Unusual locations of intraabdominal hydatid cysts including gynecological organs: Clinical features and surgical outcomes of double center experience

Karın içi kisthidatiklerin jinekolojik organları içeren alışılmamış yerleşim yerleri; Çift merkez deneyimi, klinik bulguları ve cerrahi sonuçları

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ABSTRACT

Objective: Hydatid disease (HD) is a parasitic infestation that most commonly caused by the larval stage of Echinococcus granulosus. Peritoneal echinococcosis (13%) is usually secondary and primary peritoneal HD is very rare and only sporadic cases have been reported.

Methods: The demographic data, imaging findings, indirect hemaglutination test (IHAT) levels, surgical approaches, pathological findings, complications and outcomes were analysed.

Results: The localization of the hydatid cyst are spleen, as an intraabdominal cys, right adrenal gland, mesentery of the transvers colon, omentum and left tube, as a pelvic cys, right tube, and uterus. The surgical procedures includes splenectomy, total cystectomy, partial cystectomy and omentoplasty, total cystectomy and omentektomy, and right adrenalectomy.

Conclusion: Isolated primary peritoneal cyst without the presence of cysts in the other intraabdominal organs is very rare and has been reported about 2% of all abdominal HD. The differential diagnosis of primary peritoneal echinococcosis also includes soft tissue tumors, intraperitoneal abscess, cystic lymphangioma, embryonal cyst, ovarian neoplasms, teratoma, and other cystic and necrotic solid tumors. The management of extrahepatic HD is based on the size and location of the cysts and the health status of the patient. The goal of the surgery is removal of the cyst without any spillage.

Key words: Abdominal cavity, albendazole, anaphylaxis, cystectomy, Echinococcus granulosus

ÖZET

Amaç : Kisthidatik, sıkılık Echinokokus granulözus'un larva formunun yol açtığı parazitik bir enfeksiyondur. Karın içi ekinokok enfeksiyonu (%13) genellikle ikincli kincil oluşmaktadır ve primer ekinokok enfeksiyonu oldukça nadir görülmektedir ve sadece sporadik vakalar bildirilmiştir.

Metodlar: Demografik veriler, görüntüleme bulguları, indirekt hemaglutinasyon testi (IHAT), cerrahi prosedürler, patolojik bulgular, kompleksiyonlar ve sonuçlar değerlendirildi.

Sonuçlar: Hidatik kistin yerleşim yerleri; dalak, kazı içi kist, sağ sürellen bez, transvers kolon mezenteri, omentum, sol tube, pelvik kist, sağ tube ve uteurstu. Cerrahi prosedür olarak; splenektomi, total kistektomi, parsiyel kistektomiyle beraber omentoplastı, total kistektomiyle beraber omentektomi ve sağ adrenalektomi uygulandı.

Tartışma: Diğer karın içi organlarda kistik lezyon olmadan izole primer karın içi kist hidatik oldukça nadirdir ve tüm kazı içi kist hidatiklerin %2’si olarak raporlanmaktadır. Primer kazı içi kist hidatiklerin aykırı
INTRODUCTION
Hydatid disease (HD) is a parasitic infestation that most commonly caused by the larval stage of Echinococcus granulosus (1,2). It is prevalent in the Middle East, the Mediterranean region, particularly in sheep-raising countries, Australia, Argentina, and Africa (1,3). The main hosts are dog, wolf, fox and jackal that pass eggs into their feces while cattle, horses and goats are intermediate hosts. Human is the accidental intermediate host in the life cycle of Echinococcus granulosus (3,4). The annual incidence of HD has been reported as 18-20 cases per 100,000 inhabitants (5). Humans acquire the HD either by direct contact with a main host or by ingestion of food contaminated by the eggs. After ingestion, the eggs loses its protective chitinous layer, enters to the lymphatic or venous circulation via penetrating the intestinal mucosa and transported to the liver, lungs, and other organs (6,7). Although the disease can occur anywhere, the most frequently involved organs are the liver (70%) followed by the lungs (25%) (7,8). Other organs including spleen (0.9-8.9%), kidneys (1-4%), pancreas (0.25-0.75%), brain, heart, ovum, bone and abdominal wall are in a small proportion (9). Peritoneal echinococcosis (13%) is usually secondary and primary peritoneal HD is very rare and only sporadic cases have been reported (6,10,11). Extrahepatic HD is usually secondary to spontaneous or traumatic rupture of the primary hepatic hydatid cyst or surgical inoculation of a hepatic cyst (1,6). The spontaneous asymptomatic microruptures of hepatic cyst into the peritoneal cavity are not uncommon. 85% to 90% of patients with Echinococcus granulosus infection have single-organ involvement and more than 70% of patients have only one cyst. The cysts may be uni or multiloculated and thin or thick walled. HD is seen more frequently at the hepatic locations (10). The demographic data, imaging findings, pathological findings, complications and outcomes of all these patients were collected from the database of the department of surgery and obstetric and gynecology. The diagnostic workup included ultrasonography (USG) and computed tomography (CT) of the abdomen. We isolated the abdominal cavity with gauzes soaked in 20% hypertonic saline solution for preventing the spillage and anaphylaxis and Albendazole is given both in preoperative and postoperative period and the dose duration is five days before operation and one month after operation in serology positive group routinely.

RESULTS
In this retrospective study, 18 patients with extrahepatic intraabdominal hydatid cysts treated with surgery from January 2008 to January 2014 in Tepecik Teaching and Research Hospital were evaluated. The medical records of all these patients were collected with International Classification of Disease code (ICD-10) from the database of the department of surgery and obstetric and gynecology. The diagnostic workup included ultrasonography (USG) and computed tomography (CT) of the abdomen. We isolated the abdominal cavity with gauzes soaked in 20% hypertonic saline solution for preventing the spillage and anaphylaxis and albendazole is given both in preoperative and postoperative period and the dose duration is five days before operation and one month after operation in serology positive group routinely.

The demographic data, imaging findings, indirect hemaglutination test (IHAT) levels, surgical approaches, pathological findings, complications and outcomes were analysed. Medical records of all these patients were evaluated with respect to the data in the literature.
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Table 1: The demographic data of patients.

<table>
<thead>
<tr>
<th>Patients</th>
<th>Age</th>
<th>Sex</th>
<th>Serology</th>
<th>Localization</th>
<th>Operation</th>
<th>Complication (Major)</th>
<th>Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>1.</td>
<td>44</td>
<td>F</td>
<td>Negative</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>2.</td>
<td>51</td>
<td>F</td>
<td>Negative</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>3.</td>
<td>32</td>
<td>F</td>
<td>Positive</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>4.</td>
<td>58</td>
<td>M</td>
<td>Negative</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>5.</td>
<td>46</td>
<td>F</td>
<td>Negative</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>6.</td>
<td>57</td>
<td>F</td>
<td>Positive</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>7.</td>
<td>39</td>
<td>F</td>
<td>Positive</td>
<td>Spleen</td>
<td>Splenectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>8.</td>
<td>40</td>
<td>F</td>
<td>Positive</td>
<td>Intraabdominal</td>
<td>Total cystectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>9.</td>
<td>45</td>
<td>F</td>
<td>Positive</td>
<td>Intraabdominal</td>
<td>Total cystectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>10.</td>
<td>56</td>
<td>F</td>
<td>Positive</td>
<td>Intraabdominal</td>
<td>Total cystectomy+ Omentectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>11.</td>
<td>44</td>
<td>M</td>
<td>Negative</td>
<td>Mesentry of transvers colon</td>
<td>Partial cystectomy+ Omentoplasty</td>
<td>No</td>
<td>Yes</td>
</tr>
<tr>
<td>12.</td>
<td>32</td>
<td>M</td>
<td>Positive</td>
<td>Right adrenal gland</td>
<td>Right Adrenalectomy</td>
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<tr>
<td>13.</td>
<td>32</td>
<td>F</td>
<td>Unknown</td>
<td>Left tuba and omentum</td>
<td>Total cystectomy+ Omentectomy</td>
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<td>No</td>
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<tr>
<td>14.</td>
<td>31</td>
<td>F</td>
<td>Unknown</td>
<td>Pelvic</td>
<td>Partial cystectomy+ Omentoplasty</td>
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<td>Yes</td>
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<tr>
<td>15.</td>
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<td>F</td>
<td>Unknown</td>
<td>Right tuba</td>
<td>Total cystectomy</td>
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<td>No</td>
</tr>
<tr>
<td>16.</td>
<td>54</td>
<td>F</td>
<td>Unknown</td>
<td>Pelvic</td>
<td>Total cystectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>17.</td>
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<td>Unknown</td>
<td>Uterus</td>
<td>Total cystectomy</td>
<td>No</td>
<td>No</td>
</tr>
<tr>
<td>18.</td>
<td>56</td>
<td>F</td>
<td>Unknown</td>
<td>Pelvic</td>
<td>Total cystectomy</td>
<td>No</td>
<td>No</td>
</tr>
</tbody>
</table>

**DISCUSSION**

Hydatid cyst consists of three layers. The outer layer (pericyst) is formed from the host tissue as a result of inflammatory reaction to the parasite. The inner layer (ectocyst) is elastic and made of gelatinous material and the innermost germinal layer (endocyst) is cellular and secretes hydatid fluid. The high secretuar pressure is responsible for the enlargement of the cyst (4). Primary peritoneal echinococcosis is very rare and has been reported about 2% of all abdominal HD (12). Intraabdominal hydatid cysts usually develop due to spontaneous or iatrogenic rupture of hepatic, splenic, or mesenteric cysts. Isolated primary peritoneal cyst without the presence of cysts in the other intraabdominal organs is very rare (10). Primary peritoneal echinococcosis accounts for 2% of all abdominal hydatidosis (7,10). Dissemination occurs either by lymphatic or systemic circulation (10). The hydatid cyst is usually asymptomatic and clinical presentation of HD depends on the location and the diameter of the cyst. HD may present with complications including the rupture of the cyst that leads to anaphylactic shock and bacterial infection-related complications (1,8,13). The main symptom is pain and occurs as an acute onset when the cyst ruptures (4). The differential diagnosis of primary peritoneal echinococcosis also includes soft tissue tumors, intraperitoneal abscess, cystic lymphangioma, embryonal cyst, ovarian neoplasms, teratoma, and other cystic and necrotic solid tumors. Especially in endemic regions such as Turkey, the hydatid cyst must always keep in mind in the differential diagnosis of cystic lesions (1). The definitive diagnosis of HD requires a combined
assessment of clinical, radiological, and serological findings (1). Routine laboratory tests are commonly nonspecific but eosinophilia occurs in 25% of cases. Different serological tests are used for the diagnosis of HD including enzyme-linked immunosorbennt assay (ELISA), indirect hemaglutination test (IHAT), latex aglutination and immunoelectrophoresis (1,6,8,14). ELISA and IHAT has a sensitivity of 95% and 87.5%, retrospectively (1).

Imaging tools are very important because of the nonspecific clinical features of HD (3). Ultrasonography (USG) and computed tomography (CT) are helpful for the diagnosis. When the diagnosis is clear, USG is cost-effective in endemic areas. However, USG is less accurate in localising and delineating the extend of the cysts (4). The sensitivity of USG ranges from 93% to 98% and demonstrate the stage of the cyst and associated complications (1). Type II and type III cysts are pathognomonic but the other types are generally resemble other cystic lesions such as ovarian cysts or pelvic malignancies when located on the pelvic region (3). The sensitivity of CT ranges from 90% to 97% and is superior to USG in detecting the extrahepatic cysts and gives more details about the size, location, neighbourhood and number of the cyst (Figure 1)(1,7).

**Figure 1:** The axial CT image of a splenic hydatid cyst.

The management of extrahepatic HD is based on the size and location of the cysts and the health status of the patient. Asymptomatic small cysts and the patients who are not candidate for surgery can be treated with antihelminthic drugs with a usage of 28 days in one to eight cycles, seperated with 2-3 weeks of drug-free intervals (1,7). Surgical resection is the only curative treatment in both symptomatic and large hydatid cysts (Figure 2). Surgical treatment can be radical or conservative related to the health status of the patients. Total cystectomy is the gold standard. If the resection is not possible, unroofing and drainage are recommended for peritoneal cysts which were attached to the intraperitoneal viscera. The goal of the surgery is removal of the cyst without any spillage. The most important thing is to isolate the abdominal cavity with gauzes soaked in 20% hypertonic saline solution for preventing the spillage and anaphylaxis (1).

**Figure 2:** The operative image of a hydatid cyst invading the mesentery of transvers colon.

Albendazole is given both in preoperative and postoperative period and the dose duration is five days before operation and one month after operation (3,4). The use of antiparasitic drugs can be used for reducing the risk of anaphylactic reaction preoperatively and recurrence rate postoperatively (6,8). The use of hypertonic saline or 0.5% silver nitrate solutions before opening the cysts intraoperatively provides to kill the daughter cysts and prevents the spillage and anaphylactic reaction (10). Although there have been a lot of scolicidal agents, there is no consensus on which agent is the best. Some experimental studies showed that hydrogen peroxide and 10% povidone-iodine have strong scolicidal activity. Percutaneous aspiration, injection, and reaspiration (PAIR) technique is also used as a nonsurgical treatment (1). In PAIR, USG-guided percutaneous aspiration of the cyst is performed, followed by injection of scolicidal agent. The agent is left on for a minimum 15 minutes and reaspiration of the cyst's content is performed. The indications are large, multiple cysts of the spleen, kidney, bones, and the liver, and also inoperable and recurrent patients (4). There are some limitations for this technique and only suitable for predominantly fluid cysts (1). Medical therapy combined with percutaneous drainage and laparoscopic resection of the cyst have also been reported (14, 15). The recurrence rate of surgery is 2% (10). The results of only medical treatment without surgery are in up to 40% of cases (10,11). A postoperative long term follow-up is needed and repeated imaging studies are also essential (4). In the present study, we have only 2 patients with the recurrence because of the partial cystectomy at the mesenteric and the pelvic localization cysts. The cause of the partial cystectomy was the adjacent of the cysts which complicated the dissection to the transverse colon mesentery in one patient and rectum in the other patient. We isolated the abdominal cavity with gauzes soaked in 20% hypertonic saline solution for prevent-
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ing the spillage and anaphylaxis and albendazole is given both in preoperative and post-operative period and the dose duration is five days before operation and one month after operation in serology positive group. There were no recurrence in the patients with total cystectomy group.

**Conclusion**

It is difficult to diagnose extrahepatic intraabdominal HD, as it usually is not suspected. The surgeon have to take care for preventing the spillage during surgery. Total cystectomy must be the aim of the treatment, if possible.

**REFERENCES**