

CASE REPORT

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# Case report: a rare case of intradural and pleural wall cystic echinococcosis

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## Abstract

**Background** Cystic echinococcosis (CE) is prevalent in livestock farming regions around the world. However, it remains relatively rare compared to other infectious diseases. CE typically affects the liver, lungs, brain, and kidneys. Spinal and pleural wall involvement is exceedingly rare. We report a unique case of intradural and pleural wall CE in a young male, successfully treated with surgery and postoperative medication.

**Case presentation** A 19-year-old Tibetan male from the Qinghai-Tibet Plateau was diagnosed with intradural and pleural wall CE through imaging, serology, and surgical pathology. According to the Dew/Braithwaite & Lees (BL) classification, his condition was an exceptionally rare form of spinal echinococcosis, compounded by an even rarer pleural wall involvement. Prompt surgical intervention and postoperative medication resulted in significant improvement in spinal cord compression symptoms.

**Conclusions** This case highlights the diagnostic and therapeutic challenges of rare CE locations. MRI proved superior to CT in diagnosing bony cystic echinococcosis. Early surgical intervention combined with medication facilitates spinal cord function recovery, providing valuable insights for managing similar cases.

**Keywords** Case report, Spinal cystic echinococcosis, Pleural wall cystic echinococcosis, *Echinococcus granulosus*, Hydatid disease

## Background

Cystic echinococcosis (CE) is a parasitic disease with a relatively low incidence in China compared to other infectious diseases [1]. *Echinococcus granulosus* is a small cestode parasite with canid as its definitive hosts. Humans and livestock become accidental hosts by

ingesting contaminated food or water. CE is the most common form in endemic areas [2, 3]. Some literature points out that the World Health Organization (WHO) has classified echinococcosis as one of the 17 neglected diseases targeted for control or elimination by 2050 [4]. CE is widely distributed in livestock farming regions worldwide, while AE is predominantly found in livestock areas of the northern hemisphere, and both diseases have a relatively insidious onset [5]. So compared to alveolar echinococcosis (AE), CE is more widely distributed, and like AE, it is easily overlooked by the general public and healthcare professionals.. CE primarily affects solid organs like the liver, kidneys, and lungs, with bone involvement accounting for less than 4% of cases [6, 7]. In humans, cysts primarily form in the liver and lungs, with bone and pleural wall involvement being rare. Intradural and pleural wall cystic echinococcosis (CE) occurring simultaneously in a single individual is exceedingly

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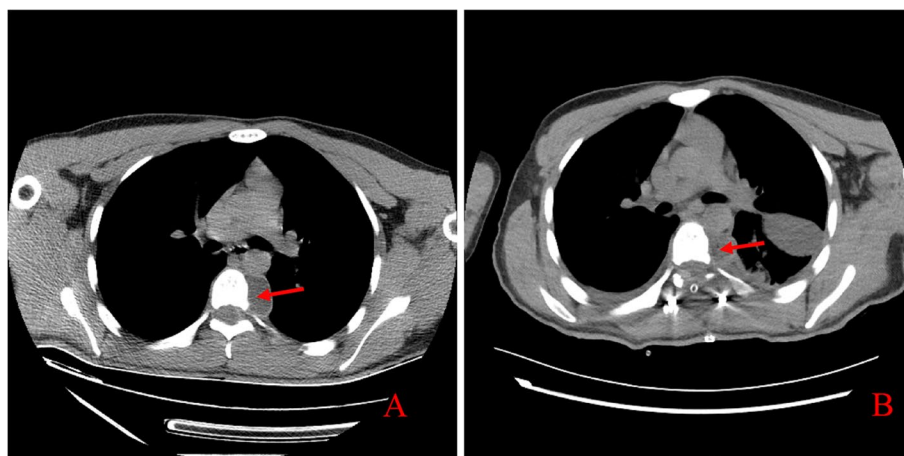
rare, and to our knowledge, there have been no previous reports of such a case. This report presents a rare case of intradural and pleural wall CE diagnosed using MRI, enzyme-linked immunosorbent assay (ELISA) from Haitai and histopathological examination and treated successfully with surgery and medication.

### Case presentation

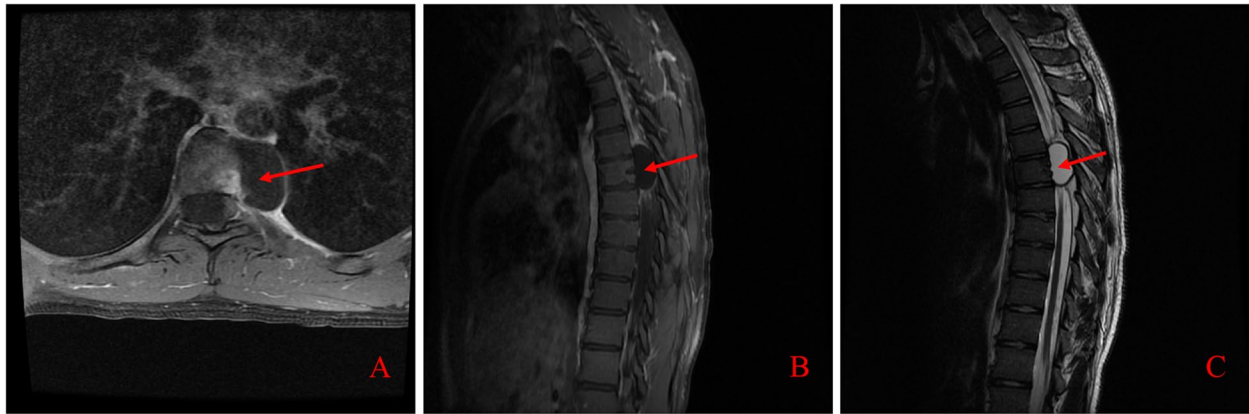
The patient is a 19-year-old male herder living in a pastoral area with a history of contact with cattle and sheep. His family keeps Tibetan mastiffs, a large breed of dog; therefore, he also had close contact with dogs. About six months before admission, he began experiencing dull mid-back pain without any obvious cause, which he did not take seriously. He self-medicated with analgesics (specific medications unknown) with poor results. It was not until four months before admission, when he started experiencing intermittent dizziness, numbness in his limbs, and worsening mid-back pain, that he sought medical attention at a local hospital. The patient has a history of pulmonary tuberculosis diagnosed 11 years ago, which he claims has been "cured" through Tibetan medicine. The local hospital suspected "tuberculous meningitis" and "secondary spinal tuberculosis," but due to the limited medical capabilities of the local hospital, the patient came to our hospital for further diagnosis and treatment. Upon admission, the patient was conscious, showed no significant weight loss, had no night sweats, and maintained normal bowel movements, urination, and appetite. On admission, physical examination revealed tenderness in the thoracolumbar region, positive paraspinal percussion pain, and severe lower limb weakness (0/5 muscle strength according to Medical Research Council grading)

with increased muscle tone (4/5 on the Ashworth scale). Reflexes were diminished, and Babinski, Chaddock, Oppenheim, and Gordon signs were positive.

We conducted abdominal ultrasound, as well as CT scans of the upper, middle, and lower abdomen, chest, and head, to rule out tuberculosis or related cranial diseases in the patient. The results showed only mild inflammation in the lower lobes of both lungs on the chest CT scan, with no abnormalities in the abdominal organs or head. CT scans for tuberculosis often show vertebral body destruction, vertebral dissolution, and bone destruction, while chest CT reveals a soft tissue mass adjacent to the T6 vertebra, connected to the spinal canal, without bone destruction (Fig. 1). Additionally, the patient does not exhibit systemic symptoms of tuberculosis, such as low-grade fever, night sweats, or weight loss. Therefore, we have initially ruled out tuberculosis-related diseases. Differential diagnosis included CE and neurogenic tumors. Further MRI revealed irregular cystic long T2 signal lesions in the posterior spinal canal and left paraspinal area at T6, compressing the thoracic spinal cord (Fig. 2). In the Haitai ELISA procedure, soluble antigen is mixed with the patient's serum, where echinococcal antibodies (IgG) in the serum bind to target molecules in the sample. An enzyme-labeled secondary antibody is added to form an "antigen-antibody-enzyme" complex. A substrate for the enzyme is added, and the intensity of the resulting color reaction correlates with the concentration of the target molecule in the sample. A spectrophotometer measures the color intensity, and the optical density (OD) value obtained is compared to a standard curve to quantify the concentration of the target molecule. S/C0 stands for "Sample/Control 0," representing the ratio



**Fig. 1** **A** Preoperative CT; The image shows paravertebral cystic soft tissue nodules, but it is impossible to accurately determine whether they communicate with the spinal canal (red arrow). **B** Postoperative CT; The image shows postoperative changes in the vertebral body and excision of cystic soft tissue (red arrow)

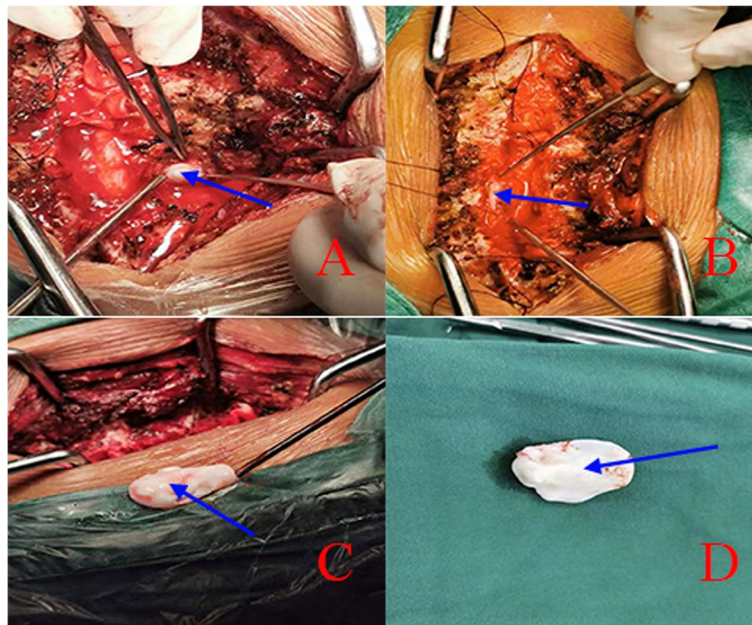


**Fig. 2** Coronal and Sagittal MRI images of the spine. **A** Coronal T1-weighted MRI image (red arrow). **B** Sagittal T1-weighted MRI image (red arrow). **C** Sagittal T2-weighted MRI image (red arrow)

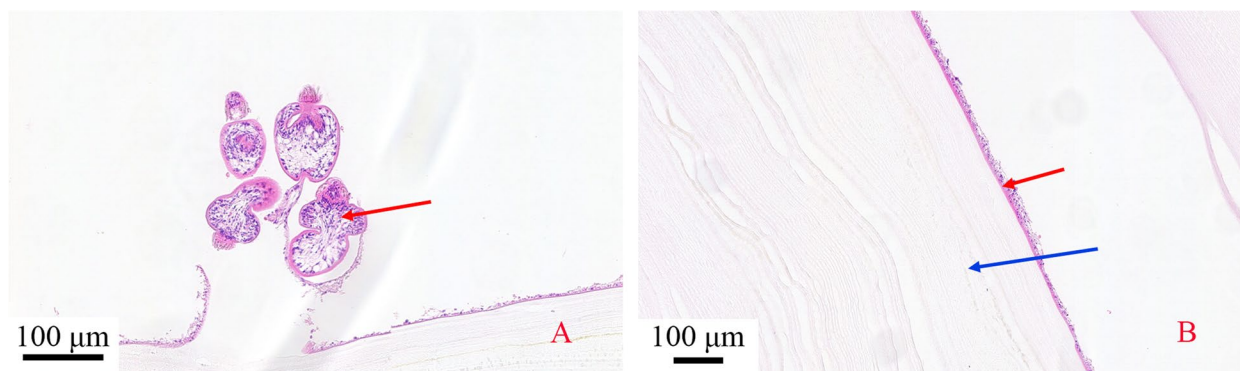
of the sample signal (S) to the control zero signal (C0). A higher S/C0 value typically indicates a higher concentration of echinococcal IgG in the sample. Haitai ELISA uses 0.9(S/C0) as the cutoff for a positive test result. Haitai ELISA for *Echinococcus* IgG was positive (13.75 S/C0). Based on his lifestyle (History of exposure to dogs, cattle, and sheep) and diagnostic results, spinal CE was highly suspected.

After thorough preoperative evaluation, surgery was performed the next day. A midline incision was made from T4 to T5, with removal of T5-T7 spinous

processes and laminectomy. A 22×36×20 mm cystic mass was found adherent to the dura, severely compressing the spinal cord. We used pads soaked in povidone-iodine to prevent spillage of parasitic contents, aspirated the cyst contents, and completely resected the cyst wall. No cyst fluid leakage occurred during the procedure. Additionally, a 30×20×32 mm cystic mass was found on the pleural wall, which was similarly excised after aspiration. Intraoperative images (Fig. 3) and pathological specimens (Fig. 4) were taken. The pathological image clearly shows the characteristic



**Fig. 3** Intraoperative images. **A** Cyst in the Intradural space (blue arrow). **B** Cyst in the pleural wall (blue arrow). **C** Excised hydatid cyst from the Intradural space (blue arrow). **D** Excised hydatid cyst from the pleural wall (blue arrow)



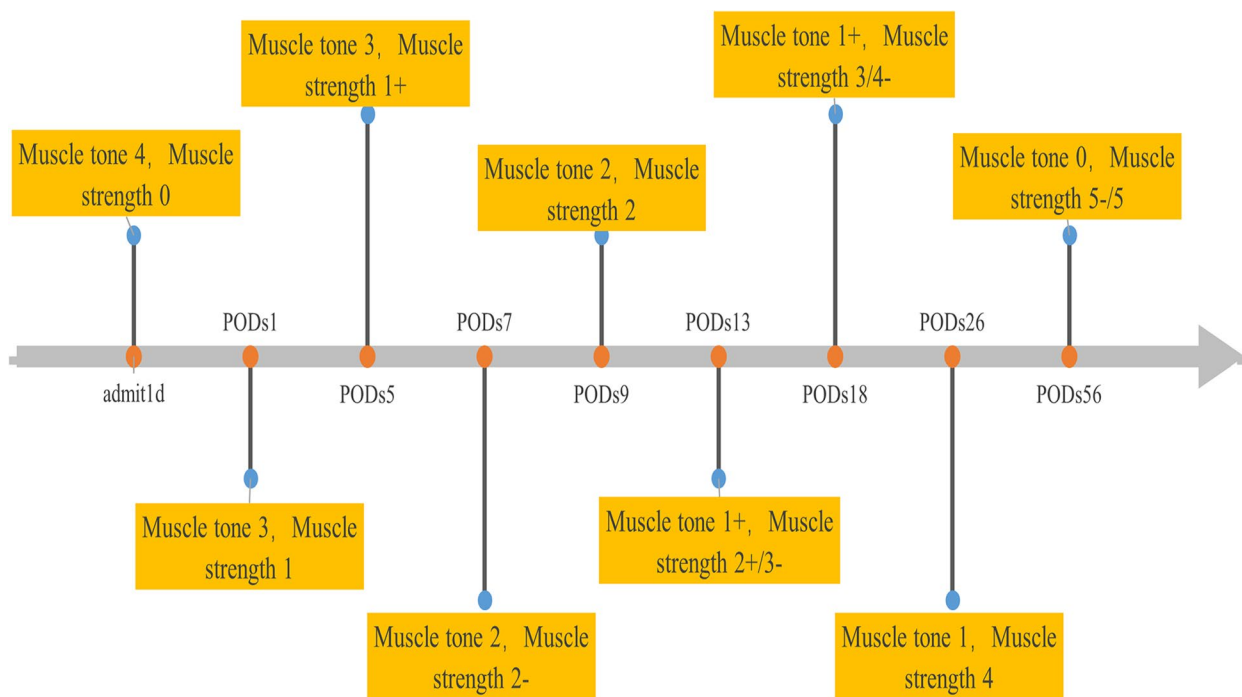
**Fig. 4** Histopathology image. **A** In the periphery of the germinal layer, distinct protoscolices are visible, with prominent small hooks (red arrow) inside the scolex. **B** In the middle laminated layer, a distinct lamellar structure is visible (blue arrow); the germinal layer consists of a single or multiple layers of germinal cells (red arrow)

features of hydatid cysts. The cyst wall includes a distinct lamellated structure of the middle laminated layer and a germinal layer composed of germinal cells. Inside the cyst, the protoscolices are visible, appearing as oval or round structures with hooks, which is typical for CE. The surgical site was irrigated with povidone-iodine and saline, and the wound was closed in layers. The operation lasted three hours. On the first day after surgery, the patient experienced some relief from numbness in the limbs but still felt dizziness and discomfort in the surgical area of the back. On the third day post-surgery, the patient developed mild abdominal discomfort. By around the tenth day after surgery, the dizziness had significantly subsided. On the 32nd day post-surgery, the dizziness completely disappeared, but the patient developed symptoms of indigestion and started taking omeprazole on their own. By the 56th day post-surgery, the numbness in the limbs had almost completely disappeared, and the pain in the chest and back was entirely gone, though occasional abdominal discomfort persisted. The patient did not take albendazole (ABZ) preoperatively because a definitive diagnosis of CE (cystic echinococcosis) had not been established. On postoperative day one, the patient was treated with ABZ 400 mg twice daily and will continuously take this medication for 2 months. The patient experienced only mild gastrointestinal symptoms such as abdominal discomfort, bloating, and dyspepsia following ABZ administration, without any other notable adverse effects. His recovery, including muscle strength and tone, was monitored over time (Fig. 5). One year post-surgery, follow-up at 1, 3, 6, and 12 months showed excellent recovery according to the American Spinal Injury Association (ASIA) scale (Table 1).

## Discussion and Conclusions

According to BL classification [8], this case represents a rare BL2 type of spinal CE, combined with pleural wall CE, both being independent cysts. Compared to some other cases [9], we further utilized MRI to enhance diagnostic accuracy. While performing adequate laminectomy and cyst removal, we also applied pedicle fixation to maintain spinal stability.

The slow growth of hydatid cysts and long latency [10] contribute to delayed diagnosis until significant symptoms like spinal cord compression occur. The rarity of CE complicates differential diagnosis, often requiring imaging and serological tests [11], and is ultimately confirmed through histopathological examination. The CT scan of this case shows a cystic lesion with clear boundaries and homogeneous content. On MRI, it appears as a water-like cyst with clear margins. In contrast, AE often present with unclear boundaries and heterogeneous content on CT, and on MRI, they show infiltrative growth with poorly defined margins and surrounding edema [12]. Therefore, we preliminarily excluded AE. Pathologically, this case displays the typical lamellated structure of cystic echinococcosis with a well-defined laminated layer and germinal layer, with minimal eosinophil and macrophage infiltration. In comparison, alveolar echinococcosis often lacks a distinct lamellated structure, shows significant necrotic tissue around it, and has a poorly defined germinal layer with difficulty in observing protoscolices. Surrounding tissue may exhibit substantial eosinophil and macrophage infiltration, along with granuloma formation and fibrous tissue proliferation [13]. Thus, we confirmed the diagnosis of CE for this patient. Diagnostic and therapeutic experience for these rare locations is scarce. While ultrasound is an effective tool



**Fig. 5** Timeline of recovery of muscle strength and tone. MRC was used as the grading standard of muscle strength, and Ashworth scoring system was used as the scoring standard of muscle tension; Abbreviation: Postoperative days (PODs) .

**Table 1** The recovery of spinal cord function in ASIA patients was evaluated during one year

ASIA Classification	PODs/d	SF	MF	Muscle strength
B	30	+-	+-	4
D	90	+	+	5-/5
D	180	+	+	5
E	360	+	+	5

Abbreviation: ASIA American Spinal Injury Association, PODs Postoperative days, SF Sensory function, MF Motor function

for common CE sites, deep tissue involvement like the spine and pleural wall often requires CT or MRI for accurate diagnosis. MRI is preferred for its higher accuracy in assessing cyst diagnosis [14–16].CT initially suggested a single lesion, making it difficult to determine whether it was a neurogenic tumor, but MRI identified two distinct cysts, indicating a higher accuracy of MRI for bony cystic echinococcosis [17]. Some studies show that ELISA has higher specificity compared to IHA and IFA detection methods [18]. Other studies indicate that ELISA has higher sensitivity compared to Siemens IHA and Fumouze IHA [19]. ELISA is helpful for diagnosing patients positive for echinococcosis. However, some studies suggest [20] that the sensitivity

of ELISA for hepatic echinococcosis is 80–100% with a specificity of 88–96%, whereas for spinal echinococcosis, the sensitivity is only 25% and the specificity is only 56%.Therefore, compared to hepatic echinococcosis, serological testing has lower sensitivity and specificity for spinal echinococcosis. Surgical pathology provided definitive confirmation.

The standard treatment for CE primarily involves a combination of surgery an BMZ antiparasitic drugs [20–22]. The standard treatment involves surgery combined with benzimidazole (BMZ) chemotherapy, though techniques like puncture, aspiration, injection, and reaspiration (PAIR) are used for inoperable cases [23]. Of course, there is also a considerable amount of research suggesting that PAIR technology can not only serve as an alternative for cases where surgery is not possible but also results in fewer adverse events for uncomplicated hydatid cysts [24]. Therefore, for patients who refuse surgery, those with uncomplicated hydatid cysts, patients who experience recurrence after surgery, and those who are resistant to drug treatment, PAIR technology will also be an excellent option [25]. The procedures of debridement and decompression in surgery can quickly alleviate symptoms of spinal cord compression, and these techniques are widely applied in the treatment of spinal echinococcosis [26]. ABZ is preferred due to its lower recurrence rate and higher efficacy [27, 28]. Recent studies highlight

the potential of ABZ-loaded polymer nanoparticles for enhanced cysticidal effects [29]. In this case, immediate decompressive surgery followed by ABZ therapy resulted in significant symptom improvement and a favorable prognosis, with no recurrence observed to date.

Intradural and pleural wall CE is an extremely rare infectious disease with low early detection rates and potential for misdiagnosis. MRI is an important imaging modality, and ELISA is considered a relatively reliable diagnostic tool when positive, while histopathology is the gold standard for diagnosing cystic echinococcosis. The mainstay treatment combines surgery and ABZ, demonstrating excellent outcomes in this case, providing valuable insights for managing similar rare cases.

#### Abbreviations

CE	Cystic echinococcosis
BL	Dew/Braithwaite & Lees
MRC	Medical Research Council
CT	Computed tomography
MRI	Magnetic resonance imaging
ELISA	Enzyme-linked immunosorbent assay
ABZ	Albendazole
ASIA	American Spinal Injury Association
PODs	Postoperative days

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Not applicable.

#### Authors' contributions

CL participated in the editing, reviewing, and revising of the manuscript. JZ collected data and wrote the original draft. PX and XL participated in the creation of the charts. YH participated in the data collection. YY and XD participated in the data organization.

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#### Availability of data and materials

No datasets were generated or analysed during the current study.

#### Declarations

#### Ethics approval and consent to participate

As a case report, our study did not require a referral from our institutional clinical ethics committee.

#### Consent for publication

Written informed consent was obtained from the patient for the publication of this case report and any accompanying data and images.

#### Competing interests

The authors declare no competing interests.

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