

## Primary splenic hydatidosis

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**Abstract** Primary extrahepatic hydatid cysts are rare, and primary splenic hydatid cysts even more so, constituting 2% to 3.5% of all hydatid cysts. We report here a case series of isolated splenic hydatid cysts. Case records of 382 adult patients of abdominal hydatid cysts were analyzed; eight of them (2%; aged 23 to 45 years, 5 women and 3 men) had primary splenic hydatid cysts. Seven patients presented with dull dragging pain in the left hypochondrium. Four patients had splenomegaly. Diagnosis was made at ultrasonography and/or contrast-enhanced computed tomography. Enzyme linked immunosorbent assay for hydatid antibodies tested positive in all patients. One patient presented with hemoperitoneum. All patients underwent splenectomy. Primary splenic hydatid cyst is rare but should be considered in patients with cystic lesions of the spleen.

**Keywords** *Echinococcus* · Splenectomy

### Introduction

Cystic lesions of the spleen are rare. These include parasitic cysts, benign neoplastic cysts such as lymphangiomas, cavernous hemangiomas or dermoid cysts, and non-neoplastic cysts (pseudocysts) resulting from hemorrhage or an area of infarction [1–4]. Among parasitic cysts, hydatid cysts are the most common (Table 1).

Classic hydatid cysts are caused by *Echinococcus granulosus*. These grow very slowly, at the rate of 1–2 cm

per year [1, 2], depending on the host-parasite relationship and resistance offered by the surrounding structures. Spleen is the third most common site for echinococcosis after liver and lungs. Splenic hydatid cysts, first described as an autopsy finding by Berlot [1], are rare. Hydatid cysts in spleen may occur as a part of disseminated disease or may be isolated. Hydatid disease involving only the spleen, in the absence of disease in other organs is known as primary splenic hydatidosis.

Splenic hydatid cysts occur at all ages and in both sexes. These may be detected incidentally or may present with non-specific complaints [3, 4]. Occasionally, these get infected, fistulize to adjacent organs or rupture into the peritoneal cavity, or may present with massive lower gastrointestinal bleeding. Serology, abdominal ultrasound (US) and computed tomography help in diagnosis. Management varies from spleen preservation to splenectomy [2, 4].

We report here our experience with primary splenic hydatid cysts in adults.

### Case series

We retrospectively reviewed the case records of patients attending our institution to identify those diagnosed as abdominal hydatidosis between January 1998 and December 2007. Details of medical history and examination of these patients and results of relevant investigations were recorded. Of 382 patients with abdominal hydatid cysts, eight (2%) had isolated splenic hydatid cysts. These patients form the basis of this report; patients with coexistent cyst in another organ were excluded.

The eight patients (5 women, 3 men) with primary splenic hydatid cysts were aged 23 to 45 years. Seven patients presented with dull dragging pain in the left

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**Table 1** Clinical presentation of patients with isolated splenic hydatid cyst

Age/gender	Clinical manifestations	Hemoglobin (g/dL)	Platelets (X10 <sup>9</sup> /L)	Radiological features
45 M	Abdominal pain, palpable spleen	11.0	170	Multilocular, calcified splenic cyst, 8×7 cm
36 M	Abdominal pain, palpable spleen	11.6	195	Multilocular splenic cyst, 6×8 cm
39 F	Abdominal pain	10.8	170	Multilocular splenic cyst, 6×6 cm
47 M	Abdominal pain, palpable spleen	13.0	188	Unilocular, calcified splenic cyst, 9×7 cm
23 F	Abdominal pain	12.0	220	Multilocular, calcified splenic cyst, 6×7 cm
30 M	Abdominal pain	10.6	197	Multilocular splenic cyst, 7×7 cm
30 F	Abdominal pain, palpable spleen	11.0	167	Multilocular splenic cyst, 8×9 cm
32 M	History of trauma, tachycardia, abdominal tenderness	12.0	177	Hemoperitoneum, lacerated spleen, multilocular splenic cyst, multiple daughter cysts in peritoneal cavity

M Male, F Female

hypocondrium; of these, 2 had also noticed an enlarging mass in the left upper abdomen. There were no other complaints. On examination, the vital signs were normal in all the seven patients. Four patients had palpable spleen. Preoperative hemogram, renal and liver function tests, and chest X-ray were within normal limits. All seven patients had well-defined, rounded soft-tissue opacities with calcified margins in the left upper abdomen on abdominal X-rays. Diagnosis was made at abdominal ultrasound, contrast-enhanced CT scan or both. These showed single well-defined rounded loculated cysts with multiple septae in the superior pole of the spleen in six patients (Fig. 1) and a unilocular cystic lesion in the inferior pole of spleen in one patient. All patients underwent splenectomy. Surgery revealed multilocular cysts in 5 patients and unilocular cysts in 2 patients. All patients had marked perisplenic adhesions.

One patient presented to the emergency department of our hospital with bullet injury to the abdomen and hemoperitoneum. On exploration, the spleen was lacerated

and had a ruptured hydatid cyst. Peritoneal cavity had several daughter cysts. Splenectomy, evacuation of daughter cysts from the peritoneal cavity and thorough peritoneal lavage with normal saline and a scolicedal agent (*Betadine* 1% [Wockhardt, Aurangabad, India]) were done. Multiple drains were left in the peritoneal cavity. The postoperative period was uneventful.

Histopathology confirmed the diagnosis of hydatid cyst in all the eight cases. ELISA for anti-hydatid antibodies was positive in all cases; in the patient with splenic laceration, it was done after surgery.

All the patients received albendazole (10 mg/kg/day) for one month prior to surgery (except the patient with splenic injury) and in the postoperative period as three cycles of 4 weeks each with one-week intervening drug-free intervals. All the seven patients undergoing elective splenectomy received pneumococcal vaccine 2–3 weeks before the surgery, to allow development of protective antibodies. The patient with history of trauma was vaccinated immediately after surgery.



**Fig. 1** CT scan showing hydatid cyst in spleen

## Discussion

Hydatid cysts represent nearly two-thirds of cystic lesions of the spleen [5–7]. However, splenic hydatid cysts account for only 0.5% to 8% of all hydatidosis [4, 6]. Splenic hydatid cysts occur in 1.5% to 3.5% of all cases of abdominal hydatidosis [8, 9]. In our experience, isolated splenic hydatid cysts constituted 2% in our patients with abdominal hydatid disease. Splenic hydatid cysts are mostly solitary [8] but may sometimes be multiple [4], and may be associated with cysts in other body parts in up to 30% of cases [3, 9, 10]. The parasite may reach the spleen through blood stream, through lymphatics [4] and by reflux into the spleen from portal vein at the time of raised intra-abdominal pressure [1]. Isolated involvement of spleen is rare. Primary infestation of the spleen usually takes place

by the arterial route after the parasite has bypassed the liver and lungs.

Approximately 30% of splenic cysts are asymptomatic. The most common finding is incidentally discovered splenomegaly [1, 2]. The symptoms are non-specific [11–13], and include an abdominal lump, dull dragging ache, dyspepsia, constipation due to pressure on colon, and dyspnea due to pushing up of the left diaphragm. Most of our patients had such a presentation. Some patients may present with complications such as infection of the cyst, rupture of the cyst into the peritoneal cavity, fistula formation into hollow viscera like colon or stomach [14], bronchopleural fistula, sympathetic pleural effusion, calcification [15], hypersplenism, and severe urticaria [16].

The blood counts were normal in all our patients. Eosinophilia (above 3%) is the most common laboratory finding. Serum immunoelectrophoresis is currently the most reliable serological test, with a sensitivity of approximately 90%. It stays positive for up to one year after the infection has been eradicated. Indirect hemagglutination test has a sensitivity of 85% but remains positive for several years [4]. Complement fixation, enzyme linked immunosorbent assay (ELISA), western blot analysis and Casoni [14] skin test have also been used. In our study, all patients tested positive by ELISA.

Plain X-ray of the abdomen may show a soft tissue shadow, with or without calcification, in the left upper quadrant, or a displacement of the left hemidiaphragm, stomach or transverse colon. Intravenous urography may show downward displacement of the left kidney. US and CT scan (Fig. 1) are reliable imaging tools for diagnosis and screening, but are not specific since similar findings may occur in other cystic lesions of spleen, such as abscess, hematoma, epidermoid cyst, pseudocyst or neoplasms [9, 11].

The treatment is principally surgical. Splenectomy has been the treatment of choice for splenic hydatid cysts since it is easy, rapid and effective. Pre- and postoperative administration of albendazole used to sterilize the cyst, reduce the risk of anaphylaxis, decrease the tension in the cyst wall and to reduce the postoperative recurrence rate [7]. Intra-operatively, hypertonic saline or 0.5% silver nitrate is instilled into the cyst before opening it; this tends to kill the daughter cysts, thereby preventing further spread and anaphylactic reaction [7].

Conservative surgery like partial splenectomy, cyst enucleation, deroofting of the cyst with omentoplasty or

external drainage of the cyst have been done for superficial cysts, cysts located in one pole of the spleen or cysts which are deemed unresectable due to enormous size and perisplenic adhesions [3, 10, 16].

We recommend that the diagnosis of hydatid cyst should be considered in all patients with cystic lesions of the spleen. Splenectomy is the gold standard treatment for splenic hydatidosis and use of albendazole both preoperatively and postoperatively may reduce the risk of recurrence.

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