

# Splenic recurrence of liver hydatid cyst and spleen preserving therapy

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## ABSTRACT

Hydatid cyst disease remains a considerable public health problem, especially in pastoral and farming regions. Although the spleen is the third most commonly affected organ after the liver and lungs, splenic hydatid cyst is an uncommon entity even in areas that are endemic for echinococcosis. The recurrence rate after surgical therapy of liver hydatid cyst is reported as 6.8–22.3 percent. Recurrences most frequently occur in the liver. Extrahepatic recurrences occur in the lung or peritoneum and the serosa of the abdominal organs. Splenic recurrence of liver hydatid cyst has not previously been reported. The most common surgical therapy is splenectomy, and the other option is spleen preserving surgery. We report the first case of recurrent splenic hydatid cyst in the spleen and liver synchronously after surgical therapy for liver hydatid disease. The patient was treated with liver resection and spleen preserving surgery.

**Keywords:** hydatid disease, recurrence, spleen, spleen preserving surgery

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## INTRODUCTION

Hydatid disease is a zoonotic infection caused by the larval stage of the genus *Echinococcus*, also known as *E. granulosus*, or less frequently, *E. multilocularis*, which occurs worldwide. However, it remains a considerable public health problem in several Mediterranean countries. Hydatid cyst disease has been described in almost every organ or tissue of the human body, but approximately 70% of the cysts are situated in the liver, followed by the lung (15%–47%). The disease is less frequently found in the kidneys, spleen, bile ducts, mesentery, brain and soft tissues.<sup>(1,2)</sup> Although the spleen is the third most commonly afflicted organ after the liver and the lung, splenic hydatid cyst is an uncommon entity, even in areas endemic for echinococcosis.

The first case of hydatid cyst of the spleen was reported by Bertelot in 1790 from an autopsy.<sup>(3)</sup> The frequency of splenic involvement within abdominal



**Fig. 1** Abdominopelvic CT image shows a cyst originating from the spleen and a second cyst originating from the left lateral lobe of the liver. The cyst in the spleen appears to displace the left kidney to the inferomedial of the abdomen.

hydatidosis is reported to be 0.5%–6%,<sup>(4)</sup> and it can either be isolated or concomitant with liver hydatid disease.

The recurrence rates after surgical therapy of liver hydatid cysts have been reported to be 6.8%–22.3% in different case series.<sup>(5–7)</sup> The liver is the most common site of recurrence. Extrahepatic recurrences occur in the lung or peritoneum and the serosa of the abdominal organs due to intraoperative dissemination of germ elements. Splenic recurrence of liver hydatid cysts, however, has not been previously reported in the literature.

Surgical treatment has traditionally been the mainstay of therapy of splenic hydatid cyst. The most common technique is splenectomy, another option being spleen preserving surgery. Two different studies, each with a small number of patients, reported no significant difference between patients who underwent splenectomy and those who had spleen preserving surgery with regard to the median hospital stay, recurrence rate and postoperative complication rate.<sup>(8,9)</sup>

We report a patient with a recurrent splenic hydatid cyst in the spleen and liver synchronously after surgical therapy for liver hydatid disease. He was treated with liver resection and spleen preserving surgery.

## CASE REPORT

A 55-year-old Turkish man was admitted to our gastrointestinal surgery clinic with left upper quadrant

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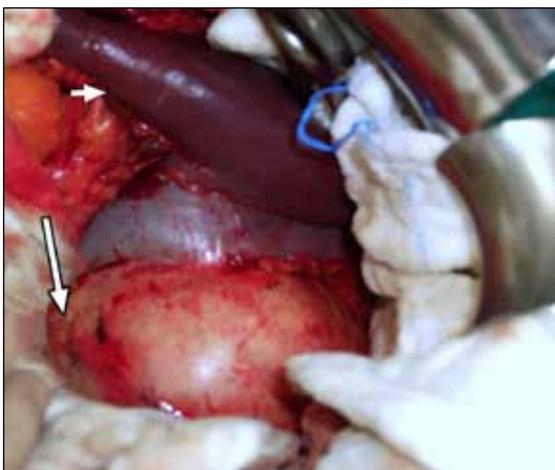
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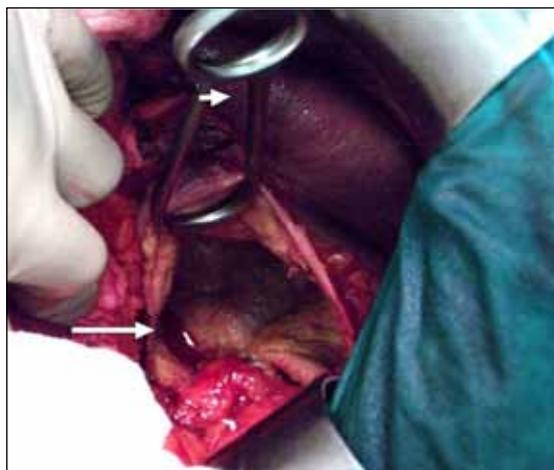
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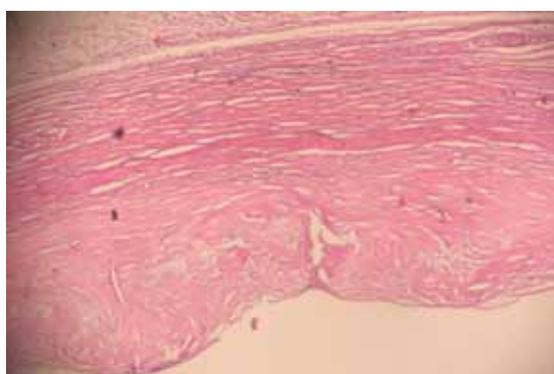
**Fig. 2** Intraoperative image shows the spleen (arrowhead) and the huge cyst (arrow) originating from it. The cyst is isolated with sponges before the cavity is opened.



**Fig. 3** Intraoperative image shows the spleen (arrowhead) and cyst wall (arrow) after partial resection and removal of the contents.

abdominal pain. Physical examination revealed a palpable mass in the left upper quadrant of the abdomen. The patient had midline and right subcostal incision scars, and his medical history included two operations for liver hydatid cyst. Biochemical and haematological tests, including renal and liver function tests, haemoglobin level, total leukocyte count and differentiation, were all found to be within the normal range. Cyst hydatid indirect haemagglutination (IHA) test was also negative. Abdominopelvic dynamic computed tomography (CT) showed a non-enhancing regular cyst of 13 cm × 12 cm originating from the spleen, as well as a second cyst of 5.4 cm × 3.5 cm in size originating from the left lateral lobe of the liver (Fig. 1). CT of the chest was unremarkable. A diagnosis of recurrent splenic and liver hydatid cysts was made, and surgery was planned. The patient was put on oral albendazole 14 days prior to surgery.

On further examination, a hydatid cyst was found in the spleen, which extended to the transverse mesocolon, pushing the intestines and the left kidney (Fig. 2). A second hydatid cyst was found in the left lateral section of the liver. Splenic vessels were found at the splenic hilum and were controlled with vessel loops. The cyst was isolated from the other parts of the abdomen to prevent intraoperative dissemination of germ elements. It was subsequently instilled with scolicedal solution and maintained. The cyst cavity was opened and the contents of the splenic cyst aspirated. The expanding part of the cyst wall outside the spleen was then resected, and the cyst cavity was cleaned in order to remove all endocysts (Fig. 3). Finally, external drainage was performed, followed by left lateral sectionectomy for the liver lesion. Histopathologic examination confirmed the diagnosis of hydatid cyst in both specimens (Fig. 4). The patient's postoperative



**Fig. 4** Photomicrograph shows the histological section of the splenic hydatid cyst wall.

period was uneventful, and he was discharged with albendazole treatment on postoperative Day 6.

## DISCUSSION

Hydatid cyst disease remains a considerable public health problem, especially in pastoral and farming regions. Different recurrence rates have been reported after surgical treatment of liver hydatid cysts in various studies.<sup>(5,6,9)</sup> About 84% of these recurrences occur in the liver due to smaller cysts being misdiagnosed during the operation. Extrahepatic recurrences that occur in the lung or peritoneum and the serosa of the abdominal organs as a result of intraoperative dissemination of germ elements have also been reported. To our knowledge, splenic recurrence of liver hydatid cyst has not been previously reported in the literature, and this is the first case of concomitant recurrence in the spleen and liver following previous surgical therapy of liver hydatid cyst.

Echinococcosis IHA is used for the diagnosis of the *Echinococcus* infection, with a specificity of 90%–100% and a sensitivity of 68.4%.<sup>(10,11)</sup> In our case, IHA was

negative despite the presence of a hydatid cyst confirmed by histopathology. This demonstrated that the diagnosis of echinococcosis cannot be excluded with a negative IHA test.

Splenic involvement of hydatid cyst has a reported frequency of 0.5%–6% within abdominal hydatidosis.<sup>(4)</sup> Complications related to splenic hydatid cyst such as infection or rupture,<sup>(12)</sup> spontaneous cutaneous fistula,<sup>(13)</sup> bronchosplenic fistula<sup>(14)</sup> and perforation into the stomach or colon<sup>(15,16)</sup> have previously been reported. This makes surgical treatment of splenic hydatid cyst mandatory. In most centres, splenectomy is the standard treatment for splenic hydatid disease. Spleen preserving surgery has been reported as an alternative method in a few studies. Atmatzidis et al reported no significant difference between patients in the splenectomy and spleen-preserving groups with regard to median hospital stay, recurrence rate and postoperative complication rate. The study included 19 patients (11 for splenectomy and eight for spleen preserving surgery), with a mean follow-up of 52 months.<sup>(9)</sup> Meimarakis et al reported three patients treated with spleen preserving surgery who were recurrence-free during the mean 8.8-year follow-up period.<sup>(8)</sup>

In our patient, partial cystectomy and drainage for the splenic hydatid cyst was performed. Bleeding is the most severe complication of spleen preserving surgery. Splenic hilum control before opening the cyst makes splenectomy easier and faster. Our patient did not experience any potential complications following splenectomy and spleen preserving surgery.

In conclusion, we report the first case of hydatid cyst recurrence in the spleen and liver synchronously, following surgical therapy of liver hydatid cyst. We also opine that spleen preserving surgery can be safely performed for splenic hydatid disease if the location of the cyst is appropriate.

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