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CASE REPORT

GIANT ISOLATED SPLENIC HYDATID CYST: A CASE REPORT

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ABSTRACT

Hydatid disease is a parasitic infection caused by the genus Echinococcus. Giant isolated splenic hydatid cyst is a very rare occurrence. In this case report, we present a 26-year-old female with giant splenic hydatid cyst from Yekatit 12 Medical College Hospital, Addis Ababa, Ethiopia. The clinical, imaging and laparotomy findings are discussed.

Key words: *Echinococcus, Splenic hydatid cyst, Splenectomy*

INTRODUCTION

Hydatid disease is a zoonotic infection. It is caused by the genus Echinococcus. Of the four known species of echinococcus, 3 are of medical importance in humans. These are *Echinococcus granulosus, Echinococcus multilocularis* and *Echinococcus vogeli*. *Echinococcus granulosus* is the most common species that causes cystic echinococcosis. The dog is the definitive host and humans are incidental host who contract the disease by ingesting highly infective eggs of adult echinococcus. The infection is usually acquired in childhood and mostly remains asymptomatic. The cyst grows slowly at a rate of 1-3 cm per year and sometimes it may take 5-20 years to grow into size to cause symptoms of abdominal discomfort. The liver is affected in approximately two-thirds of patients, the lungs in approximately 25 percent, and other organs including the brain, muscle, kidney, bone, spleen, heart, and pancreas in a small proportion of patients. Eighty-five to 90 percent of patients with *E. granulosus* infection have single-organ involvement, and more than 70 percent have only one cyst. Splenic involvement in hydatid disease is a rare occurrence with the worldwide incidence of 0.5- 4%. Secondary infection, fistulisation to adjacent organs and rupture into the peritoneal cavity are some of the complications observed with splenic hydatid cyst. Spontaneous or traumatic rupture of a hydatid cyst may cause a life threatening systemic anaphylactic reaction. Open splenectomy is the standard procedure for splenic hydatid cyst(1-6, 8-15).

The first case of splenic hydatid cyst was reported by Berlot in 1790 from an autopsy (7). Humphreys et al reviewed 6 cases of splenic hydatid cyst (5). Christos Limas reported a case of splenic hydatid cyst with numerous unilocular cysts in the peritoneal Cavity (15). We report a rare case of giant primary splenic hydatid cyst in a 26-year-old female patient managed by total splenectomy from Yekatit 12 Medical College Hospital, Addis Ababa, Ethiopia.

CASE REPORT

A 26 years old female patient came to Yekatit 12 Medical College Hospital with a complaint of dull aching left upper quadrant abdominal pain of five months duration which was associated with a slow growing mass over the same area. She had no cough or abdominal trauma. Her past medical history was unremarkable. She has two dogs at home. On physical examination, patient is comfortable and her vital signs were within normal limits. Abdominal physical examination revealed a palpable mass 10 cms below the left hypochondrium along the line of splenic growth. It was a non tender mass the upper border of which was not possible to reach. Complete blood count and organ function tests revealed no abnormality. Abdominal ultrasound showed a complex cystic mass at the left upper quadrant of the abdomen extending to the left Para renal space. The mass had double wall layer with areas of interrupted wall calcification. It measured 19cm×13.5cm. The spleen was only partially seen. All other abdominal organs were normal (Figure 1).

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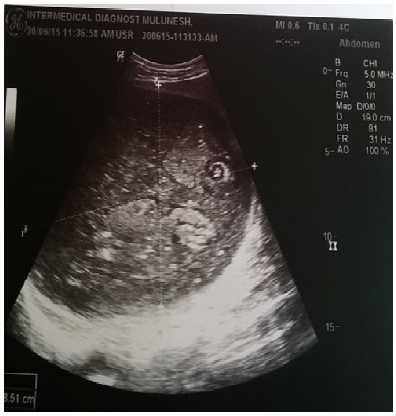

Figure 1: Ultrasound of the abdomen demonstrating a complex cystic mass at the left upper quadrant

CT scan of the abdomen revealed 14.7 cm×18.5 cm well defined cystic mass with interrupted areas of wall calcification in the spleen. There was a focal fat density area within the cyst. There were multiple daughter cysts with detached membranes and matrix. All other abdominal and pelvic organs were normal. The CT scan impression was splenic hydatid cyst (Figure 2).

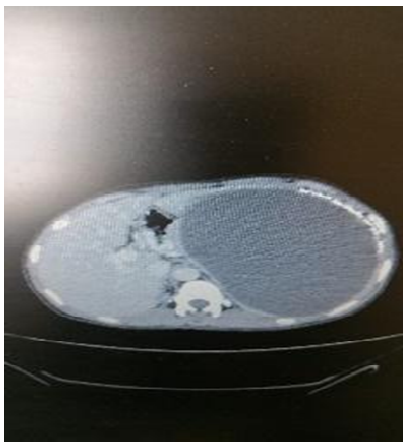

Figure 2: CT scan of the abdomen demonstrating splenic hydatid cyst

Patient was admitted and started on albendazole 400 mg PO bid for 5 days. Consent was obtained for exploration and laparotomy was performed through upper midline incision. The intra operative finding was a giant cyst occupying the whole splenic parenchyma and only thin small rim of splenic tissue was present in inferior surface. It measured about 25 x 30 cm with fibrous capsule. The cyst was adhered to the left diaphragm, liver and omentum. The adhesions were released and the cyst was resected en bloc with the spleen (Figures 3a and 3b).

Figure 3a : Intraoperative view of delivered giant splenic hydatid cyst with small splenic tissue

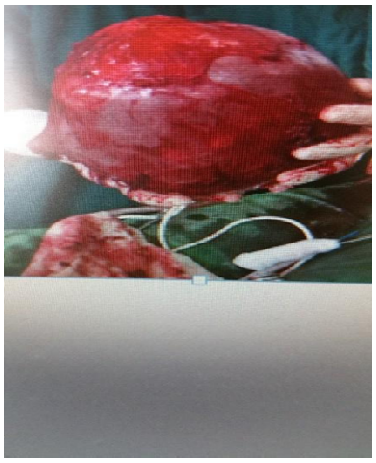

Figure 3b: Removed giant splenic hydatid cyst

Hemostasis was secured and drainage tube was left in the splenic fossa. The rest of the abdominal organs including the liver were normal. Abdominal wall was closed in layers. Cut section of the cyst revealed 4.5 liters of fluid. There were protoscolices which were attached to the cyst wall and were freely floating in the milky fluid contents giving it a sandy consistency (Hydatid sand) (Figure 4).

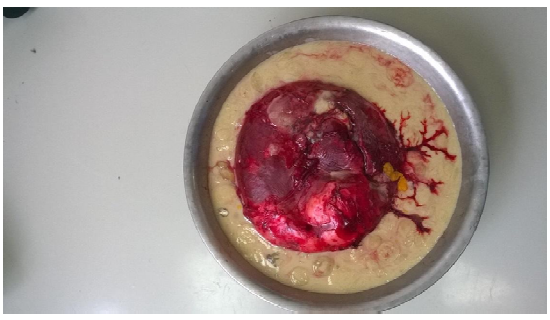

Fig 4. Cut section of the hydatid cyst showing hydatid sand with floating protoscolices

The drainage tube was removed on the 5th postoperative day. The postoperative course was uneventful and the patient was discharged on the 10th postoperative day. Albendazole was continued in a dose of 400 mg PO bid for 4 weeks. Computerised tomographic follow up at 6 months demonstrated the absence of recurrence.

DISCUSSION

Hydatid disease is common in sheep and cattle raising areas of the world where dogs have access to animal offal. It is a significant public health problem in South and Central America, the Middle East, some sub-Saharan African countries and China. Hyadatid cysts are usually single, but multiple cysts may occur.

The colony of hydatid scolices increases in size over many years displacing the surrounding parenchyma attaining a larger diameter cyst. Surrounding the cyst is an outer layer of adventitia, pericystic layer, which does not belong to the parasite but represents host parenchyma compressed into a fibrovascular layer. This layer gives some mechanical support to the cyst and facilitates exchange of nutrients and metabolic products between host and parasite.

Hydatid cysts may occur virtually anywhere in the body. Seventy per cent of all hydatid cysts are found in the liver which acts as a first filter. The right lobe of the liver is most frequently involved, probably because most of the portal blood from the upper gastrointestinal tract reaches this area. The lungs act as a second filter and pulmonary hydatid cysts account for 10-40%.

Hydatid cysts grow by expansion and tend to make their way towards regions of less resistance. In the liver they protrude from the depth of the organ towards the peritoneal surface, particularly beneath the diaphragm. The growth rate of cysts depends on their location. Five to 10 years may take for a hydatid cyst to attain a size of a tennis ball in the liver, whereas a similar size in the lung is reached in about a year. Splenic involvement is an uncommon event because cyst embryos are trapped in the liver and lungs, with only 15% entering systemic circulation. The eggs of parasite escape the liver-lung barrier and cause primary infestation of spleen through the arterial route. Splenic hydatid disease may also arise with retrograde spread of parasites via the portal and splenic veins bypassing the lung and liver.

These situations make splenic hydatid disease a rare clinical condition the frequency of which is reported to be 0.5%- 4% of all abdominal hydatid diseases even in the endemic areas. Secondary splenic hydatid disease usually follows systemic disseminated or intraperitoneal spread following ruptured hepatic hydatid cyst (1-6, 8-17).

Our patient presented with left upper quadrant abdominal pain and abdominal swelling over the same area. The presentation of splenic hydatid disease can vary greatly. Splenic hydatid cysts are usually asymptomatic, solitary, slow growing and incidentally diagnosed. The main symptoms associated with the disease are abdominal discomfort and palpable mass in the left upper quadrant. Tarccoveanu E. reported 38 cases of splenic echinococcosis and abdominal pain was the most common symptom among these patients (9).

Growth may cause compression of the segmental vessels of the spleen, which results in extensive pericystic splenic atrophy and the hydatid cyst may entirely replace the splenic parenchyma. The main differential diagnosis of splenic hydatidosis are splenic abscesses, epidermoid cysts, hematomas, posttraumatic pseudocyst and neoplasms like lymphangioma and hemangioma. Pre-operative diagnosis may be difficult due to the similarity of the presenting symptoms and the radiological findings to those of other more commonly encountered lesions of the spleen. The Casoni skin test is sensitive but not specific.

Ultrasonography and computed tomography are the major diagnostic tools for splenic hydatid cyst. Serological tests are highly sensitive and specific for echinococcosis. Drug therapy alone has limited efficacy in the management of hydatid cyst. Owing to the risk of spontaneous or traumatic rupture of hydatid cyst, splenectomy is still accepted as standard as complete resection removes all parasitic and pericystic tissues(1-6,8-15).

During surgical treatment extreme caution must be taken to avoid rupture of the cyst and content spillage. Total splenectomy, partial splenectomy, cyst enucleation and unroofing with omentoplasty are the various surgical techniques to treat splenic hydatid disease. Partial splenectomy carries a risk of poor vascular control when incising the splenic tissue while unroofing the cyst wall leaving behind a residual cavity carries the risk of postoperative infection. For the above reasons and the possibility of multiple splenic cysts, total splenectomy should be the method of choice, especially in the presence of adhe-

sions between the spleen and nearby organs, such as the stomach and diaphragm (12,13).

Albendazole is an effective adjuvant therapy in the treatment of hydatid cyst. The chance of recurrence is less in patient who received albendazole therapy (1 -6, 8-15). We performed total splenectomy along with excision of part of the adhered diaphragm. After total splenectomy, albendazole was continued for 4 weeks in a dose of 400 mg PO bid. The patient was also given prophylactic vaccination. After splenectomy, no infection was encountered. According to our knowledge this is the first case report of primary giant solitary hydatid cyst of the spleen of the size 25X30cm containing hydatid sand fluid of around 4.5 litters.

In conclusion, hydatid disease should be considered in the differential diagnosis of cystic masses in the spleen, especially in the geographical regions where the disease is endemic. Proper preoperative evaluation should be carried out. Ultrasonography and computerized tomography are important investigative modalities of which computerised tomographic scan is the most sensitive investigation for diagnosis. Splenic hydatid cyst may be a challenging surgical problem but surgical resection is the best curative procedure. Even a giant splenic hydatid cyst can be removed without breaching its wall and spilling its contents into the peritoneal cavity, a crucial focus in preventing disseminated peritoneal echinococcosis. Postsurgical albendazole treatment is necessary to ensure complete recovery.

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