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Primary splenic hydatidosis: a case report

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Abstract

We herein report a case of primary splenic hydatidosis to provide data regarding the diagnosis, treatment, and epidemiological statistics of this disease. The patient was from a pastoral area and was diagnosed with primary splenic hydatidosis with chronic atrophic gastritis. The patient had no history of surgical treatment of hydatidosis. The diagnosis was mainly based on possible exposure to endemic areas, imaging findings, serological test results, and operative and pathological examination findings. Laparoscopic splenectomy was performed, and regular albendazole therapy was given after the operation. The patient was admitted to the hospital for gastrointestinal bleeding 3 months postoperatively, and she was successfully treated and discharged. No recurrence of hydatid foci has been observed since the follow-up.

Keywords

Hydatidosis, spleen, primary disease, gastrointestinal bleeding, splenectomy, albendazole

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Introduction

Hydatidosis is a parasitic disease that is mainly prevalent in pastoral and semi-pastoral areas. The main primary site is the liver; *Echinococcus* eggs infect the host by entering the portal vein through the mesentery to form hydatid foci.[1] Patients with extrahepatic hydatidosis generally have *in situ* liver lesions; however, a few of these patients have no original focus.[2] The incidence of such patients is relatively low. The

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analysis and collation of data are not only of great significance to the diagnosis and treatment of these patients but is also helpful in studying the pathogenesis and possible mechanism of dissemination of hydatidosis throughout the body. We herein present a rare case of primary splenic hydatidosis that was treated by laparoscopic splenectomy.

Case report

A 66-year-old woman of Hui nationality living in Hualong County, Qinghai Province presented with a 20-day history of upper abdominal distension and discomfort with no obvious cause. The patient's spirit, diet, sleep, and weight had not changed substantially. Fallopian tube ligation had been performed 30 years previously, and she had a history of chronic atrophic gastritis. No abdominal tenderness or hepatosplenic mass was found.

After admission, abdominal contrast-enhanced computed tomography showed cystic lesions with thick-walled annular calcification slightly protruding from the superior spleen to the spleen–stomach space (Figure 1(a), (b)). Primary splenic hydatidosis was considered. Pulmonary scans showed chronic bronchitis. No obvious abnormalities were found in the craniocerebral or pelvic scans.

The biochemical examination results (Table 1) showed negativity for

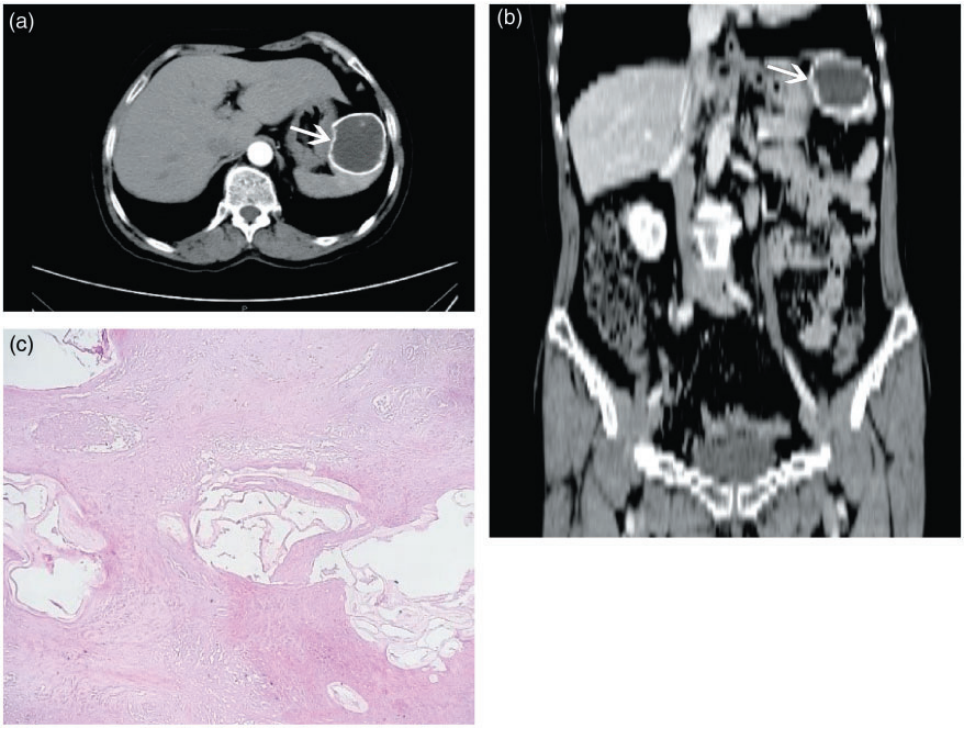

Figure 1. Computed tomography and pathologic images. (a) Contrast-enhanced computed tomography showed a cyst (arrow) occupying the superior part of the spleen with a clear boundary, wall thickening, and circular calcification. Punctate calcification foci could also be seen. (b) The lesion (arrow) was also seen in the coronal reformatted image. (c) Hematoxylin–eosin staining of the lesion showed pink material containing hydatidosis-affected tissue.

Table 1. Patient's diagnostic and treatment data.

		Reference range
Basic and clinical data		
Age (years)	66	–
Sex	Female	–
Nationality	Hui	–
History of contact in epidemic areas	Yes	–
Operative method	Laparoscopic splenectomy	–
Intraoperative blood loss (mL)	50	–
Lesion location	Superior spleen	–
Lesion diameter (cm)	6.0×5.0	
Tissues or organs adhered to the lesion	Omentum and diaphragm	–
Postoperative complications	None	–
Laboratory examination		
ALT (U/L)	21	0.0–38.0
AST (U/L)	23	0.0–38.0
TBIL (μmol/L)	24.4	0.0–17.0
DBIL (μmol/L)	8.3	0.0–3.4
IBIL (μmol/L)	16.1	0.0–14.4
HBsAg (ng/mL)	0.001	0.0–0.5
HBsAb (ng/mL)	10.0	0.0–10.0
White blood cell count ($\times 10^9$/L)	6.13	3.5–9.5
Neutrophil count ($\times 10^9$/L)	4.12	1.8–6.3
Lymphocyte count ($\times 10^9$/L)	1.64	1.1–3.2
Red blood cell count ($\times 10^{12}$/L)	4.88	4.3–5.8
Platelet count ($\times 10^9$/L)	133	125–350
Hemoglobin (g/L)	147	130–175
Anti-*Echinococcus* IgG antibody	Neg	Neg
ABO blood type	O	–
Rh blood type	Pos	–

ALT: alanine aminotransferase, AST: aspartate aminotransferase, TBIL: total bilirubin, DBIL: direct bilirubin, IBIL: indirect bilirubin, HBsAg: hepatitis B surface antigen, HBsAb: hepatitis B surface antibody, Pos: positive, Neg: negative.

anti-*Echinococcus* IgG antibody. The total, direct, and indirect bilirubin concentrations were increased, indicating occult jaundice. The alanine aminotransferase, aspartate aminotransferase, sodium, and potassium concentrations were within the reference ranges. The patient had no antibodies to hepatitis B, syphilis, or human immunodeficiency virus. Her blood type was O, Rh+. She had no significant abnormalities in her blood cell count and no history of vaccination before the operation.

Considering that the patient had no special contraindications for an operation, laparoscopic splenectomy was performed on the eighth day after admission. First, pneumoperitoneum was established using four puncture locations: the navel, 2 cm below the xiphoid process, the midline of the left clavicle, and the intersection point of the anterior axillary line and the left inferior border of the rib. We observed mild adhesions of the spleen with the omentum and diaphragm. Next, the perisplenic ligament was dissected and the upper edge of the pancreas was dissociated. The splenic artery was then separated and ligated. We used an ultrasonic knife to cut the

perigastric vessels and then carefully separated the pancreatic tail around the splenic hilum. An Endo GIA stapler (Medtronic, Dublin, Ireland) was used to disconnect the splenic pedicle while protecting the tail of the pancreas from damage. Finally, we removed the spleen through an incision extending from the midline of the left clavicle to the anterior axillary line. The intraoperative blood loss volume was about 50 mL, and no transfusion was needed.

We encountered some difficulties during the operation. Moving the spleen was difficult because of the large lesion. The tail of the pancreas was close to the splenic hilum, making performance of the operation inconvenient. We also had to consider avoiding compression during removal of the spleen.

After the surgery, we found that the spleen measured about $10 \times 8 \times 4$ cm and was hard and rounded. The cyst was located at the superior aspect of the spleen, and its dimensions were about 6×5 cm. Pathological examination showed calcification of fibrocystic walls and infiltration of chronic inflammatory cells (Figure 1(c)). A diagnosis of hydatidosis was confirmed.

After 14 days of medical therapy and careful nursing, the patient recovered well. Although her total bilirubin and direct bilirubin concentrations were slightly higher than before treatment, all other laboratory parameters were within the reference ranges. She was treated with albendazole at 15 mg/kg per day (divided into two daily doses) for 6 months after discharge from the hospital.

After 32 months of follow-up, the patient still showed no evidence of lesion recurrence. However, gastrointestinal bleeding as confirmed by gastroscopy occurred 3 months postoperatively. The specific cause was unknown, but the patient was uneventfully treated.

Discussion

Hydatidosis is distributed worldwide. Although it is regarded as a benign lesion, it has a certain mortality rate because of its potential complications, including rupture, implantation, and invasion of important organs. Hydatidosis is most commonly located in the liver and lungs; the spleen is the third most commonly affected organ.[2] The incidence of hydatidosis originating in the spleen without liver involvement is very low. According to the current statistical data, the incidence of hydatidosis originating from the spleen is $<2\%$.[3]

Our patient presented with upper abdominal pain and discomfort. Her symptoms were basically consistent with those reported by Culafić et al.[4] The diagnosis of hydatidosis was mainly based on a contrast-enhanced abdominal computed tomography scan; a serological test was negative. Of course, we also identified other cysts, hematomas, and tumors in the spleen. Laparoscopic splenectomy was performed without preoperative contraindications, and the patient recovered well.

Splenectomy is considered to be the standard procedure for the treatment of splenic hydatidosis because the lesion is susceptible to rupture and implantation. Splenectomy may minimize the postoperative recurrence of hydatid cysts.[3,5] However, Culafić et al.[4] are skeptical about the effectiveness of splenectomy to some extent because among 20 patients with splenic hydatidosis, 13 underwent splenectomy and developed complications, while the patients who underwent partial splenectomy developed no complications. Complications such as sepsis, pneumonia, and hemorrhage may readily occur after splenectomy, and some complications may be fatal.[6] Our patient was diagnosed with gastrointestinal bleeding 3 months postoperatively. Although we

found no evidence indicating that this bleeding was directly related to the splenectomy, the patient certainly did not develop gastrointestinal bleeding due to her preoperative chronic atrophic gastritis. Whether splenic hydatidosis should be treated by splenectomy may require more studies with longer follow-up periods.

Compared with open splenectomy, laparoscopic splenectomy has the advantages of less trauma, less pain, and a shorter hospitalization time; however, the risks of cyst rupture and cystic fluid leakage are higher. In the present case, the cyst wall was obviously thickened and calcified, the patient's overall condition was good, and no severe abdominal adhesions were present. Considering these various factors, we performed laparoscopic splenectomy.

In conclusion, the present report describes a case of hydatidosis originating from the spleen. A review of the patient may be helpful for clinical diagnosis and treatment and future research on the pathogenesis of this condition.

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Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Ethical approval

This article was approved by the Ethics Committee of the Affiliated Hospital of Qinghai University located at No. 29 Tongren Road, Xining City, Qinghai Province, China.

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Patient consent

The patient agreed to the use of her imaging and clinical data for publication and academic research and provided written informed consent.

References

1. Menghi C, Gatta C and Arias L. Human cystic and alveolar echinococcosis. *Curr Treat Options Infect Dis* 2017; 9: 210–222.
2. Ilica AT, Kocaoglu M, Zeybek N, et al. Extrahepatic abdominal hydatid disease caused by: imaging findings. *AJR Am J Roentgenol* 2007; 189: 337–343.
3. Durgun V, Kapan SM, Karabicak I, et al. Primary splenic hydatidosis. *Dig Surg* 2003; 20: 38–41.
4. Culafić DM, Kerkez MD, Mijac DD, et al. Spleen hydatidosis: clinical manifestations and treatment. *Scand J Gastroenterol* 2010; 45: 186–190.
5. Celebi S, Basaranoglu M, Karaaslan H, et al. A splenic hydatid cyst case presented with lumbar pain. *Intern Med* 2006; 45: 1023.
6. Ellison EC and Fabri PJ. Complications of splenectomy: etiology, prevention, and management. *Surg Clin North Am* 1983; 63: 1313–1330.