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Echinococcosis of the Spine and Retroperitoneum†

Case Report and Review of the Literature
Eugene Meyer, MD* and Raphael Adar, MD**

Echinococcosis is a zoonosis transmitted from sheep dogs to humans. It occurs most frequently in sheep and cattle-raising countries, and children who come in contact with sheep dogs are the most likely victims. The liver and the lungs are the most common sites of the hydatid cysts. Their presence in other organs is less common, and primary hydatidosis of the retroperitoneum is extremely rare. In the case reported here, a 65-year-old man had secondary retroperitoneal echinococcosis that recurred three times in 23 years. On the third occasion, marsupialization was performed on a large cyst found in the right retroperitoneum.

Humans may accidentally substitute as the intermediary host in the life cycle of the helminth echinococcus granulosus. An individual may be infected by eating food contaminated by excrement of the infected animal, usually a dog. Children in sheep and cattle-raising countries are the most frequent victims since they are in close contact with sheep dogs, the primary host. After the ova are excreted and hatched in the duodenum of the intermediary host, they spread through the intestinal wall into the portal circulation (1). Further spread of the parasite is blocked first in the liver and then in the lung. Hydatid disease beyond the hepato-pulmonary blockade is rare. Here we present a case of spinal and retroperitoneal hydatid disease that recurred three times in 23 years.

Case Report
A 65-year-old man, born in Iraq and settled in Israel in 1950, presented in the Emergency Room* with vomiting, generalized wasting, and a large mass that filled the entire right abdomen. The patient had been operated upon in 1955 and again in 1957 when he presented with cauda equina syndrome due to lumbar echinococcus. During the first operation, a laminectomy (L2-L5) was performed, and cysts were removed from the epidural space. In the second operation, the epidural space was reopened, and more cysts were removed. In both operations, 1% formalin was injected into the main cyst cavity, but live scoleses spilled locally. During the next 21 years the patient remained asymptomatic except for paresis and pain in the right leg.

At his work-up, the abdominal flat plate revealed a large mass filling the right side of the abdomen, and the ultrasound film demonstrated a huge mass with echo reflections and multiple small cystic lesions at its apex suggesting an enlarged infected echinococcus cyst with multiple small daughter cysts (Fig. 1). Transaxillary angiography indicated that the aorta, left iliac artery, and normal right kidney were displaced to the left by a large avascular mass and the right ureter overriding the mass (Fig. 2). The blood count was normal. There was no eosinophilia, and the Weinberg compliment fixation test was negative.

During surgery, one giant cyst was found in the right retroperitoneum that had displaced the small bowel and

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*This case was observed at the Haim Sheba Medical Center, Tel-Hashomer, Israel.
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right colon to the left (Fig. 3). Seven liters of yellow, turbid, odorless fluid were removed (Fig. 4). The cyst extended into the right groin under the inguinal ligament and subcutaneously posterior to the lumbar vertebrae. Spinal bone fragments were removed from the medial dorsal area of the cyst. The cavity was washed with 3% formalin even though daughter cysts were not seen. As it was impossible to remove the entire cyst wall, marsupialization was performed, and drains were placed. Cultures for aerobe, anaerobe, fungus, and tuberculosis were all negative. Echinococcosis was verified by microscopic examination of the cyst wall and bone fragments.

The patient’s postoperative course was uneventful, and he was eventually discharged.

**Literature review**

The incidence of hydatid disease in various organs differs considerably. It is most commonly found in the liver (55-70%), the lung (15-30%), and the systemic circulation (10-15%), but it can settle in any body organ such as the kidney (2.5%), heart (2.5%), bone (2%), muscle (1%), brain...
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(0.5%), and in other organs (1.5%) (2-4). Osseous hydatidosis is most frequently localized in the vertebrae, the lumbar and thoracic vertebrae, in particular (5,6), which may be due to the rich vascularity of the vertebra (7). Hydatidosis may begin in bodies, pedicles or lamina of the vertebrae and may remain confined to the bone for many years before extraosseous spread occurs. Symptoms may vary from paraparesis, paresthesia, and sphincter disturbances to paraplegia. Suri, et al found the incidence of paraplegia to be as high as 25-50% (4). Radiologic appearances are variable. The patient afflicted with vertebral echinococcosis undergoes an average of 2.6 operations, mainly laminectomy with extirpation of the cyst. The early postoperative results are good, but later results are poor due to recurrences. Recently, more aggressive surgery has been advocated that would completely remove the affected vertebrae and provide for further exploratory operations and a close follow-up. Suri, et al stressed the importance of preoperative diagnoses to plan the proper surgical approach to prevent recurrence due to contamination. Paravertebral lesions are almost invariably secondary to vertebral hydatidosis (4-8).

Retroperitoneal echinococcosis is rare and difficult to diagnose. Primary hydatidosis of the retroperitoneum is extremely rare, with only eight cases reported in the literature, but secondary retroperitoneal echinococcosis, as in our case, is not uncommon (2). Porat and Joseph reported a case of osseous hydatidosis with secondary retroperitoneal recurrence with a 14-year follow-up (8). Mosavy, et al reported ten cases of echinococcal cysts beyond the pulmonary sieve, one of which was retroperitoneal (1). Preoperative diagnosis was difficult, although radiography was most useful. Chatterjee, et al reported one case in the retroperitoneum among their collection of hydatid cysts in unusual sites (12). Others have reported cysts in specific retroperitoneal structures such as the kidney and pancreas (3,9,11). Recently, it has been reported that mabendazole has been used to replace or supplement surgical treatment. Preliminary reports are encouraging but longer follow-up will be needed for evaluation (13,14).

Our case is unusual for its ultra-sonographic and angiographic findings, negative eosinophilia and compliment fixation tests, prolonged morbidity, and for its clinical course.

References