

# Primary Intradural Extramedullary Hydatid Cyst

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**ABSTRACT:** Spinal hydatid cysts account for 1% of all cases of hydatid disease; primary intradural hydatid cysts are uncommon. We present a case of pathologically confirmed intradural spinal cyst hydatid in an otherwise healthy patient who showed no other evidence of systemic hydatid cyst disease. The patient presented with back pain, paraparesis, and weakness. An intradural extramedullary cystic lesion was identified

with magnetic resonance imaging and was shown to be a hydatid cyst by histopathologic examination after surgical removal. To our knowledge, this is the 25th case of hydatid cyst at an intradural extramedullary location reported in the literature. **KEY INDEXING TERMS:** Hydatid cyst; Spine; Surgery. [Am J Med Sci 2005; 329(4):202-204.]

**H**ydatic disease is an infestation caused by the larval stage of the tapeworm *Echinococcus granulosus*. The parasite is prevalent in most sheep and cattle raised in Mediterranean countries.<sup>1-7</sup> Humans and sheep are the intermediate hosts of this parasite, and the dog is the final host. Humans acquire the parasite by ingesting water or food contaminated with eggs that were excreted in the infected dog's faeces. Larvae migrate to the portal system from the intestine. Hydatid cyst most commonly affects liver and lung and affects bone in only 0.5% to 2% of all patients. Hydatid cyst of the spine constitutes 1% of all cases of hydatidosis and most commonly is localized to the thoracic spine and, in decreasing order of frequency, to the lumbar, sacral, and cervical spine.<sup>6,8</sup> Spinal hydatid cyst can occur by direct extension from pulmonary or pelvic foci or, less commonly, begins primarily in the body of the vertebrae.<sup>6,8</sup> Primary intradural extramedullary hydatid disease is extremely rare.<sup>1,6,9-14</sup>

We present a case of intradural extramedullary hydatid cyst. To our knowledge, this is the 25th case of intradural extramedullary hydatid cyst in the literature.

## Case Report

A 32-year-old woman presented with difficulty walking. She had a 6-month history of back pain and had recently developed bilateral leg weakness. Her physical examination findings were normal. Neurologic examination revealed bilateral lower extrem-

ity weakness and hypoesthesia in L4, L5, and S1 dermatomes bilaterally. Routine laboratory investigations and chest radiography findings were normal.

Magnetic resonance imaging revealed a 5×2×2 cm cystic lesion that was located intradurally at the L5-S2 level. The cystic lesion contained fluid that was the same density as cerebrospinal fluid and had no apparent mural nodule (Figure 1). Electromyography revealed a right-sided L5 radiculopathy. Hydatid cyst hemagglutination test was positive (up 1/8192).

The patient underwent L5-S1 laminectomy. Two intradural cysts were removed with their capsules. The postoperative course was uneventful, without neurologic deterioration. The patient's back pain resolved and the leg weakness improved. The patient was started on a course of albendazole treatment.

Histopathologic examination confirmed the working diagnosis of hydatid cyst.

## Discussion

*E granulosus* is a parasite, distributed mostly in sheep- and cattle-raising countries. Humans can be infected by ingesting the eggs. Oncospheres, which migrate to the portal circulation from the intestine, are trapped by liver and lungs. Spinal hydatid cysts account for 1% of all cases of hydatid disease.<sup>10</sup> Spinal hydatid cyst can occur by direct extension from pulmonary or pelvic foci or, less commonly, begin primarily in the vertebral body.<sup>6,8</sup> Also, *Echinococcus* embryos can reach the spine through portovertebral venous shunts. According to Braithwaite and Lees, spinal hydatid disease is classified into five types: primary intramedullary, intradural extramedullary, extradural intraspinal, vertebral, and paravertebral.<sup>2</sup> Although secondary intradural involvement can occur as a consequence of spinal dural injury or also via spread through the subarachnoid space of a ruptured intracranial cyst, primary intradural hydatid cysts are extremely uncommon.<sup>6,12,14</sup> In the most recent reviews by Chakir et al,<sup>11</sup> 23 cases of intradural spinal hydatid cysts are

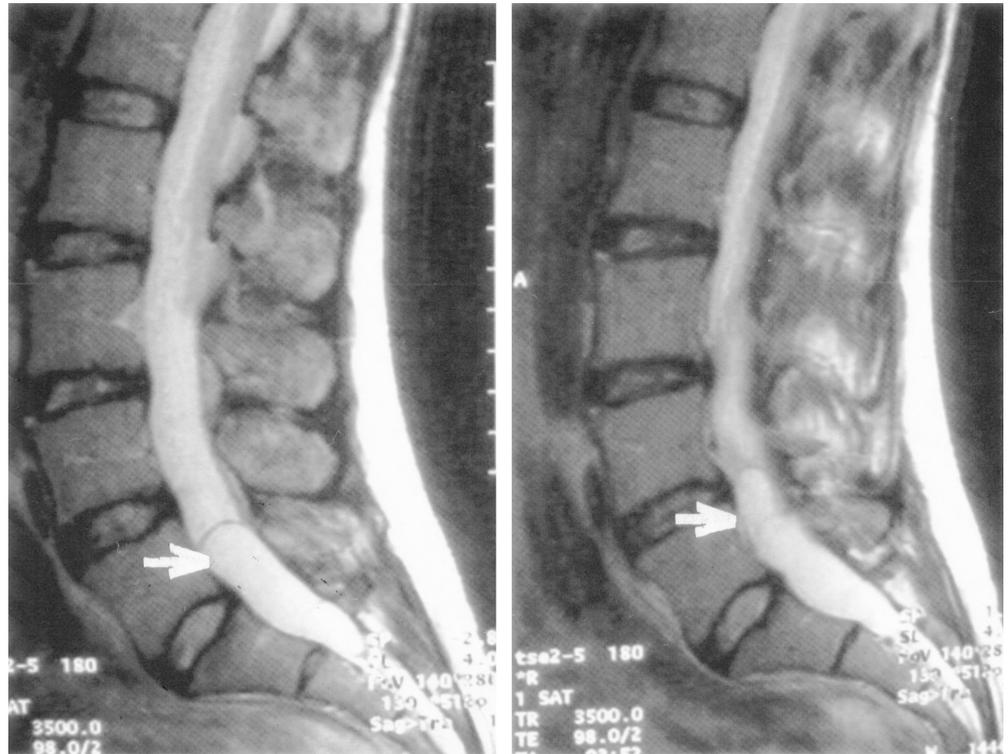
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**Figure 1.** Sagittal T2-weighted magnetic resonance images showing an intradural, sausage-shaped, cystic lesion that is almost isodense with cerebrospinal fluid, located at L5-S2.



recorded in the world literature. Hilmani et al<sup>15</sup> recently published an additional case of intradural spinal hydatid cyst.

There are no pathognomic signs or symptoms of spinal hydatid disease other than the symptoms related to compression.<sup>1,6</sup> Generally, the first symptoms are backache and radicular pain.<sup>8</sup> Weaknesses of the limbs occurs in the later phase of the disease,<sup>8</sup> and paraplegia is reported in 25% to 84% of cases.<sup>8</sup> Because hydatid cyst of the spine can be a lethal disease, early diagnosis or suspicion is of great importance, especially in endemic areas.

Because there are no distinguishing radiologic findings, preoperative diagnosis is difficult. Simple radiographic findings are nonspecific and may show bone destruction and a soft tissue mass. Myelography is an invasive procedure and is contraindicated to avoid dissemination to intradural space.<sup>14</sup> Before widespread use of magnetic resonance imaging (MRI), computed tomography had a major role in the evaluation of osseous and paraspinal soft tissue involvement, but it cannot delineate the border between the cyst and the dural sac.<sup>2,14,16</sup> MRI is the best imaging tool for diagnosing spinal hydatid cyst because it can show its relationship to normal structures and the spinal cord and its extension into soft tissue.<sup>14,16</sup> Appearance of a hydatid cyst on the MRI scan is quite characteristic in that it has two dome-shaped ends, has no debris in its lumen, and looks

like a sausage.<sup>14</sup> Cyst walls are very thin and regular and have no septation.

Serologic diagnosis is available but not reliable. A Casoni test to diagnose hydatid disease is positive in 95% of cases but can also have 40% false-positive results.<sup>4</sup>

The treatment of choice in spinal hydatid disease has been surgery for nearly all cases.<sup>5-7</sup> The preferred operative procedure is usually a laminectomy.<sup>6</sup> Total removal of the cysts without rupture should be the surgical goal,<sup>12</sup> because operative rupture of the cyst may cause dissemination of the disease and recurrence. Adjuvant antihelminthic therapy with mebendazole and albendazole has been claimed successful if integrated with surgical treatment.<sup>5,7,10,17</sup> There is a now a trend in hydatid cyst treatment to progressively rely more on chemotherapy and percutaneous drainage methods, such as percutaneous puncture, aspiration, injection of scolicalid agents and reaspiration, instead of surgery.<sup>18-20</sup>

In conclusion, a high index of suspicion combined with good-quality neuroimaging is important for early and correct diagnosis. Total surgical removal of the cysts without rupture should be aimed for. Antihelminthic drug therapy should be advised both perioperatively and well into the postoperative period. Hydatid cyst is a benign disease; however, it has a malignant prognosis because of its high risk of recurrence.

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