

Concomitant Splenic and Hepatic Hydatidosis: Report of Two Cases and Review of the Literature

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Abstract- Hydatid disease has a worldwide spreading. The most common site of the disease is the liver, lungs, kidney, bones, and brain. Splenic hydatid disease has been reported to constitute 2% to 6% of patients with abdominal hydatid disease. Because of the rarity of splenic hydatid disease, the probable concomitance of the liver and splenic hydatid cysts should be taken into consideration by clinicians, especially in nonendemic areas. In this report, we present two patients with concomitant splenic and liver hydatid cysts that underwent splenectomy and cystostomy–capitonage.

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Introduction

Hydatid disease is a zoonotic infection caused by the larval stage of the genus *Echinococcus* (1). It is an endemic disease in the Middle East and the Mediterranean countries. Now, it has worldwide spreading pattern even in developed countries (2).

The hepatic hydatid cyst is a major health problem in endemic areas especially in sheep-raising areas and farming regions (2, 3).

The most common sites of disease are the liver (approximately 70%), the lungs (15%–47%), while the kidney (2-4%), bones and brain are less to be involved. Reports indicate very rare incidence of the disease in other sites such as the heart, spleen, pancreas and muscles (3-5). Simultaneous involvement of two organs is about 5-13% of cases (6).

The splenic involvement in hydatid disease is uncommon, representing less than 2%- 6% of all human infestations by *Echinococcus* (3-8). The infestation of the spleen usually occurs either by arterial route through the hepatic and pulmonary filters or retrograde venous route in portal hypertension (9). Thus, hydatid cysts in spleen may occur as a part of disseminated disease or may be isolated (6,7). Because of the rarity of splenic hydatid disease, the probable concomitance of the liver and splenic hydatid cysts should be taken into consideration by clinicians, especially in nonendemic areas (4).

In this report, we describe two patients with

concomitant splenic and liver hydatid cysts.

Case Report

Case 1

An 18-year-old woman was admitted to Imam Reza Hospital (Mashhad, Iran) with right upper quadrant and left hypochondrium 's symptoms had started from one year ago and exacerbated from two months ago. She had no history of fever or nausea and vomiting. Routine laboratory tests including renal and liver function tests, hemoglobin level, total leukocyte count and differentiation, were all within the normal range. Cyst hydatid indirect haemagglutination (IHA) test was also negative. Abdominal examination revealed a palpable mass in the left upper quadrant of the abdomen and fullness in epigastric region.

Abdominopelvic computed tomography (CT) scan showed a non-enhancing regular cyst of 16×15×14 cm without any septation, originating from the spleen, filling the left quadrant of the abdominal cavity and a second loculated cyst of 7×6×5 cm in size originating from the left medial lobe of the liver, indicating concomitant splenic and liver hydatid cysts (Figure 1).

On exploration, in the operating room we identified a huge hydatid cyst in the spleen filling the entire left side of the abdomen pushing the intestines and a second hydatid cyst in the left lateral segment of the liver (Figure 2).

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Figure 1. a) CT scan reveals a giant cystic mass in the spleen and a normal sized loculated cyst in the liver. b) CT scan showing a large cyst of the spleen and a second cyst in the right lobe of the liver



Figure 2. Intraoperative images of a huge splenic hydatid cyst (Case 1)

We decided to perform a splenectomy. The splenic vessels at the splenic hilum were controlled and after packing off the abdomen with hypertonic (20%) saline-soaked gauze to guard against spillage, we performed a splenectomy and then a cystostomy–capitonnage to the cyst in the left lateral segment of the liver. Histopathologic examination confirmed the disease. In the postoperative period, the patient's condition was good, and no complications were observed. She was discharged with albendazole treatment.

Case 2

A 30-year-old woman was referred to our clinic with symptoms of right upper abdomen pain and left hypochondrium discomfort. In the physical examination she had a nontender, palpable mass in the left upper quadrant of the abdomen. Routine laboratory tests including liver function tests, hemoglobin level and leukocyte count, were in normal ranges. Abdominopelvic CT scan showed a 15×14×13-cm cyst with septation originating from the spleen, and a second

cyst of 10×9 cm in size originating from the right lobe of the liver. The IHA test showed a positive result.

She underwent open drainage and capitonnage of the liver cyst along with a splenectomy. The postoperative course was uneventful (good or uncomplicated), and the patient was discharged with albendazole treatment.

Two years later, she came back with a complaint of dull pain in epigastrium. The investigations showed a new cyst in the right lobe of the liver. So she underwent another surgery for the recurrent cyst. During the procedure, the cyst was evacuated, and the cavity was then irrigated with a scolecidal agent. Four days after operation she was discharged on albendazole therapy.

Discussion

Hydatid disease caused at the larval or cyst stage of infection by the tapeworm *Echinococcus granulosus*, which exists in the dogs (1).

Human can be infected by ingestion of contaminated water or vegetables. The parasite involves the liver

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through the portal venous or lymphatic system and then lung, kidney, bones, and brain (9).

Other sites such as the heart, spleen, pancreas and muscles are very rarely affected. Splenic hydatid disease has been reported to constitute less than 2% up to 6% of patients with abdominal hydatid disease (3-8). In patients with portal hypertension or rupture of hepatic cyst, splenic *echinococcosis* may develop by retrograde spread from the liver via the hepatic portal and splenic veins into the peritoneal cavity (5).

Nearly 30% of splenic hydatid cysts are asymptomatic. They usually grow very slowly (2-3 cm each year) and the patient may remain asymptomatic for years before diagnosis; so they may be detected during the investigation for symptoms incidentally (4,6). The patients usually have non-specific symptoms such as the abdominal mass, dull dragging ache, dyspepsia, constipation due to pressure on the colon, and dyspnea due to pushing up of the left diaphragm. They may also present with complications such as infection of the cyst, rupture of the cyst into the peritoneal cavity, fistula formation into hollow viscera like colon or stomach, bronchopleural fistula, sympathetic pleural effusion, calcification, hypersplenism, and severe urticaria. Splenomegaly is the very common finding in these patients (4-8). Current patients reported a long history of abdominal pain and discomfort at the time of admission.

Echinococcosis IHA with a specificity of 90-100% and a sensitivity of 68.4 % is used for the diagnosis of the infection, (1) as in present patients it was positive. The test results may be negative if the cyst has not leaked or does not contain scolices, or if the parasite is not viable (1).

Ultrasonography and CT scanning of the abdomen are both quite sensitive for detecting hydatid cysts. The imaging appearances of the cysts depend on the stage of cyst development (1). Some imaging characteristics of splenic hydatid cysts include calcification of the cyst wall, presence of daughter cysts and membrane detachment (4). CT scanning provides additional structural details than ultrasound; it shows the precise location and depth of the cyst within the liver (10).

Other splenic cystic lesions such as epidermoid cysts, pseudocysts, splenic abscesses, hematomas and cystic neoplasms of the spleen should be considered in the differential diagnosis of splenic hydatid cysts (4).

Medical treatment with albendazole can be administered as initial treatment for small and asymptomatic cysts (1). Complications related to splenic hydatid cyst such as spontaneous or traumatic rupture of the cyst make surgical treatment mandatory.

One surgical approach is percutaneous aspiration along with irrigation with hypertonic saline to wash out the cyst contents and to kill the protoscolices; however splenectomy is the treatment of choice in most centers (3,10-13). Because of the risk of spillage and anaphylaxis, other approaches such as laparoscopic and percutaneous drainage have not been widely accepted (10). In some studies, partial cystectomy is recommended when the cysts are located at the margins of the spleen (14).

A 10-year randomized clinical trial in Shiraz, Iran to investigate the long-term outcome of spleen-preserving surgery versus splenectomy for splenic hydatid cyst showed no differences between the two groups concerning the median hospital stay, postoperative complications and recurrence. The study included 20 patients (10 for splenectomy and 10 for spleen preserving surgery), with a mean follow-up of 52 months (7). Ozdogan *et al.*, study on 14 patients compared the results of partial cystectomy and omentopexy versus splenectomy and reported that partial cystectomy and omentopexy can be considered as a procedure of choice for the treatment of splenic hydatidosis (15). Meimarakis *et al.*, reported three patients treated with spleen preserving surgeries which were recurrence-free during the mean 8.8-year follow-up period (14).

Current patients underwent total splenectomy, and drainage and capitonage of the liver cyst were performed for both of them. The second case came back two years later with a new cyst and underwent a second surgery.

In conclusion, although in some studies spleen preserving surgery is recommended in selected small cysts located in spleen margins, present investigation indicated that in concomitant splenic and hepatic hydatidosis, splenectomy is a safe procedure with minimal risk of recurrence and complications such as spillage and anaphylaxis after surgery.

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