Primitive Isolated Hydatid Cyst of the Spleen: Total Splenectomy Versus Spleen Saving Surgical Modalities

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Abstract

Background

This study aims to describe the clinical features of the isolated primitive splenic hydatid cyst, discuss and compare the different surgical approaches of this uncommon disease.

Methods

This is a descriptive retrospective study carried out over a period of 7 years extended from January 2013 until December 2019 reporting eight cases of isolated primitive splenic localization of hydatid disease. Data were collected from the register of the general surgery department of the Jendouba regional hospital. Files concerning another associated hydatid localization were excluded. Four patients underwent total splenectomy and four of them underwent different spleen preserving surgical techniques including resection of the protruding dome, partial splenectomy and pericystectomy.

Results

The discovery of the pathology was incidental in 50% of cases, while pain in the left upper quadrant of the abdomen and renitent mass in the same quadrant revealed the pathology only in 25% and 12,5% respectively . None of patients who underwent total splenectomy had fever or sings of postoperative sepsis. Compared to those who had total splenectomy, patients who underwent spleen preserving surgery had a longer average hospital stay ( 9 vs 6,25 days) related to post-operative complications including abscess in the residual cavity after protruding dome resection in one patient and post-operative haemorrhage in one patient.

Conclusions

The current case series argues in favor of total splenectomy, preferably by laparoscopic route whenever the technical platform allows it, associated with some specific peri-operative therapeutic measures, as the safest way that helps to avoid post-operative complications of spleen saving surgical modalities. These complications are usually difficult to manage in poor countries with limited technical resources. Total splenectomy guarantees at least a decreased hospital stay, reduced healthcare costs, and the absence of recurrence in highly endemic underdevelloped countries.

Background

The splenic localization of the hydatid cyst is exceptional and the clinical presentation lacks specificity. Therefore, this cosmopolitan parasitosis should always be considered as a possible differential diagnosis of every splenic cyst. The aim of this study is to describe the clinical features, discuss and compare the different surgical modalities of this rare localization of the hydatid disease.

Methods

This is a descriptive retrospective study carried out over a period of 7 years extended from January 2013 until December 2019 concerning patients hospitalized for abdominal hydatidosis. The retrospective analysis was done by collecting data from the register of the general surgery department of the Jendouba regional hospital and the clinical records of patients. Of the 391-hydatid disease files, only eight cases of isolated splenic localization were collected and all files concerning another associated hydatid localization were excluded. The various details of medical history, clinical examination, further examinations, treatment modalities and follow-up visits were reported. Once the diagnosis was established, all the patients underwent surgical treatment based on total splenectomy or another surgical method preserving the spleen adapted to each patient according to the clinical and radiological data. All surgical resection specimens were sent for pathological
examination. Ethical approval for the study was obtained from the medical ethics committee of the Jendouba regional hospital.

Results

Among the eight patients involved in this study, there were three men (37.5%) and five women (62.5%). The average age was 42.62 years with extremes ranging from 17 to 62 years. Six patients lived in rural areas or had contact with cattle dogs. Pain in the left upper quadrant of the abdomen was the revealing symptom of the pathology in only two patients (25%). A patient presented with a painless retentive mass gradually increasing in size and located in the left upper quadrant of the abdomen. One patient always complained of dyspepsia with bothersome early postprandial fullness. For the four other patients, the discovery was fortuitous based on abdominal ultrasounds requested in framework of assessment of biliary colic (2 patients), viral hepatitis B (1 patient) and renal colic (1 patient). Routine laboratory tests were normal in all patients. Hydatid serology using the enzyme-linked immunosorbent assays (ELISA) technique was performed in all patients but was positive in only three cases (37.5%). Chest X ray failed to demonstrate any concomitant cysts in the lungs. All patients had abdominal ultrasound and abdominal CT. The cysts were solitary in 100% of cases, their diameter varied from 2.9 cm to 16 cm with an average of 7.91 cm. According to Gharbi’s classification, two of them were classified as type I (25%), one cyst type II (12, 5%), four cysts type III (50%) and one cyst type IV (12, 5%). The location of the cyst was superior polar in one case (12, 5%), inferior polar in five cases (62, 5%), centro-parenchymal in two cases (25%). An anti-pneumococcal and anti-meningococcal vaccine was given to all patients approximately 15 days before surgery. All patients were operated on via a left subcostal approach. Four patients had a total splenectomy for two giant cysts almost destroying all the spleen parenchyma and two cysts near the hilum (Fig. 1 + 2). The remaining 4 patients were divided into a resection of the protruding dome in 2 cases for easily accessible cysts on the surface of the spleen, a partial splenectomy in 1 case for an inferior polar cyst measuring 4 cm and one patient had a pericystectomy for a 2.9 cm diameter cyst. Meticulous protection of the operating field using surgical swabs soaked with a scolicidal solution (20% hypertonic saline solution) was performed in all patients. One of the patients who had total splenectomy presented on postoperative day 5 with extreme reactive thrombocytosis with a platelet count exceeding 1000 K/µL requiring him to be put on Acetyl salicylic Acid with Enoxaparin therapy. The iterative check-up blood tests showed a decreased platelet count with a return to normal on post-operative day 12. The remaining three patients who had total splenectomy had a slight transient thrombocytosis with a platelet count below 650 K/µL. Oral antibiotic prophylaxis based on phenoxymethylpenicillin (1 g/day) was administered to all patients undergoing total splenectomy. None of them had fever or signs of postoperative sepsis. One of the patients treated with protruding dome resection leaving behind a non-declivitous residual cavity had fever on post-operative day five associated with vomiting and a biological inflammatory syndrome. An abdominal CT scan performed the same day revealed a suppurative collection occupying this cavity. The decision was to combine CT-guided percutaneous drainage and intravenous antibiotic therapy with a good outcome. For the other patient, the residual cavity left was declivitous and the postoperative course was uneventful. The patient who underwent partial splenectomy had previous systemic hypertension. On postoperative day 1, his blood pressure remained in the normal range but he was noted to be mildly tachycardic. The hemoglobin did not decrease and 75 ml sanguinous fluid was collected during the preceding 24 hours via the redon’s suction drain. A CT scan performed within 12 hours after the onset of the symptoms showed a hemoperitoneum in the perisplenic space and the Douglas pouch without active extravasation of contrast enhanced arterial blood. A revision surgery was required on postoperative day 2 to improve hemostasis and perform peritoneal lavage with drainage. The postoperative outcome was good. Scrupulous wound care helped prevent wound infections in all patients. The hospital stay ranged from 5 days to 13 days with an average of 7.6 days. All patients received albendazole based medical therapy (15 mg/kg/day) for 3 months after surgery. Mean follow-up period was 38 months. No case of recurrence was observed till now.

Discussion

Tunisia is a North African country with wide sheep-raising areas, making it endemic for hydatid disease. Spleen involvement in hydatid disease is relatively rare even in endemic zones\(^1\). The isolated and primitive spleen localization of the hydatid
disease is much more exceptional, representing only 2 to 3% of cases. This corroborates with data in the current study showing that isolated primary splenic localization represents only 2.04% of abdominal hydatid disease. The mechanisms responsible of splenic localization may clarify the rarity of this entity. Only the minority of Echinococcus Granulosus eggs that manage to escape both hepatic and pulmonary barriers may reach the spleen via systemic route. Portal, lymphatic and colonic trans-parietal passage are other infrequent ways that explain the disease.

The very slow growth rate explain the long period of clinical latency, which can reach up to 20 years. In our study, the average age of discovery was over 40. It also explains the fact that in many studies including the current one, the fortuitous discovery is a considerable revealing circumstance. The main clinical symptoms are dominated by left upper quadrant abdominal pain (25% in our series) and the appearance of an abdominal palpable mass in the left upper quadrant which is the other most common complaint. Complications such as compression of adjacent organs and blood vessels, infection, fistulisation and rupture are rare but possible diagnostic situations. Physical examination reveals in most cases, nothing except a painless splenomegaly.

The routine blood tests are often normal. Hydatid serology is contributive in extrahepatic localizations in only 65% of cases. In our series it was associated with a false negative rate of 62.5%. Therefore, it represents an unreliable diagnostic clue. Ultrasound is the best first-line examination for the diagnosis of hydatid cyst of the spleen thanks to its sensitivity of 90–95%. Abdominal CT can directly visualize the image of the cyst, specify its size, location and detailed anatomic contacts with surrounding vessels and organs, and it can look for other concomitant intra-abdominal localizations. The diagnosis is strongly guided by some suggestive signs such as parietal calcifications, floating membrane, daughter cysts and the presence of other concomitant extra-splenic cystic images.

The treatment of hydatid spleen cysts remains primarily surgical. The therapeutic modalities vary between total splenectomy allowing a radical treatment of the parasite and spleen-saving methods including deroofing with omentoplasty, pericystectomy, partial splenectomy, and internal cysto-jejunal anastomosis.

Total splenectomy certainly allows avoiding recurrences and complications related to the residual cavity. However, in addition to its technical difficulty due to adhesions to neighboring organs induced by chronic pericystic inflammation, the risk of gastric or pancreatic injuries and sometimes the difficulty of splenic pedicle control, it exposes to overwhelming post splenectomy infections (OPSI) and thromboembolic complications with a mortality rate ranging to 1.9% in adults. For these reasons, authors recommend keeping this method only for multiple cysts, those occurring on a pathological spleen, those not offering a clear protruding dome, centroparenchymal cysts, near hilum cysts and those with adhesions to surrounding organs. Total splenectomy is also the treatment of choice for giant spleen cysts destroying more than 75% of the spleen parenchyma because in this case the splenic parenchyma is very reduced and almost completely destroyed by the pericystic fibrosis. In the current series, the giant volume of the cyst destroying almost all of the splenic parenchyma made it necessary to choose this method in 2 cases. Whereas in the two other cases, the cysts were very close to the spleen hilum.

As for the spleen-saving approaches, they are dominated by deroofing and omentoplasty, which is a simple classical conservative technique described first by Largot. It is suitable for polar cysts with an accessible protruding dome or in the presence of adhesions between the spleen and surrounding organs making total splenectomy risky. It is associated with a low risk of hemorrhage compared to partial splenectomy, however, it exposes to the risk of postoperative suppuration due to the persistence of a residual cavity. This risk is considerable (25% in our study) and it cannot be overlooked. Whereas, total splenectomy allows avoiding this major disadvantage. Although some studies show no significant difference in term of recurrence between this surgical procedure and total splenectomy, in the case series reported by Ousadden et al, all recurrence cases (two cases) occurred following this surgical technique. In our series, no recurrence case was reported among all patients. Partial splenectomy and pericystectomy are other alternatives that make it possible to alleviate the risk.
of septic post-operative complications by preserving the immune function of the spleen. Partial splenectomy is suitable for polar cysts and requires leaving behind at least 25% of the splenic tissue. However, it exposes to a significant hemorrhagic risk as shown in the current study. The hospital stay is clearly extended with not went to plan conservative surgical methods (Table 2).

Punction aspiration injection and reaspiration (PAIR) is an emerging curative option that has been increasingly advocated due to its minimally invasive nature. It is known to be a safe procedure with much shorter hospital stay and lower morbidity than total splenectomy. It also offers a good alternative to surgery for patients with high anesthesia risk. However, it can proposed only for simple type I or II and small cysts (< 5 cm).

Whichever technique is chosen, the laparoscopic approach remains an increasingly advocated method because of its minimally invasive nature and its good results, but other authors have reservations about such an approach because of the risk of accidental cystic rupture with an anaphylactic shock, which can be a source of spillage of cystic fluid and postoperative recurrences. Albendazole-based perioperative therapy is recommended for at least one month before and after the surgical procedure in order to reduce the volume of the cyst to facilitate its resection and to reduce the risk of postoperative secondary hydatid cysts by sterilizing the disseminated protoscoleces.

**Conclusion**

Despite the fact that currently many authors from all over the world have become a little reluctant to perform total splenectomy because of the risks incurred by this procedure dominated by some serious postoperative infections, this method remains an option of choice especially in underdeveloped countries like ours.

Indeed, the technical platforms available there do not offer the possibility to easily perform more sophisticated surgical methods such as laparoscopic or robotic partial splenectomy, pericystectomy or protruding dome resection in the daily surgical routine.

In addition, the management of immediate postoperative complications of conservative methods such as hemorrhage or abscess formation in the residual cavity is difficult as a consequence of the unavailability of interventional radiology thus, requiring another surgical intervention as demonstrated in this series. All these reasons push the surgeon to reconsider the subject under a different angle and argue in favor of total splenectomy especially in adults. Total splenectomy associated with some crucial and mandatory perioperative therapeutic measures, including preoperative vaccination, subcutaneous prolonged postoperative anticoagulant therapy, antibiotherapy, complete blood count twice a week, thoughtful surveillance and adjusted care according to each patient's associated illnesses, seems to be the most convenient choice for highly endemic and poor countries with often limited technical resources. It guarantees at least a decreased hospital stay, reduced healthcare costs, and the absence of recurrence. Laparoscopic route for total splenectomy should always be advocated as it has the advantages of minimizing postoperative pain, large wound infections, postoperative complications related to bed rest and hospital stay period.

**Abbreviations**

ELISA
enzyme-linked immunosorbent assays
CT
computed tomography
OPSI
overwhelming post splenectomy infections
PAIR
Punction aspiration injection and reaspiration

Declarations

- Ethical approval: An ethical approval was obtained from the Jendouba Regional Hospital Medical Ethics Committee N°: HRJ.53/20
- Consent for publication: A written consent for the publication of personal/clinical data has been obtained from each patient.
- Competing interests: no competing interests to declare
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Tables
Table 1
the main clinical features corresponding to each hydatid cyst

| Patient | sex  | age | discovery     | Cyst location     | Gharbi classification | maximal diameter (cm) | Surgical technique                                        | Post-operative complication requiring surgical/ percutaneous treatment |
|---------|------|-----|---------------|-------------------|-----------------------|-----------------------|----------------------------------------------------------|------------------------------------------------------------------------|
| 1       | Male | 17  | Dyspepsia     | Inferior polar    | Type III              | 7,2                   | Protruding dome resection                                | none                                                                   |
| 2       | Male | 54  | Incidental (Renal colic) | Inferior polar | Type II              | 4                     | Partial splenectomy                                      | hemorrhage                                                             |
| 3       | Male | 62  | Abdominal pain | Superior polar    | Type I               | 5,7                   | Protruding dome resection                                | Abscess in the residual cavity                                       |
| 4       | Female | 46  | Incidental (biliary colic) | Centro-parenchymal | Type III              | 7,5                   | Total splenectomy                                        | none                                                                   |
| 5       | Female | 39  | Palpable Mass | Inferior polar    | Type III              | 14                    | Total splenectomy                                        | none                                                                   |
| 6       | Female | 43  | Abdominal pain | Inferior polar    | Type IV              | 16                    | Total splenectomy                                        | none                                                                   |
| 7       | Female | 29  | Incidental (Hepatitis B) | Inferior polar | Type I               | 2,9                   | pericystectomy                                           | none                                                                   |
| 8       | Female | 51  | Incidental (biliary colic) | Centro-parenchymal | Type III              | 6                     | Total splenectomy                                        | none                                                                   |

Table 2
average hospital stay for total splenectomy and spleen preserving modalities

| Surgical technique       | The average hospital stay (day) |
|--------------------------|---------------------------------|
| Total splenectomy        | 6.25                            |
| Conservative methods     | 9                               |
**Figure 1**

CT scan sagittal view showing a splenic hydatid cyst of 16cm diameter