Long-term result of treatment for paraspinal and extradural hydatid cyst: a case report

Paraspinal ve ekstradural yerleşimli spinal kist hidatik tedavisinde uzun dönem takip sonucu: Olgu sunumu

Ahmet SALDUZ, Lutfu Ozgur KOYUNCU,1 Fatih DIKICI, Ufuk TALU

Istanbul University, Istanbul Faculty of Medicine, Department of Orthopaedics and Traumatology;
1American Hospital, Department of Orthopaedics and Traumatology


Anahtar sözcükler: Albendazol/terapötik kullanım; Echinococcus granulosus; laminektomi; lomber vertebra; omurga hastalığı/surgery.

Hydatid cyst is a zoonosis affecting any part of the body and presenting difficulty in diagnosis and treatment. Primary bone involvement seen in about 2% of the cases is accompanied by spinal involvement in 50%. A 41-year-old female presented with low back pain radiating to the left leg. After physical examination, radiologic and laboratory investigations, a diagnosis of spinal hydatid cyst was made with intraspinal extradural, vertebral, and paravertebral involvement. The patient was treated with two-staged posterior and anterior surgical resections with addition of pre- and postoperative antihelminthic therapy. No recurrences developed within a follow-up of 5.5 years. Recurrences can be reduced by initiation of antihelminthic therapy preoperatively, perioperative injection of scolicidal agents into the cyst, and careful excision of the cyst without spread to neighboring tissues.

Key words: Albendazole/therapeutic use; Echinococcus granulosus; laminectomy; lumbar vertebrae/surgery; spinal diseases/surgery.

Correspondence / Yazaşma adresi: Dr. Ahmet Salduz.Istanbul University, Istanbul Faculty of Medicine, Department of Orthopaedics and Traumatology, 34093 Çapa, İstanbul. Phone: +90212 - 635 12 35 e-mail: ahmet_salduz@yahoo.com
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findings in spinal hydatid cyst cases. Root pressure and paraplegia has been reported at the 25-84 % of these patients.\textsuperscript{[8-10]}

According to radiological classification of Braithwaite and Lees\textsuperscript{[11]}, spinal hydatid cases are described in five groups. These are intramedullary, intradural extramedullary, extradural, vertebral and paravertebral lesions. In our case, extradural (intraspinal), vertebral and paravertebral involvements were seen together.

**Case report**

Forty-one years old female patient, presented to our clinic with hip and leg pain at the left side ongoing for last three years that had increased in the last two months. Laseque test was positive at 60 degrees on the left side. Hypoesthesia was detected below knee level on the left side and bilateral muscle strength was 5 / 5. Anteroposterior and lateral lumbar sacral plain radiographs were normal. Magnetic resonance images demonstrated benign multiloculated cystic lesions in the spinal canal extending to the retroperitoneal area at second lumbar vertebra level with no gadolinium uptake (figure 1). Computerized tomography (CT) of thorax was normal. Abdominal CT demonstrated horseshoe kidneys and cystic lesions at the retroperitoneal area related with bilateral psoas muscles, which might be compatible with hydatid cyst. The ELISA test for *E. granulosus* resulted positive. Patient was consulted with department of infectious diseases and antihelmintic albendazol treatment (2x400 mgr tablets) was given before the surgical treatment.

Two months after the beginning of medical treatment, surgery was scheduled. Total laminectomy of L\textsubscript{2} and partial laminectomy of L\textsubscript{1} and L\textsubscript{3} with posterior approach were performed. Two extradural cysts (one 1x2 cm and other 0.5x2 cm) which were juxtaposed anteriorly to medulla spinalis were explored. Hypertonic (20%) saline solution was injected into these cysts. Cysts were removed after waiting a sufficient period of time. One of the cyst was ex-
tending to the L₂ vertebra body through a 2x2 mm fistula hole and had a continuity with paravertebral cysts. Posterior instrumentation and fusion were performed from T₁₂ through L₄ and operation was terminated. Approach to the retroperitoneal space for the complementary second stage of surgery was achieved with left abdominal incision one week later. Cysts over the iliopsoas muscle were dissected without being damaged. Cysts were removed after hypertonic saline injection. Total corpectomy of L₂ vertebra was performed after total discectomy of L₁-2 and L₂-3 (figure 2). The cysts at right retroperitoneal area became approachable after corpectomy and they were also removed meticulously after hypertonic saline injection. A titanium cage filled with 60 cc cancellous allogreft was placed at corpectomy site. The operation was terminated after the surgical site was irrigated with hypertonic saline solution. Cultures of the samples taken at surgery resulted negative. Results of the specimens’ histopathological examinations obtained at first and second surgical interventions revealed hydatid cyst (Figure 3). Albendazol treatment was continued for three years. Patient had no complaints at last follow-up, 5.5 years after surgery. Her neuromuscular examination was normal and Laseque test was found negative. No radiological recurrence was detected (Figure 4).

**Discussion**

Hydatidosis is a difficult to diagnose disease without obvious clinical symptoms. Mortality risk is more than 50% when the spine is involved. History, physical examination, radiological evaluation and serological tests are used for diagnosis. MR and CT imaging techniques are good guides for diagnosis and follow-up. ELISA, Western blotting, indirect haemagglutination test and polymerase chain reaction tests are used for the serological diagnosis of hydatidosis. Sensitivity of serological methods for liver involvement is 80-100% and specificity 88-96%. Sensitivity for the lung and other organ involvements has been reported as 50-56% and 25-56% respectively. Echinococcus granulosus specific IgE level used for serological diagnosis of the disease vary depending on the location of cysts. The sensitivity of this test is reported 92% for liver cyst and 61.5% for bone hydatidosis. Although sensitivity of ELISA test for bone hydatidosis is low, it was found positive.
in our case.

Aneurysmal bone cyst, giant cell tumor, solitary bone cyst, arachnoid cyst, neurofibromatosis, fibrocystic disease, chondrosarcoma and tuberculosis should be considered at the differential diagnosis of hydatid cyst. Although hydatidosis is an infection disease, clinical behavior is like a local malignant tumor. In our case, local (back pain) and reflected (reflected pain to the leg) symptoms were seen together. Systemic antihelmintic therapy alone is not enough for local control. The main aim of treatment is curative surgical removal of the cysts. Oral or parenteral medical therapy, have been suggested to reduce the risk of recurrence and systemic spread. In our case albendazole treatment began before the operation to prevent recurrence and avoid systemic spread.

Radical excision of spinal hydatid cysts is very difficult due to anatomical structures and possible damage to neural elements. Karray et al. formerly proposed markedly increase of the survival rates with combined anterior and posterior surgery plus adjuvant antihelmintic chemotherapy. Early surgical decompression and stabilisation followed by adjuvant chemotherapy for patients with motor deficit provides an opportunity for healing. In our case posterior decompression and stabilisation was performed primarily for preventing the deterioration of neurological status and cysts were removed completely with an anterior surgical exposure as a second stage. Govender et al. suggested radical total excision of the involved vertebra. Filling PMMA (polymethylmethacrylate) to the defects occurring after resection of nonspinal bone hydatidosis was suggested by Yildiz et al. In our case, total vertebrectomy was performed even though a part of the second lumbar vertebra was involved. Easier and less traumatic exposure was achieved in this way and cysts at both sides of the vertebral body were removed without rupturing them. Irrigation of the cyst fluid contaminated tissues with formalin, nitrate of silver (5%) or hypertonic saline is suggested for preventing recurrence. Hypertonic saline injected into the cyst acts as a scolicidal agent because the difference of osmolality between the hypertonic saline and the interior of the cyst causes the sclerosis of the cyst. Saline solution has to be applied at least 6 minutes with a minimum concentration of 20% to be effective.

In our case, cysts were explored and removed without being ruptured after scolexes were inactivated by hypertonic saline injection into the cysts.

Recurrence rates of spinal hydatidosis were reported markedly high in different series. In a survey study from Turkey which consist the hydatid cyst cases from 1944 - 96, 84 patients with spinal hydatidosis had had a recurrence rate of %18 in a 1 week to 3 years follow-up period. Moreover, when a comparison is made between the recurrence rates of the cases who had only surgery and who had surgery with adjuvant chemotherapy, significant difference had been found in favor of the cases treated with chemotherapy. Recurrence rate of another serie with 20 patients followed at a mean of 4.8 years by Herrera et al. is 60%. No recurrence was observed in our case at 5.5 years. According the guidelines of ASTM (The American Society for Testing and Materials) titanium implants are compatible with MR. Diagnostic images can be obtained because these implants cause less artefact. Titanium’s resistance against corrosion is stronger than stainless steel and cobalt alloys. Usage of titanium implants in surgeries of these patients is important for recurrence control with MRI. We used titanium implants for these reasons.

Albendazol and mebendazol are frequently used antihelmintic agents for the treatment of hydatidosis. Absorption of orally taken mebendazol is low, so high doses are required to be effective. Mebendazol can cause neutropenia. Absorption of orally taken albendazol is better than mebendazol and it also reaches higher therapeutic levels. Hepatotoxicity is the most important side effect of albendazol. Treatment with albendazol for 2 month intervals before and after the definitive surgery decreases viability of the cysts and thereby decreases the recurrence rate. Albendazol was the choice for treatment in our case and liver function tests were routinely controlled during the treatment period.

Eventually being successful at the treatment of hydatidosis is possible with complete cyst removal and taking care of no dissemination during surgery.

Using of antihelmintic agents before the surgery and continuing afterwards and injection of scolicidal agents into the cysts are important procedures which reduce the recurrence rate.
References


