

Case report

BEYOND THE LIVER AND LUNGS: AN UNCOMMON CASE OF ISOLATED SPLENIC ECHINOCOCCOSIS

НАДВОР ОД ЦРНИОТ ДРОБ И БЕЛИТЕ ДРОБОВИ: РЕДОК СЛУЧАЈ НА ИЗОЛИРАНА ЕХИНОКОКОЗА НА СЛЕЗИНАТА

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Abstract

Hydatid disease is an infection caused by the parasite *Echinococcus granulosus*, present almost worldwide, but mostly in countries where sheep are cultivated. It can occur in any organ, but most often in the liver and the lungs, while in the spleen it is very rare from 0.5 to 4% of all abdominal cases.

The diagnosis of echinococcosis of the spleen can be challenging due to its rare occurrence and nonspecific presentation.

A 62-year-old patient presented to us with persistent and nonspecific abdominal pain localized in the epigastric region and left upper quadrant, accompanied by obstipation and weight loss for more than a year.

Serological testing for echinococcus was positive. Abdominal ultrasound and computed tomography showed a cystic lesion of the spleen without involvement of other abdominal organs.

The patient underwent a surgical intervention where the spleen totally was removed. Histopathological results confirmed an hydatid cyst of the spleen.

This rare case emphasizes the importance of considering hydatid cysts of the spleen as one of the possible diagnoses in patients presenting with abdominal pain for a longer period of time accompanied by a palpable abdominal mass, especially in endemic places. It also expresses the need for rapid and adequate treatment to minimize the complications while achieving the best possible outcomes for the patient.

Keywords: Echinococcus, spleen, surgery, Hydatid Cyst, case report

Апстракт

Хидатидната болест е инфекција предизвикана од паразитот *Echinococcus granulosus*, присутна речиси насекаде во светот, но почеста во земјите каде што се одгледуваат овци. Може да се појави во кој било орган, но најчесто ги зафаќа црниот дроб и белите дробови, додека зафатеноста на слезината е многу ретка и изнесува од 0,5 до 4% од сите абдоминални случаи.

Дијагнозата на ехинококозата на слезината може да биде предизвикувачка поради нејзината ретка појава и неспецифичната клиничка презентација.

Кај нас беше примена 62-годишна пациентка со перзистентна и неспецифична абдоминална болка локализирана во епигастричната регија и левиот горен квадрант, проследена со опстипација и губење на телесна тежина во период подолг од една година.

Серолошкото тестирање за ехинокок беше позитивно. Абдоминалниот ултразвук и компјутеризираната томографија покажаа цистична лезија на слезината без зафатеност на други абдоминални органи.

Кај пациентката беше изведена хируршка интервенција со тотална спленектомија. Хистопатолошките резултати потврдија хидатидна циста на слезината.

Овој редок случај ја нагласува важноста од вклучување на хидатидната циста на слезината во диференцијалната дијагноза кај пациенти со долготрајна абдоминална болка, особено кога е присутна палпабилна абдоминална маса и во ендемски подрачја. Исто така, ја потенцира потребата од навремен и соодветен третман со цел минимизирање на компликациите и постигнување на најдобар можен исход за пациентот.

Клучни зборови: Ехинококус, слезина, хирургија, хидатидна циста, приказ на случај

Introduction

Hydatid disease, caused by the parasite *Echinococcus granulosus*, is a cystic infection that remains common globally, especially in areas where sheep are being cultivated mostly [1,2]. It could affect any organ, and the liver is most frequently involved (50-70%), followed by the lungs (about 25%) [3]. Involvement of the spleen, known as splenic echinococcal disease (SED), is uncommon, representing only 0.5-4% of all abdominal hydatid cases [4].

Diagnosing SED can be difficult because symptoms are often vague and nonspecific, and the condition itself is rare. If the diagnosis is delayed, several complications may occur. Short-term issues can include

rupture of the cyst, allergic reactions, or spread of the infection within the abdominal cavity, potentially leading to fistulous connections between organs [1,4].

Case Presentation

A 62-year-old woman presented with persistent, dull abdominal pain localized mainly in the epigastric region and left upper quadrant, accompanied by constipation and weight loss lasting for over a year. She had previously undergone evaluation by gastroenterologists and hepatologists, including abdominal ultrasonography and colonoscopy.

Serology for *Echinococcus*, using indirect hemagglutination (IHA), was positive.

All routine laboratory parameters, including hemoglobin, platelet count, coagulation profile, inflammatory markers, and liver function tests, were within normal limits (Table 1).

Table 1. Laboratory findings

Parameter	Result	Unit	Reference/Interpretation
Hemoglobin	122	g/L	Within normal range (no anemia)
Platelet count	319,000	/mm ³	Normal
Prothrombin time (PT)	10.7	Seconds	Normal
C-reactive protein (CRP)	2.5	mg/L	No inflammatory response
Alanine aminotransferase (ALT)	12	U/L	Normal liver function
Aspartate aminotransferase (AST)	14	U/L	Normal liver function
Total bilirubin	11.6	μmol/L	Normal

Colonoscopy was largely unremarkable except for grade II internal hemorrhoids.

Abdominal ultrasonography demonstrated a well-defined, oval mass in the spleen, measuring at least 10 cm, with a complex structure characterized by septations and mixed hypoechoic and hyperechoic areas. No in-

volvement of other abdominal organs was noted. A contrast-enhanced CT scan of the abdomen and pelvis was subsequently performed, confirming a splenic cystic lesion measuring 85 × 85 mm that caused mass effect on surrounding tissue (Figure 1 and Figure 2).

Figure 1 and 2, showing cystic formation with calcifications and inner hyperdense zones suspected to be calcifications

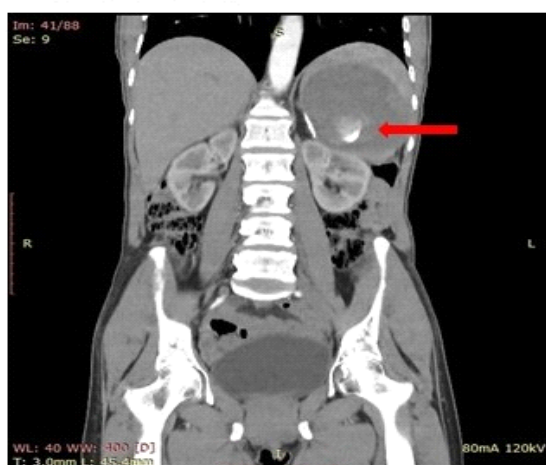


Figure 1



Figure 2

Fig. 1 and 2. CT showing a cystic formation with calcifications and inner hyperdense zones suspected to be calcifications

The patient underwent elective surgery via midline laparotomy. Intraoperative findings revealed a hydatid cyst

located at the upper pole of the spleen. A total splenectomy was performed, and an abdominal drain was placed.

Gross examination of the removed spleen was consistent with a hydatid cyst (Figure 3).

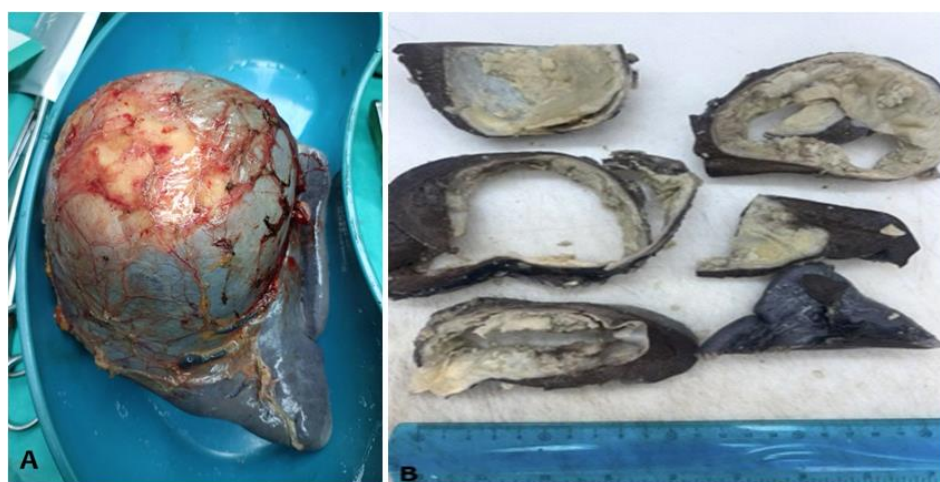


Fig. 3. A. Gross view of surgical specimen B. Cut section of the splenic hydatid cyst

Histological examination showed luminal eosinophilic necrotic debris with polymorphonuclear leukocytes and numerous cholesterol crystals, as well as foci of calcification. Peripherally, a pericyst of collagenous connective tissue with chronic inflammatory infiltrate and occasional multinucleated foreign body-type giant cells, as well as clusters of macrophages, was observed.

In some sections, remnants of the hydatid cyst wall composed of lamellar chitinous material were visible within the lumen. No vital germinal epithelium or scolex was identified. Compressed splenic tissue with stagnant changes was noted peripherally. The submitted material was consistent with an echinococcal cyst of the spleen (Figure 4).

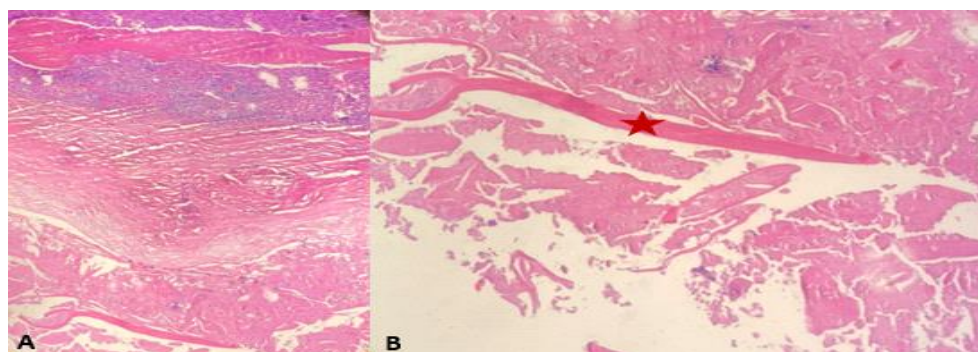


Fig. 4. A. Histology of splenic hydatid cyst HEX40 B. Laminated wall cyst (red star)HEX100

The patient was discharged from the hospital on the fifth postoperative day with recommendations for albendazole (400 mg twice daily) for three cycles of 28 days, with a two-week pause between cycles. No local recurrence was detected during postoperative follow-up for one year.

Discussion

This is a rare instance of a solitary splenic hydatid cyst with no other organ involvement, depicting an uncommon manifestation that constitutes merely 0.5-4% of abdominal hydatid cases [4]. These situations present a diagnostic challenge due to the nonspecific clinical manifestations and the need to consider a wide array of differential diagnoses. Hydatid disease typically has no

symptoms and can persist for several years until the cyst enlarges, at which time symptoms start to manifest [5]. Pulmonary cysts can lead to cough, shortness of breath, and chest discomfort, whereas hepatic cysts result in right upper quadrant abdominal discomfort, nausea, and general malaise [6,7].

The main symptoms of hydatid cysts primarily arise from the pressure effects they exert on nearby structures, including blockages in the lymphatic and venous systems. Primary cysts can become secondarily infected by bacteria, or they may rupture, potentially leading to an anaphylactic hypersensitivity reaction [8]. Serological tests can additionally assist in diagnosing this disease by measuring IgG antibodies. The sensitivity of serological tests is 80-100% for the liver, 50-56% for the lungs, and 25-56% for the spleen [9]. Diagnosis

of hydatid disease relies on clinical suspicion supported by imaging techniques like ultrasound and computed tomography for better anatomical visualization, along with serological testing as a crucial component. Histopathological examination remains the definitive diagnostic method.

Administering albendazole therapy before and/or after surgery is advised to lower the chance of recurrence. While total splenectomy remains the most frequently performed surgery, spleen-preserving procedures are gaining support for certain patients to preserve splenic immune function. It encompasses traditional open surgical procedures and laparoscopic techniques, comprising partial or complete splenectomy. Spleen-sparing surgery encompasses cyst unroofing, partial cystectomy, partial splenectomy, and cystojejunal Roux-en-Y anastomosis [10,11].

In our patient, effective surgical management was accomplished, highlighting the importance of early diagnosis and prompt intervention to avert complications.

Conclusion

This rare case highlights the importance of considering splenic hydatid cysts as a possible diagnosis in patients presenting with abdominal pain and a palpable mass, especially in endemic regions. Potential complications such as cyst rupture, and the need for prompt and appropriate management to prevent further complications and ensure the best possible outcome for the patient.

Conflict of interests: None declared.

References

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