Primary extrahepatic abdominal hydatidosis

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KEYWORDS
Hydatid cyst; Extrahepatic; Echinococcus

Abstract
Introduction: Most of the abdominal hydatid cysts occur in liver. Extrahepatic hydatid cyst is usually secondary to rupture (operative and non-operative) of the hepatic hydatid cyst. Primary extrahepatic hydatid cysts are rare and only a few sporadic cases have been reported.

Material and methods: One hundred and eighty-three patients with abdominal hydatid cysts managed surgically from January 1998 to December 2003 were evaluated retrospectively. Twelve (6.5%) patients had only extrahepatic abdominal involvement.

Results: The cysts were present in spleen (2.2%), pancreas (1.1%), peritoneum and pelvis (1.6%), gallbladder (0.6%), mesocolon (0.6%) and adrenal (0.6%).

Discussion and conclusions: It is difficult to diagnose extrahepatic echinococcosis as it usually is not suspected. Symptoms are related to size, location or ensuing complication of the cyst. It should be strongly suspected in differential diagnosis of all abdominal cysts especially in an endemic area.

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Introduction

Echinococcosis represents a common condition in many parts of the world where livestock is raised in association with dogs. Besides Kashmir, it is prevalent in Australia, Argentina, Chile, Africa, Eastern Europe, Middle East, New Zealand and the Mediterranean region particularly Lebanon and Greece. With immigration, the prevalence of the disease has increased in Europe and North America in recent years.1–3 The hydatid cyst has been reported in virtually all organs and tissues. However, exact extrahepatic incidence is not known.

The purpose of this study was to present our experience of some rare location and presentation and management of extrahepatic abdominal organs.

Materials and methods

In this retrospective and descriptive study, 183 patients with abdominal hydatid cysts managed
surgically from January 1998 to December 2003 in Sher-i-Kashmir Institute of Medical Sciences, Srinagar (SKIMS) were evaluated. Twelve (6.5%) patients had only extrahepatic abdominal involvement (Table 1). The records of all these patients were collected from the medical records department of SKIMS and were evaluated with respect to localization, mode of presentation and management. The diagnostic workup included ultrasonography (USG) abdomen and CT scan abdomen. ELISA for hydatid cyst was only done when there was a strong suspicion preoperatively. However, most of the times diagnosis was made only on laparotomy when it was located at a rare site.

Results

One hundred and sixty-seven (91.3%) cases were found in liver alone. Hydatid cyst of right lobe of liver predominated (73.8%); 12.5% patients had hydatid cyst of left lobe of liver only while 13.7% patients had hydatid cysts of both lobes. Four (2.2%) patients had disseminated hydatidosis involving liver and other abdominal organs.

The localization of the hydatid cysts of remaining 12 (6.5%) patients is depicted in Table 1.

In our study, 4 (2.2%) patients had hydatid cyst of spleen. Three patients presented with abdominal discomfort. Two patients had palpable spleen. Ultrasonography and CT scan of abdomen revealed splenic cyst (Fig. 1). On exploration hydatid cyst of spleen was found and splenectomy was performed. One patient was explored for a bullet injury in abdomen. Operative findings revealed a lacerated spleen along with a ruptured hydatid cyst. A splenectomy was performed.

We found 2 (1%) cases of hydatid pancreas. One patient presented with epigastric pain while another had an epigastric mass. Ultrasonography and CT scan revealed cystic mass in tail of pancreas in one, and body and tail of pancreas in another case. However, the diagnosis was made on laparotomy in both cases. A distal pancreatectomy was performed in each case.

There was one case of hydatid gallbladder in our series. The patient presented with pain in right hypochondrium. Ultrasonography and CT scan revealed cholangitis and a 4.5 cm × 4 cm cyst near porta hepatis. ERCP was normal. On laparotomy, a hydatid cyst arising from gallbladder was found which was loosely attached to inferior border of liver.

There were three primary peritoneal hydatidosis with different presentations. One of the patients presented as retention of urine. Cystoscopy revealed a bulge in posterior wall of bladder. Another patient presented with a mass hypogastrum. Ultrasonography and CT scan revealed a retrovesical cyst. On exploration, retrovesical hydatid cyst was confirmed. The third patient was admitted for evaluation of ascites. Ultrasonography revealed free fluid in peritoneal cavity. During the hospital stay, the patient developed features of peritonitis after an ascitic tap. An exploratory laparotomy was performed and a huge hydatid cyst occupying whole of peritoneal cavity was found. All these cases were managed by cystectomy and tube drainage.

A patient presented with a swelling in right hypochondrium. Ultrasonography and CT scan revealed a cystic mass in relation with inferior surface of liver. On exploration a hydatid cyst in the mesocolon of hepatic flexure invading the colon and loosely attached to inferior surface of liver was found. The cyst had to be excised.

A patient presenting with pain in right flank was found to have a cystic mass in right adrenal on ultrasonography which was confirmed by CT scan. On exploration right adrenalectomy was performed.

<table>
<thead>
<tr>
<th>Table 1</th>
<th>Unusual cases of abdominal hydatid cysts</th>
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<tr>
<td>Case no.</td>
<td>Age (years)/Sex</td>
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<tr>
<td>1</td>
<td>36/F</td>
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<tr>
<td>2</td>
<td>39/F</td>
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<tr>
<td>3</td>
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<td>4</td>
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<td>30/F</td>
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<tr>
<td>12</td>
<td>49/M</td>
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</table>
Histopathology revealed hydatid cyst of the adrenal gland. The postoperative period was without any major event. All these patients are on regular follow-up and no recurrence has so far been noted.

**Discussion**

Echinococcosis is a human disease caused by the larval form of *Taenia echinococcus*, which lives in the gut of the dog, wild canines and other carnivorous animals, which represents the definitive host and involves both domestic and wild animals. Humans become the accidental intermediate hosts by ingesting *Taenia* eggs. The main species pathogenic for man in Kashmir is *Echinococcus granulose* causing cystic echinococcosis. It is endemic in Kashmir. We have not encountered any case of *Echinococcus multilocularis* during the study period.

After ingestion of the contaminated food, hexacanth embryos migrate by portal system to liver where majority lodge. Those escaping this hepatic filter pass through the heart to lungs, and in most intermediate hosts these two sites account for majority of cysts. Others, however, may return to the heart and be distributed by systemic circulation to wide range of organs in which hydatid cysts have been recorded. There is some evidence that lymphatic system may be implicated in the transport of oncospheres from gut to the site of cystic development.

Extahepatic abdominal involvement may be primary or secondary. By far, hydatid spleen is the most common site after liver involvement in abdomen. 1.5—3.5% of all cases of abdominal hydatidosis have been reported due to splenic hydatid cysts.5—7 There are a few sporadic reports of hydatid cyst pancreas.8,9 Only one case of hydatid cyst of gallbladder has been reported so far.10 Peritoneal hydatidosis is usually secondary to liver hydatidosis.11 Hydatid cyst of colon has rarely been reported.12 Hydatid adrenal is very rare. But with the wider application of ultrasonography (US) and computed tomography (CT) more adrenal cysts are detected incidentally.13 No recurrence has been found in these patients till date. Our usual policy for hydatid cyst (if diagnosis confirmed preoperatively) is to give albendazole at a dose of 10 mg/kg/day for two months followed by surgery. Postoperatively the patient is again put on albendazole. However, in patients where diagnosis is made on laparotomy, the albendazole is given soon after the surgery once the orals are tolerated, for two months. With this protocol the recurrence rate has reduced to about 4% for all abdominal hydatid cysts.

It is difficult to diagnose extrahepatic echinococcosis, as it usually is not suspected. The surgeon must be aware of this possibility so as to follow the policy to avoid spillage during surgery. Symptoms are related to size, location or ensuing complication of the cyst. It should be strongly suspected in differential diagnosis of all abdominal cysts especially in an endemic area — where it can be a usual parasite at an unusual site.

**References**