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Case Report

Spinal intramedullary cysticercosis with syringomyelia: a case report

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Abstract: Intramedullary cysticercosis of the cervical spine is extraordinarily rare; prior reports are limited to single cases. We review cases of intramedullary cysticercosis, and summarize the features and outcome. Herein, we report a 38 year old woman with progressive quadriplegia, paresthesia in bilateral upper limbs, neck pain and headache for 1 month. She had dyspnea for 1 week. A gross total resection was performed, and after the surgery, the patient was given prednisolone per day orally, for 2 weeks. Oral albendazole 400 mg/day was started 2 days after the start of prednisolone. Ag-ELISA was performed 2 months after the completion of treatment and no residual lesion was seen. At the 6-month postsurgical follow-up, no recurrence of the cysticercosis was noted. Cysticercosis of the cervical spine is extraordinarily rare. Preoperative identification of intramedullary cysticercosis is challenging, and the exact diagnosis depends on histopathological evidence and Ag-ELISA. With symptoms of the nervous system, surgical resection should be performed in time.

Keywords: Cysticercosis, intramedullary, diagnosis, surgical resection

Introduction

Cerebral cysticercosis is a common intracranial disease, but its incidence is very low in the spinal cord [1]. In patients with neurocysticercosis, the incidence of spinal cysticercosis is 0.7%-3.0%, and ordinarily, patients with spinal cysticercosis also have intracranial cysticercosis [2]. Isolated intramedullary cysticercosis of the cervical spine is extraordinarily rare, with only 85 cases reported. Because of its rarity, its radiologic and clinical characteristics and natural medical history are not well understood. To our knowledge, all formerly described intramedullary cysticercosis of the spine cases did not have clinical, radiological and location characteristics described.

Here we report a case of cervical intramedullary cysticercosis. The clinical, radiological, and location characteristics are presented, and related literature is reviewed.

Case report

History and examinations

A 38 year old woman presented to the Department of Neurosurgery with progressive quadriplegia, paresthesia in bilateral upper limbs, neck pain, and headache for 1 month. She had dyspnea for a week. She had a history of consumption of pork. There were no bowel or bladder symptoms. Neurological examination revealed the muscle strength of extremities and nervous reflexes were all normal, while high muscle tension in both lower extremities. MRI of the cervical spine (**Figure 1A**, **1B**) showed an intramedullary cystic solid mass at the C1-C2 vertebral level, which was hypointense on T1 weighted and hyperintense on T2 weighted images with peripheral. The lesion had an intracystic hypointense on T2 weighted. Contrast-enhanced MRI (**Figure 1C**) showed a clear, peripherally enhanced cystic lesion and a solid lesion with heterogeneous enhancement.

Surgery and pathology

C1-C2 laminoplastic laminotomy was performed under general anesthesia. Intraoperatively, we found that the spinal cord was swollen. Masses were all located in the intramedullary portion. When a midline myelotomy was performed several white cystic solid masses were seen and clear fluid was then sucked out.

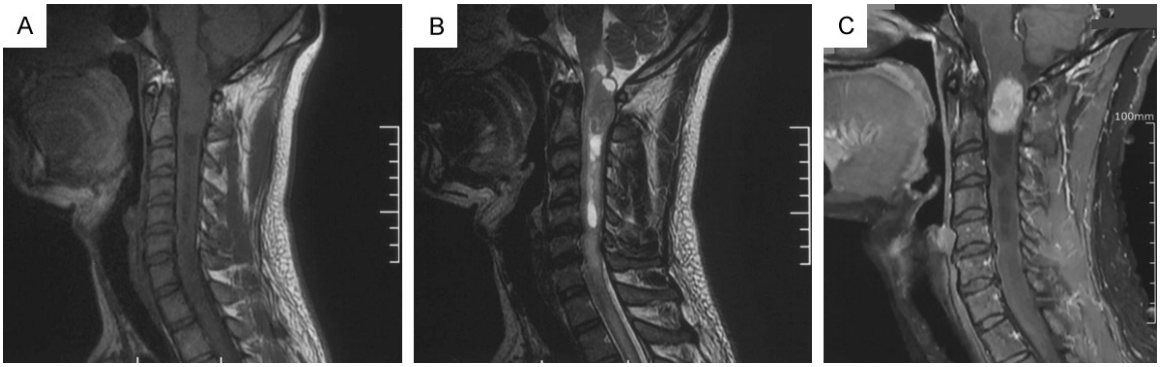

Figure 1. MRI of the cervical spine. (A, B) an intramedullary cystic solid mass at the C1-C2 vertebral level, which was hypointense on T1 weighted and hyperintense on T2 weighted images with peripheral. The lesion had an intracystic hypointensity on T2 weighted. Contrast-enhanced MRI (C) showed a clear, peripherally enhanced cystic lesion and a solid lesion with heterogeneous enhancement.

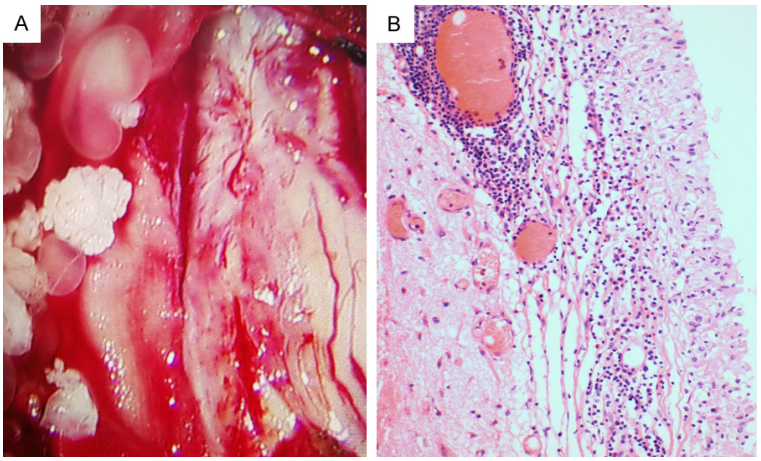

Figure 2. A. General examination of the lesion revealed several ovals, white, smooth, semitransparent, and friable cystic solid masses. B. Pathologic section (original magnification ×200) of the cysticercus is composed of loose fibrous connective tissue, eosinophilic lining, and filled with clear fluid with chronic inflammatory cells.

General examination of the lesion revealed several ovals, white, smooth, semitransparent, and friable cystic solid masses (**Figure 2A**). The cyst wall and solid part stuck slightly to the surrounding spinal cord. In order to dissect the cyst with minimal injury to the surrounding tissue, we withdraw a part of the cystic liquid first. Liquid was yellowish and transparent; then the cyst wall became slackened, and the cyst wall and solid part were removed totally.

Histopathological examination of the cyst showed (original magnification ×200) the cysticercus composed of loose fibrous connective tissue and the eosinophilic lining. It also had clear fluid with chronic inflammatory cells (**Figure 2B**). After surgery, the serum Ag-ELISA was performed, the result was positive.

Postoperative course

Neck pain and headache disappeared after surgery, muscle strength improved significantly, and there was no sphincter dysfunction. Muscle tension of lower extremities was lower than before. Paresthesia was alleviated partially. The patient was given prednisolone per day orally, for 2 weeks. Oral albendazole 400 mg/day was started 2 days after the start of prednisolone. Ag-ELISA was performed after 2 months of finishing the treatment and was negative suggestive of no residual lesion. At the 6-month postsurgical follow-up, no recurrence of the cysticercosis was noted (**Figure 3A-C**).

Summary of reported cases

In the literature, 85 cases of intramedullary cysticercosis were identified in 57 articles. There were only 51 cases of intramedullary cysticercosis that contained lesion's location, clinical symptoms, treatment, laboratory examination, follow-up, and MRI reported in 41 articles [1, 3-42]. Together with our case, there are 52 cases that can be analyzed.

The clinical and radiological characteristics of reported cases are summarized in **Table 1**.

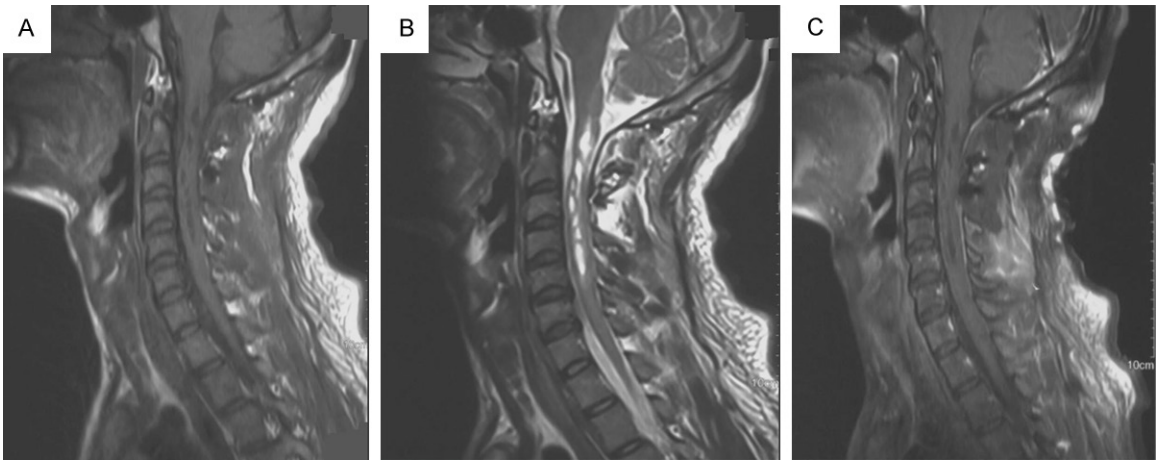

Figure 3. A-C. MRI of 6-month postsurgical follow-up: no recurrence of the cysticercosis was noted.

Discussion

Rockitansky reported the first case of intraspinal cysticercosis in 1856 [1]. Intramedullary spinal cysticercosis was considered extremely rare, up to now, less than 100 cases have been reported. Compared to intramedullary infestation, the greater incidence of brain involvement is postulated to be because 100 times more blood flows into the brain than the spine [1]. Our case is isolated intramedullary cysticercosis.

According to clinical and radiologic characteristics documented in literatures, most intramedullary spinal cysticercosis were located in the thoracic segment. There are four cases reported at the lumbosacral level (including the conus medulla) [15, 20, 32, 34].

From the literature review, we found that 52 patients (including our case) of intramedullary cysticercosis were described in detail so that we can accurately collect its clinical characteristics, imaging performance, treatment methods, and prognosis.

Intramedullary cysticercosis has no typical signs and symptoms, which may include myelopathy or radiculopathy. Specific manifestations depend mainly on the location of cysts and sometimes mimic more common neuropathology [1]. Homans [10] and Singh [15] reported patients presented with the conus medullary syndrome. Noguera [35] and Rice [31] reported patients presented with the Brown-Sequard syndrome. The weakness of limbs, impairment of sensory modalities, and back pain were prominent complaints.

The etiology of intramedullary cysticercosis is well known. Most patients of intramedullary cysticercosis are found in the thoracic segment of the spinal cord. The main reason is that cysticercosis spreads into the spinal cord parenchyma through the blood is through main blood vessels supplying the spinal cord called Adamkiewicz.

Radiologic manifestations of intramedullary spinal cysticercosis are specific. All the reported cases lesions and the current case appeared as fluid-filled cysts. 81.25% of the reported cases were hypointense on T1WI and 93.55% of them were hyperintense on T2WI sequences. We also found that most of them were surrounded by edema, causing focal enlargement of the spinal cord. Most of them had an intramedullary ring-enhancing lesion. There was an internal high-intensity point surrounding the scolex of the parasite that was seen in some patients, which is a characteristic MRI manifestation of intramedullary cysticercosis.

Due to the low incidence of intramedullary cysticercosis, the most appropriate treatment strategy remains unclear. 33 of the 52 patients had their intramedullary cysts removed by laminectomy and subsequent myelotomy.

Conclusion

Intramedullary cysticercosis is extremely rare but has serious clinical symptoms. The preop-

Table 1. Summary of previous cases with intramedullary cysticercosis

Characteristics	Case number (N)	Percentage
Gender	n=52	
Male	n=35	67.31%
Famle	n=17	23.69%
Areas	n=52	
cysticercosis-endemic areas	n=46	88.46%
India	n=35	67.31%
Brazil	n=5	9.62%
China	n=3	5.77%
Mexico	n=1	1.92%
Guatemala	n=1	1.92%
Thailand	n=1	1.92%
developed countries	n=6	11.54%
USA	n=5	9.62%
Korea	n=1	1.92%
Duration of clinical symptoms	n=43	
acutely	n=9	20.93%
chronically	n=34	79.07%
Main clinical symptoms	n=52	
Limb weakness	n=26	50.00%
Limb pain or paraesthesia	n=23	44.23%
Bladder symptoms	n=23	44.23%
Back pain or paraesthesia	n=19	36.54%
Paraparesis	n=12	23.08%
Bowel symptoms	n=8	15.38%
Neck pain	n=4	7.69%
Seizure	n=3	5.77%
Locations	n=52	
Cervical	n=15	28.85%
Cervicothoracic	n=3	5.77%
Thoracic	n=30	57.69%
Thoracolumbar	n=2	3.85%
Lumbar	n=1	1.92%
MRI T1-weighted imaging	n=32	
Hypointensity	n=26	81.25%
Isointensity	n=4	12.5%
Hyperintensity	n=2	6.25%
MRI T2-weighted imaging	n=31	
Hypointensity	n=1	3.23%
Isointensity	n=1	3.23%
Hyperintensity	n=29	93.55%
MRI enhancement	n=31	
enhancement was “ring-like”	n=26	83.87%
enhancement was “nodular”	n=5	16.13%

erative identification is challenging, and accurate interpretation of MRI findings is helpful for diagnosis. Gold standards for diagnosis are histopathologic evidence and Ag-ELISA. We think that patients with neurological symptoms should be treated with surgery, otherwise they should be treated with drugs first. Surgical resection can help relieve the symptoms, whereas glucocorticoids and anti-cysticercosis drugs (include albendazole and praziquantel) should be used in combination after surgery to reduce the rate of temporary clinical deterioration due to parasite destruction causing secondary inflammatory changes in the spinal cord.

Disclosure of conflict of interest

None.

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