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Case Report

Giant Isolated Splenic Hydatid Cyst: A Case Report

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Abstract
Hydatid cyst disease is a common disease in humans and animals. In livestock and agricultural areas, it is caused by the tapeworm Echinococcus. Canines serve as the definitive host, while livestock act as the intermediate host for this parasite. Humans can also be infected with the eggs of this parasite as accidental hosts, and in most cases, the liver and lungs are the primary sites of infection with this parasite and its cysts. In rare cases, other organs such as the spleen can also be infected with this cyst. Its rarity as well as the nonspecific clinical manifestations of splenic hydatid cysts lead to delays in its diagnosis and treatment. Ardabil province in northwestern Iran is one of the largest livestock-breeding areas in Iran, contributing to the spread of Echinococcosis in this region. This case report discusses a 47-year-old female farmer from Ardabil Province who, following 2 years of chronic pain in the left upper quadrant of her abdomen and splenomegaly, was diagnosed with a 13×10×10 cm isolated hydatid cyst of the spleen without involvement of other organs according to paraclinical examinations (e.g., ultrasound, CT scan, Indirect hemagglutination test). Finally, she underwent total splenectomy due to the large size of the cyst and the risk of rupture without any complications, and a histological examination confirmed the diagnosis.

Introduction

Hydatid cyst disease (HCD) or cystic Echinococcosis is a zoonotic parasitic disease caused by the larval stages of cestodes belonging to the genus Echinococcus and the family Taeniidae. Six species of the Echinococcus tapeworm are known; four are pathogenic to humans, and of these, Echinococcus granulosus is the most common cause of HCD. Hydatid cysts are the larval stage of the Echinococcus parasite, which forms in

various tissues after the oncospheres of an Echinococcus tapeworm grow there (1, 2).

The adult Echinococcus granulosus parasite, which is 3-6 mm long, resides in the small intestine of canines (dogs and wolves), which are its definitive host, and becomes infected by ingesting hydatid cysts containing viable protoscolices. The eggs are released in the canine's feces and spread throughout their living environment, where a suitable intermediate host ingests them. Cattle, sheep, pigs, horses, camels, and horses act as intermediate hosts for this parasite. In the small intestine of these intermediate hosts, the eggs hatch and an oncosphere develops, which passes through the intestinal wall and travels through the circulatory system to various organs, mainly the liver and lungs. The oncosphere develops into a cyst that slowly grows and produces daughter cysts. The cycle is complete when the intermediate host dies, and the canines ingest the cysts. Humans, as accidental intermediate hosts, become infected via the fecal-oral route by consuming organs contaminated with cysts of intermediate hosts, by direct contact with infected animal hosts, or by eating food or water contaminated with carnivore feces containing parasite eggs (2-4).

In this case report, we report a 47-year-old female farmer with an isolated primary hydatid cyst of the spleen, treated with total splenectomy.

Case Presentation

A 47-year-old woman from Ardabil Province, Iran, was referred to the surgical clinic for evaluation of chronic abdominal pain. On initial examination, she had been experiencing mild pain in the left upper quadrant (LUQ) for about 2 years, which she had ignored and had not been investigated, and worsened about 5 d earlier. She also felt a heaviness in this area. Pain was continuous, non-radiating, and aggravated by bending over. She lived in a rural area, engaged in cattle and sheep farming, and kept several dogs on her farm, with close and frequent contact. She had no history of surgeries or other significant diseases and was not taking any medication. She was hemodynamically stable and did not complain of any other particular problems. Her vital signs were within normal limits (temperature 36 °C, blood pressure 120/80 mm Hg, respiratory rate 16 cycles per min, heart rate 65 beats per min, and O2 saturation 98%). There was no weight loss, digestive or urinary symptoms, jaundice, fever, or respiratory symptoms. Splenomegaly was also palpated abdominally, dull on percussion without tenderness, resistance, or rebound. Routine laboratory studies, including complete blood count (CBC), liver function tests (LFTs), biochemistry analysis, electrolyte profile, and renal function tests, were normal.

Therefore, an abdominal ultrasound was performed for further evaluation. The ultrasound showed a spleen with a diameter of 129 mm and a well-defined, giant, solitary, non-septate, and hypoechoic cyst with a relatively thick (3mm) and regular wall without internal solids, measuring 105 × 97 mm throughout the length of the spleen, which to a large extent replaced the parenchyma of the spleen (Fig. 1). Subsequently, a thoracoabdominal CT scan was performed, which confirmed the ultrasound findings and demonstrated a large, well-defined, non-calcified cystic spleen lesion with uniform fluid content measuring 130×105×97 mm in anterior-posterior, craniocaudal, and transverse dimensions. The thin rim of splenic parenchyma covers the lesion, displacing the stomach to the opposite side (Fig. 2). There were also no signs of HCD involvement in other organs within the chest or abdomen, particularly the liver and lungs, nor were there any free fluids.

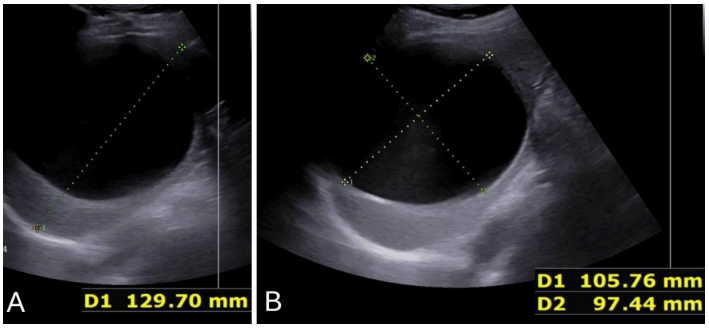

Fig. 1: An abdominal ultrasound demonstrates a solitary giant lesion with pure fluid with no internal septations or internal solids

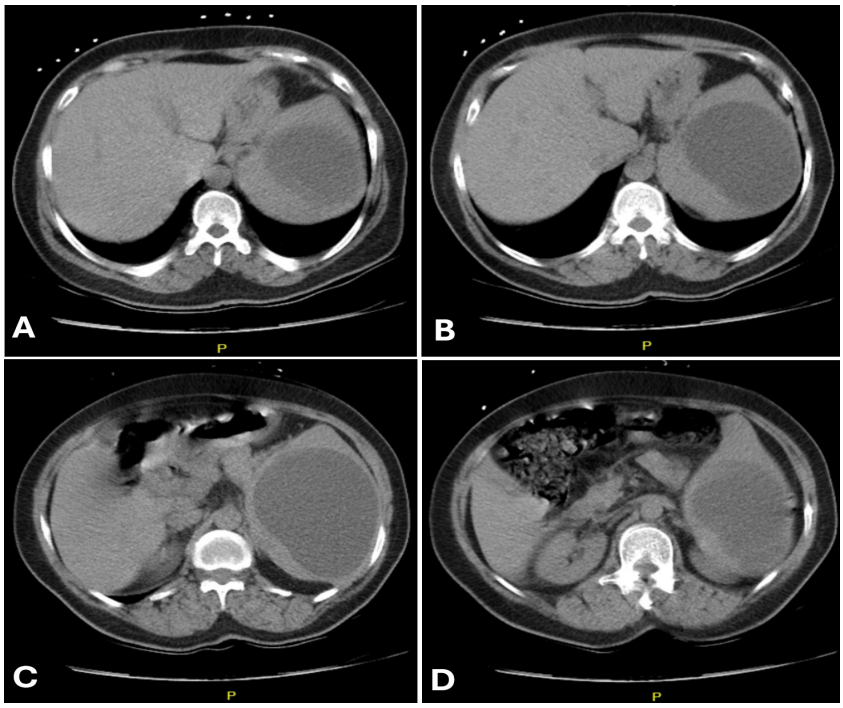

Fig. 2: Axial (transverse) CT scan of the abdominal cavity shows an isolated, large, well-defined, round, hypodense and uniform fluid-density cystic lesion throughout the length of the spleen, without calcifications, a solid tissue component, or septations, suggestive of a splenic hydatid cyst. The germinal layer is intact, and there is no sign of cyst rupture. There are no signs of other organ involvement, importantly, the liver and lungs

A hydatid cyst was strongly considered the primary differential diagnosis for this patient. Therefore, a positive Indirect hemagglutination (HAI) test for Echinococcus (titer > 160) confirmed the diagnosis of splenic hydatid cyst in this patient. The patient was considered for total splenectomy surgery. The standard approach in dealing with similar patients is that patients are treated with albendazole (400 mg twice daily) for 3 wk before surgery and then undergo surgery. However, given the size of the cyst and high risk of cyst rupture, and the high probability of the patient dropping out of follow-up and not returning for surgery, it was decided to forgo preoperative drug therapy and have the patient undergo total splenectomy as soon as possible.

After general anesthesia, Laparotomy was performed through a midline incision, which gave access to the abdominal cavity and showed inflammation and fibrosis around the spleen, leading to severe adhesion of the spleen to the intestine, stomach, diaphragm, and other adjacent structures. There was no evidence of cyst perforation or bleeding. Extensive lysis was performed to separate the spleen from surrounding structures and provide complete access to it. The spleen, most of which was a cyst extending from the upper to the lower pole, was dislocated and removed from the abdominal cavity without rupturing, measuring 13×10×10 cm. A unilocular cyst occupied the spleen, characterized by a yellowish to white internal surface and a firm wall surrounded by splenic tissue (Fig. 3). The surgery was performed without any major incidents, and the patient tolerated it well in a favorable condition with stable vital signs. Postoperatively, she did not experience any complications, and the inserted drain output was suitable.

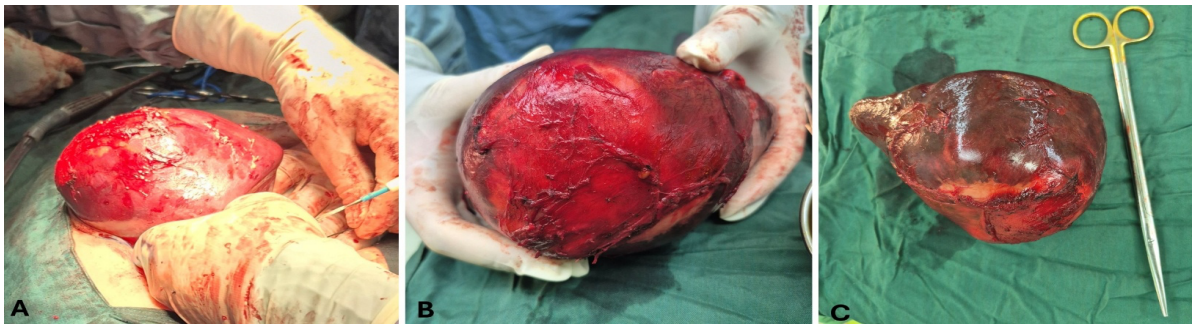

Fig. 3: Intraoperative (A) and postoperative (B&C) gross views of the spleen measuring 13×10×10 cm, without rupture

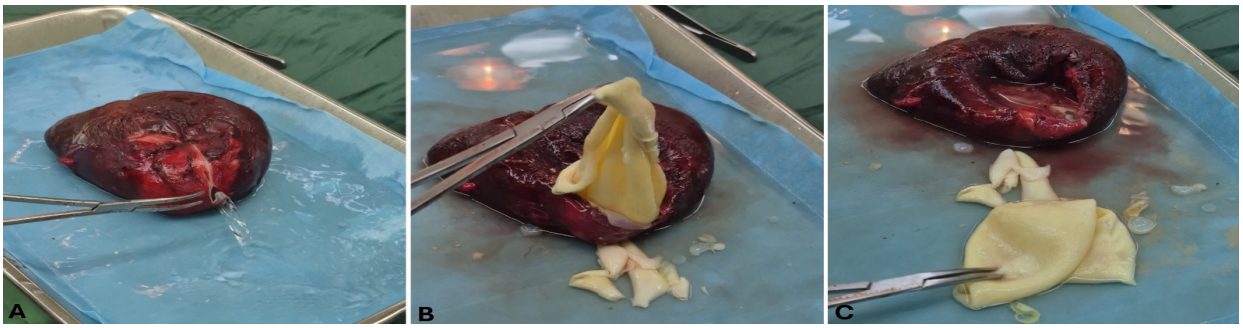

Fig. 4: Clear fluid ejects from the incision site with high pressure (A). Specimen displaying yellowish-white membranous tissue, which is the hydatid cyst's germinal layer (B&C)

After obtaining the patient's consent, the specimen was dissected in the operating room under sterile conditions, in accordance with hygiene standards. After a small transverse incision was made at the cyst site, approximately 300 ml of clear fluid was ejected from the incision site under high pressure (Fig. 4A). Moreover, the yellowish-white, folded, membranous tissue with a wall thickness of 3 mm, consistent with the germinal layer of a hydatid cyst, was pulled out from inside the cyst (Fig. 4 B&C). All specimens were subjected to histopathological analysis, which confirmed the diagnosis of splenic hydatid cyst (Fig. 5).

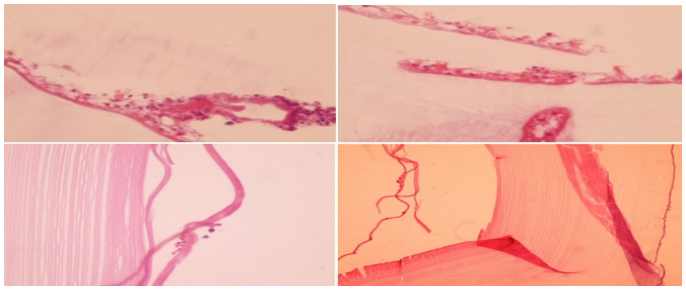

Fig. 5: Histopathological examination of a splenic hydatid cyst, identifying a laminated layer

The patient was hospitalized for a duration of seven days, not because of medical complications, but to address transportation issues and guarantee thorough follow-up, given the rural setting and restricted access to healthcare services. She was monitored with a normal

clinical examination and without inflammatory manifestations. As recommended, to prevent the risk of OPSI, patients receive vaccines against Streptococcus pneumoniae (pneumococcal), Neisseria meningitidis (meningococcal), and Haemophilus influenzae type b. She was discharged with a prescription of albendazole (2 × 400 mg/day) for three months with regular monitoring of liver function tests, which remained within the normal range, to avoid recurrence of HCD. At the three-week follow-up, she was clinically stable. She underwent clinical evaluation with periodic abdominal ultrasonography scheduled at 3 and 6 months after surgery, as well as chest CT scans in month 6. No signs of local recurrence, liver or lung involvement, or other notable complications were observed. At the patient's last visit, 18 months after surgery, she had no specific complaints, and a liver ultrasound and lung CT scan were normal and free of any cysts.

Ethical approval

The present study was approved by the Ethics Committee of Biomedical Research, Ardabil University of Medical Sciences, under ethical code IR.ARUMS.REC.1404.331. Written informed consent was obtained from the patient for publication of this case report. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Discussion

The global prevalence of HCD appears to be endemic in rural and cattle-rearing areas, with livestock farming and the presence of dogs (for guarding livestock or feeding on their carcasses) as the cause, but it is found worldwide. In endemic regions of HCD, such as the Mediterranean, Australia, New Zealand, North and East Africa, Eastern Europe and the Balkans, Central Asia, South America, and other regions where sheep-raising is prevalent, HCD remains a major public health concern (5, 6). Cystic echinococcosis is also a major human and animal health problem in the Middle East and Iran, considered an endemic area for Hydatidosis (7). In a systematic review and meta-analysis study, the overall seroprevalence of human CE in Iran's general population was 4% (8). Ardabil province in northwestern Iran is one of the largest livestock-breeding areas in Iran, contributing to the spread of Echinococcosis in this region. Heydari et al. reported that the seropositivity of human Hydatidosis by ELISA in Ardabil Province and its suburban area was 4.4%, a rate roughly similar to that of other parts in Iran (7).

HCD can involve various organs, but in 60%-70% of cases it infects the liver, and in 30% of cases the lungs. Rarely, involvement of the kidneys, spleen, muscles, heart, pancreas, eye, ovary, salivary glands, peritoneal and pelvic cavities, breast, bone, thyroid, and central nervous system (CNS) has also been reported, considered rare and unusual locations (9). Although the liver is the most common site of hydatid cysts, splenic hydatid cysts are rare, with a global prevalence of 0.5%-8% of all hydatid cyst cases. In other words, although HCD is common in endemic areas, splenic HCD is rare even there. The splenic hydatid cyst can be primary (or isolated), in which only the spleen is infected, or secondary, in which the spleen and other organs, such as the liver and lungs, are infected. Primary splenic hydatid cyst accounts for less than 2% of all cases, often presenting diagnostic challenges due to its rarity (5, 6, 10).

A splenic hydatid cyst is mostly asymptomatic and may be detected incidentally on imaging or present with nonspecific complaints. Its symptoms may include dull pain in the left hypochondrium, a palpable abdominal mass, gastrointestinal symptoms, etc. If left untreated, these cysts can lead to complications such as rupture, infection, fistula formation, cyst calcification, and, most importantly, anaphylactic shock. The fluid inside the cyst is highly allergenic, and any traumatic or spontaneous rupture of the cyst can lead to the release of

these allergens, leading to a potentially life-threatening anaphylactic shock (11-13).

In cases of splenic hydatid cyst, the parasite typically reaches the spleen through an arterial route after passing through the liver and pulmonary filters. It may also occur in patients with portal hypertension due to retrograde spread from the liver to the spleen via the hepatic portal and splenic veins. The spleen may become infected with a hydatid cyst due to rupture of a hydatid cyst from another abdominal organ into the peritoneal cavity. There are other theories about the causes of the spread of infection to the spleen. Such passage from the intestine to the inferior vena cava without passing through the liver; passage through the chyliferous ducts to the superior vena cava and systemic circulation after penetrating the intestinal wall; direct entry into the spleen from the stomach and colon; and lymphatic dissemination. However, the most logical route seems to be the bloodstream, through which a percentage of the parasites escape liver and lung filtration and settle in the spleen (10).

The evaluation and diagnosis of splenic hydatid cysts rely on imaging techniques such as ultrasound and CT. The ultrasound often shows well-defined cystic lesions with daughter cysts. A CT scan can also show calcification of the cyst wall and its relationship to adjacent structures. However, these imaging modalities lack specificity for noncalcified cysts and cysts without daughter cysts, making it difficult to distinguish a splenic hydatid cyst from other splenic lesions. Serological tests, especially ELISA, which detect antibodies against Echinococcus antigens, can also be used as a complementary diagnostic method. However, these tests are often inconclusive, and in many cases, splenic hydatid cyst is only confirmed during surgery or histopathological examination (5).

There are various treatment approaches for splenic hydatid cysts, such as total splenectomy, spleen-preserving techniques, PAIR (puncture, aspiration, injection, reaspiration), and Anthelmintic therapy. Total splenectomy without cyst perforation is considered the standard treatment for splenic hydatid cysts because it has a lower mortality and morbidity rate and reduces complications from cyst rupture. Total splenectomy should be used in cases of adhesion or invasion of the cyst to adjacent structures, a large cyst that occupies more than 3/4 of the splenic parenchyma and has destroyed its parenchyma, multiple and symptomatic cysts, and cysts located in the splenic hilum. The advantages of this procedure include preventing the spread of parasites to the surgical site, reducing the risk of anaphylactic shock, and preventing recurrence and complications related to the residual cavity. However, this surgical procedure has risks such as bleeding, more difficult surgery due to adhesions to adjacent organs and structures, damage to the stomach and pancreas, the occurrence of thromboembolic events, subphrenic abscess, chest infection, and OPSI, especially in children under 4 yr of age (11,13,14).

Spleen-preserving approaches such as partial splenectomy (removal of the cyst along with part of the spleen), cyst enucleation (without removal of part of the spleen), deroofing of the cyst with omentoplasty, and external drainage are also used to treat splenic hydatid cysts. This approach is suitable for the treatment of polar cysts with an accessible protruding dome, the high risk of total splenectomy due to adhesion to surrounding structures, and pediatric patients with small, isolated peripheral cysts. The advantages of this technique include the preservation of splenic function and a reduction in complications associated with total splenectomy. However, this method has complications such as the risk of poor control of splenic bleeding during surgery, residual cavity infection, cyst recurrence, intra-abdominal abscess, etc. (11, 13-15).

A newer, less invasive approach is the PAIR technique. This technique is effective for single non-septated thin-walled cysts, cysts smaller than 5 cm, and patients whose general

condition is unsuitable for surgery and anesthesia. This technique allows for the avoidance of total splenectomy and reduces the length of hospital stay and complications. However, this method is less successful in calcified cysts, multiseptated cysts, cysts containing multiple daughter cysts, and cysts containing a predominantly solid component (5, 6, 11, 13, 14).

Although surgical intervention remains the mainstay of treatment for splenic hydatid cysts, Anthelmintic therapy is recommended as a palliative measure in patients who are not candidates for surgery, for asymptomatic cysts smaller than 5 cm, or in conjunction with surgery to prevent cyst spread and recurrence. Albendazole administration before surgery sterilizes the cyst by destroying the germinal layer. It also reduces intracystic pressure and subsequently reduces tension in the cyst wall, which reduces the risk of intraoperative rupture and anaphylaxis. Preoperative and postoperative administration of this drug is recommended to reduce the risk of postoperative recurrence (5, 6, 13).

The choice of treatment method should be based on the size and location of the cyst, its relationship to other surrounding structures, the patient's comorbidities, and the patient's general condition, etc. In our patient, due to the high risk of spontaneous or traumatic cyst rupture because of its large size, as well as the occupation of a large volume of the spleen and the fragility of the spleen, total splenectomy with an open approach was chosen as the treatment method.

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Conflict of interest

The authors declare that there is no conflict of interests.

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