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CASE REPORT

Minimally manipulative extraction of polycystic cervical neurocysticercosis

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Abstract
Clinical history Intradural, extramedullary cervical spinal involvement is an uncommon manifestation of neurocysticercosis.
Case report A case of a middle-aged man with neurocysticercosis in the intradural extramedullary cervical spine and brain who originally presented with bilateral paresthesias of his extremities, with a progressively unsteady gait. Magnetic resonance imaging revealed cystic enhancing lesions in the brain and cervical region of the spine, with the largest cyst extending from the posterior fossa through C2, causing spinal cord compression. The patient underwent surgical resection of the intradural extramedullary cervical spinal lesions, and he has continued to improve clinically, with no recurrence of cystic lesions.
Conclusion When examining patients with clinical signs of a spinal mass lesion, the differential diagnosis should include neurocysticercosis of the spine.

Keywords Neurocysticercosis · Racemose · Cervical · Spine · Polycystic · Intradural · Extramedullary

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Introduction

Neurocysticercosis is a parasitic disease of the central nervous system acquired through fecal-oral contamination with the cestode, *Taenia solium*. Humans are an intermediate host. The *Taenia solium* eggs are ingested, typically through infected pork. The eggs are digested and develop into larvae that can then travel to any part of the body, including the central nervous system. Cysticercosis is prevalent in less industrialized nations, particularly in Latin America where there are 400,000 symptomatic patients and another 75 million people at risk for infection [1, 2]. Neurocysticercosis is classified based on its location, which includes, in order of decreasing frequency, subarachnoid-cisternal, parenchymal, intraventricular, and spinal [3, 4]. Based on radiologic findings, CT scan and MR imaging, neurocysticercosis has five stages: non-cystic, vesicular, colloidal vesicular, granular nodular, and calcified nodular [2, 5].

Intradural, extramedullary cervical spinal involvement with large cystic or racemose structures and spinal cord tethering is an uncommon manifestation of neurocysticercosis. We present our experience with the diagnosis and operative technique in treating this unique presentation of neurocysticercosis.

Clinical presentation

History and exam

A 49-year-old male originally from Guatemala, who had been in the United States for the past 3 years with a history of pulmonary tuberculosis and seizures presented to the Emergency Department with paresthesias

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predominantly in his fingers bilaterally, which had been slowly progressing to his arms over the previous 2 months. The patient noticed that paresthesias had also been spreading to his lower extremities over the preceding week. In addition, he complained of an increasingly unsteady gait. He was found to be clear of active tuberculosis on this hospital admission. On physical examination, he had patchy loss of sensation in the upper extremities bilaterally. There was also decreased sensation noted in the lower extremities bilaterally below the L1-L2 dermatomes. He had bilateral proximal muscle weakness of the upper and lower extremities at 4 to 4+/5. Deep tendon reflexes were brisk at 3+ and symmetric throughout. He had bilateral Babinski signs. The patient had dysmetria on the finger-to-nose testing and decreased amplitude of rapid alternating movements. His gait was wide-based and unsteady. His bowel, bladder, and sexual function were intact. Ophthalmologic exam was normal.

Magnetic resonance (MR) imaging of the cervical spine (Fig. 1) demonstrated large, enhancing polycystic lesions prominent dorsally, compressing the spinal cord, most prominent at the posterior fossa through C2. There was an additional area of stenosis at C6-C7. A computed tomography (CT) scan of the brain demonstrated multiple calcific nodules throughout the parenchyma, with multiple small enhancing lesions throughout the brain parenchyma on MR imaging (Fig. 2). MR imaging of the thoracolumbar spine demonstrated diffuse enhancement along the cord surface, with large nodules in the lower lumbar region. Laboratory work included western blot analysis which demonstrated positive antibodies for *Taenia solium*. Antibodies were negative for *coccidioides* and *histoplasma*.

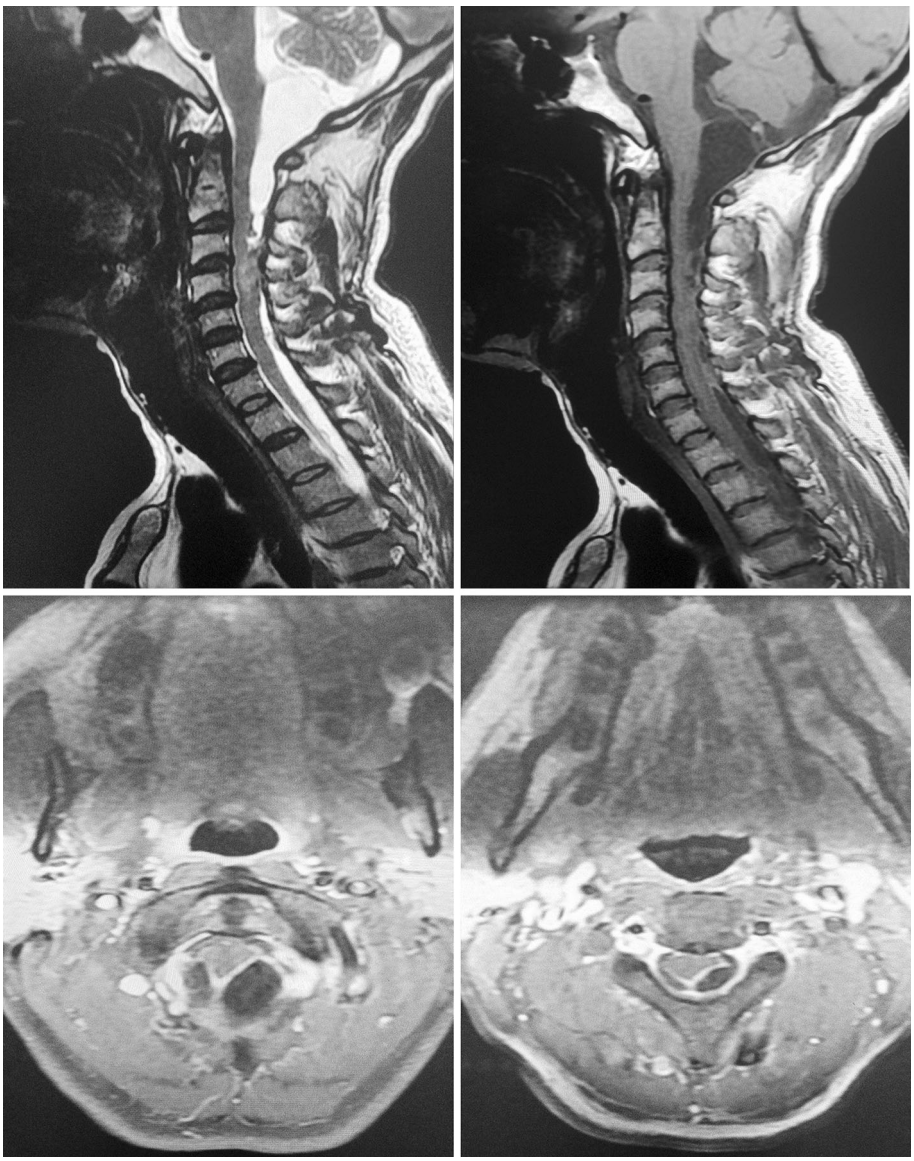

Fig. 1 Preoperative imaging: sagittal fast relaxation fast spin-echo T_2 (*Upper left*) and sagittal T_1 (*Upper right*) MR imaging demonstrating a septated lesion of the cervical medullary junction causing spinal cord compression. Axial T_1 MR imaging with contrast at C1 (*Bottom left*) and at C2 (*Bottom right*) demonstrating midline septation and tethering behind C2 lamina

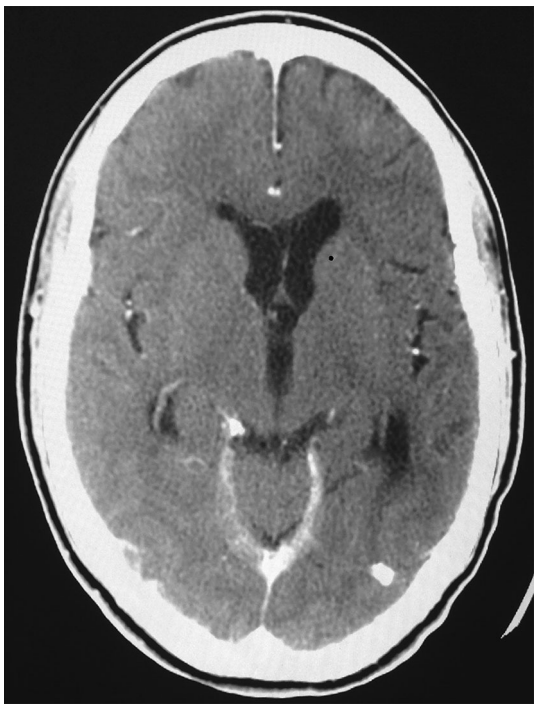

Fig. 2 CT of the head demonstrating multiple calcified nodules throughout the brain parenchyma

Operation

The patient elected surgical treatment for a suboccipital craniectomy, C1 laminectomy, resection of the intradural extramedullary lesions, and untethering of the cervical spinal cord. Upon opening the dura at the level of C1, a markedly thickened and opacified arachnoid was appreciated. Large cystic lesions spontaneously expressed themselves through this dural defect and were extracted with forceps (Video 1). Several of these cysts were submitted for microbiologic study and for frozen section and permanent pathology. The dura was then opened widely with extraction of several smaller cysts. Just below the level of C1, there was a large, abnormal pedicle of tissue, adherent to the arachnoid of the spinal cord. The arachnoid at this site was excised from this tether, and the tethering pedicle was then resected to remove this area of spinal cord tethering. Intraoperatively the patient received dexamethasone, albendazole, and cefazolin.

Pathological findings

On pathologic examination, there were cysts of *Taenia solium* with tegument with fine, basophilic microvilli on the surface of the tegument. Basophilic calcareous bodies were scattered in the stroma of the organisms, as were enlarged, cystic spaces. No scolex was apparent on any of the specimens submitted. A small focus of necrosis of a portion of a cyst, with inflammation, including scattered eosinophils, was present. The pedicle and adjacent arachnoid had fibrosis with collagen and chronic inflammation with foreign body giant cells (Fig. 3).

Postoperative course

Postoperatively the patient continued to receive albendazole and dexamethasone. His paresthesias, dysmetria, and gait improved. Postoperative cervical MR imaging the following day (Fig. 4) demonstrated that all cystic lesions in the cervical spine region had been removed. Imaging 5 months (Fig. 5) postoperatively demonstrated no recurrence of cystic lesions, persistent tethering at C4, and myelomalacia. The patient improved during a 5 month hospital course, prolonged in part due to issues related to placement. His paresthesias continued to improve, and he slowly regained strength through daily physical and occupational therapy. Therapy consisted of gait training and use of assistive devices to help with balance due to poor proprioception. He also had some strength training performed for deconditioning and proximal muscle group weakness. The patient was discharged on gabapentin and baclofen for neuropathic pain and spasticity. The patient did not attend scheduled follow-up in clinic with neurosurgery service. He did have several follow-up visits with neurology which included electromyography of upper extremities, performed approximately 1 year postoperatively for ongoing neuropathic pain in hand, to rule out ulnar neuropathy versus lower cervical radiculopathy. This study was negative. After discharge from hospital he reportedly was in the process of renewing his passport for return to his home country.

Discussion

Intradural, extramedullary cervical spinal neurocysticercosis is a rare disease. In particular, MRI findings of large polycystic lesions in the cervical spine are not typically seen in the presentation of neurocysticercosis. While intramedullary neurocysticercosis is thought to occur through hematogenous dissemination [6], intradural extramedullary neurocysticercosis is the result of direct cerebrospinal fluid dissemination from the cerebrum to the subarachnoid space [4]. Although CT is advantageous in detecting calcifications of neurocysticercosis, MR imaging is superior in detecting spinal lesions [7]. In addition to neuroimaging studies, cerebrospinal fluid analysis, antibody detection, and ultimately histological analysis are used to aid in the diagnosing of neurocysticercosis [8]. The racemose form of neurocysticercosis consists of cysts without a scolex, with the cysts expanding by contiguous

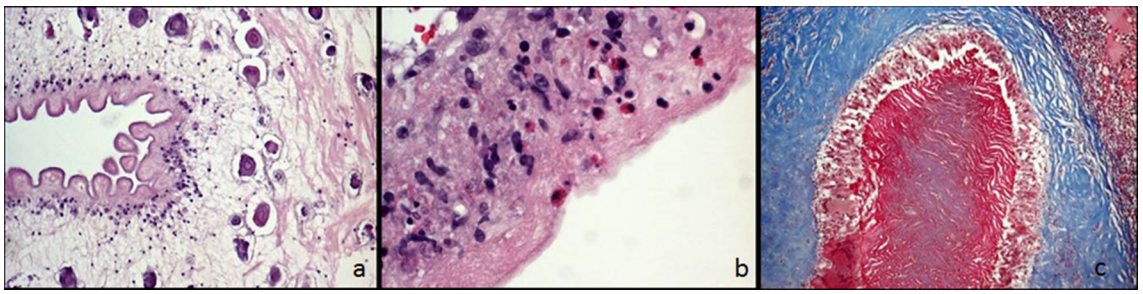

Fig. 3 **a** Large posterior fossa cyst: tegument, with calcareous bodies in stroma. H&E, ×10. **b** Early necrosis of a portion of the organism, with inflammatory reaction, with polymorphonuclear leukocytes, fibrin, and eosinophils, with remnant of tegument at the inferior edge on this image. H&E, ×25. **c** Fibrosis with granulomatous reaction and chronic inflammation. Foreign body giant cells around necrotic material give a "Mandorla" effect. Masson trichrome, ×4

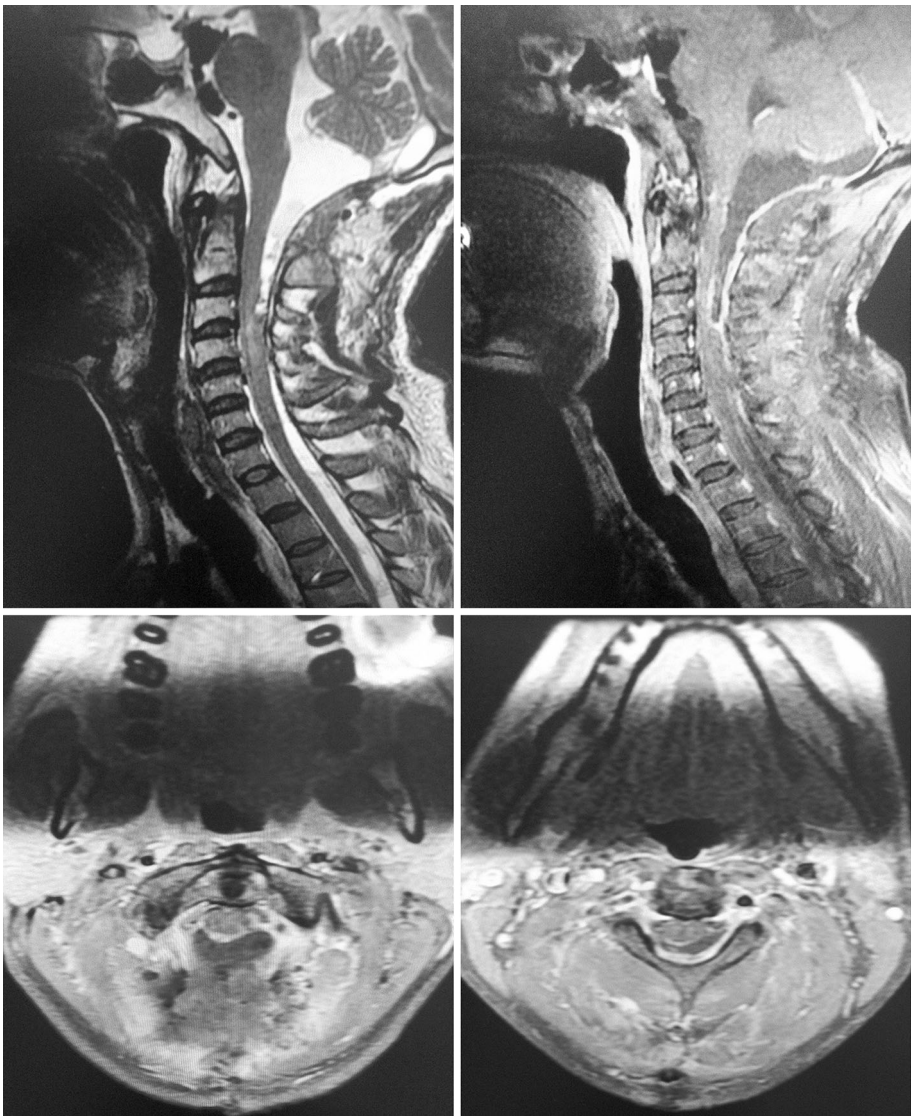

Fig. 4 Postoperative imaging, day 1. Sagittal fast relaxation fast spin-echo T_2 (*Upper left*) and sagittal spin-echo T_1 post gadolinium (*Upper right*) demonstrating removal of the cyst with C4 tethering. Axial T_1 MR imaging with contrast at C1 (*Bottom left*) and at C2 (*Bottom right*) demonstrating removal of the cystic lesion removal posterior to the respective lamina

growth. Such cysts are often large in the posterior fossa, as seen in this patient, and they can extend downward into the spinal subarachnoid space, as also seen in this case [9]. Viable cysts typically do not incite an inflammatory reaction unless the organism dies, which was present focally in this case, with eosinophils involved in the inflammatory reaction. In addition, there were older lesions with fibrosis and a foreign body giant cell reaction, noted in

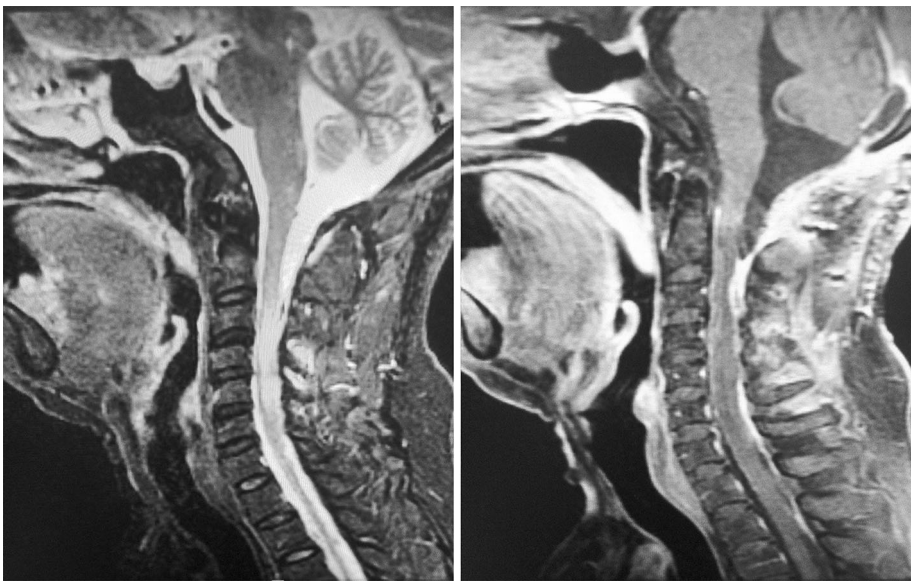

Fig. 5 Postoperative imaging, month 5: sagittal short T_1 inversion recovery (*left*) and sagittal spin-echo T_1 post gadolinium (*right*) demonstrating no recurrence of the cystic lesion, persistent tethering at C4, and myelomalacia. *Note* the foramen of Magendie is widely decompressed

the tethered lesion. Neurocysticercosis of the brain parenchyma is typically treated medically with albendazole and possibly corticosteroids for inflammation [10].

More aggressive surgical intervention may be considered for the treatment of spinal neurocysticercosis [10, 11]. This is true in cases with spinal cord compression and large cystic lesions, as described here. The severe inflammatory processes caused by the cysts, and in particular, from cyst rupture makes gentle manipulation of the intrathecal contents of great importance. Gentle handling of the cyst to avoid spillage, as there may be as in our case adhesions between the cord and the dura or arachnoid, is essential. Gentle manipulation of these membranes during exposure is also vital. Following decompression of the spinal contents, inspection of the spinal canal to ensure that areas of tethering do not exist should be part of standard part of the surgical exploration. Administration of steroids and extensive irrigation of the spinal canal following decompression should help to minimize postsurgical inflammatory processes.

Conclusion

Although intradural extramedullary cervical spinal neurocysticercosis is a rare disease, it should be considered in the differential, especially in patients from endemic areas who have MRI findings of polycystic lesions. Additionally, surgical intervention should be considered as a viable treatment option, particularly in cases with large, complex cystic structures that may be readily evacuated with minimal manipulation of the surrounding neural elements.

Compliance with ethical standards

Conflict of interest statement The authors have no personal or institutional interest with regards to the authorship and/or publication of this manuscript.

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Disclosure The authors have no personal financial or institutional interest in any of the drugs, material, or devices described in this article.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

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