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## **Spleen-Preserving Surgery Versus Splenectomy for Splenic Hydatid Cyst: Ten Years Experience.**

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### **Abstract:**

**Background:** Although rare, the spleen is the third most common location of hydatid disease after liver and lung. The aim of this study was to analyze the long-term outcome of surgical treatment of patients with splenic echinococcosis comparing spleen-preserving surgery with splenectomy.

**Materials and Methods:** During a period of 10 years (1996-2006), 20 patients with splenic echinococcosis entered our study as test and control groups. In our series the spleen was the only organ involved. Splenectomy was performed in the control group and spleen preserving drainage of cyst in test group.

**Results:** There was no significant difference between the splenectomy and spleen-preserving groups concerning median hospital stay, postoperative complication rate and recurrence. ( $p < 0.5$ ) The median follow-up in 15 patients was 52 (range 6-300) months.

**Conclusion:** In the present series it was possible to preserve the spleen affected by hydatid cyst without significant increase of recurrent echinococcosis.

**Key Words:** spleen, echinococcus, cyst, splenectomy, drainage.

### **Introduction:**

Splenic echinococcosis or hydatid cyst is a rare disease even in endemic regions such as Iran<sup>(1,2,3)</sup>. The most commonly affected organ is the liver (70%) followed by the lung (15-20%), spleen (3-8%) and kidney (2-4%), although hydatid cysts has been described in almost every organ of the human body including the subcutaneous tissue, muscles, brain and the heart. Only rather small clinical series or case reports have addressed the issue of splenic echinococcosis<sup>(1-7)</sup>. It is unclear whether total splenectomy should be used in all cases of splenic echinococcosis or if preservation of at least part of the spleen can be performed without a high risk of recurrence. This study describes a series of 20 patients with splenic echinococcosis treated in a single unit in which the spleen was preserved in 10 cases and followed for 52 months.

### **Materials and Methods:**

In the context of a randomized clinical trial 20 patients with splenic echinococcosis which the spleen was the only affected organ entered the study. There were 12 males and 8 females with a median age of 52 (range 32-78) years. The preoperative diagnosis was established using CT, ultrasonography or MRI as imaging techniques and specific immunological tests (ELISA) have been applied replacing the old diagnostic methods.

All patients underwent laparotomy via a left subcostal or a mid-line incision. Randomization was done during the operation by the operating surgeon who determined the spleen and the cyst. In centrally located or hilar cysts splenectomy was done. Under the circumstances where the cyst was not located centrally or in the hilum, then randomization would take place and the patients would enter the study as test (spleen preserving) and control (splenectomy) groups. In the spleen preserving group after enucleating of cysts and extraction of the germinal layer, the cavity was irrigated by Silver-Nitrate soaked gauze and the cavity was drained externally. We kept the drain in place until the drainage volume reached under 100 ml<sup>3</sup>.

In all cases the surgical field was protected by Silver-Nitrate soaked gauzes.

All resected specimens were examined histologically and the diagnosis confirmed. Patients that underwent splenectomy received pneumococcal vaccine in the immediate postoperative period. The patients were followed yearly with physical examination, serological tests and ultrasonography or CT for the detection of recurrence. The splenectomy group was compared to the spleen-preserving group for duration of hospital stay, postoperative complications and recurrence. Statistical analyses were performed using the  $\chi^2$  and Mann-Whitney U tests. Statistical significance was assumed if  $p < 0.05$ . The study protocol was approved by Institutional Review Board of Shahid Beheshti University of Medical

Sciences (Tehran, Iran). Patient consent was obtained in all cases.

### Results:

The main symptomatology included mild discomfort or pain in the left hypochondrium (15 patients), palpable mass (9 patients), postprandial nausea and vomiting (5 patients) and weight loss (2 patients). 13 patients had mild to severe eosinophilia at the time of diagnosis. ELISA test was positive in 7/8 patients. The diagnosis was established preoperatively in all cases.

The spleen was the only affected organ in our series. In 18 patients there was a solitary echinococcal cyst of the spleen, while the remaining patient had two cysts. Pus was present in two cysts. In the first patient, bacteriology revealed *Staphylococcus aureus*, while in the other case the culture was negative. The median size of the cysts was 10.5 × 7.7 × 6.9 cm. The weight of the resected spleen with the unruptured cyst ranged between 1 and 1.5 kg. In 15 patients the wall of the cyst was partially calcified and 11 cysts contained at least one daughter cyst.

The surgical procedures were total splenectomy in 10 patients and cyst enucleation and tube drainage of the cavity in other 10 patients. The diagnosis of echinococcal cyst was confirmed by histopathology in all of the resected specimens.

The median hospital stay was 16 (range 14-41) days. The difference between the splenectomy and spleen-preserving groups (15.3 vs. 16.4 days respectively) was not

statistically significant. ( $p < 0.5$ ). None of our patients received pre or post operative albendazole. The drain was extracted after 10 days (range 8-20 days) in the spleen preserving group.

Fifteen patients were followed for a median of 52 (range 6-300) months and the other 5 patients were non-compliant, of whom 3 belonged to the splenectomy group and 2 to the spleen-preserving group. No patient developed post-splenectomy sepsis or serious infectious complications. There was no recurrence of the disease in our series.

### Discussion:

The main interest of our series lies in the relatively long period of postoperative follow-up. The development of echinococcal cysts in the spleen is uncommon because hexacanth embryos are usually trapped in the liver (first Lemman's filter) and/or lung (second Lemman's filter) but will be trapped in the splenic capillaries once in the systemic circulation<sup>(8)</sup>. Splenic echinococcosis may also arise by retrograde spread from the liver to the spleen via the hepatic portal and splenic veins in portal hypertension. The spleen may also be affected by rupture of an hepatic echinococcal cyst into the peritoneal cavity<sup>(1,5)</sup>. As the hydatid cyst increases in size it may lead to compression of the hilar vessels of the spleen, resulting in pericystic splenic atrophy. Eventually the cyst may completely replace the splenic parenchyma. Chronic pericystic inflammation may cause adhesion with adjacent organs or even fistulization between the cyst and nearby or-

gans such as the stomach, pancreas, left colon, left kidney, or bronchus<sup>(3,4,9)</sup>. Secondary infection of splenic cysts is rare and usually occurs by haematogenous spread. In the present series only 1 (5%) splenic cyst was infected.

Echinococcal cysts of the spleen usually grow very slowly (approx. 2-3 cm each year)<sup>(1,2,5,7)</sup>. Patients may be asymptomatic for 5-20 years before diagnosis. The median size of the echinococcal cysts in the present series was 10.5 × 7.7 × 6.9 cm, consistent with other series<sup>(1,2,5,6)</sup>. The cyst is usually single without daughter cysts and partially calcified (87% in this series). The symptomatology is usually mild and is mainly caused due to pressure of adjacent viscera or the presence of complications. The patients usually complain for mild discomfort or pain in the left hypochondrium. Severe pain may occur following infection or intraperitoneal rupture of the cyst. Physical examination may reveal either a large palpable spleen or a hard, round and smooth mass, which follows the respiratory movements of the diaphragm. Nine of the patients in this series presented with palpable mass in the left hypochondrium or epigastrium.

Before the introduction of the cross-sectional imaging techniques<sup>(10,11)</sup>, the preoperative diagnosis was established using the presence of eosinophilia, plain films of the abdomen and chest, Casoni's skin test, Weinberg's indirect haemagglutination test and scintiscanning of the spleen. Eosinophilia is the least reliable method being present in only 20-30% of

the patients<sup>(12)</sup>. Plain X-ray of the abdomen may show a soft tissue mass with peripheral calcification in the left upper quadrant or displacement of the stomach, left kidney and left colon flexure. Plain X-ray of the chest may reveal an elevated left hemidiaphragm or pleural effusion. Casoni's and Weinberg's tests have been replaced by enzyme-linked immunosorbent and immunoelectrophoresis assays which have significantly higher sensitivities (95-99%)<sup>(12)</sup>. Splenic echinococcosis needs to be differentiated from non-parasitic cysts, epidermoid cysts, haemangiomas, sarcomas, pseudocysts and tumours of the diaphragm, stomach, colon, left kidney or pancreas. The diagnosis is made by the history, physical examination, the presence of peripheral calcification or daughter cysts within a large cystic lesion or coexistent cystic lesions in the liver or other organs<sup>(13)</sup>.

Total splenectomy is advocated by the majority of the surgeons, since it provides a minimal risk of recurrence<sup>(1,2,5,7,9)</sup>. Splenectomy however is associated with sepsis related deaths in 1.9% in adults and 4% in children<sup>(14)</sup>. None of the 10 patients in our series who underwent total splenectomy developed postoperative sepsis. Thus conservative surgical procedures have been increasingly proposed for children including partial splenectomy, enucleation, de-roofing with omentoplasty, internal drainage with cystojejunal anastomosis or external drainage<sup>(18)</sup>. This series showed that the recurrence rate after total splenectomy did not differ significantly compared to

spleen-preserving surgery and the complication rate and hospital stay were also comparable between the two groups. An alternative to surgery is percutaneous drainage and administration of sclerosing agents such as alcohol 96% under sonographic guidance <sup>(19)</sup>. Laparoscopic or laparoscopically assisted splenectomy has also been successfully used for echinococcal disease of the spleen <sup>(20)</sup>. Medical treatment with albendazole has a place as an adjuvant therapy in selected cases after surgical treatment in order to reduce the incidence of recurrence, especially after inadvertent intraoperative rupture of the cyst or the presence of disseminated intra-abdominal disease <sup>(21)</sup> Indications for albendazole prescription after surgery at the time of our study design were:

- 1-concurrent pulmonary or disseminated hydatid disease
- 2-multiple recurrent or inaccessible hydatid liver cysts
- 3-spontaneous intraperitoneal or intrathoracic rupture
- 4-accidental contamination of the peritoneum with cyst contents during operative cystectomy
- 5-when the patient is unfit for the definite surgical procedure.

In conclusion, splenic echinococcosis is a rare but relatively benign disease. The selection of the surgical technique should be made according to the size of the cyst and its location. Splenic preservation should be undertaken if possible.

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