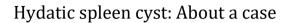


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(CASE REPORT)



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# Abstract

**Introduction**: Isolated cystic echinococcosis of the spleen (Splenic hydatid cyst) is extremely rare, ranking third after liver and lung involvement. It represents approximately 5% of all hydatid cyst localizations.

**Aim**: To report a new case of splenic hydatid cyst and conduct a literature review on its pathophysiology, diagnostic circumstances, and therapeutic options.

**Observation**: A 21-year-old male, professionally a student, residing in an urban area with no medical history, presented with left hypochondrial pain of a dragging nature evolving over several days. Abdominal examination revealed a visible and palpable bulge in the left hypochondrium.

Ultrasonography, abdominopelvic CT scan, and hydatid serology confirmed the diagnosis of splenic hydatid cyst. Total splenectomy via laparotomy was performed in our patient. The postoperative course was uneventful.

**Conclusion**: Isolated splenic hydatid cyst is rare. Several routes of contamination are possible for the spleen. Diagnosis is often incidental. Treatment remains primarily surgical.

Keywords: Cystic echinococcosis; Echinococcus granulosus; Hydatic cyst; Splenic cyst

# 1. Introduction

Hydatid cyst is the development of the larval form of *Echinococcus granulosus* in humans [1]. This disease is currently designated as "Cystic Echinococcosis" to comply with the recommendations of the International Society of Parasitology [1].

Isolated cystic echinococcosis of the spleen is extremely rare [2], ranking third after the liver and the lung [3]. It represents approximately 5% of all hydatid cyst localizations [4]. Isolated spleen involvement can occur via blood or lymphatic routes [2].

The majority of patients are asymptomatic [5], often incidentally discovered as incidental findings [7]. Thus, preoperative diagnosis can be challenging as there is no specific symptomatology [8]. Treatment consists of partial or total splenectomy [6], and in some cases, some authors propose cyst puncture aspiration, combined with albendazole [5].

Our aim is to report a new case of Cystic Echinococcosis (hydatid cyst) of the spleen and to review the literature to discuss the pathophysiology of this localization, its clinical manifestations, and its therapeutic modalities.

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# 2. Observation

A 21-year-old young man, a student by profession, living in an urban area, with no medical history, consulted for left hypochondrium pain of a heavy type evolving for a few days.

Clinical examination revealed a patient in good general condition, afebrile, with a body mass index (BMI) of 21.

Abdominal examination revealed a visible bulge in the left hypochondrium and left flank upon inspection. On palpation, the abdomen was soft, with the presence of a firm and painless mass in the left hypochondrium, mobile with respiratory movements, and with the lower edge dull located at the level of the umbilicus, all suggestive of type III splenomegaly. The rest of the clinical examination was unremarkable.

Abdominal ultrasound and CT scan favored an enlarged spleen with a septated cystic formation measuring 07 cm x 06 cm, suggestive of a stage III hydatid cyst according to the Gharbi classification and CE2 according to the World Health Organization (WHO) classification.

Serology using the ELISA technique was positive (4.01 UI/L).

We operated on the patient via a left subcostal approach, and intraoperative exploration revealed a spleen with a partitioned cystic formation measuring approximately 08 x 07 cm, occupying the hilum of the spleen. After protecting the peritoneal cavity with hydrogen peroxide-soaked drapes, we performed a total splenectomy.

Postoperative recovery was uneventful, and the patient was discharged on the 3rd postoperative day.



Figure 1 Perioperative photo of the hydatid cyst of the spleen



Figure 2 Photograph of the operative specimen (Total splenectomy) seen from the hilar aspect



Figure 3 Photograph of the operative specimen (Total splenectomy) viewed from the external lateral aspect."

# 3. Discussion

Cystic echinococcosis (Hydatid cyst) most commonly affects the liver (60 to 70%), lungs (30%), and rarely other organs such as the kidneys, spleen, bones, thyroid, breast, and pancreas [7, 8]. Splenic involvement is rare, representing 3% to 5% of all hydatid cyst locations [4, 8]. It is often associated with hepatic and pulmonary involvement; isolated splenic

involvement is extremely rare [7, 9-11]. The first case of splenic hydatid cyst was described post-mortem by Berlot in 1971 [8]. Cystic echinococcosis primarily affects adult subjects, with a peak incidence between 30 to 40 years old and a slight female predominance [12].

On a pathophysiological level, Echinococcus granulosis larvae are typically trapped by hepatic and pulmonary filters, with only 15% of larvae possibly evading these filters and entering the systemic circulation [8]. Thus, isolated involvement of another organ without associated hepatic and/or pulmonary involvement would be rare.

Several routes of contamination have been proposed to explain isolated involvement of other viscera. Firstly, the hematogenous route; after bypassing the hepatic and pulmonary filters, *Echinococcus granulosus* larvae are carried via the systemic circulation, primarily to highly vascularized organs such as the spleen and muscles [8, 13]. The second route of contamination is the lymphatic route or gastrointestinal tract shunt [13, 14]. For our patient, both of these contamination routes are plausible and could explain this primary and isolated localization in the spleen. Other routes of contamination, such as contiguous spread, have been reported [13, 15]. However, this latter route of contamination is not applicable to our patient.

The development of splenic hydatid cyst is pauci-symptomatic and slow, with a clinical latency phase ranging from 2 to 20 years [12]. Incidental discovery is the most common mode of presentation [2, 6, 8]. The most frequent reasons for consultation are pain followed by the detection of a left hypochondrial mass (or splenomegaly) [6, 12]. Dyspepsia, constipation due to colon compression, and dyspnea due to diaphragmatic compression may also prompt consultation [2]. Cases of hematemesis have also been reported in the literature [2]. Finally, revelation may occur through complications such as arterial compression and systemic hypertension, abscess formation, rupture into the peritoneal cavity or neighboring organs (rupture into the pleura, stomach, colon, or skin) with anaphylaxis [3, 6, 7]. Physical examination typically reveals a palpable mass in the left hypochondrium [8]. In our patient, discovery was made following right hypochondrial pain with splenomegaly.

Currently, ultrasound and computed tomography are the most useful imaging techniques for diagnosing and evaluating focal splenic diseases [2, 3]. Findings range from purely cystic lesions to completely solid appearances and are classified based on appearance. One may observe a simple cyst without internal architecture, a cyst with daughter vesicles and a matrix, a calcified cyst, or a complicated cyst [2]. Combined with hydatid serology, these imaging examinations allow for the diagnostic confirmation of splenic hydatid cyst [3]. Various serological tests exist such as hydatid immunophoresis, ELISA, and latex tests; indirect hemagglutination tests are useful for diagnosis, screening, and postoperative recurrence monitoring [8]. Indirect hemagglutination test and ELISA have a sensitivity rate of 85 to 90%. This was the case with our patient where abdominal-pelvic ultrasound and CT scan, as well as hydatid serology (ELISA technique), were concordant in favor of an isolated splenic hydatid cyst.

The differential diagnosis of a cystic splenic mass includes epidermoid cysts, pseudocysts, splenic abscesses, hematomas, and cystic splenic neoplasms [5, 6].

Treatment of splenic hydatid cysts is primarily surgical [3]. Both laparotomy and laparoscopy are viable options. The laparoscopic approach is easy to apply, safe, and effective for splenic hydatid cyst surgery [2]. It is intended for patients with single, small-sized cysts located superficially [2]."

Robotic surgery is feasible; however, to date, there have been only a few reports on robotic partial or total splenectomy [2]. In our patient, we opted for laparotomy due to the large volume of the cyst, and also because our laparoscopic column lacks a crusher to crush the spleen before extraction.

Total splenectomy, partial splenectomy, cyst enucleation, and resection of the protruding dome, with omentoplasty, are the preferred surgical techniques for treating splenic hydatid disease [8]. Currently, there is no consensus regarding the choice of a specific surgical technique [7]. Total splenectomy should be favored in large splenic hydatid cysts because typically, the remaining splenic parenchyma is reduced and laminated [2]. Additionally, it helps to avoid the risk of peritoneal cavity contamination caused by cyst opening, thus minimizing the risk of recurrence [3]. Advocates of conservative surgery argue that total splenectomy predisposes to septicemia and should be avoided, especially in children [2]. Conservative techniques are employed for superficial cysts confined to one pole of the spleen and cysts with extensive adhesions. Small asymptomatic cysts may be treated with anthelminthic drugs. However, splenic cysts require close monitoring [8].

The puncture, aspiration, injection, and reaspiration (PAIR) technique using hypertonic saline solution or 0.5% silver nitrate solutions before opening the cavities tend to sterilize daughter vesicles [7]. Medical treatment constitutes the

cornerstone of postoperative follow-up [7], and it is prescribed for multi-visceral forms even though the results are inadequate [3]. In our patient, we opted for total splenectomy due to the near-hilar location and significant volume of the cyst, as well as to avoid any risk of parasite dissemination into the peritoneal cavity upon cyst opening.

For our patient, we initiated medical treatment one month before surgery.

Prevention relies on public health education, veterinary control of livestock slaughter, culling of stray dogs along with census and deworming of domestic dogs [12].

### 4. Conclusion

Isolated splenic hydatid cyst is rare. The hematogenous route, having bypassed the hepatic and pulmonary filters, the lymphatic route or gastrointestinal tract shunt, contamination by contiguity are possible contamination routes of the spleen. Diagnosis is often incidental as an incidentaloma. Treatment remains primarily surgical. With the advancement of surgical techniques, the current trend is conservative treatment through minimally invasive approaches.

### **Compliance with ethical standards**

### Disclosure of conflict of interest

No conflict of interest to be disclosed.

### Statement of informed consent

Informed consent was obtained from all individual participants included in the study.

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