



Primary splenic hydatidosis: Case series

Primer dalak hidatik kist hastalığı: Olgu serisi

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Abstract

Aim: Primary hydatid cysts of the spleen are very rarely seen in endemic regions. We report here our experience with primary splenic hydatid cysts in adults.

Methods: We retrospectively analyzed eleven isolated spleen hydatid cyst cases that applied to our clinic and underwent surgery.

Results: Among 11 patients, who underwent operations for primary splenic hydatid cyst disease, six patients (54.5%) were male and five patients (45.5%) were female. The average spleen size was 14.2 cm (10–22 cm) in length. The average cyst diameter was 10.1 cm (5–20 cm). Nine (81%) patients underwent splenectomy as the surgical procedure and two (18%) patients underwent cystotomy, partial cystectomy, and tube drainage processes as a spleen protective surgery. Two patients could not undergo splenectomy because the cyst was localized in the lower pole of the spleen in one patient and in another patient, it was centrally localized.

Conclusion: Primary splenic hydatid cyst is a rare condition. Abdominal tomography is the best method for diagnosing splenic hydatid cyst. The treatment of splenic hydatid cysts is surgically. No exact consensus has been reached regarding the selection of an optimal surgical procedure.

Keywords: Splenic hydatidosis, Splenectomy, Splenomegaly

Öz

Amaç: Dalağın izole hidatik kist hastalığı endemik bölgelerde nadiren görülür. Biz bu çalışmada yetişkinlerde primer splenik hidatik kist hastalığı ile ilgili deneyimimizi sunuyoruz.

Yöntemler: Kliniğimize müracaat eden ve ameliyat edilen on bir izole dalak hidatik kist hastasını retrospektif olarak inceledik.

Bulgular: Primer splenik hidatik kist ameliyatı yapılan 11 hastanın altısı (%54,5) erkek, beşi (%45,5) kadın idi. Ortalama dalak büyüklüğü 14,2 cm (10-22 cm) uzunluğunda idi. Ortalama kist çapı 10,1 cm (5-20 cm) idi. Dokuz hastaya (%81) cerrahi girişim olarak splenektomi yapıldı. İki hastaya (%18) dalak koruyucu cerrahi olarak kistotomi, parsiyel kistektomi ve tüp drenaj işlemleri uygulandı. Splenektomi yapılmayan iki hastanın birinde kist dalak alt pol, diğerinde ise santral yerleşimli idi.

Sonuç: Dalağın izole hidatik kist hastalığı nadir görülebilen bir durumdur. Abdominal tomografi hastalığı saptamada en etkin yöntemdir. Dalağın hidatik kist hastalığının tedavisi cerrahidir. En uygun cerrahi yöntemin seçimi ile ilişkili bir uzlaşma sağlanamamıştır.

Anahtar kelimeler: Splenik kist hidatik hastalığı, Splenektomi, Splenomegali

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Introduction

Cystic hydatid disease, or cystic echinococcosis (CE), is an important parasitic zoonosis caused by the larval cyst stage of the dog tapeworm *Echinococcus granulosus*. This disease affects both healthy people and people who are involved in animal production in endemic regions such as South America, Mediterranean countries, and Middle Asia [1]. It is a significant disease problem in Turkey, particularly in the East and Southeast Anatolian Region [2]. Humans become infected by accidentally ingesting the eggs of the tapeworm [1].

Echinococcosis can be seen in almost every organ or tissue of the human body. Hydatid cysts are most frequently seen in the liver, followed by the lung. The brain, spleen, bile ducts, mesentery, and soft tissues are the organs where hydatid cyst is seen less frequently [3,4]. Hydatid cysts of the spleen are very rarely seen in endemic regions [5]. The prevalence of splenic hydatid cysts varies from series to series, ranging from 0.5–4%. The mean prevalence was reported as 3% [3-5].

We report here our experience with primary splenic hydatid cyst disease in adults.

Materials and methods

We retrospectively reviewed the case records of the patients attending our institution to identify those diagnosed as abdominal hydatidosis between July 2008 and March 2014 in the General Surgery Clinic of Erzurum Regional Education and Research Hospital. Details of the medical history and examination of these patients and results of relevant investigations were recorded. Of 193 patients with abdominal hydatid cysts, eleven (5.6%) had isolated splenic hydatid cysts. These patients form the basis of this report; patients with coexisting cysts in any other organ were excluded. The clinical findings, diagnostic methods, therapeutic measures undertaken, and histological data were retrospectively analyzed for all eleven patients. Abdominal computed tomography (CT) and abdominal ultrasound (US) were used as radiological diagnostic tools in the patients (Figure). After diagnosis, the patients underwent radical surgical resection of the splenic hydatid cyst, including elective splenectomy or spleen-preserving surgery (cystotomy, partial cystectomy, and omentoplasty), according to the number, location, and diameter of the cysts in each patient. All patients were routinely used with 20 % saline solution as a scolicial agent into the cysts in the operation. Each specimen of the resected spleen and the cysts was histopathologically analyzed.

Results

Among 11 patients who underwent operations for primary splenic hydatid cyst disease, six patients (54.5%) were male and five patients (45.5%) were female. The average age was 45.1 years (23–80 years). Two patients complained of left upper quadrant pain, eight patients had nonspecific stomach ache, and one patient had dyspeptic complaints. Preoperative complete blood counts, renal and liver function tests, and chest X-rays were within normal limits.

A hydatid cyst serology test was performed on three patients: one showed positive and two showed negative responses. Abdominal CT and US were used as the radiological diagnostic tools in ten patients, and abdominal US alone was used in one patient. According to the Gharbi Classification, six of the cysts were type 2, and five of the cysts were type 3. No hydatid cyst disease in another organ was detected in any of the cases by radiological scanning, and none of the patients underwent operations due to hydatid cyst disease in another organ previously.

All patients were administered 15 mg/kg albendazole for five days in the preoperative period, and 15 mg/kg albendazole for 90 days in the postoperative period.

The average spleen size was 14.2 cm (10–22 cm) in length. Five patients had splenomegaly. The average cyst diameter was 10.1 cm (5–20 cm). Only one cyst was found in each case. Six patients (54%) had a cyst in the upper pole of the spleen, two patients (18%) had a cyst in the lower pole of the spleen and three patients (27%) had a cyst localized in the central part of the spleen. Nine (81%) patients underwent splenectomy as the surgical procedure. Two patients underwent cystotomy, partial cystectomy, and tube drainage processes as a spleen protective surgery. One of the patients who underwent spleen preservation surgery had a cyst at the lower pole and the other one had a cyst at the center (Table).

Diaphragm rupture and subsequent pneumothorax developed in one patient during the operation as the cyst in the spleen had adhered to the diaphragm. The patient underwent a left tube thoracostomy intra-operatively and the tube was drawn postoperatively on the 3rd day. One of the patients died due to pulmonary emboli on the 1st day postoperatively. The average postoperative hospitalization period was 3.9 days (1–7 days). All nine patients who underwent elective splenectomy received pneumococcal and meningococcal vaccines 2–3 weeks before the surgery to allow the development of protective antibodies.

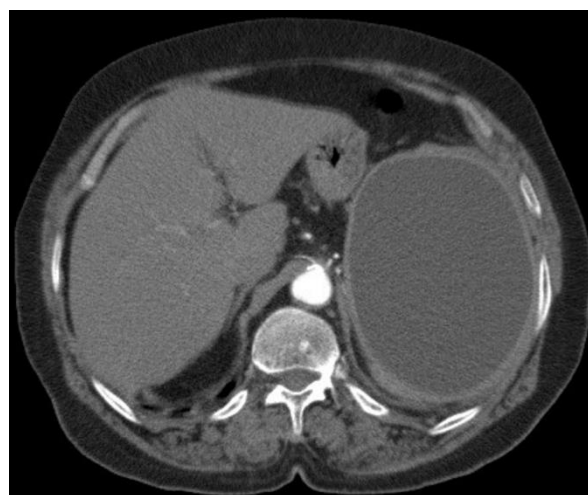


Figure: Hydatid cyst in the spleen

Table: Characteristics of the patients with splenic hydatid cyst disease. (M: Male, F: Female *: mm)

No	Sex	Age	Symptom	Spleen diameter*	Cyst diameter*	Operative procedure	Postoperative complications	Hospitalisation periods (day)
1	M	23	Abdominal pain	155	100x90	Splenectomy	-	4
2	F	27	Abdominal pain	140	70x55	Splenectomy	-	4
3	F	28	Abdominal pain	165	100x60	Splenectomy	Diaphragm rupture	4
4	M	60	Abdominal pain	120	60x40	Splenectomy	-	4
5	F	80	Abdominal pain	160	130x100	Splenectomy	-	3
6	M	25	Abdominal pain	220	200x150	Spleen-preserving surgery	-	3
7	M	52	Abdominal pain	160	130x80	Splenectomy	Pulmonary emboli	1
8	F	36	Abdominal pain	110	50x40	Splenectomy	-	3
9	M	35	Abdominal pain	100	100x80	Splenectomy	-	7
10	M	76	Abdominal pain	115	80x50	Spleen-preserving surgery	-	5
11	F	55	Abdominal pain	120	100x80	Splenectomy	-	5

Discussion

Hydatidosis is a disease caused by the larva belonging to the genus *Echinococcus*, with *E. granulosus* being the most commonly found [6]. Hydatid cyst disease has a wide prevalence in the world. The disease is frequently encountered in Turkey, particularly in Southeast and Eastern Anatolia regions involved in livestock breeding. The degree of prevalence of the infection depends on the multitude of herds raised in that region [7]. Although hydatid cyst disease can be found in almost all organs and tissues of the human body, it is most frequently seen in the liver (50–77%), the lungs (15–47%), the spleen (0.5–8%), and the kidneys (2–4%) [8,9]. In our experience, isolated splenic hydatid cysts constituted 5.6 % of our patients with abdominal hydatid disease.

The hydatid cyst factor, *E. granulosus*, can cause the disease by reaching the spleen via several means. The parasite can directly reach the spleen because the portal blood flow turns in the opposite direction in human beings with portal hypertension. Another possibility is that the parasite reaches the spleen by means of the lymphatics or blood flow, and a third is that it reaches the spleen as a result of reflux from the portal vein due to the increase in intra-abdominal pressure [10].

Clinically, nearly 30% of the patients with splenic hydatid cysts are asymptomatic. Splenomegaly is the most frequent finding, which is incidentally determined [11]. The clinical symptoms caused by splenic hydatid cyst mostly depend on the pressure effect of the cyst on the neighboring organs and the replacement of the neighboring organs. The symptoms are few, non-specific, and comprise mainly an abdominal mass mostly located in the left hypochondrium and less frequently in the epigastrium, pain that is usually a dull, dragging ache, dyspepsia, constipation due to pressure on the colon, and dyspnea due to pushing up of the left diaphragm [1,3,8]. A pain in the lumbar region constitutes a clinical sign in a few patients [12]. Hypertension induced by renal artery pressure is another clinical symptom that occurs due to the pressure of the cyst on the neighboring organs [4, 11]. In our study group, eight and two patients (totally 91%) had stomach ache and left upper quadrant pain, respectively, and while one case had dyspeptic complaints.

Several serological tests are specific to hydatidosis and are used to confirm the diagnosis. Enzyme-linked immunoelectrotransfer blotting, where available, is the test of choice. In some studies, it has greater than 95 percent of sensitivity and specificity [13,14]. In addition, ELISA has up to 84 percent sensitivity. The determination of specific IgG1 and IgG4 antibodies, which develop against *Echinococcus* in the human body might increase the specificity of ELISA test [15]. In our study, a hydatid cyst serology test was performed on three patients and one patient showed a positive response. We do not use serological tests routinely at our clinic. Özdoğan et al [16] emphasized that serological tests were not necessary for diagnosing hydatid cyst disease.

US and CT scans, alone or in combination, can establish a definite diagnosis of splenic hydatid cysts in almost all cases. Today, US is the primarily preferred monitoring method because it is inexpensive, easy, and has a high diagnostic value. It is diagnostic because it shows the cystic structure of the lesion, the presence of daughter vesicles, and hydatid sand [17]. CT is usually the next step after an US diagnosis has been made. The main purpose is to visualize the relation between the hydatid cyst and the surrounding tissue. Although CT scan is more sensitive than abdominal US, but non-calcified benign cysts without daughter cysts cannot be differentiated per se from other benign cysts either by CT or by US [15]. Direct imaging can also be exploited in diagnosing splenic hydatid cysts. The calcifications

on the cyst wall are visible with direct imaging [18]. In our study, abdomen CT was used in ten patients and abdomen USG alone was used in one patient.

The primary nonparasitic cysts, pseudocysts of the spleen, splenic abscesses, cystic neoplasia, and traumatic spleen cysts should be kept in mind in the differential diagnosis [19].

The treatment of splenic hydatid cysts is made conservatively or surgically. Small and asymptomatic splenic hydatid cysts require close follow-up, although they can be treated by anthelmintic medications [20]. Surgical operations vary from aspiration to total splenectomy [21]. No exact consensus has been reached regarding the selection of an optimal surgical procedure. Symptomatic or large cysts should be treated surgically because they can rupture spontaneously or traumatically [20]. Total splenectomy is preferred by most of the surgeons because of its very low or no recurrence rate [22]. It is the preferred approach undertaken in cases with larger, multiple, and symptomatic cysts of central or hilar location or in cases with simultaneous involvement of other organs [23]. However, sepsis-associated mortality rates of total splenectomy are 4% in children and 1.9% in adults and this is the greatest disadvantage of total splenectomy. For this reason, the number of surgeons in favor of spleen-saving surgery has increased. Spleen-saving approaches are preferred for small and single cysts that are settled in the periphery of the spleen. Spleen-saving approaches include partial splenectomy, enucleation, deroofting with omentoplasty, internal drainage with cystojejunostomy, or external drainage [24, 25]. In our study, nine patients (81%) underwent splenectomy as the surgical procedure and two patients (19%) underwent cystotomy, partial cystectomy, and tube drainage processes as a spleen-saving approach. No difference was found between total splenectomy and spleen-saving approaches in terms of the recurrence rate of the splenic hydatid cyst disease, postoperative hospitalization period, and complications [24]. Surgical treatment can also be made by laparoscopic or robotic methods [21,26,27]. However, the cyst can be torn during surgical treatments made by laparoscopic or robotic methods and this can result in anaphylactic shock, intra-abdominal dispersion, and recurrence of the cyst. Clinical experience and costs are also included among the significant problems [20,27,28]. If the splenic hydatid cyst is torn during the surgery, either spontaneously or traumatically, anaphylactic shock is a rare but severe condition that can occur. The patient can die if anaphylactic shock is not diagnosed, not immediately treated, or is resistant to treatment [29]. We did not observe any surgical operation-induced anaphylactic reactions in our patients.

In summary, splenic hydatid cyst is a rare condition that can be found in isolation or together with cysts that affect other organs. Abdominal CT is the best method for diagnosing splenic hydatid cyst. Although laparoscopic or robotic methods can be used in selected patients, the most prevalent treatment method is total splenectomy by open surgical methods. More spleen-protective methods should be preferred, particularly in children.

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