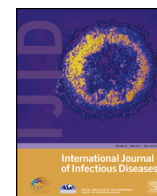


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Spleen-preserving surgery is effective for the treatment of spleen cystic echinococcosis



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SUMMARY

Objective: This study evaluated two surgical procedures, total splenectomy and spleen-preserving surgery, for the treatment of spleen cystic echinococcosis (CE).

Methods: A total of 21 patients who underwent surgery for removal of spleen CE were evaluated retrospectively. Patients were divided into two groups, those who received a total splenectomy ($n = 7$) and those who underwent spleen-preserving surgery ($n = 14$).

Results: Total splenectomy surgery took 127.1 ± 11.1 min and spleen-preserving surgery took 104.3 ± 25.3 min ($p < 0.05$). The length of hospital stay was the same for both patient groups (10.6 days on average). No patient suffered from recurrence during follow-up and all of the patients made a good recovery. No patient developed post-splenectomy sepsis or serious infectious complications. No patient suffered recurrence.

Conclusions: The spleen-saving approach is likely an effective method for the removal of CE spleen cysts given that it is a quick procedure and preserves the function of the spleen. Total splenectomy procedures may be performed particularly for larger, centrally located cysts.

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1. Introduction

Cystic hydatid disease, or cystic echinococcosis (CE), is an important parasitic zoonosis caused by the larval cyst stage of the dog tapeworm *Echinococcus granulosus*. This disease has impacts on both population health and animal production in high endemic areas, such as Central Asia, the Mediterranean countries, and South America.¹ Humans become infected by accidentally ingesting the eggs of the tapeworm.¹ The disease can develop in any organ, but most frequently affects the liver (60–70%) and the lungs (20–30%).² Isolated splenic involvement is quite rare.³

Splenectomy is advocated by many surgeons for the treatment of spleen CE due to the minimal risk of recurrence,^{4–6} although splenectomy is associated with sepsis between

0.18 and 0.42 person-years.⁷ Spleen-preserving treatment is considered technically simpler and safer to perform compared with the splenectomy method. However, this procedure has some postoperative complications such as a residual cavity and recurrence.^{8,9} There is no consensus regarding the selection of surgical procedure and most reports on spleen cysts have been case reports or clinical series with small numbers of cases.^{5,8–11} In this study, we found spleen-preserving surgery to be effective and safe for the surgical removal of cystic hydatid cysts in the spleen.

2. Materials and methods

From January 2002 to December 2012, a total 3003 patients underwent surgery for the removal of hydatid cysts in our hospital (unpublished data). Among these patients, 21 (0.7%) had undergone treatment for isolated spleen hydatidosis; these patients were reviewed retrospectively in the present study. Seven of the 21 patients had undergone a total splenectomy, while 14 had

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undergone spleen-preserving surgical treatment. Patient age, gender, symptoms, surgical procedure performed, postoperative complications, mean operation time, and mean hospital stay were recorded. The preoperative diagnosis was established by history, physical examination, abdominal ultrasonography (US) or computed tomography (CT), and serological tests. Cyst size and number were measured by US and CT to determine the location, number, and morphology.

All patients underwent laparotomy via a left subcostal incision. Spleen-preserving treatment included simple drainage, partial cystectomy plus omentopexy, cystectomy plus external drainage, and segmental splenectomy. At the end of the procedure, a drain was placed in the left subphrenic area in all cases.

After surgical removal of cystic hydatid cysts, all patients took 10 mg/kg/day albendazole orally for 21 days. During this period, liver function was closely monitored.

Patients were followed up every 3 months for at least 1 year using a questionnaire, and ultrasound imaging was performed every 6 months to evaluate the recovery status.

2.1. Statistical analysis

All data were analyzed using SPSS version 17.0 (SPSS Inc., Chicago, IL, USA). The Student's *t*-test and non-parametric test (Mann–Whitney *U*-test) were used for comparisons of patient age, size of the cyst, and duration of surgery for the two surgical procedures. A *p*-value of less than 0.05 was considered statistically significant.

3. Results

A total 21 patients with solitary spleen hydatidosis underwent surgery in our hospital. For 19 patients this was the first surgery; two patients had previously undergone an operation at another hospital. In terms of symptoms, 14 patients experienced left upper abdominal pain, while seven patients were without symptoms and were diagnosed incidentally during a medical checkup. US and CT were performed for all patients preoperatively and only one patient was misdiagnosed with a hematoma. A rapid dot immunogold filtration assay (DIGFA) was used for all patients and 17 (80.9%) were seropositive. The demographic and clinical characteristics of the 21 patients are presented in Table 1.

A total of 14 patients (nine males and five females) underwent spleen-preserving surgery treatment, including two patients who had a partial splenectomy. This group of patients had a mean age of 42.1 ± 15.6 years (range 21–62 years). The total splenectomy group comprised seven patients (one male and six females) with a mean age of 36.4 ± 6.9 years (range 29–48 years). Age was not significantly different between the two groups ($p > 0.05$). In the spleen-preserving group, solitary cyst formation was identified in 13 patients and multiple cysts were found in only one patient. The cysts were located in the upper pole of the spleen in five patients, in the lower pole in seven patients, and in middle pole in two patients; mean size was 7.5 ± 1.9 cm (range 5–10 cm). In the total splenectomy group, all patients had a solitary cyst. The cysts were located in the hilus in five

patients, in the lower pole in one patient, and in the upper pole of the spleen in one patient; mean size was 8.10 ± 2.34 cm (range 5–11 cm). The size of the cysts was not significantly different between the total splenectomy and spleen-preserving groups ($p > 0.05$). There was only one patient with a cyst infection in the splenectomy group and no cyst complications occurred in the spleen-preserving group. The duration of surgery was 127.1 ± 11.1 min for the total splenectomy group and 104.3 ± 25.3 min for the spleen-preserving surgery group ($p < 0.05$). The length of hospital stay was the same for the patients in the two groups (10.6 days on average).

A total of five patients (23.8%) had complications during their hospital stay, including two (2/7) in the total splenectomy group and three (3/14) in the spleen-preserving group (Table 2). Of the two cases treated with a total splenectomy, one had a left pleural effusion on the fifth postoperative day and one patient had an increased platelet count to $1000 \times 10^9/L$ on the 20th postoperative day. His platelet count returned to normal after taking low-dose aspirin for 3 weeks (300 mg daily). Of the three cases with complications in the spleen-preserving group, one had undergone a partial cystectomy and drainage and had an adhesive partial small-bowel obstruction on the seventh postoperative day; this resolved with effective gastric compression and medical treatment. Another patient had pneumonia secondary to left pleural effusion and the final patient suffered from a residual cavity effusion.

All of the cases were followed-up for a period of 1 to 2 years with US every 6 months. No postoperative recurrence of hydatid cysts was detected in these patients by US. No patient developed post-splenectomy sepsis or serious infectious complications.

4. Discussion

Splenic echinococcosis is rare, about 0.5–4% of total CE cases.^{8,12} Over the 11-year study period, 3003 patients underwent surgical treatment of CE in our hospital, but only 21 (0.7%) had an isolated spleen CE infection. Splenic hydatid cysts generally develop by means of systemic dissemination or intraperitoneal spread from a ruptured liver cyst, and isolated splenic involvement is very uncommon.² However, in our study, we had two relapse cases who had undergone surgical treatment for splenic echinococcosis at local hospitals. The cysts were likely due to a secondary infection from the primary splenic hydatid cysts. There was no evidence that the primary splenic cysts had arisen from ruptured cysts in other organs in the patients involved.

Clinical manifestations of splenic hydatid disease are usually mild and non-specific.^{4,9} Symptoms occur when complications are present, such as impingement on neighboring organs, secondary infection, and intracystic hemorrhage. A dramatic clinical presentation can be seen when a cyst ruptures into the peritoneal cavity, with or without an anaphylactic reaction.¹³ In our study, 14 (66.7%) patients experienced left upper quadrant pain and seven cases were asymptomatic and diagnosed incidentally during investigations for other reasons.

Table 1
Demographic and clinical characteristics of the study patients

Surgical features	Splenectomy, n (%)	Spleen-preserving treatment, n (%)	Total
Number of patients	7 (33.3)	14 (67.7)	21
Male	1 (10)	9 (90)	10
Female	6 (54.5)	5 (45.5)	11
No symptoms	2 (28.6)	5 (71.4)	7
Left upper abdominal pain	5 (35.7)	9 (64.3)	14
Serological tests positive	7	10	17

Table 2
Description of the postoperative complications

Surgical features	Splenectomy	Spleen-preserving treatment
Pleural effusion	1	0
High platelet syndrome	1	0
Adhesive partial small-bowel obstruction	0	1
Pneumonia	0	1
Residual cavity effusion	0	1
Recurrence	0	0
Total	2	3

In the majority of cases, a definitive diagnosis can be established on the basis of a combination of imaging techniques and serological techniques. US and CT evaluations are the most valuable diagnostic tools for detecting hydatid cysts,¹⁴ and are not only useful for determining the location of the cyst in the spleen and its relationship with the surrounding organs, but also for planning the optimal approach, especially when minimally invasive surgery is being considered. In our group, all of the patients had US and CT scans. One patient was misdiagnosed with a hematoma. In this patient the CT scan showed multiple irregular high density shadows in the spleen, which was considered to indicate hematoma of the spleen, while the hydatid cyst was confirmed intraoperatively.

Serological tests are frequently used in the diagnosis of hydatid disease and the follow-up of recurrent cases. The Casoni and Weinberg tests are not used in our hospital for the diagnosis of cystic echinococcosis due to the high false-positive rate.¹⁵ A rapid DIGFA has been used to diagnose human echinococcosis in our hospital, which has the advantages of rapidity, convenience, and the ability to provide an initial diagnosis.¹⁶ In our study, all patients had their serum tested with the DIGFA and only four patients (19.0%) had negative results; this is similar to previously reported data.¹⁶

To date, surgery remains the cornerstone of treatment to cure CE, including splenic echinococcosis. Reports of splenectomy have indicated the advantages in terms of reducing recurrence and fewer complications.^{4–6,11} However, splenectomy has been observed to have a substantial case fatality rate, reported at 50% to 90%.^{17,18} In our study the total splenectomy procedure was used only for those patients with a particularly large (occupying more than half of the spleen) or centrally located cyst. In the total splenectomy group of seven patients, three had cysts larger than 10 cm in size and four patients had cysts located in the central spleen parenchyma or close to the hilum of the spleen. None of the seven patients developed postoperative sepsis.

In our center, we prefer spleen-preserving procedures for the removal of splenic CE cysts in most cases, especially for younger patients. During the operation, we are likely to remove the pericystic membrane extending outside the spleen as much as possible after removal of the inner cyst, leaving only the outer layer attached to the spleen parenchyma. This procedure can minimize the incidence of recurrence and residual cavity complications.¹⁹ Only one patient in this study suffered a residual cavity effusion and no patient developed postoperative recurrence.

In conclusion, the involvement of the spleen in hydatid disease is rare, and considering its low complication rate and benign

nature, spleen-saving surgical approaches should be performed in most cases. This procedure is safe and preserves the immune function of the spleen. However, total splenectomy procedures should be performed to remove large, centrally located cysts.

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References

- McManus DP, Zhang W, Li J, Bartley PB. Echinococcosis. *Lancet* 2003;**362**:1295–1304.
- Aydin U, Yazici P, Onen Z, Ozsoy M, Zeytinlu M, Kilic M, et al. The optimal treatment of hydatid cyst of the liver: radical surgery with a significant reduced risk of recurrence. *Turk J Gastroenterol* 2008;**19**:33–9.
- Karakaya K. Primary hydatid cyst disease of the spleen; a rare entity: report of two cases. *Trakya Univ Tip Fak Derg* 2007;**24**:256–8.
- Durgun V, Kapan S, Kapan M, Karabicak I, Aydogan F, Goksoy E. Primary splenic hydatidosis. *Dis Surg* 2003;**20**:36–41.
- Safioleas M, Misiakos E, Manti C. Surgical treatment for splenic hydatidosis. *World J Surg* 1997;**21**:374–8.
- Ibrarullah M, Sreenivasa D, Sriram P, Haragopal M. Hydatid cyst of spleen. *Trop Gastroenterol* 1999;**20**:55–6.
- Kyaw MH, Holmes EM, Toolis F, Wayne B, Chalmers J, Jones IG, et al. Evaluation of severe infection and survival after splenectomy. *Am J Med* 2006;**119**:276.e1–7.
- Atmatzidis K, Papaziogas B, Mirelis C, Pavlidis T, Papaziogas T. Splenectomy versus spleen-preserving surgery for splenic echinococcosis. *Dig Surg* 2003;**20**:527–31.
- Culafić DM, Kerkez MD, Mijac DD, Lekić NS, Ranković VI, Lekić DD, et al. Splenic cystic echinococcosis: clinical manifestations and treatment. *Scand J Gastroenterol* 2010;**45**:186–90.
- Arikanoglu Z, Taskesen F, Gumus H, Onder A, Aliosmanoglu I, Gul M, et al. Selecting a surgical modality to treat a splenic hydatid cyst: total splenectomy or spleen-saving surgery? *J Gastrointest Surg* 2012;**16**:1189–93.
- Rahmani SH, Mohammadi Tofigh A. Spleen-preserving surgery versus splenectomy for splenic hydatid cyst: ten years experience. *Shiraz E Medical Journal* 2008;**9**:82–7.
- Rodríguez-Leal GA, Morán-Villota S, Milke-García Mdel P. Splenic hydatidosis: a rare differential diagnosis in a cystic lesion of the spleen. *Rev Gastroenterol Mex* 2007;**72**:122–5.
- Polat FR, Sezer A, Polat S. Laparoscopic treatment of hydatid cyst of the spleen without splenectomy: report of a case. *Surg Laparosc Endosc Percutan Tech* 2009;**19**:e215–6.
- Franquet T, Montes M, Lecumberri FJ, Esparza J, Bescos JM. Hydatid disease of the spleen: imaging findings in nine patients. *AJR Am J Roentgenol* 1990;**154**:525–8.
- Zhang W, Li J, McManus DP. Concepts in immunology and diagnosis of hydatid disease. *Clin Microbiol Rev* 2003;**16**:18–36.
- Feng X, Wen H, Zhang Z. Dot immunogold filtration assay (DIGFA) with multiple native antigens for rapid serodiagnosis of human cystic and alveolar echinococcosis. *Acta Trop* 2010;**113**:114–20.
- Cullingford G, Watkins D, Watts A, Mallon D. Severe late postsplenectomy infection. *Br J Surg* 1991;**78**:716–21.
- Brigden M. Overwhelming post-splenectomy infection—still a problem. *West J Med* 1992;**157**:440–3.
- Wen H, Aji T, Shao YM. Diagnosis and management against the complications of human cystic echinococcosis. *Front Med China* 2010;**4**:394–8.