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Original study

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 tuba, as a pelvic cys, right tuba, and uterus. The surgical procedures includes splenectomy, total cystectomy, partial cystectomy and omentoplasty, total cystectomy and omentectomy, 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.

Keywords: Abdominal cavity, albendazole, anaphylaxis, cystectomy, *Echinococcus granulosus*

ÖZET

Amaç : Kisthidatik, sıklıkla *Ekinokokus granülozus*'un larva formunun yol açtığı parazitik bir enfeksiyondur. Karın içi ekinokok enfeksiyonu (%13) genellikle ikincil oluşmaktadır ve primer ekinokok enfeksiyonu oldukça nadir görülmektedir ve sadece sporadik vakalar bildirilmiştir.

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

Sonuçlar: Hidatik kistin yerleşim yerleri; dalak, karın içi kist, sağ sürrenal bez, transvers kolon mezenteri, omentum, sol tuba, pelvik kist, sağ tuba ve uterustu. Cerrahi prosedür olarak; splenektomi, total kistektomi, parsiyel kistektomiyle beraber omentoplasti, 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 karın içi kist hidatiklerin %2'si olarak raporlanmaktadır. Primer karın içi kist hidatiklerin ayırıcı

tanısında; yumuşak doku tümörleri, karın içi abse, kistik lenfanjiom, embriyonel kist, over tümörleri, teratom ve diğer kistik ve nekrotik solid tümörler yer almakatadır. Karaciğer dışı kist hidatiklerin tedavi yönetiminde, kistlerin çap ve sayısı ve hastanın genel sağlık performansı oldukça büyük rol oynamaktadır. Cerrahinin ana hedefi karın içi yayılıma yol açmadan kisti tamamen çıkarmaktır.

Anahtar kelimeler: Karın boşluğu, albendazol, anaflaksi, kistektomi, Ekinokokus granülozus

INTRODUCTION

Hydatid disease (HD) is a parasitic infestation that most commonly caused by the larval stage of Echinococcus granulosus (1,2). It is prevelant 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 ot thick walled. HD is seen more frequently at the ages of 20 to 40 years and usually occurs in childhood and grows so slowly about 1-3 cm per year (1). HD may be asymptomatic or present with complaints and complications (8). The most common complaint is abdominal pain but the clinical features may be nonspecific and generally depends on the location of the cyst (1). In extrahepatic locations HD is usually remains asymptomatic unless the cyst grows and causes symptoms due to pressure, rupture of the peritoneal or pleural cavity, secondary infection, or an allergic reaction (1,9).

MATERIAL AND METHOD

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 databese 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.

RESULTS

In this retrospective study, 18 patients with extrahepatic intraabdominal hydatid cysts treated with surgery from 2008 to 2014 in Tepecik Teaching and Research Hospital were evaluated (Table 1). The medical records of all these patients were collected with International Classification of Disease code (ICD-10) from the databese 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.

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Table 1: The demographic datas of patients.

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

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 gelatinuous 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). Intraperitoneal 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 immunosorbent 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).

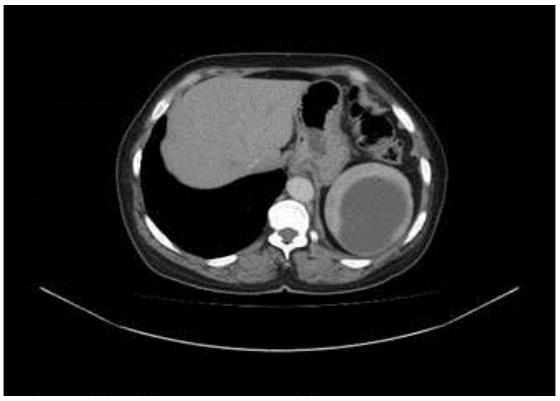

Figure 1: The axial CT image of a splenic hydatid cyst.

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 standart. 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).

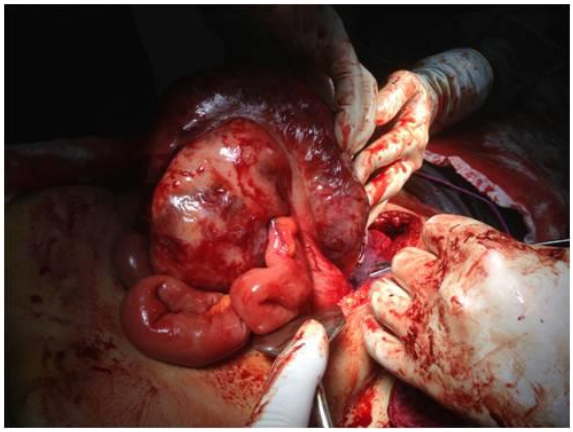

Figure 2: The operative image of a hydatid cyst invading the mesentery of transvers colon.

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 scolocidal 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 reperated 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 preventing 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 intra-abdominal 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.

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