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Case Report

A Rare Parasitic Infection: Primary Intradural Extramedullary Hydatid Cyst

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ABSTRACT

Echinococcus granulosus infrequently induces spinal hydatid cysts, and intradural hydatid cysts are extremely rare among these spinal hydatid cysts. We report a 30-year-old man with a history of progressive back pain caused by a previous back injury. Magnetic resonance imaging revealed a spinal intradural cystic lesion. After surgical removal, histopathological diagnosis was a hydatid cyst. The patient had no other symptoms of systemic hydatid cyst disease. Diagnosis of hydatid cyst should be considered prior to surgery, especially in young patients with spinal intradural cystic lesions, as leakage of the hydatid cyst's fluid during surgery is a frequent case of recurrence.

KEYWORDS: Hydatid cyst, Echinococcus, Albendazole, Intradural

INTRODUCTION

Among cases of hydatid disease, cystic hydatidosis is frequently found in the liver (75%), although the lungs (15%), brain (2-4%), genitourinary tract (2-3%), and spine (1%) may also be involved (13). Spinal involvement has been classified into five groups by Braithwaite and Lees: (i) primary intramedullary, (ii) intradural extramedullary, (iii) extradural, (iv) hydatid disease of the vertebra, and (v) paravertebral (2). An intradural disease without vertebral column involvement is extremely rare, and the thoracic spine (63.9%) is the most commonly affected region, as *Echinococcus* typically become lodged in the vascular part of the spine (8). Secondary intradural involvement can occur through dural spinal injury, or spread through the subarachnoid space of a ruptured intracranial cyst, although primary intradural extramedullary hydatid cysts are extremely uncommon (9).

CASE REPORT

A 30-year-old man presented to the Neurosurgery Department with a history of progressive back pain and a fever, following a back injury sustained 5 days prior. Physical examination detected no abnormalities. Neurological examination revealed numbness at the right T8-9 dermatomes, and there were no other neurological abnormalities. Hematologic analyses (including total blood cell count and, erythrocyte sedimentation rate) were in the normal ranges.

Magnetic resonance imaging (MRI) revealed a 24 x 9 mm cystic lesion, reported as arachnoid cyst, invading the spinal canal posterior to the cord. The lesion was isointense on T1 and hyperintense on T2-weighted images (Figures 1; 2A, B). After contrast enhancement of the posterior aspect of the lesion, there was a significant thickening observed at the left posterolateral border of the dura. There were no abnormalities in the spinal column, and no other lesions were detected when the patient was examined for a systemic disease. Serological test for *Echinococcus* was negative.

When the patient complained that the pain was unbearable, a surgery with posterior approach was planned. The patient was laid prone and a midline longitudinal incision was made from T7-T10, and posterior decompression with T8-T9

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laminectomy was performed. After the dural incision, the cystic lesion spontaneously rose with the capsule (Figure 3). Histopathological examination led to a diagnosis of a hydatid cyst, given the visible scolexes. Postoperatively, the patient was afebrile with no further back pain, and the patient was prescribed albendazole.

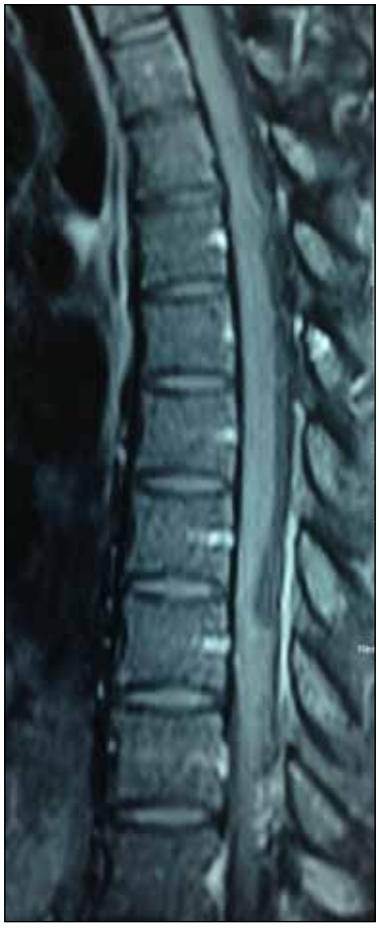

Figure 1: T1-weighted MRI scan shows an isointense cystic lesion.

■ DISCUSSION

Hydatid cysts are a rare parasitic infection, caused by the larval form of *Taenia* or *Echinococcus granulosus*, which are typically found in young patients of both genders (11).

Hydatid cyst in the spine can be detected after direct extension from pulmonary infestation, or more frequently in the vertebral body, where intradural involvement can be primary or secondary (14). The pathological mechanism of primary intradural involvement has been reported by some authors to be hematic dissemination (7). Haddad et al. have also reported that the larval form of the parasites (oncospheres) may pass these barriers, allowing them to reach any part of the body, including the intradural space resulting in primary lesions (5).

Groen et al. have divided the vertebral venous plexus into three compartments: the internal vertebral venous plexus, external vertebral venous plexus, and basal vertebral veins (3). The internal vertebral venous plexus communicates with the intraspinal and radicular veins, and also communicates freely with the external vertebral plexus via the intervertebral veins (4). Tadie et al. have reported a narrowing of the radicular veins on penetrating the dura, which functioned as an anti-reflux valve mechanism. Although the radicular veins allow for draining of blood from the intradural to extradural compartment, there are valveless, which might explain the rare incidence of primary intradural hydatid in the spine (15).

Unfortunately, there may be no pathognomonic symptoms of infection, although spinal cord compression tends to cause radicular pain and segmental neurologic deficits. Hancı et al. have reported an epidural hydatid cyst case with low back pain and sciatica on the right side (6). Conventional myelography is contraindicated, as it may cause intradural dissemination of the disease (16). In these cases, computed tomography myelography can be effective in demonstrating the communication of the cyst with the subarachnoid space (2,10). However, MRI is the gold standard for diagnosis of vertebral hydatid cysts. On T1-weighted images, the cyst may show an isointense signal, with hyperintensity on T2-weighted

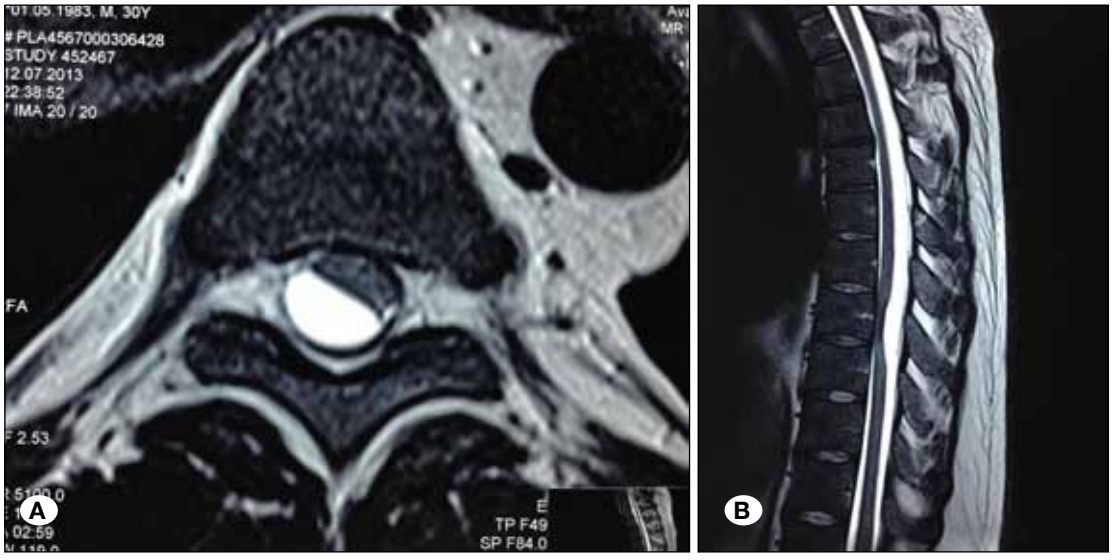

Figure 2: Axial **(A)** and sagittal **(B)** T2-weighted MRI scans show a cystic lesion invading the spinal canal at the posterior of spinal cord.

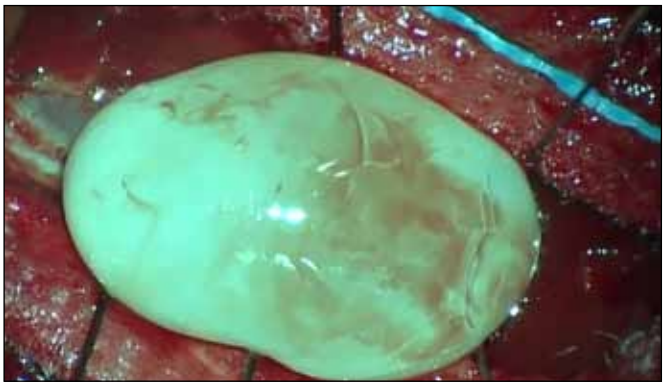

Figure 3: Total excision of the cystic lesion with the capsule.

images (8). In addition, cerebrospinal fluid flow-sensitive MRI techniques may be used in the diagnosis of the cyst and its communication with subarachnoid space (10). Berk et al. found slight contrast enhancement of the cyst, which we also observed in our patient (in the posterior aspect), which is likely the result of reactive fibrosis and degeneration around the parasitic membrane (1).

If a patient does not have a positive anamnesis for a previous *Echinococcus* infection, it can be difficult to differentiate between an arachnoid cyst and a hydatid cyst when performing a differential diagnosis for an intradural cystic lesion in the thoracic region. Nabors et al. (12) have classified the extramedullary cysts of the spinal canal into three main groups. The first group is meningeal cysts, which has three subgroups: type 1-extradural meningeal cysts without neural tissue (type 1A: extradural arachnoid cysts, type 1B: sacral meningocele), type 2-extradural meningeal cysts with neural tissue (perineural cysts or Tarlov cysts), and type 3-intradural meningeal cysts (arachnoid cysts). The second group is non-meningeal epidural cysts, including non-neoplastic lesions (juxta-articular cysts, synovitis) and neoplastic lesions (dermoids, cystic nerve sheath lesions and metastases). The third group is neuroenteric cysts (12). Acquired arachnoid intradural cysts are typically observed in the thoracic region.

When intradural cystic lesions are detected in the thoracic region, a strong suspicion of a hydatid cyst is important. The surgical procedure should be planned for the total removal of the cysts without rupture, to avoid dissemination and recurrence (9). As well, adjuvant antihelmintic therapy (mebendazole and albendazole) should be combined with the surgical treatment.

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