

## An Uncommon Primary Splenic Hydatid Cyst in Human: A Case Report Study

Haitham Elaadli<sup>1</sup>, Hany El Adly<sup>2</sup>, Raafat M. Shaapan<sup>3\*</sup> and Mohamed Bessat<sup>4</sup>

<sup>1</sup>Department of Animal Hygiene and Zoonoses, <sup>4</sup>Department of Parasitology, Faculty of Veterinary Medicine, Alexandria University.

<sup>2</sup>Department of Internal Medicine, Damanhur Medical National Institute (DMNI),

<sup>3\*</sup>Department of Zoonotic Diseases, National Research Center, Giza, Egypt.

\*Corresponding Author: Raafat M. Shaapan, Mobile: (+20) 01005280571, E-mail: rmshaapan2005@yahoo.com ,

ORCID: <https://orcid.org/0000-0002-2620-4189>

### ABSTRACT

**Background:** Cystic Echinococcosis (CE) is one of the most prevalent important neglected zoonotic diseases caused by the larval form of the tapeworm, *Echinococcus granulosus*, and it represents a significant public health threat in many Mediterranean countries like Egypt. Human acts as the accidental intermediate host where the larval stage, hydatid cyst is formed. While liver and lung constitute the most commonly involved sites for primary cysts, other organs are occasionally affected. Primary hydatid cysts of the spleen are rarely present.

**Patients and methods:** This study reported an uncommon case of multiple primary hydatid cysts of the spleen in a 55-year-old man presented with pain in the left hypochondrium. A history of close contact with dogs was reported. The initial diagnosis was based on the abdominal ultrasonography and confirmed by the computed tomography (CT) scan and the serological detection using indirect hemagglutination test (IHAT) in the clinic.

**Results:** The ultrasonography and CT scan revealed subcapsular hydatid cyst in the spleen with peripheral small daughter cysts. Serologically, hydatid cyst positive patient serum was recorded at 1:1280 titer by IHAT. Total splenectomy was successfully done, and the patient was discharged and prescribed with a prophylactic albendazole.

**Conclusions:** ultrasonography, computerized tomography and serology are useful diagnostic tools for hydatidosis and the disease is considered a public health challenge and needs accurate differential diagnosis from any cystic mass/s in the abdomen.

**Keywords:** Hydatid cyst, Echinococcus, Spleen, Ultrasonography, CT scan.

### INTRODUCTION

Hydatidosis (Cystic Echinococcosis) is a zoonotic parasitic disease caused by the larval form of the cestode *Echinococcus granulosus* <sup>(1)</sup>.

The adult worm occurs in the small intestine of dogs, who serve as its primary host. Cattle, sheep, and camels are the most frequent intermediate hosts, where the larval stages (metacestodes), that can develop into advanced hydatid cysts, can be found <sup>(2)</sup>. In the life cycle, humans serve as an incidental intermediate host. The fecal-oral pathway or eating food contaminated with dog feces that contain Echinococcus eggs cause infection <sup>(3)</sup>. In areas where the disease is prevalent, such as the Middle East, North Africa, India, Turkey, South America, New Zealand, and Australia, in terms of public health, cystic echinococcosis is still a major concern <sup>(4)</sup>.

Hydatid cysts can regularly multiply in any organ, however they typically affect the liver (60–70%) and lungs (30%) and are infrequently detected in the kidney, spleen, brain, heart, pancreas, breast, bone, and thyroid. Even in areas where the disease is endemic, spleen cysts account for less than 3% of cases <sup>(5)</sup>. Rarely, after the parasite has passed the hepatic and pulmonary filters, does a primary infection take place, and it typically does so via the arterial pathway <sup>(6)</sup>. Due to the release of the extremely allergic hydatid fluid, any spontaneous or violent rupture could result in a life-threatening anaphylactic shock <sup>(7)</sup>.

Investigation of hydatidosis/echinococcosis currently relies on a variety of imaging technologies,

including computerised tomography, ultrasonography, and X-rays <sup>(8)</sup>. Evaluation and antigen purification are required to improve serological techniques' sensitivity to identify and confirm disease in its early stages because they currently lack diagnostic specificity, especially in endemic areas <sup>(9)</sup>.

For the accurate diagnosis of hydatidosis in humans by immunoblotting, hydatid fluid and protoscoleces crude antigens of camel and sheep hydatid cysts may both be helpful <sup>(10)</sup>. When coupled with a confirmatory ELISA test, the latex agglutination test (LAT) is a suitable and useful diagnostic tool for the diagnosis of cystic echinococcosis <sup>(11)</sup>. In effectively diagnosing hydatidosis in both camels' and humans' sera with ELISA, the diagnostic antigens, made from camel hydatid cysts, may be able to control the infection and lessen human transmission <sup>(12)</sup>.

### PATIENT AND METHODS

A 55-years-old male patient who is a farmer from a rural area located in the Western part of Behera province, Egypt, was admitted to a private clinic of the internal medicine and nephrology with left hypochondrium dull aching pain and recurrent fever. He had no history of icterus, respiratory problems, abdominal complaints, weight loss, and medical history record only indicated a Bilharzial Hepatic Fibrosis (BHF). On examination, his vital parameters were within the normal limits. The physical study revealed an asymmetric abdomen and splenomegaly with smooth surface in the left upper abdominal quadrant. Routine

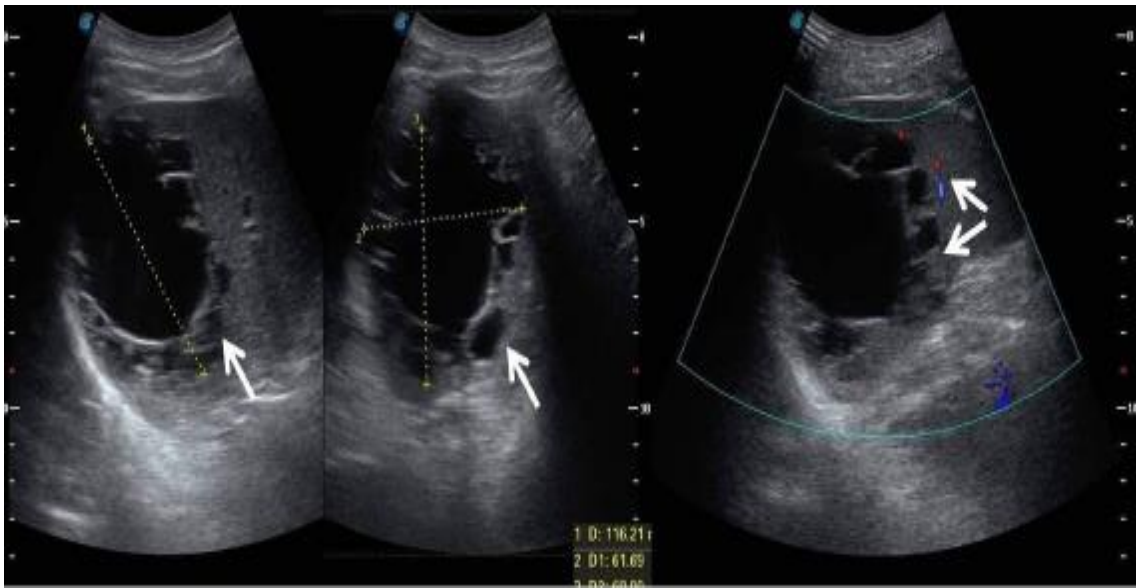
laboratory investigations such as the complete blood picture, biochemistry analysis, electrolyte profile, and the renal function tests showed no abnormalities.

**Ethical consent:**

As a routine procedure, a written consent from the patient was obtained throughout the diagnosis and treatment process. An additional consent related to the disclosure of data for the scientific and research benefits was also approved by Medical National Institute Ethics Committee and the study's protocol complied with the Helsinki Declaration, which is the World Medical Association's code of ethics for research on humans.

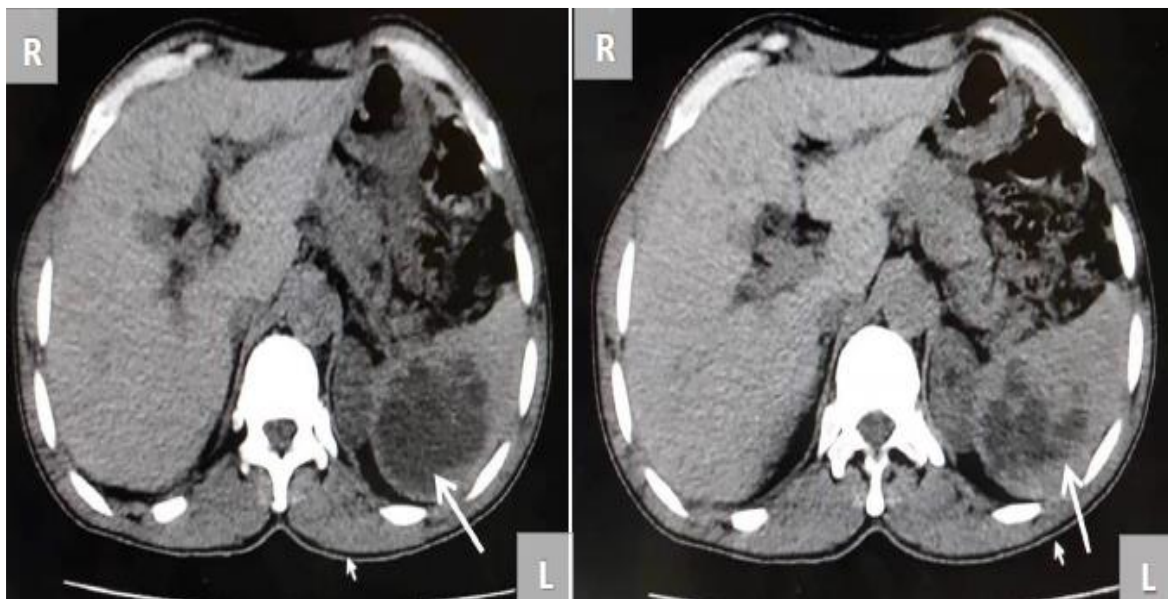
**RESULTS**

The abdominal ultrasonography showed a 62x68x77mm upper polar subcapsular cystic lesion in the spleen with innumerable peripheral small interconnecting splenic cysts (daughter cysts). There was no solid internal vascularity component in the splenic tissue, with no solid focal lesions or visible hilar collaterals (Figure 1). The Damanhour Medical National Institute (DMNI) was then recommended for the patient. After special consultations on ultrasonography data with medical specialists from the Internal and the Tropical Medicine Departments, a non-tumor growth idealistically of parasitic origin was suspected.



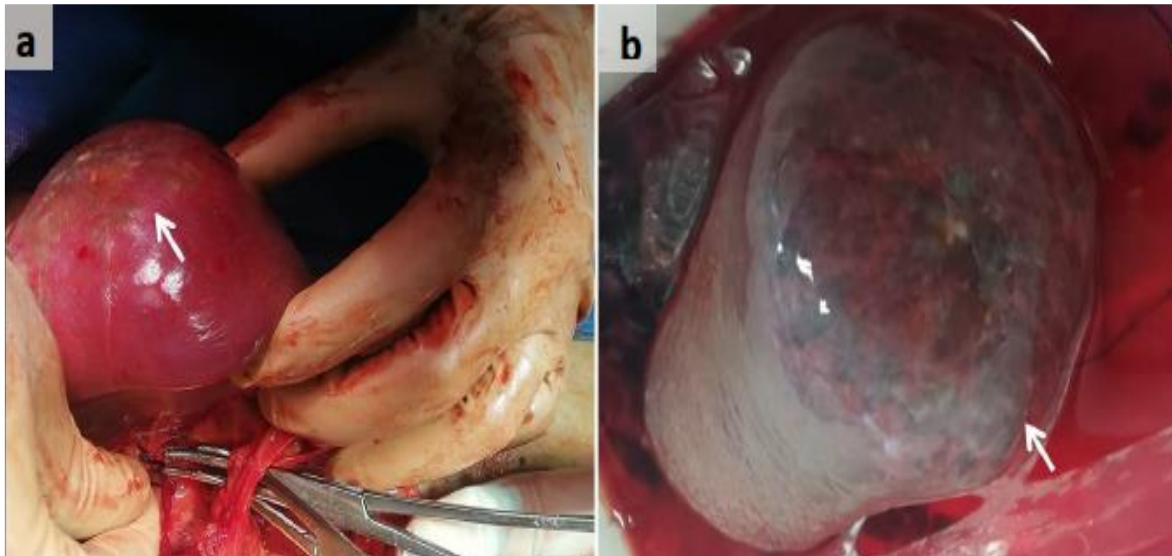
**Figure (1):** An abdominal ultrasonography demonstrating a multiloculated splenic hydatid cyst (arrow)

A computed tomography (CT) scan of the pelvis and abdomen was performed, revealing a mildly bulky spleen with upper polar hypodense lesion averaging 60x75x47mm being of relative fluid density and showing fine internal septae and few tiny spots of calcification and the other abdominal viscera were normal (Figure 2).



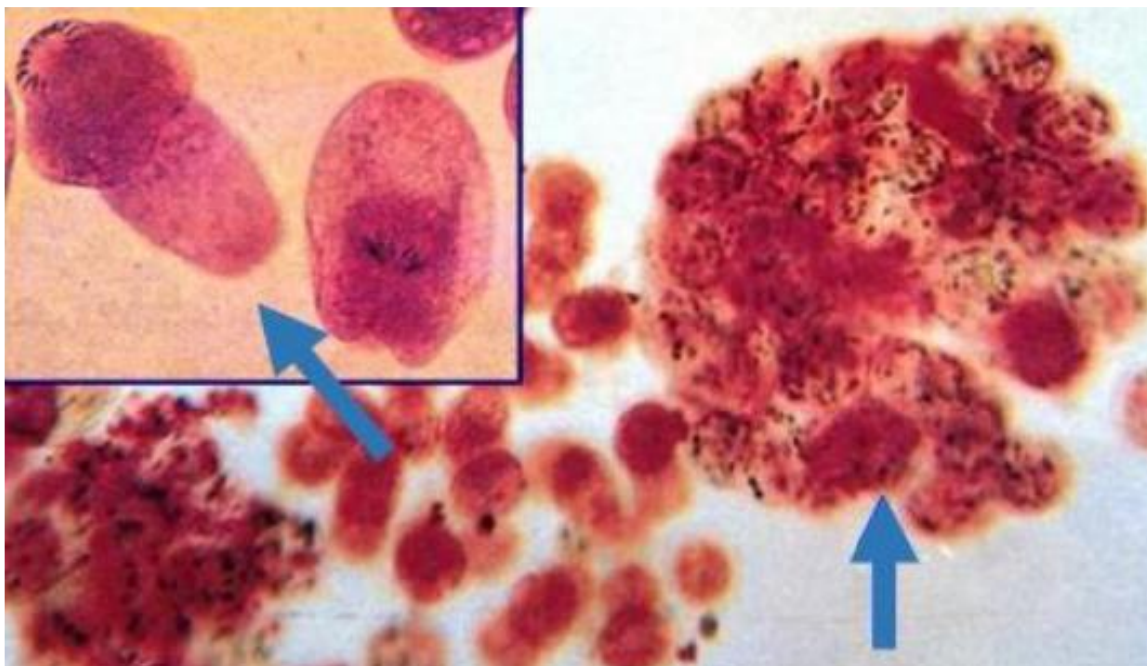
**Figure (2):** CT abdomen showing a well-defined hydatid cyst with many daughter cysts (arrow). (R, right; L, left)

Serological examination based on the indirect hemagglutination test (IHA) using the commercial IHA kit (Fumouze Laboratories, France) specific for human cystic Echinococcosis detected a positive titer of 1:1280. Together, the imaging and serology data further confirmed the question's lesion as a splenic hydatid cyst that required a splenectomy. Laparotomy was done through a left paramedian incision, and total splenectomy was successfully performed in which the spleen was extirpated with intact cysts (**Figure 3**).



**Figure (3):** Intraoperative (a) and postoperative (b) views showing a well-demarcated cystic mass of hydatid cyst in the spleen (**arrow**)

The histopathological analysis has confirmed the hydatid cyst, with germinal layer, scolices and hooklets. Microscopic examination of a stained smear prepared from the cyst content detected the hydatid sand with broad capsules and scolices (**Figure 4**). This was further confirmed by the postoperative oral communications with the patient, who admitted his close and frequent contacts with at least two domestic shepherd dogs in his farm premises.



**Figure (4):** Microscopic detection of a stained smear showing content of hydatid cyst. Hydatid sand (**arrow**); broad capsules and scolices (**inset**)

A postoperative course of albendazole and antibiotic treatment was initiated after the surgical operation, and the patient was discharged a week later. Four weeks had passed since the operation, with no notable complications were registered.

## DISCUSSION

The cystic echinococcosis parasitic disease, which is prevalent all over the world, poses a serious threat to both public health and the economy. Recently, this condition was included in the WHO's strategic plan to combat neglected tropical diseases<sup>(13)</sup>. Splenic hydatid cyst is a rare clinical condition as its total incidence is recorded to be 0.5- 4% of hydatid cysts in the abdomen, even in the endemic areas<sup>(14)</sup>. In this case study, the patient suffered from left hypochondrium dull aching pain with an asymmetric abdomen, recurrent fever and splenomegaly with smooth surface in the upper left abdominal area. Previous research revealed that the primary clinical complaint was abdominal discomfort<sup>(5,15,16)</sup>. Accordingly, because splenic hydatid disease is slowly progressing and frequently misdiagnosed, patients with it rarely presented a clinical picture. The most frequent clinical signs are upper abdominal pain and a palpable mass<sup>(17)</sup>.

In this study, the serological examination based on the commercial indirect hemagglutination test (IHA) specific for human cystic Echinococcosis indicated a positive patient serum titer of 1:1280. Because the clinical picture was vague and the radiological characteristics were non-pathognomonic, it was challenging to diagnose a case of a splenic hydatid cyst. Sensitive, Casoni's intradermal allergy test lacks specificity<sup>(18)</sup>. ELISA is the preferred serological test for the diagnosis of CE in domestic humans and animals. It is typically inexpensive, quick, has good sensitivity and specificity, is non-sophisticated and requires fewer skilled workers<sup>(19)</sup>. Incomplete satisfaction exists with serological diagnostic procedures using crude hydatid cyst antigen. Consequently, antigen purification is essential to exclude additional cross-reactive proteins<sup>(20)</sup>. Ultrasonography and CT scanning are the most fundamental investigation tools for the diagnosis of splenic cystic echinococcosis and CT scan has a higher sensitivity (95-100%) than ultrasonography and more helpful in detecting the cyst's number, size, and location<sup>(21)</sup>. In current case, the imaging data as, abdominal ultrasonography and computed tomography (CT) were used to confirm that the lesion of our patient was a splenic hydatid cyst and also, could determine the cystic mass, hydatid membrane, small daughter cysts and hydatid sands.

The surgical excision of the cyst during the full splenectomy, which eliminates all the parasitic and pericystic tissues, constitutes the gold standard method due to the limited efficiency of anti-parasitic drugs and the risk of traumatic or spontaneous rupture<sup>(22)</sup>. Thus, in our case study, total splenectomy was successfully done, and a postoperative prophylactic albendazole treatment was prescribed. Without therapy, secondary inflammatory reaction, irritable abdomen, rupture inside the patient's abdomen, anaphylaxis, intestinal fistula and even serious intestinal hemorrhage development could exacerbate splenic cystic

echinococcosis<sup>(23)</sup>. It is advised that the patient get preoperative preventive medication with albendazole and praziquantel for 30 days and 14 days, respectively. This lowers intra-abdominal pressure, lowers the risk of recurrence, and also aids with hydatid cyst stability<sup>(24)</sup>. If the hydatid cyst is large or has adhesions to adjacent organs, total splenectomy is preferred, and this is what was done in our patient case. Additionally, technically, whole splenectomy is easier than partial splenectomy. However, to maintain the spleen's immunological function, surgeons conduct partial splenectomy, or cyst de-roofing<sup>(25)</sup>. The different splenic cystic masses, such as hematomas, abscesses, dermoid cysts, pseudocysts, hemangiomas, and lymphangiomas, must be considered in the differential diagnosis of splenic hydatid disease<sup>(26)</sup>.

## CONCLUSION

Despite the rarity of the splenic hydatid cyst, the physician should suspect and consider it in the differential diagnosis of the spleen's pathological cystic lesions, particularly in the geographical areas where cystic Echinococcosis is endemic. Abdominopelvic ultrasonography and computerized tomography and serological detection are the most sensitive and useful diagnostic tools. In adults, total splenectomy remains the treatment of choice, but the spleen preserving surgical exploration should be considered in childhood to avoid any postoperative complications.

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**Author contribution:** Authors contributed equally in the study.

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