Large solitary retroperitoneal echinococcal cyst: A rare case report

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Abstract

Echinococcal disease remains a problem within some endemic areas. Echinococcal cysts usually involve the liver and lungs, but any other organ can potentially be involved. Extrahepatic localization is reported in 14%-19% of all cases of abdominal hydatid disease. We report the case of a large echinococcal cyst localized in the lower pelvis. A 28-year-old woman was admitted to a surgical ward with lower abdominal pain and discomfort lasting for a month. Ultrasonography and computed tomography scanning revealed a large retroperitoneal cystic mass (9 cm × 4 cm) in contact with the left ovary and left ureter. There were no cysts in any other location. Serological tests were positive for Echinococcus. The patient was operated on and the entire cyst was excised intact. Histopathological results confirmed the diagnosis of echinococcosis. Anthelmintics were administered postoperatively and the patient was discharged after 6 d, and is now being closely followed up. Total cystectomy when possible represents the treatment of choice for large extrahepatic echinococcal cysts.

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Key words: Echinococcus cysts; Ultrasonography; Extrahepatic location; Seropositivity; Anthelmintics; Total cystectomy

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lesions, is an infection of humans caused by the larval stage of *Echinococcus granulosus*. It is prevalent in the Middle East, the Mediterranean region, particularly in Greece and Lebanon, Australia, Argentina and Africa. Confirmed hosts are dogs that pass eggs into their feces. Intermediate hosts, e.g. sheep, cattle, humans, goats and horses, ingest the eggs and develop cysts^[1]. Echinococcal cysts are mostly found in the liver (60%-70% of cases), followed by the lungs (10%-25%), spleen, ovaries, kidneys, brain, bones and heart, but rarely elsewhere in the body^[1]. Hydatid disease in extrahepatic locations usually remains asymptomatic unless the cyst grows and produces symptoms due to pressure, rupture to the pleural or peritoneal cavity, secondary infection, or an allergic reaction^[2]. We report the rare case of a 28-year-old woman with a large hydatid cyst in her lower pelvis, without hepatic or any other involvement.

CASE REPORT

A 28-year-old woman was admitted to our surgical ward with vague abdominal pain localized in the hypogastrium, and severe constipation, which had lasted for the previous 20 d. Her gynecologist had diagnosed a large cystic mass adjacent to the left ovary. She did not report episodes of fever, vomiting or diarrhea. Neither did she note any alterations in her menstrual cycle. Blood tests showed mild leukocytosis (12000 white blood cells). Plain abdominal X-rays did not show any specific diagnostic findings. Abdominal ultrasonography (US) revealed a large cystic mass (9 cm × 4 cm) in contact with the left ovary and left ureter. Computed tomography (CT) confirmed the US findings (Figure 1). There were no cysts in any other location. Serological tests were positive for echinococcal disease. The patient underwent a laparotomy, and a large cystic mass was identified retroperitoneally, firmly attached to the sacrum and in contact with the left ovary, left ureter and the sigmoid colon. Total cystectomy was performed en block with a portion of the mesosigmoid. The sigmoid vasculature remained intact, and there was no evidence of intraoperative colon ischemia. Postoperatively, anthelmintics were administered and the patient was discharged after 6 d. She had been closely followed up for 3 mo, without any abdominal pain and with significant improvement of constipation.

INTRODUCTION

Echinococcal disease, which produces unilocular cystic

DISCUSSION

Echinococcosis remains a problem in endemic areas. *E.*

Figure 1 CT image of a large retroperitoneal echinococcal cyst in contact with the left ovary and left ureter.

granulosus is a 5-mm long worm, with a lifespan of 5-20 mo within the jejunum of dogs. When eggs are ingested by an intermediate host, the embryos escape, penetrate the intestinal mucosa, enter the portal circulation, and are then trapped in the liver^[3]. A small number escape the hepatic filter, enter the systemic circulation, and are scattered to other organs. Larvae develop into fluid-filled unilocular hydatid cysts that consist of an external membrane and an inner germinal layer. Daughter cysts originate from the inner layer^[3,4]. Slowly enlarging echinococcal cysts generally remain asymptomatic. Five to 20 years elapse before cysts enlarge sufficiently to cause symptoms. Abdominal pain, hepatomegaly or a palpable mass in the right upper quadrant are the most common symptoms for patients with liver echinococcosis^[1]. Bile-duct compression or leakage of fluid into the biliary tree may mimic cholelithiasis. Biliary obstruction can result in jaundice. Generalized toxic reaction due to hydatid cyst rupture and secondary infection are among the most common complications^[5].

Cysts in the peritoneal cavity account for 10%-16% of cases and are mainly the result of the rupture of concomitant hepatic cysts^[2]. Extrahepatic locations of the echinococcus include the lungs (10%-15%), spleen (0.9%-8%), kidneys (1%-4%), pancreas (0.25%-0.75%), brain, heart, ovaries, bones and abdominal wall. Symptoms in such cases occur because of pressure or complications including rupture, allergic reaction and secondary infection^[6]. Radiography, US and CT studies are important for a diagnosis of echinococcal disease. Plain abdominal X-rays may show calcifications of the cystic wall^[7]. US is the method of choice for the detection of hepatic and extrahepatic echinococcal cysts. Hydatid cysts are classified by ultrasound into six categories.

Type I are defined as univesicular and < 50 mm in diameter. Type II are univesicular with a prominent laminated layer, and tend to be seropositive. Type III are subdivided into IIIa, defined as cysts with a prominent lamination that contains daughter cysts, and IIIb, characterized by lamination but a lower number of daughter cysts. Both IIIa and b are highly seropositive. Type IV appear as solid masses. Type V are characterized by degeneration with calcifications. Type VI are defined as multiple cysts that may be univesicular and laminated,

with daughter cysts involving one or more organs^[8]. The sensitivity of US ranges from 93% to 98%^[5].

CT confirms the diagnosis by revealing the presence of daughter cysts and plaque-like calcifications in the cystic wall. It is important as it provides information regarding the exact location of extrahepatic cysts in relation to neighboring structures. CT sensitivity ranges from 90 to 97%^[5,9].

Serological tests contribute to diagnosis. Immunoglobulin G antibody detection by ELISA has a sensitivity of 95% and a specificity of 94%^[8]. The sensitivity of indirect hemagglutination test has been found to be 87.5%^[8,10].

Therapy for extrahepatic echinococcal disease is based on considerations regarding the size, location and manifestations of the cysts, and the overall health status of the patient. Asymptomatic small cysts once diagnosed can be treated with antihelminthic drugs, administered for 28 d in one to eight repeating cycles, separated by drug-free intervals of 2-3 wk^[11].

For symptomatic or large hydatid peritoneal cysts, surgery, when feasible, is the principal method of treatment. Surgical treatment can be either radical or conservative. Total cystectomy, whenever possible, is the gold standard. For peritoneal cysts firmly attached to intraperitoneal viscera, unroofing and drainage has been proven to be a safe method^[11,12]. It is important that the abdominal cavity is isolated with gauzes soaked in 20% hypertonic saline solution to avoid secondary hydatosis and allergic reaction^[11].

In our case, to eradicate the disease, total cystectomy was carried out with meticulous dissection of the left ureter, gonadal vessels, hypogastric nerve plexus and the mesosigmoid vasculature. Thus, the sigmoid remained intact, free from compression, and constipation was greatly improved postoperatively.

In conclusion, a diagnosis of extrahepatic echinococcal disease is more accurate today because of the new imaging techniques available. The treatment of choice for small asymptomatic hydatid cysts is conservative by administration of antihelminthic drugs. Large or symptomatic echinococcal cysts need to be treated by total cystectomy, or unroofing and drainage followed by adjuvant antihelminthic therapy.

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