



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*

Kist hidatik, değişik organları tutabilen, tanı ve tedavisi zor bir zoonozdur. Primer kemik tutulumu olgularının %2'sinde gözlenir. Kemik tutulumu olan olguların yaklaşık yarısında ise omurga tutulumu vardır. Kırk bir yaşında kadın hasta, bacağa vuran kalça ağrısı yakınmasıyla başvurdu. Fizik muayene, radyolojik ve laboratuvar incelemeleri sonucu intraspinal ekstradural, vertebral ve paravertebral tutulumlu spinal kist hidatik tanısı kondu. Ameliyat öncesi ve sonrası dönemde antihelmintik tedavi uygulanan hastaya iki aşamalı posterior ve anterior cerrahiyle medikal eksizyon uygulandı. Hastanın 5.5 yıllık izleminde nüks saptanmadı. Nüks oranlarını azaltmak için antihelmintik tedaviye cerrahi öncesinde başlamak, cerrahi sırasında kist içine skolisidal madde enjekte etmek ve komşu dokulara yayılımı önleyici cerrahi prensiplere özen göstermek gerekir.

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

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 scolical 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.

Hydatid cyst is a zoonosis, which is caused by a parasite of cestode group, Echinococcus granulosus. The incidence of the cyst hydatid disease ranges from 3 to 50 cases per 100,000 inhabitants which is endemic in the South America, Far East Middle and East Mediterranean.^[1,2]

Hydatid cyst can infest all organs and tissues especially liver (%50-70) and lungs (%20-30). Spinal involvement is usually seen in the thoracic region with spread from pulmonary, abdominal or pelvic involve-

ment. Primary bone involvement in the nonendemic regions differs between 0.5 - 2%, but this ratio can be up to 4% in the endemic regions.^[3-5] Approximately 50% of the cases with bone involvement had spinal involvement.^[1, 6] Primary spinal involvement is being explained by direct portovertebral venous shunt theory.^[1,3, 7] 50% of spinal involvement was seen in thoracic region, 10% in cervical region, 20% in lumbar region and 20% in sacral region.^[8] Weakness of the extremities and back pain are the most common

Correspondence / Yazışma 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

Submitted / Başvuru tarihi: 17.11.2008 **Accepted / Kabul tarihi:** 20.02.2009

© 2009 Türk Ortopedi ve Travmatoloji Derneği / © 2009 Turkish Association of Orthopaedics and Traumatology



Figure 1. Preoperative magnetic resonance images at (a) axial and (b) sagittal planes.

findings in spinal hydatid cyst cases. Root pressure and paraplegia has been reported at the 25-84 % of these patients.^[8-10]

According to radiological classification of Braithwaite and Lees^[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 lumbosacral plain radiographs were normal. Magnetic resonance images demonstrated benign multilocula-

ted 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 antihelminthic 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 L₂ and partial laminectomy of L₁ and L₃ 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-

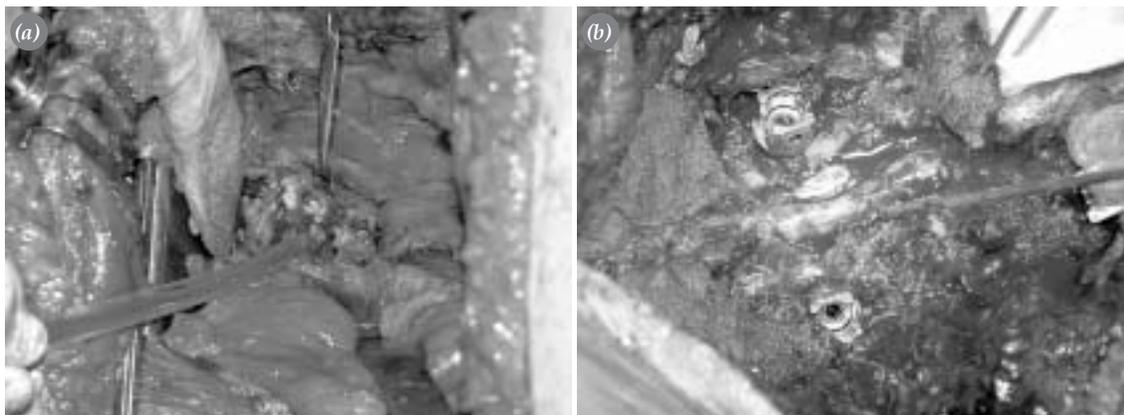


Figure 2. (a) The image of the fistula at corpus of L₂ vertebra extending posteriorly, (b) the image of the cyst after laminectomy.

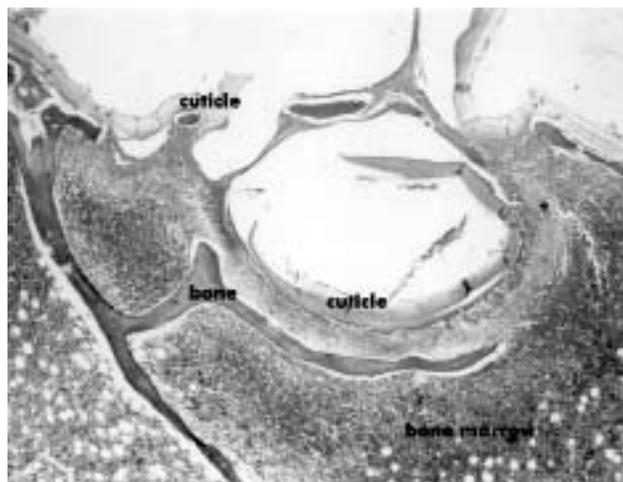


Figure 3. Histopathologic cross-section of an excised hydatid cyst.

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 allograft 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.^[12] 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.^[3] 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.^[3] *Echinococcus granulosus* specific IgE level used for serologic 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.^[13] Although sensitivity of ELISA test for bone hydatidosis is low, it was found positive

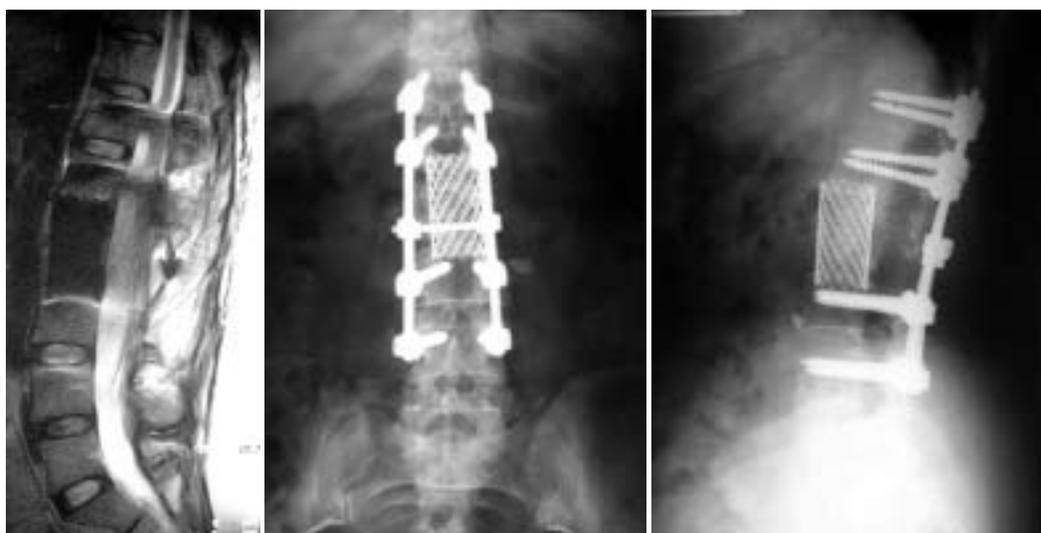


Figure 4. Magnetic resonance image and plain films after 5.5 years postoperatively.

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.^[14, 15] 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 antihelminthic therapy alone is not enough for local control.^[16] 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.^[17] 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.^[12] formerly proposed markedly increase of the survival rates with combined anterior and posterior surgery plus adjuvant antihelminthic 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.^[6] suggested radical total excision of the involved vertebra. Filling PMMA (polymethylmetacrylate) to the defects occurring after resection of nonspinal bone hydatidosis was suggested by Yıldız et al.^[18] 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.^[5, 6, 9] Hypertonic saline injected into the cyst acts as a scolicial 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.^[19]

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.^[9] 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.^[5] 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.^[20,21] 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 antihelminthic agents for the treatment of hydatidosis. Absorbtion of orally taken mebendazol is low, so high doses are required to be effective. Mebendazol can cause neutropenia. Absorbtion 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 decrease the recurrence rate.^[22] 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 antihelminthic agents before the surgery and continuing afterwards and injection of scolicial agents into the cysts are important procedures which reduce the recurrence rate.

References

1. Sapkas GS, Machinis TG, Chloros GD, Fountas KN, Themistocleous GS, Vrettakos G. Spinal hydatid disease, a rare but existent pathological entity: case report and review of the literature. *South Med J* 2006;99:178-83.
2. Onbas O, Kantarcı M, Alper F, Sekmenli N, Okur A. Spinal widespread intradural extramedullary hydatidosis. *Neuroradiology* 2004;46:310-2.
3. King CH. Cestodes. In: Mandell GL, Bennett JE, Dolin R. editors. *Douglas and Bennett's principles and practice of infectious diseases*. 5th ed. Philadelphia: Churchill Livingstone, 2000. p. 2962-3.
4. Özdemir HM, Oğün TC, Taşbaş B. A lasting solution is hard to achieve in primary hydatid disease of the spine: long-term results and an overview. *Spine* 2004;29:932-7.
5. Herrera A, Martínez AA, Rodríguez J. Spinal hydatidosis. *Spine* 2005;30:2439-44.
6. Govender TS, Aslam M, Parbhoo A, Corr P. Hydatid disease of the spine. A long-term followup after surgical treatment. *Clin Orthop Relat Res* 2000;(378):143-7.
7. Karadereler S, Orakdöğen M, Kılıç K, Özdoğan C. Primary spinal extradural hydatid cyst in a child: case report and review of the literature. *Eur Spine J* 2002;11:500-3.
8. Baysefer A, Gönül E, Canakçı Z, Erdoğan E, Aydoğan N, Kayalı H. Hydatid disease of the spine. *Spinal Cord* 1996; 34:297-300.
9. Turgut M. Hydatid disease of the spine: a survey study from Turkey. *Infection* 1997;25:221-6.
10. İşlekel S, Zileli M, Erşahin Y. Intradural spinal hydatid cysts. *Eur Spine J* 1998;7:162-4.
11. Braithwaite PA, Lees RF. Vertebral hydatid disease: radiological assessment. *Radiology* 1981;140:763-6.
12. Karray S, Zlitni M, Fowles JV, Zouari O, Slimane N, Kassab MT, et al. Vertebral hydatidosis and paraplegia. *J Bone Joint Surg [Br]* 1990;72:84-8.
13. Sjölander A, Guisantes JA, Torres-Rodriguez JM, Schröder H. The diagnosis of human hydatidosis by measurement of specific IgE antibody by enzyme immunoassay. *Scand J Infect Dis* 1989;21:213-8.
14. Booz MK. The management of hydatid disease of bone and joint. *J Bone Joint Surg [Br]* 1972;54:698-709.
15. Seçer HI, Anık I, Çelik E, Daneyemez MK, Gönül E. Spinal hydatid cyst mimicking arachnoid cyst on magnetic resonance imaging. *J Spinal Cord Med* 2008;31:106-8.
16. Arazi M, Memik R, Kapıcıoğlu MI. Answer please. Hydatid disease of the spine. *Orthopedics* 1998;21:912.
17. Szypryt EP, Morris DL, Mulholland RC. Combined chemotherapy and surgery for hydatid bone disease. *J Bone Joint Surg [Br]* 1987;69:141-4.
18. Yıldız Y, Bayrakçı K, Altay M, Sağlık Y. The use of polymethylmethacrylate in the management of hydatid disease of bone. *J Bone Joint Surg [Br]* 2001;83:1005-8.
19. Kayaalp C, Balkan M, Aydın C, Özgürtaş T, Tanyüksel M, Kırımlıoğlu V, et al. Hypertonic saline in hydatid disease. *World J Surg* 2001;25:975-9.
20. Kumar R, Lerski RA, Gandy S, Clift BA, Abboud RJ. Safety of orthopedic implants in magnetic resonance imaging: an experimental verification. *J Orthop Res* 2006;24:1799-802.
21. Harkess JW, Daniels AU. Introduction and overview. In: Canale ST, editor. *Campbell's operative orthopaedics*. Vol. 1, 10th ed. Philadelphia: Mosby; 2003. p. 223-42.
22. Arif SH, Shams-Ul-Bari, Wani NA, Zargar SA, Wani MA, Tabassum R, et al. Albendazole as an adjuvant to the standard surgical management of hydatid cyst liver. *Int J Surg* 2008;6:448-51.