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Anaphylaxis due to spontaneous rupture of primary isolated splenic hydatid cyst

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Abstract

Echinococcosis is a helminthic zoonosis mainly caused by *Echinococcus granulosus* and commonly encountered in endemic areas. The liver and lung are the most frequently involved organs. A primary isolated hydatid cyst of spleen is an extremely rare disease even in endemic areas. Anaphylactic reaction is a known complication of cystic hydatid disease, a parasitic infestation caused by the larval/cyst stage of *E. granulosus* that usually occurs after trauma or during interventions. To the best of our knowledge, anaphylaxis with spontaneous rupture of primary isolated splenic hydatidosis had not been reported previously. The main purpose of this report is to highlight life-threatening complications such as anaphylactic shock that should be considered due to primary isolated splenic cyst hydatid rupture in especially endemic regions.

Keywords: Anaphylactic shock, computed tomography, hydatid cyst, isolated, spleen

INTRODUCTION

Hydatid disease (HD), which is caused by *Echinococcus granulosus*, is a common parasitic infestation especially in endemic regions such as the Middle East, Mediterranean, and South American Countries. Even though it mainly involves the liver, it has been reported in nearly all parts of the body.[1] Isolated splenic involvement is an extremely uncommon condition even endemic regions.[2] The incidence of the splenic cysts in HD varies from one series to another, ranges 0.5–4% of all cases of echinococcosis.[1,2] Generally, a mean incidence of 3% is reported.[1,2,3] However, one study showed that HD is common in Jordan, but isolated spleen disease is described only in 2.5% of cases.[1] Splenic hydatid cysts are generally asymptomatic. Symptoms of splenic hydatidosis is usually mild and is mainly caused due to pressure of adjacent organs such as colon/diaphragm or the presence of complications. The patients usually complain of mild discomfort or pain in the left hypochondrium. The diagnosis is established generally during imaging for other reasons. Cysts may become infected or may rupture into the peritoneal cavity, especially subcapsularly located cysts like our case. Cyst rupture into the peritoneal cavity is a rare but serious complication. The hydatid fluid is antigenic and highly toxic and can cause a potentially fatal anaphylactic reaction in humans. Herein, we describe an acute happening of anaphylactic shock due to a spontaneous rupture of the primary isolated hydatid cyst in a 57-year-old woman.

CASE REPORT

A 57-year-old woman was admitted to the emergency department for diffuse maculopapular rashes on the whole body and anaphylactic shock. She had been complaining of sudden-onset of abdominal pain, dyspnea, pruritus, and urticaria. On physical examination, it revealed tenderness, guarding, and rigidity

all over the abdomen. Her blood pressure was 90/60 mmHg, heart rate 118/min, and temperature 37.0°C. Blood test analysis was normal and subsequent leucocytosis with eosinophil dominance. Hydration, hydrocortisone, and antihistamines were used to treat the possible anaphylactic reaction. The patient received oxygen as supportive treatment. The patient recovered after emergency medications and then abdominal computed tomography (CT) revealed a splenic cystic lesion measuring about 9 × 6 cm² in diameters and containing floating membranes. The anterior wall of the cyst showed discontinuity at a point [Figure 1]. Diffuse pelvic and minimal perisplenic fluid were detected. Our fundamental concerns were to preserve the spleen. Then, the patient underwent a laparoscopic partial cystectomy and omentoplasty, since the spread of the disease into the peritoneal cavity of lesions was strong contraindications for a radical surgical approach. At the time of laparotomy, the cyst's outer membrane was seen ruptured [Figure 2]. The postoperative course was uneventful, and the patient was discharged 10 days after the surgery. We applied albendazole, 10 mg/kg/day for 6 months, and the patient was followed up with CT that displayed no recurrence of the disease during a follow-up of 6 months [Figure 3]. She is still symptom-free in the 12-month follow-up.

DISCUSSION

Hydatid disease is a parasitic infestation caused by the larva of echinococcus, more frequently encountered in endemic areas such as Mediterranean countries, South America, Africa, and Middle East. The liver and lung are the most frequently involved organs.[1,4] Hydatid cyst of spleen is an extremely rare disease even in endemic areas.[1] Even though various theories have been published about the pathophysiology of splenic hydatidosis, the most suitable study was published by Bourgeon *et al.*[5,6] The development of echinococcal cysts in the spleen is uncommon because hexacanth embryos are usually trapped in the liver (first Lemman's filter) or lung (second Lemman's filter), but will be trapped in the splenic capillaries once in the systemic circulation. Splenic echinococcosis may also occur by retrograde reach from the liver to the spleen via the portal and splenic veins in portal hypertension. The spleen may also be affected due to rupture of a hepatic echinococcal cyst into the peritoneal cavity.[7] Echinococcal cysts of the spleen usually grow very slowly approximately 2–3 cm per year.[2] Patients remains asymptomatic for 5–20 years before the diagnosis, and approximately 30% of splenic cysts are asymptomatic.[8] In these asymptomatic patients, the diagnosis is established during investigations for other reasons. When the cyst reaches a huge size, it presents with a painful mass in the left upper quadrant. If the cyst is infected, or abscess formation has been occurred, left upper quadrant pain, fever, and leukocytosis develop.[9] Cysts may rupture into the peritoneal cavity, especially subcapsularly located. Rupture into the abdominal cavity is a rare but serious complication[10] Because the hydatid fluid is antigenic and highly toxic. Anaphylactic reaction to the hydatid cyst usually occurs after microscopic or macroscopic rupture of the cyst and leakage of contents into the peritoneum or blood circulation. The response can be vary from a mild hypersensitivity reaction to a fatal anaphylactic shock. [4,10] Rupture of a splenic hydatid cyst occurs commonly secondary to trauma, but it may also rarely occur spontaneously such as our patient.[4,10] The diagnosis is mainly historical and geographic background of the patient, physical examination, serology, and visualization of peripheral calcification or daughter cysts within the large cystic lesion or coexistent cystic lesions with radiologic examination in the liver or other organs. Ultrasound and CT imaging have been used successfully in the detection of a ruptured hydatid cyst.[1,2] At present, ultrasonography is the most valuable and most widely available imaging modality for the diagnosis, classification, and follow-up of splenic hydatid cysts. Ultrasound and CT are complementary examinations for diagnosis. The presence of collapsed membranes in a cystic lesion is pathognomonic for HD.[3] CT is more sensitive than plain films or sonograms in depicting subtle cyst wall calcification. CT is an important imaging modality to have the actual number and location of the cysts not only in the spleen, but also in the abdomen.[3,9] The presence of a cyst in the spleen of a patient who developed anaphylactic reaction with detection of membrane-like structures inside the cyst and discontinuity of the wall are considered diagnostic for ruptured hydatid cysts such as our patient. Emergency surgery remains the only effective therapy for a ruptured cyst.[8] Surgical excision is still the only curative treatment, but good results have been reported with albendazole administration. Medical treatment is indicated in cysts inaccessible for surgical removal or as a complementary therapy to prevent recurrence.[8] Albendazole is indicated for 6 months to reduce the

risk of distant recurrence.[11] The surgical procedure consists of removing the cyst and possible daughter cysts spilled into the abdominal cavity in combination with peritoneal washing.[7,8] Total splenectomy is advocated by the majority of surgeons, especially for patients with large cysts located centrally or near the hilus, since it gives rise to minimal risk of recurrence.[7,10] Laparoscopic or laparoscopically assisted splenectomy has also been successfully used for echinococcal disease of the spleen.[12] Atmatzidis *et al.* showed that the recurrence rate after total splenectomy was not significantly different compared to spleen-preserving surgery, and the complication rate and hospital stay were also comparable between these two groups.[7]

CONCLUSION

In summary, the hydatid cyst is still a serious health problem in endemic areas as well as in our country. Although the liver and lung are the most frequently involved organs, primary splenic hydatidosis is quite rare. Cyst rupture into the peritoneal cavity represents a rare but serious complication. Hydatid cyst must be considered in the differential diagnosis of patients presenting with anaphylactic shock in areas where the disease is endemic and even in nonendemic. Early diagnosis with CT and appropriate treatments are essential for favorable outcomes.

Footnotes

Source of Support: Nil

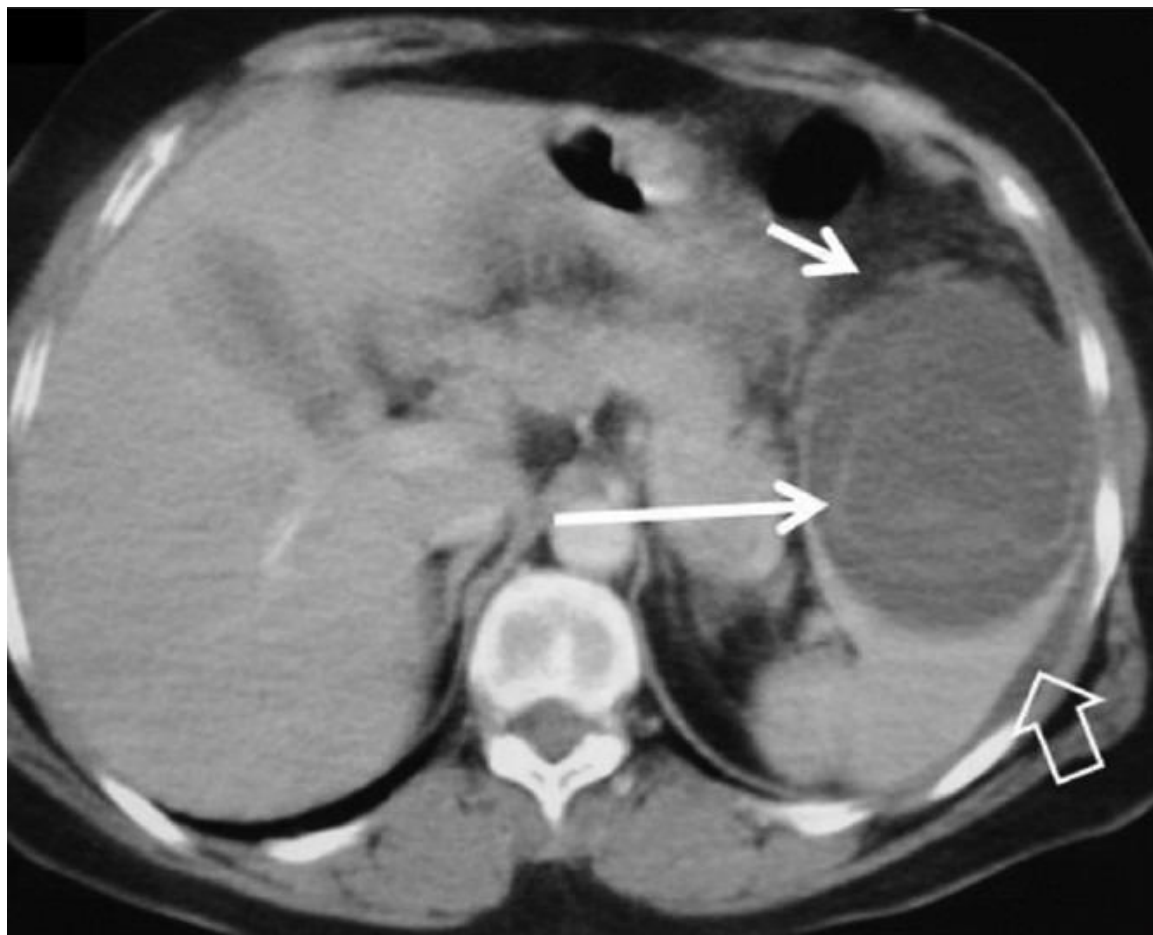
Conflict of Interest: None declared.

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Figures and Tables

Figure 1



Computed tomography showed floating membranes in the splenic cystic lesion (long arrow), rupture point (short arrow), and perisplenic fluid (open arrow)

Figure 2



Intraoperative photograph shows a ruptured outer membrane of an echinococcal splenic cyst

Figure 3



Follow-up computed tomography image shows no residue or recurrence in spleen with omentopexy (star)

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