Lumbar spinal intradural neurocysticercosis: A case report

SHIZHONG ZHANG[1,2], YANYAN HU[3], ZHEN LI[2], LI ZHAO[3] and ZHIGANG WANG[1]

[1]Department of Neurosurgery, Qilu Hospital of Qingdao Branch, Shandong University, Qingdao, Shandong 370203; [2]Department of Neurosurgery; [3]Color Ultrasonic Room, Taian Central Hospital, Taian, Shandong 271000, P.R. China

Received January 20, 2016; Accepted February 14, 2017

DOI: 10.3892/etm.2017.4403

Abstract. Neurocysticercosis is a parasitic disease that results from the larvae of *Taenia solium*. While neurocysticercosis is considered as the most common parasitic infection of the nervous system, spinal neurocysticercosis is rare. The present case study investigated a 59-year-old woman, who was admitted to our hospital with pain in the bilateral lower limbs and urinary complaints in the form of straining of micturition with increasing frequency. Magnetic resonance imaging revealed an intradural extramedullary lesion extending from L1/2 to S1 of the spine. Subsequently, a laminectomy from L1 to S1 was performed 3 days post-presentation. Intraoperatively, a thin-walled cyst with clear fluid was identified, extending from L2 to S1. The exposed, white and mucoid cysticerci was removed completely. Which on pathological examination was identified to be cysticercosis.

Introduction

Cysticercosis, caused by the tapeworm, *Taenia solium,* is the most common parasitic infection disease of the central nervous system (1). Humans are the only definitive hosts for the adult tapeworm, whereas both pigs and humans may act as intermediate hosts for the larval form, called cysticercus (2). In the normal cycle of transmission, the adult *T. solium* inhabits the small intestine of humans, and gravid proglottids are detached from the distal end of the worm and are passed with the feces to the environment (2). When pigs feed on human feces containing *T. solium* eggs, the eggs subsequently lose their coats and liberate oncospheres, which are infective to humans (2). Thus, humans who eat improperly cooked infected pork meat may themselves become infected (2).

Correspondence to: Dr Shizhong Zhang or Professor Zhigang Wang, Department of Neurosurgery, Qilu Hospital of Qingdao Branch, Shandong University, 758 Hefei Road, Qingdao, Shandong 370203, P.R. China
E-mail: 94439041@qq.com
E-mail: wzg1110@126.com

Key words: cysticercosis, intradural, neurocysticercosis, *Taenia solium*

Cysticercal involvement of the spinal cord is rare and accounts for 0.7-5.85% of all cases of cysticercosis worldwide (3). Cysticercal involvement of the spinal cord is even rare in endemic areas such as Mexico, Central and South America, and parts of Africa and Asia, particularly the Indian subcontinent, and is responsible for more than 50,000 deaths annually in these regions (4-6). Neurocysticercosis is considered a rarer event as the majority of spinal neurocysticercosis cases are accompanied by brain intrusion (1). Treatment for this disease usually includes a combination of symptomatic and cysticidal drugs. Surgical treatment of spinal neurocysticercosis is used when severe symptoms are apparent, symptom aggravation has occurred, medical treatment has failed, and acute aggravation of neurological symptoms has occurred during treatment (7).

The present case study reports an unusual case of neurocysticercosis of isolated spinal involvement. The majority of case reports have not measured the size of the cysticercosis and, to the best of our knowledge, the present case is one of the largest lumbar spinal intradural neurocysticercosis in literature, with its longest axis in magnetic resonance imaging (MRI) being ~16 cm.

Case report

A 59-year-old women was admitted to the Department of Neurosurgery of Qilu Hospital of Qingdao Branch (Qingdao, China), in May 2014 with pain in the bilateral lower limbs for the previous 22 months and gradually progressive pain of the left lower limb for 26 days. The patient presented with increasing urinary frequency. She did not complain of weakness in the upper and lower limbs. The study was approved by the Ethics Committee of Qilu Hospital of Qingdao Branch and the patient provided written informed consent.

The patient was a resident in North China, which is not an endemic region for neurocysticercosis (2). Clinical physical examination revealed the patient exhibited normal cranial nerve evaluation and intact higher mental functions. Deep tendon reflexes were brisk with bilateral positive Babinski's sign of the two legs. On sensory examination, the bilateral lower limbs suffered hypoesthesia along S2 to S5 dermatomes, with decreased perianal sensation exhibited on the left side. Examination of upper extremities did not reveal any abnormalities.

MRI of the patient's spine revealed an intradural extramedullary lesion extending from L1/2 to S1, which was hyperintense in T2- and hypointense in T1-weighted images

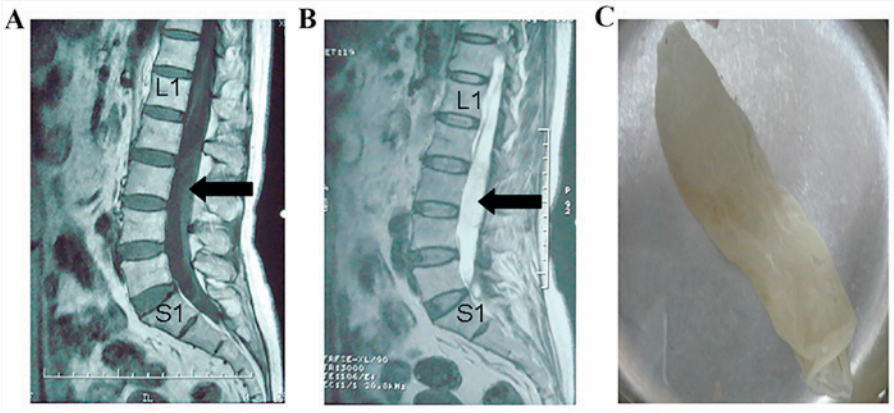

Figure 1. MRI detected a 16-cm cystic mass on L2 to S1. The MRI signal intensity of the lesion was high in the (A) T2-weighted image and low in the (B) T1-weighted image. (C) Intraoperative image of the white/cream colored mucoid cysticerci that was removed. The arrow indicates the cystic mass. MRI, magnetic resonance imaging.

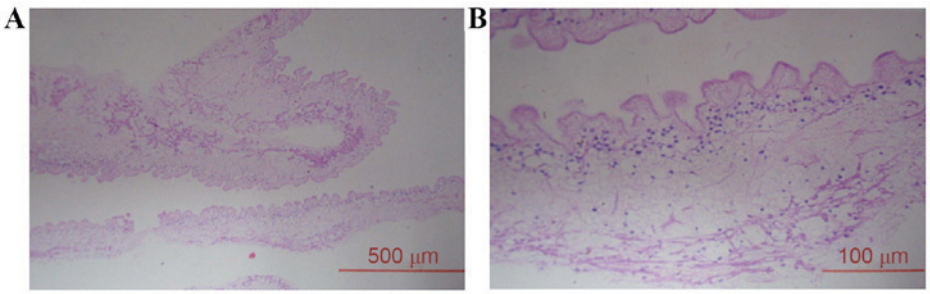

Figure 2. Hematoxylin and eosin staining indicated an eosinophilic outer cuticle layer and a single layered sub-cuticle cell, representing typical cysticercosis findings, at magnification (A) x100 and (B) x400.

(Fig. 1A and B). The patient underwent a laminectomy from L1 to S1 3 days post-presentation. Intraoperatively, a thin-walled cyst was identified with clear fluid, extending from L2 to S1, measuring ~8 cm after discharge of part of the cyst fluid. The entire cystic mass was removed (Fig. 1C) and its longest axis was ~16 cm in MRI. An ELISA test (Human Cysticercosis antibody, CYT Ab ELISA kit; Nanjing SenBeiJia Biological Technology Co., Ltd., Nanjing, China) was performed, according to the manufacturer's instructions. Results demonstrated that the cerebrospinal fluid (CSF) and cyst fluid were positive for anti-*T. solium* immunoglobulin (Ig)G. After removal, the cyst was immediately fixed in 10% formalin for ~8 h at room temperature, subjected to hematoxylin and eosin staining and sliced into 5-μm sections. The sections were viewed under a light microscope and the histopathology of the mass was consistent with a cysticercus cyst (Fig. 2A and B). Following surgery, the patient was treated with albendazole (15 mg/kg/day; GlaxoSmithKline, Tianjin, China) for 4 weeks and exhibited no additional complications. The patient recovered well in the 6 weeks following the laminectomy. No dysfunction concerned with this disease was observed in the patient 6 months subsequent to surgery.

Discussion

Spinal neurocysticercosis is an uncommon variation of cysticercosis and isolated intradural extramedullary or intramedullary is considered to be the rarest form (8). Variations in symptoms are often exhibited in spinal neurocysticercosis, depending on the location, size, number of cysts and the presence or absence of any inflammation of the lesion (1,9). Types of spinal neurocysticercosis have been identified in the vertebral, extradural, intradural and intramedullary region (10). The clinical features of spinal cysticercosis include paresis, sensory loss, autonomic dysfunction involving the bowel and bladder, radicular pain and paresthesia (5,6).

With respect to serologic diagnosis, serum and CSF ELISA tests have aided to diagnose cysticercosis (11,12). The sensitivity and the specificity of the ELISA test is ~50 and 70% for serum and ~87 and 97% for CSF, respectively (13). The present case study used an ELISA test for anti-*T. solium* IgG in the CSF and cyst fluid, which were positive and successfully indicated *T. solium*.

Furthermore, MRI is considered the optimal testing method for diagnosing spinal neurocysticercosis (14,15). MRI typically exhibits lesions as hypointense in T1-weighted images and hyperintense in T2-weighted images (13,16).

Various therapeutic options are available to treat spinal neurocysticercosis. Medical treatment with albendazole is considered effective in neurologically stable patients (13) and is the preferred treatment over praziquantel by multiple researchers, due to the higher CSF penetration of the agent and a possible drug serum concentration increase in combination with steroid administration (11,17). However, when patients present with an acute or progressively deteriorating neurological state or where diagnosis is missed or in doubt,

surgical excision is the preferential choice of the treatment. Moreover, histopathology confirms the diagnosis and early surgery provides recovery before any irreversible cord damage occurs (5).

To conclude, the present case study indicates a specific case of rare spinal neurocysticercosis. Urgent surgical decompression and medical treatment have vital roles in the management of this condition.

References

1. Alsina GA, Johnson JP, McBride DQ, Rhoten PR, Mehringer CM and Stokes JK: Spinal neurocysticercosis. Neurosurg Focus 12: e8, 2002.
2. Del Brutto OH: Neurocysticercosis: A review. ScientificWorld Journal 2012: 159821, 2012.
3. Guedes-Corrêa JF, Macedo RC, Vaitsman RP, Mattos JG and Agra JM: Intramedullary spinal cysticercosis simulating a conus medullaris tumor: Case report. Arq Neuropsiquiatr 64: 149-152, 2006.
4. Flisser A, Sarti E, Lightowlers M and Schantz P: Neurocysticercosis: Regional status, epidemiology, impact and control measures in the Americas. Acta Trop 87: 43-51, 2003.
5. Rajshekhar V, Joshi DD, Doanh NQ, van De N and Xiaonong Z: *Taenia solium* taeniosis/cysticercosis in Asia: Epidemiology, impact and issues. Acta Trop 87: 53-60, 2003.
6. Qazi Z, Ojha BK, Chandra A, Singh SK, Srivastava C and Patil TB: Isolated intramedullary spinal cord cysticercosis. J Neurosci Rural Pract 5 (Suppl 1): S66-S68, 2014.
7. Nash TE, Singh G, White AC, Rajshekhar V, Loeb JA, Proaño JV, Takayanagui OM, Gonzalez AE, Butman JA, DeGiorgio C, *et al*: Treatment of neurocysticercosis: Current status and future research needs. Neurology 67: 1120-1127, 2006.
8. Sotelo J, Guerrero V and Rubio F: Neurocysticercosis: A new classification based on active and inactive forms. A study of 753 cases. Arch Intern Med 145: 442-445, 1985.
9. Ahmad FU and Sharma BS: Treatment of intramedullary spinal cysticercosis: Report of 2 cases and review of literature. Surg Neurol 67: 74-77, 2007.
10. Salazar Noguera EM, Pineda Sic R and Escoto Solis F: Intramedullary spinal cord neurocysticercosis presenting as Brown-Séquard syndrome. BMC Neurol 15: 1, 2015.
11. Homans J, Khoo L, Chen T, Commins DL, Ahmed J and Kovacs A: Spinal intramedullary cysticercosis in a five-year-old child: Case report and review of the literature. Pediatr Infect Dis J 20: 904-908, 2001.
12. Rosas N, Sotelo J and Nieto D: ELISA in the diagnosis of neurocysticercosis. Arch Neurol 43: 353-356, 1986.
13. Garcia HH and Del Brutto OH; Cysticercosis Working Group in Peru: Neurocysticercosis: Updated concepts about an old disease. Lancet Neurol 4: 653-661, 2005.
14. Rahalkar MD, Shetty DD, Kelkar AB, Kelkar AA, Kinare AS and Ambardekar ST: The many faces of cysticercosis. Clin Radiol 55: 668-674, 2000.
15. Han SB, Kwon HJ, Choi SW, Koh HS, Kim SH, Song SH and Youm JY: Lumbar intradural neurocysticercosis: A case report. Korean J Spine 11: 205-208, 2014.
16. Ganesan S, Acharya S, Kalra KL and Chahal R: Intradural neurocysticercosis of lumbar spine: A case report. Global Spine J 5: e1-e4, 2015.
17. Sotelo J and Del Brutto OH: Review of neurocysticercosis. Neurosurg Focus 12: e1, 2002.