Surgical Resection of Isolated Intramedullary Neurocysticercosis

Neurocisticercose Intramedular Isolada Evidenciada em Ressecção Cirúrgica

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ABSTRACT

Neurocysticercosis is a parasitic disease caused by the cysticerci of Taenia solium. Central nervous system involvement is endemic in underdeveloped countries and intracranial involvement is the most common presentation. Isolated spinal cord involvement is quite rare and definitive diagnosis in most cases is obtained by anatomopathological evaluation. Case Report: 58 year-old female, from Nicaragua, with a 3-year history of dysesthesia and allodynia on right T5 dermatome, with thoracic spine magnetic resonance imaging (MRI) revealing an intramedullary cystic lesion at the level of T4 and T5 vertebrae with perilesional edema. MRI of the rest of the central nervous system was unremarkable. After initial empirical treatment with intravenous methylprednisolone without clinical or radiological improvement, surgical exploration was proposed, confirming the diagnosis of neurocysticercosis. Conclusion: Diagnosing spinal cysticercosis represents a challenge and should be considered in patients with intramedullary cystic lesions in endemic areas. Key words: Neurocysticercosis; Parasitology; Spinal Cord Diseases.

INTRODUCTION

Neurocysticercosis is a parasitic disease caused by accidental ingestion of eggs of Taenia solium, corresponding to the most common parasitic central nervous system (CNS) infection in underdeveloped countries15. The disease occurs when humans become hosts of Taenia solium, through the ingestion of eggs present in food or directly by the fecal-oral transmission5. Cysticercosis is endemic in low income regions, mainly in Africa, Latin America and Asia13,15. Brazil, Peru, Mexico, Korea and India are examples where cysticercosis is common17,18. Even in endemic areas, spinal cord involvement is unusual, about 1.0-5.8% of reported cases, and is even rarer when not associated to intracranial lesions3,18. Currently only 57 intramedullary cysticercosis cases have been reported in the literature15,16. The intramedullary cysticercosis is presented mostly in patients between 20-45 year-old11. According to the location of the cysticercus in spine, it can be...
classified anatomically as extraspinal (vertebral) or intraspinal (epidural, subdural, arachnoidal or intramedullary)\textsuperscript{1,2,10}. The intramedullary type is commonly found in the thoracic region (55.1%), followed by cervical (24.1%), and lumbosacral (20.6%)\textsuperscript{19}. Symptoms are generally associated to the inflammatory reaction and vary according to the number, morphology and the location of the cysts\textsuperscript{1,13,20}. Among the most common symptoms are pain, paraparesis, spasticity, sexual dysfunction, and urinary and bowel incontinence\textsuperscript{1}, in addition to other medullary syndromes\textsuperscript{1}. Diagnosis is suggested by neuroimaging and confirmed by histopathological examination\textsuperscript{5-7}. The authors report a case of isolated intramedullary spinal neurocysticercosis, discussing the rarity of this condition and the management.

**CASE REPORT**

A 58 year-old female, otherwise healthy, born in Nicaragua, but living in Curitiba, Brazil, presented to the Emergency Department of the Neurological Institute of Curitiba with a 3-year history of sporadic right T5 neuropathic pain. The pain worsened in intensity and frequency in the last weeks, and was recently associated with allodynia. The patient denied trauma, fever, weight loss or any other associated symptom.

General physical examination was unremarkable. Neurological examination showed normal strength and reflexes in upper and lower limbs. No signs of pyramidal syndrome were found, and only sensitive changes were noted in the right T5 dermatome.

Thoracic spine MRI showed a T4-T5 intramedullary lesion with perilesional edema (Figure 1a) and contrast enhancement in the anterior region of the lesion (Figure 1b). No other lesions were found on brain and cervical spine MRI. Routine cerebrospinal fluid (CSF) (leucocytes, glucose and proteins), and CSF infectious serology tests (including for *Taenia*) showed no abnormalities, so a 3-day course of intravenous pulsed methylprednisolone was proposed as treatment for an inflammatory lesion. Thirty-day reevaluation showed no clinical improvement and an increase of the lesion on MRI, so the patient underwent surgical exploration of the lesion. Open T4-T5 laminectomy was performed, with complete resection of the lesion. Neurological status remained stable after the procedure, without new deficits. Anatomopathological examination showed a cysticercus (Figure 2). At 6 months follow-up, the patient was asymptomatic, and neurologically intact on physical examination.

**DISCUSSION**

Cysticercosis is endemic in underdeveloped regions, affecting mainly Africa, Latin America and Asia\textsuperscript{15}. Spinal cord location corresponds to only 1.0 to 5.8% of cases of neurocysticercosis, and isolated spinal involvement is extremely rare even in endemic areas\textsuperscript{3,18}, with only 57 cases reported in the literature\textsuperscript{15,16}, making this the 58th report. Time to diagnosis may vary from one week to 10 years\textsuperscript{18}, and symptoms are highly variable, such as pain, paraparesis, spasticity, sexual
The local inflammatory response to the parasite is associated with the development of perilesional edema, which can itself lead to spinal cord injury and consequently worsening of the initial symptoms. In the spinal cord the cysticerci spread most commonly to the thoracic region, which could be explained by the abundant blood supply in this region, which reinforces the theory of hematogenous spread of the parasites. Also, there is the theory of CSF spreading to spinal cord through the ependymal lining.

Cystic lesion on MRI can be hypointense in T1WI with hyperintense scolex inside its cavity, or hyperintense in T2WI on vesicular stage and with discreet hypointensity around the intramedullary cyst on T2WI. On T1WI the contents of the cyst show hyperintensity, and in this case the scolex is not seen. A large amount of edema around the lesion can be seen. Among the differential diagnosis are tumoral and infectious lesions.

After imaging, investigation should focus on serological and cerebrospinal fluid (CSF) investigation of infectious diseases, including cysticercosis. CSF often shows increased protein, normal or low glucose, moderate lymphocytic pleocytosis and eosinophilia. Anti-cysticercus antibodies detected by Enzyme-linked Immunosorbent Assay (ELISA) or by enzyme-linked immunoelectrotransfer blot essay can have good sensitivity and specificity for the diagnosis of cysticercosis. In the case described, laboratory investigation was extensive, including cysticercus in CSF, with negative serology, nonetheless.

Isolated drug treatment in literature showed a few reports of success, and surgical results for intramedullary cysticercosis are not widely conclusive. Mohanty et al. reported a 75% surgical favorable outcome. Sharma et al. evidenced 60% of postoperative clinical improvement and 25% of worsening symptoms, with a 15% mortality rate due to procedure. According to Salazar Noguera et al. since the beginning, surgical treatment must be considered in cases with neurological deficits in order to avoid progressive compression of the spinal cord and subsequent clinical deterioration. Also, they emphasize that anti-parasitic drugs should be given in the postoperative period. Albendazole was used in these reports at a dose of 15 mg/kg/day for 4 to 6 weeks. Jung et al. reported that albendazole associated with corticosteroids may have synergistic effects. Qi et al. suggested that starting the therapy at the preoperative period could be beneficial to consolidate the lesion and to better limit the lesion during surgery. Isolated drug treatment is indicated in patients with high surgical risk or unresectable lesions.

In the reported case, the surgical approach proved essential to the diagnosis and was furthermore, curative, with no need of postoperative drug therapy.

**CONCLUSION**

Intramedullary cysticercosis represents a diagnostic challenge, which must be considered in the presence of an intramedullary cystic lesion especially in regions where cysticercosis is endemic. Surgery should be considered for symptomatic spinal cord decompression, diagnostic confirmation, and in some cases, as definite treatment.

**REFERENCES**


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