



Long-Term Disseminated Recurrence in Spinal Hydatid Cyst: A Case Report

Uzun Dönemde Yaygın Rekürrens Gösteren Spinal Hidatik Kist: Bir Olgu Sunumu

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ABSTRACT

The spinal column is rarely affected by hydatid cyst; however, when involved, it has higher rates of recurrence particularly in case of osseous and paravertebral extensions. We report a 36-year-old patient. The patient was operated previously for hydatid cyst through laminectomy 13 years ago. After 13 years of surgery, the patient was admitted to our clinic with progressive paraparesis. Radiological evaluation revealed multicystic lesions affecting T4 and T5 vertebrae as well as the posterior thoracic wall and paravertebral musculature. Serological findings were also compatible with a hydatid cyst. The patient underwent surgical treatment; the cystic lesions were removed, and vertebral stabilization was provided. The treatment of hydatid cyst in the spine is challenging. Particularly in cases with vertebral involvement, spinal instability and recurrence are the main handicap. Preoperative and postoperative antihelminthic treatment as well as close clinical, radiological and serological follow up in postoperative period is important to avoid recurrence risk.

KEYWORDS: Hydatid cyst, Paraparesis, Spinal, Echinococcus, Albendazole

ÖZ

Spinal kolon hidatik kistten seyrek olarak etkilenir ancak kemik ve paravertebral uzanımları olduğu durumlarda nüks oranı daha yüksek olur. Otuzaltı yaşında bir hasta sunuldu. Hasta 13 yıl önce laminektomi ile hidatik kist nedeniyle ameliyat edilmiştir. Ameliyattan 13 yıl sonra, hasta progresif paraparezi ile kliniğimize başvurdu. Radyolojik değerlendirme arka göğüs duvarı ve paravertebral kas ile birlikte T4 ve T5 vertebra etkileyen multikistik lezyonlar görüldü. Serolojik bulgular da kist hidatik ile uyumlu idi. Hastaya cerrahi tedavi uygulandı, kistik lezyonları çıkarıldı ve vertebra stabilizasyonu sağlandı. Omurgada hidatik kist tedavisi zordur. Özellikle vertebra tutulumu, spinal instabilite ve nüks olgularda temel sorundur. Ameliyat öncesi ve sonrası antihelmintik tedavisi yanı sıra, postoperatif dönemde yakın klinik, radyolojik ve serolojik takip nüks riski için önemlidir.

ANAHTAR SÖZCÜKLER: Hidatik kist, Paraparezi, Spinal, Ekinokokus, Albendazol

INTRODUCTION

Hydatid cyst (HC) is a parasitic disease, generally affecting the ones having close contact with animas like dogs and cats (7,9,11,12,16). The disease is prevalent in Mediterranean countries, Africa, South America, New Zealand, and Southern Australia (14).

The Liver and spleen are the main target organs in 80-90% of the cases. Primary spinal HC is quite rare with an incidence rate of 1% of all cases. Osseous involvement is also rare (0.4-5%), approximately half being in the vertebral column (2,12). Recurrence is the major handicap in spinal hydatisosis, particularly in osseous involvement, rates reaching up to 30-100% of the cases (6,12).

In this report, we discussed management of recurrent hydatid cyst with late and disseminated recurrence after more than 13 years of treatment.

CASE REPORT

A 36-year-old female patient was admitted to our clinic with progressive paraparesis. Her history revealed surgical intervention for thoracic HC disease 13 years ago at another center. The extradural cysts had been removed with laminectomy. The patient had received albendazole for 1 month postoperatively. The postoperative period was uneventful with complete resolution of paraparesis and return to normal daily activities until the last 2 months. Thirteen years later, she was admitted to our clinic with paraparesis, hypoesthesia below T10 level and increased deep tendon reflexes.

Magnetic resonance imaging (MRI) evaluation of the thoracic region demonstrated multiple cystic lesions at T4, T5 level with extensions to the right thoracic wall and paravertebral musculature. Spinal cord compression was also obvious (Figure

1A-D). The costae on the right thoracic wall were destroyed and a kyphotic deformity along with thoracic laminectomy was shown on sagittal 3-D computerized tomography (CT) scans. Besides these findings, ultrasonographic evaluation revealed big cystic lesion (8x7x5 cm) in the spleen. The serological test was highly positive for hydatid cyst [specific enzyme-linked immunosorbent assay (ELISA), Western Blot]. Albendazole treatment (400 mg twice a day) was given preoperatively.

The surgical treatment was performed in two stages. In the first stage, right thoracotomy was performed. The cysts in the right lateral thoracic wall were removed along with destroyed costae. Multiple cysts were removed from the destroyed vertebrae and extradural region. T4 and T5 vertebrae were removed and spinal decompression was achieved. Surgical field was irrigated with hypertonic saline solution.

Stabilization was performed with iliac autograft and plate-screw system between T3 and T6 (Figure 2A-E).

After 10 days, the patient was operated posteriorly in the second stage. Multiple cysts in paraspinal musculature were also removed. Posterior thoracic transpedicular screw fixation was performed through T1, T2, T7, T8, and T9 vertebrae. Posterolateral allograft was also used for bony fusion.

The patient was further operated for HC in the spleen, and splenectomy was performed by general surgeons. Histopathological evaluation confirmed the diagnosis of HC. The patient's paraparesis was relieved. Postoperative MRI and CT scans verified total removal of the cyst. Moreover, kyphotic deformity was corrected along with spinal cord compression.

The patient was discharged from the hospital with albendazole treatment (400 mg twice a day) for 6 months. No recurrence was detected after 2-year follow up.

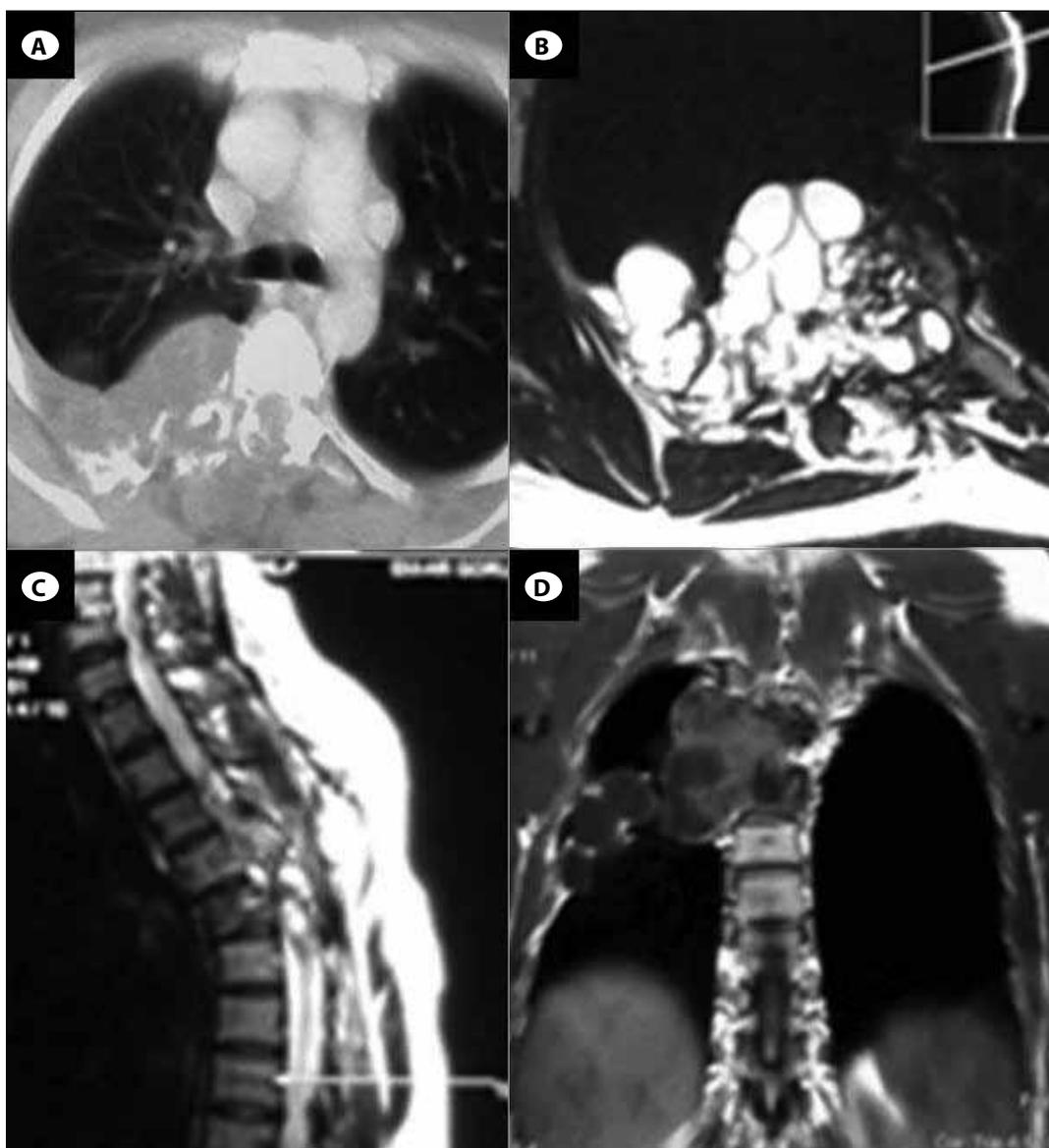


Figure 1: Preoperative images; **A)** Axial thoracic BT shows iso-hypodense posterolaterally located destructive lesion, **B)** Axial T2-weighted MRI shows hyperintense well-defined multiloculated cysts in the extradural space, **C)** Sagittal T2-weighted MRI shows destruction and kyphotic angulation of the dorsal vertebra, **D)** Coronal T1-weighted MRI shows hypointense well-defined multiloculated cysts.

DISCUSSION

Hydatid cyst is a parasitic infestation caused by larval stage of tapeworm echinococcus. *Echinococcus granulosus* is more common in humans; however, less commonly, *Echinococcus multilocularis* is also diagnosed. Human is the intermediate host for the parasite. Adult parasites live in the intestine of dogs and wolves and deliver eggs. The eggs are excreted in the stool. When water sources and plants are contaminated with the eggs, they are ingested by sheep, goats, rodents, or human. The eggs hatch in the intermediate hosts. The larva then penetrates the intestinal wall and duodenum, to gain access to the portal system. The larvae are carried by the bloodstream to distal organs like the liver, lungs, spleen, bone, and the central nervous system (1).

Hydatid cyst settles in these organs, particularly in the liver and lungs. Spinal involvement is quite rare, making up to

1% of all hydatidoses (12). Spinal column can be affected by extension of pulmonary, abdominal, and pelvic involvement. Spinal involvement is most commonly seen in the thoracic region and cervical, lumbar, and sacral areas are less common (3,8,17). Without other systemic infestations, primary spinal involvement was believed to result from porto-vertebral venous shunts (8).

Spinal hydatid cysts are classified into 5 categories as radiological: intramedullary, intradural-extramedullary, extradural vertebral hydatid cyst, and paravertebral lesions (4,5). In our case, the cysts affected the vertebrae, extradural, and paravertebral regions. In spinal involvement, vertebral body, pedicles and laminae are usually affected by multivesicular involvement of the cancellous bone. The intervertebral discs are generally spared from cyst because of periosteal barrier (10,15).

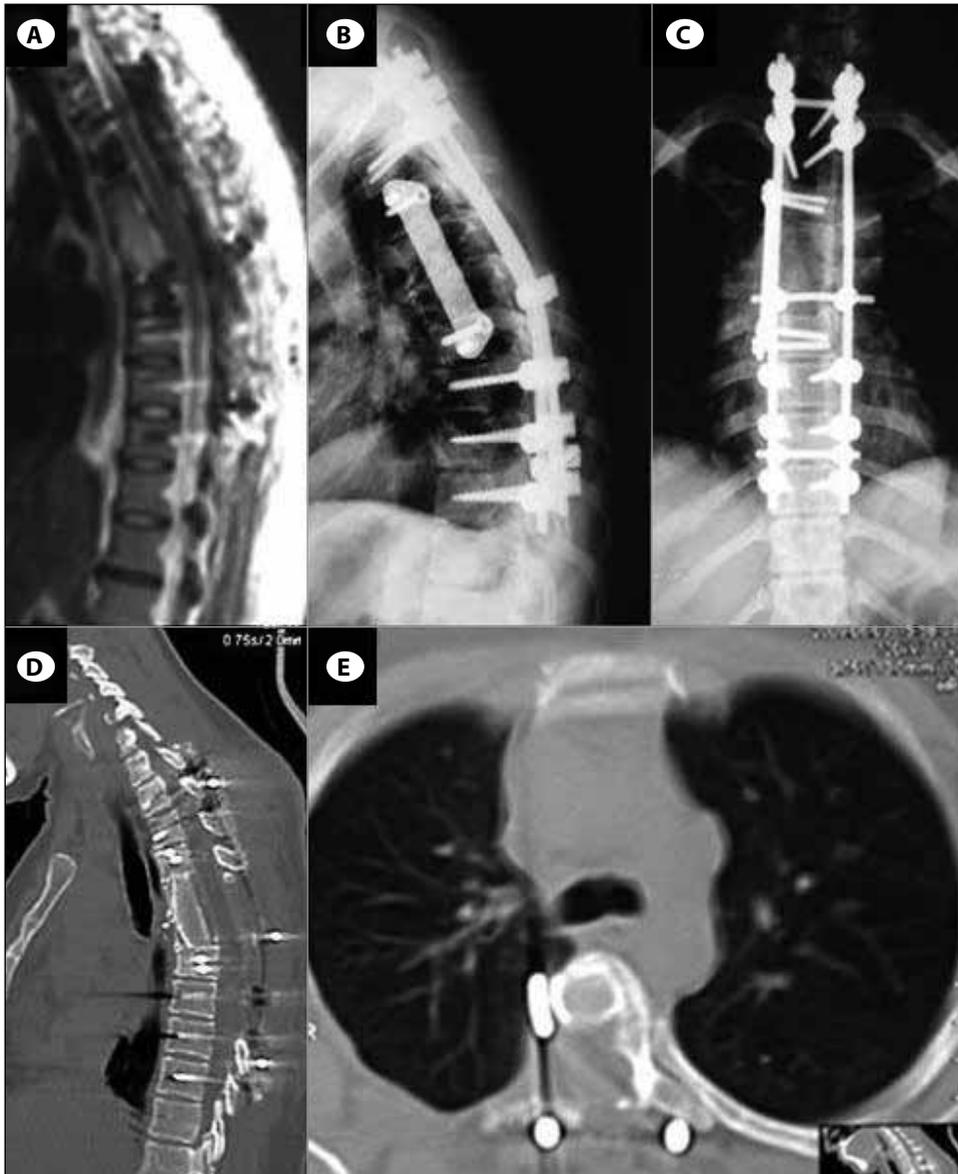


Figure 2: Postoperative images; **A)** Sagittal T2-weighted MRI shows no evidence of residual cysts, **B-C)** Lateral and anteroposterior X-rays shows plate-screw system between T3-T6 and posterior thoracic transpedicular screws through T1, T2, T7, T8, T9 vertebrae, **D)** Sagittal thoracic CT shows improving of kyphotic angulation, **E)** Axial thoracic CT shows plate-screw with bone graft.

Spinal hydatid cyst can present with back and radicular pain, and with signs and symptoms of spinal cord compression, like paraparesis (13). Our case was admitted with paraparesis.

For diagnosis, physical examination and radiological studies CT and MRI scans are helpful. Serologically enzyme-linked immunosorbent assay (ELISA), Western blot, indirect hemagglutination tests, and polymerase chain reactions are generally used for diagnosis. Treatment of spinal HC cyst is surgery accompanied by early postoperative adjuvant antihelminthic medications. Surgical goal is to remove cyst totally without rupturing the cyst, and irrigation of the surgical field with hypertonic saline. There is no consensus for the duration of antihelminthic treatment. Nevertheless, the mean duration in the literature is 3-4 months (12). However, some reports indicated in the range of 6 months to 1 year (11,12). In presented case, patient had received albendazole treatment for a month after first surgery. Currently the patient continued medical treatment for 6 months.

A recurrence is a major handicap for spinal hydatid cysts. Surgery for spinal hydatid cyst is more problematic compared to cranial ones. Recurrence may be because of multivesicular and invasive nature of the cyst in the spinal column. Moreover, spinal cases are prone to be multiple in nature. The other reason is that the cysts are located in such a limited space in the bone that there is a higher inborn risk of cyst rupture (1,3). This was also true for the presented case; particularly during resection of cysts in the bony structures, they may not be protected and some cysts are ruptured.

Subtotal removal of the cyst and rupture leads to a higher rate of recurrences. In the literature, there are various reports of duration and efficacy of antihelminthic treatment to prevent early postoperative recurrences (8,9,11,12). The majority of the studies have suggested 3-4 months of antihelminthic treatment. Prabhakar et al (11) reported 1 year of albendazole treatment, but recurrence rates were 100%. In our case, 6 months of albendazole treatment was implemented, and no recurrence was detected after 2 years of follow up.

CONCLUSION

Spinal hydatid cyst is a rare entity. In endemic regions, it should be considered in the differential diagnosis of spinal cord compression. It is important to remove all the cysts without rupturing and preventing spill to surrounding structures. Spinal instability and recurrences are a major handicap for spinal hydatid cyst particularly with vertebral and paravertebral involvement. Antihelminthic treatment should be given preoperatively and postoperatively. These patients should be followed for recurrences for long periods clinically, serologically, and radiologically.

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