Hydatid Cyst of the Spleen: Tunisian Series of 21 Cases

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Abstract Hydatid disease is a parasitic infection caused by *Echinococcus granulosus*. Splenic involvement is rare even in endemic areas. The aim of this study is to specify epidemiologic features, diagnostic tools, and therapeutic modalities of this uncommon entity. This is a retrospective study of 21 patients operated on for a splenic hydatid cyst between January 1996 and December 2011. The mean age was 43.8 years (range 15–72 years). Sixteen patients (76.2 %) had symptoms related to a splenic location of hydatid disease. While splenic hydatid disease was solitary in 8 patients (38.1 %), other locations were present in 13 patients (61.9 %), mainly in the liver. Most splenic cysts were type I (39.3 %) or III (42.8 %). Nine patients (42.8 %) underwent resection of the protruding dome with one postoperative complication (suppuration of residual cavity). Total and partial splenectomies were performed in nine and three patients, respectively, without any complication. No death was noted. After a mean follow-up period of 36 months (range 2–108 months), no recurrence was observed. Splenic hydatid cyst is a rare location. The diagnostic is usually easy, based on serology and imaging. The surgery is the mainstay of treatment. The type of surgical procedure depends on size, number, and location of the cyst. Total splenectomy is more effective but may have high morbidity and mortality and must be reserved to specific situations. Conservative procedure is safer but could increase the rate of recurrence and postoperative collection. Puncture aspiration injection reaspiration could be proposed for small cyst in inoperable patients.

Keywords Echinococcosis · Spleen · Surgery · Splenectomy

Introduction

Hydatid disease is a parasitic infection caused by *Echinococcus granulosus*. Although it can be found in any organ or soft tissue, it mostly affects the liver and lungs. Splenic involvement is rare even in endemic areas. The aim of this study is to specify epidemiologic features, diagnostic tools, and therapeutic modalities of this uncommon entity.

Materials and Methods

Between January 1996 and December 2011, the records of patients that had been surgically treated for spleen hydatid cysts at the Department of General Surgery, Habib Bourguiba's Teaching Hospital, Sfax, Tunisia, were retrospectively reviewed. The variables recorded were demographic characteristics of the patients (age, gender, geographic origin), their symptoms, results of serologic tests (enzyme-linked immunosorbent assay (ELISA) and indirect hemagglutination test), radiographic findings (abdominal ultrasound (US) and computed tomography (CT) scan), surgical methods, mortality, postoperative complications, and disease recurrence.

On US and CT scan, cysts were classified according to the Gharbi's classification (Table 1) [1]. This has been elaborated at the beginning for liver hydatid cysts on the basis of the US findings. Later, it was extended to CT scan findings and to other organs such as the spleen.

Whatever the chosen surgical technique, protection of adjacent organs with large compresses impregnated with hypertonic saline solution (as scolicide) was done. If conservative

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Table 1 Gharbi's classification for hydatid cyst

Type I	Univesicular cyst
Type II	Univesicular cyst with membrane detachment
Type III	Multivesicular cyst
Type IV	Pseudotumoral cyst
Type V	Fully calcified cyst

procedure was chosen, some common steps were always performed: decompression of the cyst by trocar, careful removal of the parasite without spillage of cyst contents, washing of the cavity with hypertonic saline solution, and external drainage of the residual cavity. Splenectomy could be performed with or without prior opening and cleaning of the cyst, depending on its size. If splenectomy (either total or partial) was performed, suction drain was placed in left subphrenic area. For patients who underwent total splenectomy, antipneumococcal vaccine was administered.

Results

This study included 21 patients operated on for splenic hydatid cyst (13 females and 8 males). The mean age was 43.8 years (range 15–72 years). Thirteen patients (61.9 %) came from a rural area. Four patients (19 %) had a history of surgery for hydatid disease.

Sixteen patients (76.2 %) had symptoms related to a splenic location of hydatid disease. These were left upper quadrant pain or heaviness in all cases, left hypochondrium mass in five cases (31.2 %), and splenomegaly in two cases (12.5 %). In five cases (23.8 %), the hydatid cyst of the spleen was discovered incidentally on imaging exams for hepatic hydatid cyst. All these patients had right upper quadrant pain, and one patient had acute cholangitis (fever and jaundice). Hydatid serologic tests were performed for 12 patients (57.1 %) and were positive in the all cases.

Abdominal US was performed in all patients, and CT scan was done in 15 patients (71.4 %). They showed hydatid cyst of the spleen in all cases. These were single in 16 cases (76.2 %) and multiple in 5 cases (23.8 %) (2 cysts: 4 cases; 4 cysts: 1 case). Then, there were 28 splenic hydatid cysts. The average size was 10.1 cm (range 4–20 cm). The most common types were III (12 cases, 42.8 %) and I (11 cases, 39.3 %) (Table 2). The spleen was the only site of the hydatid disease in eight patients (38.1 %). However, other locations were objectified in 13 patients (61.9 %): they were liver cysts in all cases, which were associated in three patients with peritoneal, lung, and chest wall cysts, respectively (Fig. 1).

All patients were operated on, using midline laparotomy in 15 cases (71.4 %), bi-subcostal incision in 5 cases (23.8 %), and left subcostal incision in one case (4.8 %). Intraoperative

Table 2 Type of splenic hydatid cyst according to Gharbi's classification

	Number (%)
Type I	11 (39.3)
Type II	2 (7.2)
Type III	12 (42.8)
Type IV	3 (10.7)
Total	28

findings were similar to imaging data. For splenic hydatid cysts, resection of the protruding dome (RPD) was performed in nine patients (42.8 %). All cysts were single with an average size of 9.5 cm (range 5–19 cm) (Table 3). Total splenectomy was performed in nine patients, also. These were single cysts in four cases and multiple cysts in five cases. The average size was 12 cm (range 7–20 cm) (Table 3). Three patients underwent partial splenectomy: a superior polar resection for a 7-cm-sized cyst and inferior polar resection for 4- and 8-cm-sized cysts (Table 3). The associated hepatic cysts are also treated (mostly by RPD) except of two type V cysts. The patient who had acute cholangitis underwent external biliary drainage through cysto-biliary fistula. The intraperitoneal cyst was treated by subtotal cystectomy.

The diagnosis of splenic hydatid cyst was confirmed in all cases by histopathological examination of resected specimens. Mortality was nil. The postoperative course was uneventful in 19 cases. Two complications were observed: biliary fistula related to a hepatic hydatid cyst which was successfully treated conservatively and purulent suppuration of residual cavity after RPD of a 19-cm-sized splenic cyst. This was successfully managed par antibiotics and CT scan-guided drainage.

Albendazole was administered to 13 patients because of multiple hydatid cysts. After a mean follow-up period of 36 months (range 2–108 months), no recurrence was observed among patients with splenic hydatid cysts.

Discussion

Hydatid disease is endemic in rural areas, particularly in sheep-raising areas, like the Middle East, North Africa, New Zealand, Australia, and South America [2]. The most common sites of disease are the liver (70 %) and the lungs (40 %). The spleen constitutes the third location but remains uncommon even in endemic countries. The frequency of splenic hydatid disease ranges from 0.9 to 8 % among overall hydatid locations and between 0.5 and 4 % within abdominal hydatid diseases [3–5]. This low incidence is related to mechanism of splenic involvement by *E. granulosus*. In fact, these may occur through three ways: systemic, portal, and local ones. In the systemic way, which seems to be preponderant, the eggs of

Fig. 1 **a** Large type I splenic hydatid cyst occupying almost all of the spleen (treated by total splenectomy) associated with multiple hydatid cyst of the liver (treated by RPD). **b** Type I medio-splenic hydatid cyst with intraperitoneal development treated by total splenectomy. **c** Type I hydatid cyst of the upper pole of the spleen treated by partial splenectomy. **d** Type II hydatid cyst of the lower pole of the spleen treated by partial splenectomy

parasite escape the liver and lung barrier before entering systemic circulation and setting at the splenic parenchyma. In the portal way, splenic hydatid disease arises from retrograde spread of parasites via the portal and splenic veins. In the local way, splenic involvement results from gastric or colonic trans-parietal passage of eggs or from intraperitoneal rupture of a hepatic hydatid cyst [2, 4]. Splenic hydatid cysts can be solitary (29 to 52 %; our series, 38.1 %) or associated with another location, more frequently liver and lungs [4, 5].

Due to its slow growth, splenic hydatid cyst may be completely asymptomatic and was then incidentally discovered during investigation for unrelated symptoms or for pre-operative assessment of another hydatid location (26 to 28.6 %; our series, 23.8 %) [2, 4–6]. Elsewhere, the main symptoms are abdominal discomfort or pain (all symptomatic patients in our series) and palpable mass in the left upper quadrant (31.2 % of symptomatic patients in our series) [2, 4, 5]. Rarely, complications can reveal the cyst (no patients in our series). They include infection, fistulisation to adjacent organs (colon, stomach, abdominal wall), intra-abdominal rupture with anaphylactic reaction or hydatidosis, renal arte-

rial compression with systemic hypertension, and splenic vein compression with segmental portal hypertension [2, 6–11].

Diagnosis is based on a combination of hydatid serology and imaging which is contributive in almost 90 % of cases [2]. Serological tests include indirect hemagglutination and an enzyme-linked immunosorbent assay (ELISA), because of their higher sensitivity and specificity, respectively. US and CT scans are the major diagnostic tools for splenic hydatid cyst. Calcifications of the cyst wall, presence of daughter cysts, membrane detachment, or concomitant cystic lesions in the liver or other organs are very suggestive to the diagnosis. However, these findings are not always present especially in the early stages of the disease (type I), and other splenic cystic lesions, such as an epidermoid cyst, abscess, pseudocyst, or cystic neoplasm, may have a similar appearance [2, 6]. In cases where the serology is negative and US and CT are inconclusive, MRI may be helpful by demonstrating a low-signal intensity rim on T2-weighted images (rim sign). However, this finding is not specific because it can also be seen with some other cystic lesions such as abscesses, necrotic tumors, and hematomas [12].

Table 3 Distribution of surgical procedure by number and size of cyst

Surgical procedure	Number of cysts	Average size of cyst (range) (in cm)
Resection of the protruding dome	Single: 9 cases	9.5 (5–19)
Total splenectomy	Single: 4 cases	12 (7–20)
	Multiple: 5 cases	
Partial splenectomy	Single: 3 cases	6.3 (4–8)

Elsewhere, the diagnosis of splenic hydatid cyst is made intraoperatively or postoperatively on histological examination of specimen.

Surgery is the mainstay of splenic hydatid cyst. Laparoscopic approach has been reported by some authors but has not yet been recommended as reference because of high risk of peritoneal spillage [13–16]. Surgical techniques are divided to radical (total or partial splenectomy) and conservative procedures (RPD).

Total splenectomy is theoretically the best treatment because it provides complete removal of the parasite and therefore prevents recurrence and complications related to the residual cavity. However, it is sometimes difficult to perform because of close adhesions with adjacent organs. On the other hand, some authors had reported high mortality (3.7 to 22.5 %) and morbidity (21 to 25 %), mainly related to overwhelming postsplenectomy sepsis (fulminant pneumonia, septicemia, etc.) especially in children [17, 18]. For these reasons, the total splenectomy is reserved to multiple cysts, giant cyst occupying more than 75 % of spleen, and central intraparenchymal cysts without a protruding dome [4, 5]. In our series, total splenectomy was performed for multiple or large cysts (average size of 12 cm).

The RPD technique is an easy, no-bleeding procedure that is nearly always feasible since the cyst is available on the surface of the spleen, preserving at least 25 % of parenchyma. In our series, this procedure was done for single and smaller cysts (average size of 9.5 cm). The disadvantage is the risk of suppuration of residual cavity (27.3 %; our series, 11.1 %) and of recurrence (18.2 %; our series, 0 %) which can be caused by reinfestation or by little missed cyst in the remaining spleen or in residual pericyst [4, 5].

Partial splenectomy is an intermediate procedure that avoids the complications of RPD and total splenectomy. It is feasible if the cyst is located in a polar position and save more than 25 % of the splenic parenchyma [4]. This limit of 25 % represents the minimum of splenic tissue necessary to preserve immune function of the spleen [4]. Three patients in our series underwent partial splenectomy.

Puncture aspiration injection reaspiration (PAIR) is an alternative to surgery. In the international literature, there is one study which was focused on PAIR for splenic hydatid cyst [19]. Eight patients with nine splenic hydatid cysts underwent injection of ethanol 95 % and povidone iodine 1 % during 10 min into the cyst cavity and after that of 2 to 5 ml of albendazole. During the follow-up period, mean cyst diameter decreased from 46 ± 16.4 mm to 13.6 ± 16.26 mm, and five cysts (55 %), all smaller than 50 mm, disappeared. Thus, PAIR seems to be more effective in small cysts (<5 cm). Further larger studies are needed to confirm these facts.

Benzimidazole carbamates (mebendazole and albendazole) allow reducing cyst viability but rarely give definite cure. Like in a hepatic location, these drugs are usually used in splenic

hydatid cyst either alone for patients who are at high risk for surgery or after surgery in order to reduce recurrence rate for multiple cysts after spontaneous intraperitoneal rupture or accidental spillage during surgery. They are also indicated as a concomitant therapy with PAIR [20].

Conclusion

The diagnosis of splenic hydatid cyst is usually easy, based on serology and imaging. Total splenectomy is more effective but may have high morbidity and mortality and must be reserved to specific situations. A conservative procedure is safer but could increase the rate of recurrence and postoperative collection. PAIR could be proposed for small cysts in inoperable patients.

Conflict of Interest The authors declare that they have no conflict of interest.

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