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Surgical Management of Splenic Hydatidosis

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Abstract

Purpose. This study was conducted to evaluate the surgical management of splenic hydatidosis in an area where the disease is endemic.

Method. Over a period of 16 years, 26 patients with splenic hydatidosis underwent surgery in our department. Preoperative investigations included plain abdominal X-ray, serology, ultrasonography, and computed tomography.

Results. In our series, splenic hydatidosis represented 3.5% of total abdominal hydatidosis. The majority of patients presented with abdominal discomfort and palpable swelling in the left hypochondrium. Twenty-two patients were operated on electively and 4 as emergency cases, including 2 with ruptured hydatids and 2 with infected hydatids of the spleen. Isolated splenic hydatid cysts were present in 21 patients, associated liver hydatid cysts in 4, and diffuse abdominal hydatidosis in 1 patient. All patients underwent splenectomy except for one patient who initially had partial splenectomy for a lower polar cyst. This patient also ended up undergoing a splenectomy for postoperative hemorrhage. One patient died on the sixth postoperative day (mortality rate 3.8%) as a result of multiorgan failure, and 4 developed minor complications (morbidity 15.4%).

Conclusion. Splenic hydatidosis, although rare, is the third most common type of hydatidosis after liver and lung hydatidosis. This entity should thus be kept in mind when encountering a splenic cyst especially in areas where the disease is endemic. A splenectomy remains the treatment of choice because it demonstrates low morbidity and mortality rates.

Key words Splenic hydatidosis · Surgical management

Introduction

Hydatid disease primarily involves the liver, followed by the lung, and has been rarely found in almost all organs of the human body. The spleen is one of the rare sites involved in this zoonotic disease. After its first report by Bertelot in 1970,[1] various studies published so far report a mean incidence of 3% in the case of splenic hydatidosis, thus placing it third after liver and lung involvement.[2,3]

The majority of the reports of this rare clinical entity are based on a small number of cases or even sporadic cases.[4] This report which is one of the largest so far consists of 26 patients operated on for splenic hydatidosis with a discussion regarding the etiopathogenesis, presentation, diagnosis, and surgical treatment.

Material and Methods

Twenty-six patients with splenic hydatidosis were treated surgically in the Department of Surgery of Sher-i-Kashmir Institute of Medical Sciences, Srinagar, Kashmir, India, from January 1983 to June 1999. During this period a total of 751 patients with abdominal hydatid disease were operated on in the department. Splenic hydatidosis represents 3.5% of all abdominal hydatidosis-treated cases. The patients included 14 male and 12 female patients, with a mean age of 41 years (range 24–60) (Table 1). The first six patients (1–6) of this series have already been reported[5] and the inclusion of these cases in present series represents the total experience of splenic hydatidosis treated at our institution. This series also includes a case report (patient 13),[6] which was previously published because of its unique presentation.

Preoperative investigations included a plain abdominal X-ray, ultrasonography (US), computed tomography (CT), and immunological tests, i.e., Casoni's, indirect hemagglutination test (IHA), and an enzyme-linked immunosorbent assay (ELISA).

A left subcostal incision was made in cases with isolated splenic hydatid cysts and a midline incision in

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Table 1. Clinical data of 26 patients with splenic hydatidosis

Patient no.	Age/ (years) sex	Symptomatology	Associated disease	Diagnostic studies	Immunological test	Splenic cyst size (cm)	Operative findings	Surgery done	Complications	Results
1.	40/M	Mass LHC	—	Abd Xr US	Casoni's +	10 × 12	Cyst in upper part of spleen adherent to diaphragm	Splenectomy	—	Recovery
2.	45/M	Vague pain LHC Swelling LHC	—	Ab Xr US	Casoni's + IHA +	8 × 10	Cyst on medial aspect of spleen near hilum adherent to pancreas descending colon, and transverse colon	Splenectomy with excision of pancreatic tail	—	Recovery
3.	28/M	Palpable mass LHC		Ab Xr	Casoni's − IHA +		Mobile spleen with hydatid cyst	Splenectomy	—	Recovery
4.	38/F	Vague pain LHC palpable masses LHC and RHC	Liver, hydatid	Ab Xr US	Casoni's + IHA +		Cyst in spleen, adherent to diaphragm, pancreas omentum. Another cyst in posterior superior surface, surface of (right) lobe of liver	Splenectomy with enucleation and drainage of liver cyst. Rent in diaphragm closed and an under-water seal kept in	—	Recovery
5.	30/F	Vague pain LHC Palpable mass LHC	—	Ab Xr US	Casoni's IHA		Cyst in spleen	Splenectomy	—	Recovery
6.	35/F	Vague diffuse pain Abdominal multiple swellings, palpable	Liver, retroperitoneum, pelvis	Ab Xr US	Casoni's − IHA +	6 × 8 10 × 12 5 × 8	Cyst in right lobe of liver, spleen, pelvis, and retroperitoneum	Splenectomy with excision of cysts from liver and pelvis	—	Recover
7.	38/F	Fever, abdominal pain, palpable spleen		Ab Xr, US, CT	Serology −	15 × 13 × 10	Infected hydatid cysts with adhesion with diaphragm and parietes	Splenectomy	Wound infection	Recovery
8.	24/F	Vague abdominal pain, palpable mass	Liver hydatid	Ab Xr	Serology +	7 × 5	Liver hydatid 12 × 10, splenic hydatid 7 × 5	Splenectomy and enucleation of liver hydatid	—	Recovery
9.	21/M	Palpable mass in left hypochondrium	—	Ab Xr, US	Serology +	12 × 10	Splenic cyst involving lower pole	Partial splenectomy then reexplored splenectomy	Bleeding	Recovery
10.	32/M	Severe abdominal pain, shock	—	US			Ruptured hydatid spleen with laminated membrane and daughter cysts in peritoneal cavity	Splenectomy with thorough peritoneal lavage	MOF	Died

Table 1. *Continued*

Patient no.	Age/ (years) sex	Symptomatology	Associated disease	Diagnostic studies	Immunological test	Splenic cyst size (cm)	Operative findings	Surgery done	Complications	Results
11.	45/F	Dyspepsis, pain RHC, palpable mass LHC	Cholelithiasis	Ab Xr, US	Serology +	8 × 7	Cholelithiasis with splenic polar cyst	Cholecystectomy with splenectomy	—	Recovery
12.	54/M	Abdominal discomfort, palpable mass RHL, liver hydatid	5 years back operation for liver hydatid	Ab Xr, US	Serology +	14 × 12	Cyst involving whole of spleen adhesion with parietes	Splenectomy	—	Recovery
13.	37/M	Abdominal discomfort, palpable mass RHC and LHC	Liver hydatid	Ab Xr, US	Serology +	8 × 7	Hydatid cyst of (right) lobe, liver, and spleen	Splenectomy, and enucleation and drainage of liver hydatid	—	Recovery
14.	25/F	Abdominal pain and fever, vomiting, tender spleen		Ab Xr, US/CT	Serology −	12 × 10 × 8	Infected hydatid spleen	Splenectomy	Pnenumonitis	Recovery
15.	38/F	Abdominal discomfort, palpable mass		Ab Xr, US	Serology +	12 × 10	Hydatid spleen body	Splenectomy	—	Recovery
16.	31/M	Abdominal discomfort, mass RHC	Hydatid liver	Ab Xr, US/CT	Serology +	8 × 6	Hydatid (right) lobe of liver and spleen	Splenectomy and enucleation, discharge of liver hydatid	—	Recovery
17.	60/M	abdominal discomfort, mass LHC	—	Ab Xr, US	Serology +	14 × 12	Splenic hydatid body and lower pole	Splenectomy	—	Recovery
18.	56/F	Abdominal discomfort, mass LHC	—	Ab Xr, US	Serology +	12 × 10	Hydatid spleen with adhesion colon	Splenectomy	—	Recovery
19.	35/M	Abdominal discomfort, mass LHC	—	Ab Xr, US	Serology +	14 × 12	Hydatid spleen with adhesions with parietes diaphragm	Splenectomy	—	Recovery
20.	48/M	Abdominal discomfort, mass LHC	—	Ab Xr, US	Serology +	14 × 12	Spleen hydatid	Splenectomy	—	
21.	55/F	Abdominal discomfort, mass LHC	—	Ab Xr, US	Serology +	16 × 14	Huge splenic hydatid, adhesion to parietes	Splenectomy	Pnenumonitis	Recovery
22.	29/F	Abdominal discomfort, mass palpable	—	Ab Xr, US	Serology +	12 × 10	Splenic hydatid	Splenectomy	—	Recovery
23.	55/M	Bullet injury left lower chest	Hydatid liver	X-ray chest			Ruptured splenic hydatid with blood and daughter cysts, enucleation of liver hydatid	Splenectomy with lavage	Left pleural effusion	Recovery
*24.	59/M	Mass LHC	—	Ab Xr, US	Serology +	13 × 11	Splenic hydatid	Splenectomy		Recovery
25.	36/F	Mass LHC	—	Ab Xr, US	Serology +	12 × 10	Splenic hydatid	Splenectomy		Recovery
26.	42/M	Pain RHC	Gallstone	Ab Xr, US	Serology +	9 × 7	Cholelithiasis with splenic hydatid	Cholecystectomy with splenectomy		Recovery

RHC, right hypochondrium; LHC, left hypochondrium; Ab Xr, abdominal X-ray; US, ultrasonography; CT, computed tomography; MOF, multiple organ failure

those with associated liver hydatids, cholelithiasis, and emergency patients. A splenectomy was performed in all except one young patient (patient 9) with a lower polar hydatid who underwent a partial splenectomy. All splenectomy specimens were sent for a histopathological examination (HPE) to confirm the diagnosis of the disease.

All of the patients were given a pneumococcal vaccine and long-term penicillin therapy. To reduce the recurrence in nine patients which included three who underwent an emergency operation and six who had associated liver, pelvic, and retroperitoneal hydatids, 400 mg of tab albendazole was given twice a day for 4 weeks. The therapy was repeated after a 2-week interval for a total of three cycles. During the therapy the liver functions were monitored.

All patients were followed up yearly, and clinical examinations, liver function tests, immunological tests, and US were performed.

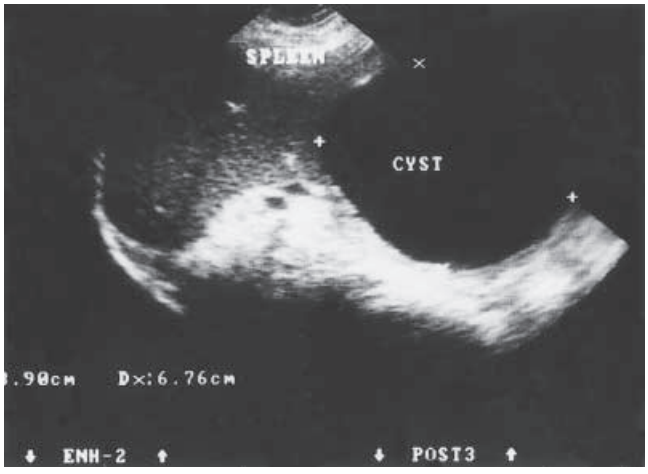

Fig. 1. Ultrasonography showing a splenic hydatid cyst

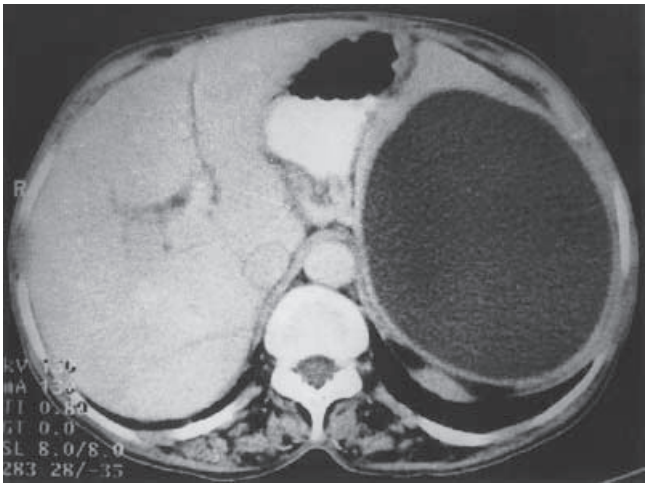

Fig. 2. Computed tomography scan showing a splenic hydatid cyst

Results

Out of the 26 patients a preoperative diagnosis was made in 22 (84.6%), and the diagnosis was confirmed at surgery. A plain abdominal X-ray was helpful in showing the elevated left hemidiaphragm in 7 patients and a calcified cyst in 4. Ultrasonography (Fig. 1) was successful in diagnosing the cystic lesions in all of them, while the serology findings were only positive in only 20. A CT scan which was done in 3 patients showed a cystic lesion in all 3 (Fig. 2). All 22 elective patients had unilocular cysts on US and surgery. The size of the splenic cyst varied from 5 to 15 cm. All except one patient had a splenectomy as the procedure of choice. Patient 2 had a cyst which adhered to the pancreatic tail which was thus excised along with the cyst. The splenectomy was laborious in 4 patients due to adhesions and one of them (patient 4) had on iatrogenic diaphragmatic tear, which was repaired, and the pleural cavity drained with chest tube.

Twenty-two patients were operated on electively while 4 had emergency operations (Table 1). The spleen was the only site of the hydatid in 19 patients (73%). An associated liver hydatid was present in 6 patients including one (patient 6) with pelvic and retroperitoneal hydatids, and gallstones were present in 2 patients. The most common symptoms among those operated on electively were vague abdominal pain or discomfort and/or a palpable mass in the left hypochondrium. Two patients had a history of biliary colic and the presence of splenic hydatidosis was incidentally found during US.

Two of the four patients who underwent emergency operations (patients 7 and 14) presented with toxemia and tender splenomegaly, and were clinically diagnosed to have splenic abscesses. On laparotomy, both patients proved to have infected hydatid cysts of the spleen. Patient 10, who had history of sudden severe abdominal pain, presented with shock and rigid abdomen. Ultrasonography revealed free fluid in the peritoneal cavity with an altered echogenicity in the lower pole of spleen. A clinical diagnosis of a ruptured spleen was made. On exploration a ruptured hydatid cyst was found in the spleen, with daughter cysts in the peritoneal cavity. This patient went into multiorgan failure and died on the sixth postoperative day. The perioperative mortality rate was 3.8%. Patient 26,[6] who presented with a bullet injury in the left lower chest, was in shock and had tenderness and guarding in the left hypochondrium. A chest X-ray of this patient showed left-sided hemopneumothorax while abdominal paracentesis revealed hemoperitoneum. An emergency laparotomy was performed,

Fig. 3. Specimen of the spleen with a hydatid cyst

revealing multiple daughter cysts and blood in the peritoneal cavity. The spleen showed multiple parenchymal tears with a huge ruptured hydatid cyst. A splenectomy and a repair of the diaphragm with chest tube drainage were done.

Patient 9 was reexplored because of hemorrhage after a partial splenectomy and eventually the spleen had to be removed. Six patients (23%) (patients 4, 6, 8, 13, 16, 23) had associated liver hydatids which were treated simultaneously with enucleation and drainage, and patient 6 also had associated pelvic and retroperitoneal hydatids which were excised at the same time. Two patients (11 and 26) who had concomitant gallstones underwent a cholecystectomy. All of the resected specimens (Fig. 3) which were sent for HPE were confirmed to have the hydatid disease of the spleen in them (Fig. 4).

During the early postoperative period four patients developed minor complications including left pleural effusion, pneumonia, and wound infection, with a morbidity rate of 15.3% (Table 1).

The mean hospital stay was 8 days with a range of 6–15 days. After being discharged from the hospital, 5 patients were lost to the follow up and the remaining 20 were thereafter followed up yearly. The mean follow-up was 7 years with a range of 13 months to 16 years. One patient (patient 8) required a second operation for recurrent peritoneal hydatid 4 years after the initial surgery for a hydatid liver and spleen. None of the remaining patients demonstrated recurrence during the follow-up. No patient had any serious septic complications.

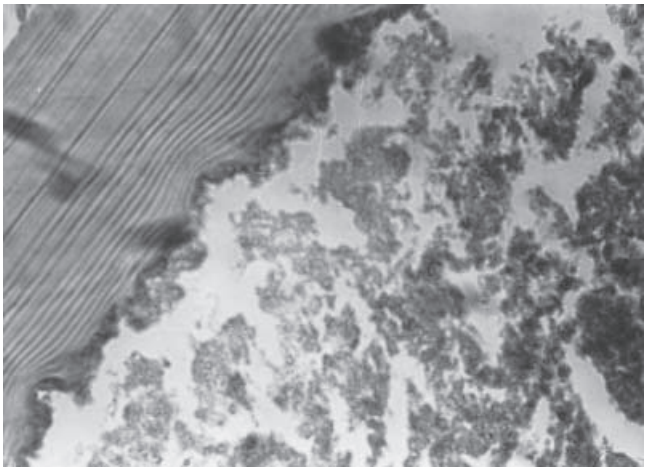

Fig. 4. Photomicrograph of the hydatid spleen showing splenic tissue with an adjacent laminated membrane of the hydatid cyst (H & E, ×400)

Discussion

The spleen is the third most common organ involved in hydatid disease after the liver and lung.[1] Its incidence in various reported series varies from 0.9% to 8%,[1,4,7] and in our series was 3.4%. Associated liver and lung involvement is seen in from 20% to 30% of all cases,[8] and in our series it was 23%.

Although various theories have been put forth regarding the etiopathogenesis of splenic hydatidosis, the first and most acceptable pathophysiological and pathologoanatomical study was published by Bourgeon et al. in 1960.[9] After crossing the two filters (i.e., liver and lung), the hexacanth embryo gets lodged in the periphery of the splenic capillaries. The incompressible mass of the growing cyst gradually crushes the segmentary vessels with extensive pericystic atrophy and the formation of regional necrotic zones. The process of aseptic inflammation of splenic parenchyma extends to the outside of the splenic capsule thus resulting in adhesion formation with the surrounding structures, such as the stomach, colon, and diaphragm. This situation may lead to the accidental rupture of the cyst into these adjacent organs or make a splenectomy very difficult, as was found in four of our patients. There have been several reports about a rupture of the cyst into the colon.[1,10,11]

The clinical presentation of the splenic hydatidosis is often vague. Most of the patients in our series presented

with mild abdominal discomfort, and a palpable mass was present in 80.7% of cases. Some of the patients presented with symptoms due to hydatid in other organs such as the liver[12–14] or due to other associated disease such as cholelithiasis, which was observed in two of our cases. Sometimes patients may present with complications such as an infection or rupture of the cyst into the peritoneal cavity, as was seen in four of our cases.

The preoperative diagnosis of splenic hydatidosis has become much easier thanks to the development of modern radiological imaging such as US and CT scan. Although similar sonographic and CT findings may be seen in other cystic lesions of the spleen, including splenic abscesses, epidermoid cysts, pseudocysts, or a cystic neoplasms of the spleen, when combined with immunological tests such as IHA, ELISA, and immunoelectrophoresis, a diagnosis of splenic hydatidosis can be successfully established in more than 90% of all cases.[15] In the presence of complications such as infections or rupture, immunological tests may either be helpful or not, and there may also be no time to perform such tests.

Regarding the surgical management of splenic hydatidosis, various techniques have been used by various authors. Conservative surgery such as a partial splenectomy, cyst enucleation, or deroofing of the cyst with omentoplasty or external drainage have been performed for superficial cysts, cysts localized to one pole of the spleen, or cysts that are unresectable due to extensive adhesions.[8,17–19] None of these conservative procedures are free of complications such as hemorrhage after a partial splenectomy or an infection of the residual cavity after deroofing the cysts. One of our patients had severe hemorrhage after a partial splenectomy and ultimately a complete splenectomy was done. We preferred splenectomy in all other cases, and low morbidity and mortality rates were obtained (15.3% and 3.8%, respectively). Splenic salvage may be justified both in children to avoid septic complications,[20,21] and in cases in which the cysts tightly adhere to the adjacent structures.[22] After a splenectomy prophylactic vaccination and a 2-year course of prophylactic penicillin was given to all of our patients, and none developed any septic complications. During the follow-up period only one patient (patient 8) developed a recurrent hydatid cyst in the pelvis, which needed surgical removal. This patient was again given a course of Albendazole.

We conclude that a splenectomy remains the procedure of choice in cases of splenic hydatidosis, since a complete cure of the splenic and perisplenic diseases can be achieved with low morbidity and mortality rates. In addition, postoperative Albendazole therapy has been proven to be effective in preventing a recurrence of hydatid disease.

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