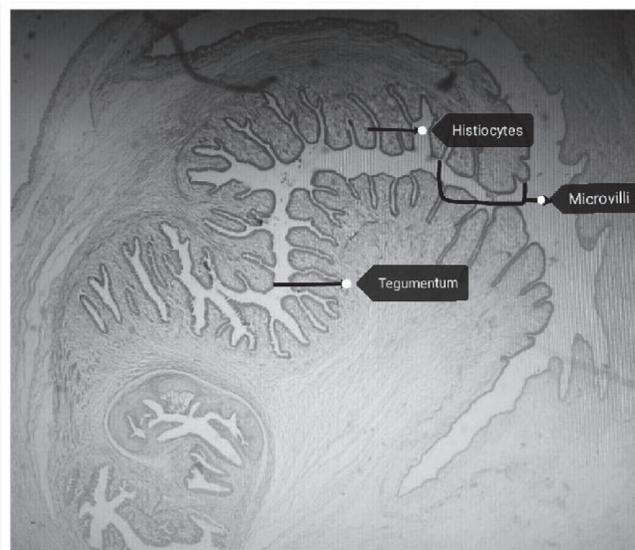
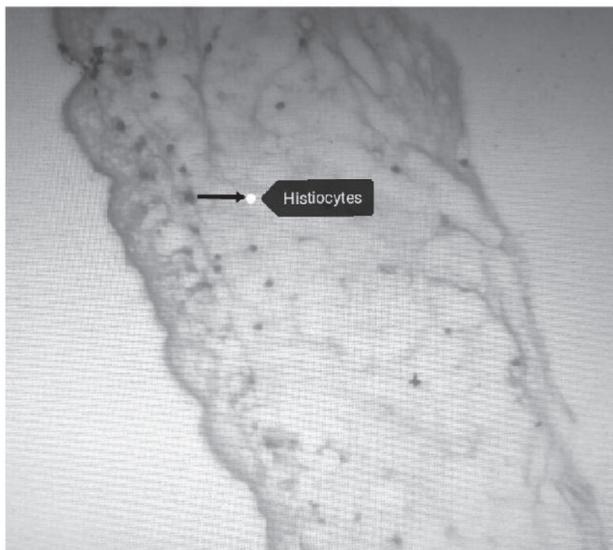


Figure 4: Microscopic slides showing cyst wall with pallisading epithelial histiocytes and chronic inflammatory cells along with parasite with tegument and bladder wall of cysticercus.

with rarely involving the cervical and the lumbar cord. Our case of cervical intramedullary neurocysticercosis is an extremely rare disease. This distributional mode of cysticercus supports the hypothesis that intramedullary cysticercus comes from the blood circulation, because thoracic cord has much more blood supply than the

Early surgery not only provides sample for early diagnosis, but it also provides recovery before irreversible cord damage accomplished.¹⁰ In our case, owing to increasing neurological deficit, surgical treatment was opted to remove the lesion which produced progressive spinal compression and also to confirm the diagnosis. Though preoperative

Cervical intramedullary neurocysticercosis

clinico- radiological findings and even intraoperative macroscopic features favored the diagnosis of spinal intramedullary ependymoma, post operative histopathological examination proved it to be neurocysticercosis. Therefore, for cystic lesion within the cord, the differential diagnosis of intramedullary neurocysticercosis should be always taken into consideration, particularly in the endemic areas of the South East Asian subcontinent.

Conclusion

Spinal cysticercosis, a rare entity, should always be kept as a differential diagnosis for cystic lesion within the cord. However, in our case, the clinician-radiologic features suggested ependymoma. Surgical excision helps in the removal of the lesion, decompression of the cord and provide histopathological diagnosis.

References:

1. Garcia HH, Del Brutto OH: Cysticercosis Working Group in Peru. Neurocysticercosis: updated concepts about an old disease. **Lancet Neurol** 4:653-661, 2005
2. Sahoo PK: Spinal intramedullary cysticercosis. **Med J Armed Forces India** 56:240-241, 2000
3. Kasliwal MK, Gupta DK, Suri V, et al: Isolated spinal neurocysticercosis with clinical pleomorphism. **Turk Neurosurg** 18:294-297, 2008
4. Mathuriya SN, Khosla VK, Vasishta RK, et al: Intramedullary cysticercosis: MRI diagnosis. **Neurol India** 49:71-74, 2001
5. Agrawal R, Chauhan SP, Misra V, et al: Focal spinal intramedullary cysticercosis. **Acta Biomed** 79:39-41, 2008
6. Gaur V, Gupta RK, Dev R, et al: MR imaging of intramedullary spinal cysticercosis: A report of two cases. **Clin Radiol** 55:311-314, 2005.
7. Sharma BS, Banerjee AK, Kak VK: Intramedullary spinal cysticercosis: case report and review of literature. **Clin Neurol Neurosurg** 89:111-116, 1987
8. Mathuriya SN, Khosla VK, Vasishta RK, et al: Intramedullary cysticercosis: MRI diagnosis. **Neurol India** 49:71-74, 2001
9. Del Brutto OH, Rajshekhar V, White AC, et al: Proposed diagnostic criteria for neurocysticercosis. **Neurology** 57:177-183, 2001
10. Agale SV, Bhavsar S, Choudhury B, et al: Isolated intramedullary spinal cord cysticercosis. **Asian J Neurosurg** 7:90-92, 2012