Original Article

Laboratory Results and Clinical Findings of Children with Hydatid Cyst Disease

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ABSTRACT

Objectives: The relationship hydatid cyst (HC) is a parasitic disease that is endemic worldwide. **Aims:** To study the clinical findings and laboratory results of patients with hydatid disease. **Materials and Methods:** Total number of patients (n = 78) who underwent surgery for HC disease from 2000 to 2017 were retrospectively evaluated. The patients' demographic characteristics, reasons for admission into hospital, lesion location and size, laboratory results, and complications were recorded. **Results:** Of the HCs, 59% and 26.9% were located in the liver and lungs, respectively. The rate of multiple organ involvement was 10.3%. A total of 16 (20.5%) cases had ruptured HCs (7 livers, 7 lungs, 1 spleen, and 1 omentum). There was no significant difference in the rate of eosinophilia between patients with and without cyst rupture (P = 0.9). Indirect hemagglutination (IHA) tests yielded negative results in 38.9% of the patients; among them, 32.1% had ruptured HCs. A negative IHA test result was significantly associated with rupture (P = 0.046). No significant difference between rupture and cyst size or location was found. **Conclusions:** HC rupture was not correlated with cyst size. The sensitivity of serological tests and the blood eosinophil count was low, even in cases of ruptured HCs. The recurrence rate can be reduced by open surgery and medical treatments.

Keywords: *Child, echinococcosis, hemagglutination test, recurrence, rupture*

Introduction

Although hydatid cyst (HC) disease is endemic worldwide, it is more frequent in societies reliant on animal husbandry and in less-developed and developing countries.[1] The liver (60–75%) and lungs (15–25%) are the most common sites for HC disease. HCs can spread to other parts of the body through the circulatory, including the brain, spleen, kidneys, pancreas, musculoskeletal system, and skin.[1,2] Patients with noncomplicated HC disease are generally asymptomatic. The symptoms of HC disease vary depending on the location and size of the cysts; bigger HC makes to compress adjacent organs. Patients with complications due to HCs typically show clinical symptoms.[1] Eosinophilia is seen in 20–34% of patients with HCs but it is not diagnostic.[3] Immunoglobulin G (IgG) enzyme-linked immunosorbent assays (ELISAs) and indirect hemagglutination (IHA) tests are frequently performed for the diagnosis of HC disease. However, the sensitivity of these tests is 50–100%.[1,4]

In this study, we aimed to present our clinical experiences by evaluating the relationship of HC cases with age, sex, location and size of the cyst, IHA test, and rupture.

Materials and Methods

Study design, setting, and ethics

The study protocol was approved by the Ethics Committee of our institution (date of approval 18/01/2018, approval number 06).

In the pediatric surgery clinic of our university hospital, the records of 78 patients treated and followed up for HC

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disease from 2000 to 2017 were retrospectively reviewed. The demographic characteristics of the patients, reasons for and duration of hospitalization, lesion location and size, laboratory characteristics, radiological imaging findings, cyst rupture, complications, and duration of follow-up were recorded. Patients with ruptured and non-ruptured cysts were compared in terms of age, sex, cyst location and size, and IHA test results. Only patients who underwent surgery were included in the analysis. Surgery was performed in patients with a ≥5-cm-diameter and/or ruptured HC. The diagnosis was confirmed by macroscopic and pathological evaluations.

Statistical analysis

For the statistical analysis, IBM Statistical Package for the Social Sciences statistics version 22 software (SPSS inc. Chicago, IL, USA) was used. The normality of the distribution of quantitative data was analyzed by the Shapiro-Wilk test. The unpaired *t*-test was used to compare independent variables and Pearson's Chi-square and Fisher's exact tests for categorical variables. Level of significance was accepted at *P* value <0.05. Patients not diagnosed with HC disease before surgery were excluded from the analysis.

Results

Of the 78 patients, 52 (66.7%) were male and 26 (33.3%) were female (M: F ratio = 2). The mean age of the patients was 10.3 ± 2.9 years (range 5–16). Of the lesions, 46 (59%) were located in the liver and 15 (32.6%) involved multiple cysts. Of the 78 HC cases, 21 (26.9%) were in the lungs (15 in the left lung, 13 in the right lung, and 1 bilateral). The locations of the lesions are listed in Table 1. The mean size of the lesions was 8.1 ± 2.4 cm (range 3–15 cm) in the liver and similar size in the lung. Cyst rupture was detected in 16 cases [Table 1]. There was no significant difference between rupture, age, sex, cyst location, and cyst size [Table 2].

Most common complain were abdominal pain (n = 31) and an abdominal mass (n = 14) for abdominal HCs; and cough (n = 18), chest pain (n = 12), fever (n = 10), and respiratory distress (n = 9) for lung HCs. Commonest finding on physical examination was tenderness and guarding for abdominal HCs and decreased respiratory sounds on the relevant side for lung HCs. A skin rash developed in only 2 of the 16 cases of ruptured HCs.

Of the patients, 17 (21.8%) had eosinophilia. There was no significant difference in the rate of eosinophilia between patients with and without cyst rupture (P = 0.9) [Table 3]. IHA tests were conducted in all patients, except for the six not pre-diagnosed with HC disease.

Table 1: Cyst location and cyst rupture rates

Cyst location	*n* (%)	Rupture *n* (%)
Liver	46 (59%)	7 (13.2%)
Lung	21 (26.8%)	7 (24.2%)
Liver + Lung	6 (7.7%)	-
Liver + Lung + Spleen	1 (1.3%)	1
Lung + Pancreas	1 (1.3%)	-
Omentum	1 (1.3%)	1
Renal	1 (1.3%)	-
Urachal	1 (1.3%)	-
Total	78 (100%)	16 (20.5%)

Table 2: The relationship between the cyst rupture, cyst location and size, and some epidemiological and IHA tests significance

Parameters	Cyst rupture (*P*)	Cyst location (*P*)	Cyst size (*P*)
Sex	0.474	0.553	-
Size	0.965	0.654	-
Age	0.677	-	-
Cyst location	0.232	-	-
Negative IHA test	**0.046***	-	-
Developed complication	-	-	0.642

**P*<0.05 is assumed significant; IHA=indirect hemagglutination.

Table 3: Eosinophil values and hospitalization duration for patients with and without cyst rupture

Variable median (min-max)	Rupture		
	No	Yes	*P*
Eosinophil values	0.25 (0.02-4.68)	0.25 (0.02-4.68)	0.90
Duration of hospitalization (day)	8 (4-39)	9.5 (6-38)	0.055

**P*<0.05 is assumed significant. min-max=minimum-maximum.

Table 4: Eosinophil values and hospitalization duration for patients with and without complications

Variable median (min-max)	Complication		
	No	Yes	*P*
Eosinophil values	0.22 (0.02-3.72)	0.32 (0.06-4.68)	0.092
Duration of hospitalization (day)	8 (4-38)	17 (7-39)	< 0.001

**P*<0.05 is assumed significant. min-max=minimum-maximum.

In 28 (38.9%) of 72 patients, IHA tests yielded negative results. HC rupture occurred in 9 (32.1%) patients with a negative IHA test result. Among these patients, twelve cysts were in the lungs; twelve in the liver; two in the liver and lungs; one in the liver, lung, and spleen; and one in the omentum. Of these cysts, six lungs, one liver, one spleen, and one omentum cyst ruptured. A negative IHA test result was significantly associated with rupture (P = 0.046) [Table 2].

In about 72 of the 78 (92.3%) patients, radiological imaging (direct radiography, ultrasonography [US],

computed tomography [CT] and magnetic resonance imaging) was performed for the early diagnosis of HC disease. Six (7.7%) patients showed nonspecific radiological imaging findings, which did not contribute to the diagnosis.

Open surgery was performed on all of the patients, except for the six who received percutaneous aspiration, injection, and reaspiration (PAIR). As a scolicidal agent, 3% NaCl was used during surgery. Albendazole treatment was started 2 weeks before elective surgery and was continued for 3–6 months thereafter.

Around 5 (9.4%) patients, in whom bile drainage continued for >13 days (range 5–20 days), underwent sphincterotomy of the ampulla of Vater with endoscopic retrograde cholangiopancreatography (ERCP). In 13 (44.8%) of the 29 patients who underwent surgery for lung HCs, a bronchoalveolar fistula was detected during the operation.

Mean duration of stay in the hospital was 8 days (range 4–35 days) after surgery for liver HCs. Biliary fistula on whom endoscopic sphincterotomy was performed, bile leakage stopped after a median of 7 days (range 5–9 days). Mean duration of stay in the hospital was 10 days (range 6–39 days) for patients with lung HCs and 13 days (range 7–39 days) for the 13 patients with an alveolar fistula. Duration of stay in the hospital differed significantly between patients with and without complications ($P < 0.001$) [Table 4]. The mean duration of follow-up was 12 months (range 6–36 months). No recurrence was detected by physical examination, IHA testing, direct radiography, US, or CT during the 6–36-month follow-up period.

Discussion

The incidence of HC disease is high in animal care professions;[1] the annual incidence in endemic areas is 1–150/100,000.[5] In this study, 61 (78.2%) of the patients resided in a rural area, and their families were engaged in farming husbandry. HC disease is reportedly more common in females.[6] However, in this study, 52 (66.7%) and 26 (33.3%) of the patients were male and female, respectively (M:F ratio = 2).

Hydatid cysts can spread to other parts of the body through the circulatory, most frequently to the liver (60–75%) and lungs (15–25%). Multiple organs are involved in 18% of cases.[1,3] However, lung HC disease is reportedly more common in children.[7] In this study, 46 (59%) of the HCs were in the liver, 21 (26.8%) in the lungs and 8 (10.3%) in multiple organs. Lung HCs are typically located on the right side and are bilateral in 32.2% of patients.[3,6,8,9] This is because the blood flow rate is higher in the lower right lobe.[10] In this study, 15 (51.7%) lung HCs were on the left, 13 (44.8%) on the right, and 1 (3.5%) was bilateral.

Hydatid cyst disease is not typically symptomatic. However, bigger HCs produce symptoms by compressing adjacent organs. A ruptured lung HC is always symptomatic.[8] The most common symptoms of abdominal HCs are abdominal pain, an abdominal mass and fever; in comparison, those of lung HCs are cough and fever.[1] In this study, all of the patients with ruptured lung HCs had a cough, respiratory distress, chest pain and fever. Abdominal pain was evident in patients with abdominal HCs. A physical examination is typically not helpful for the diagnosis of HC disease as the findings are nonspecific and depend on cyst location and complications. Here, the patients with lung HCs had decreased respiratory sounds on the related side, and those with abdominal HCs exhibited tenderness and guarding.

The rate of rupture secondary to trauma or spontaneous rupture of a liver HC is 1.75–8.6%.[11,12] Ozturk *et al*.[13] reported that the rupture of an intraperitoneal HC (3%) was secondary to trauma in adults. In our study, the incidence of a ruptured liver HC was 13.2% and rupture was secondary to trauma in only one patient. The incidence of a ruptured lung HC in children is reportedly up to 47%.[8] In this study, the ruptured lung HC rate was 24.2% ($n = 7$). A diameter of ≥10 cm and elevated internal pressure are often related to HC rupture.[2,14] In this study, there was no significant association between cyst size and rupture ($P = 0.965$) [Table 2]. Therefore, pressure in the cyst was likely responsible for rupture.

The rates of anaphylaxis and mortality after HC rupture are 25%.[12] In our study, a skin rash was in two cases with a ruptured HC, but anaphylaxis and mortality did not develop. Laboratory findings in the diagnosis of HCs are nonspecific. Although eosinophilia is present in 20–34% of patients, it is not diagnostic for HC to be.[3,15] Aydogdu *et al*.[16] reported that all 11 patients with a ruptured lung HC had eosinophilia. In this study, 21.8% of the patients had eosinophilia. There was no significant difference in the rate of eosinophilia between patients with and without cyst rupture ($P = 0.9$) [Table 3]. This may be due to the high incidence of parasitic disease in children and rural areas[17] or to differences in cyst location. Prospective studies involving a more heterogeneous group of patients are needed.

The sensitivity of IgG ELISAs and IHA tests is 50–100%.[1,4] In this study, IHA tests were performed in all of the patients, except the six who did not have a pre-diagnosis of HC disease. IHA tests yielded negative

results in 28 (38.9%) of 72 patients. The HC was ruptured in 9 (32.1%) of the patients with a negative IHA test result. The results of IHA tests were negative for 12 lungs; 12 liver; 2 liver and lung; 1 liver, lung, and spleen; and 1 omentum HC. Of them, six lungs, one liver, one spleen, and one omentum HC ruptured. The sensitivity of the IHA test was 61.1%, and a negative IHA test result was significantly associated with rupture (P = 0.046) [Table 2]. The antibody response changes according to cyst location, and serological tests have reduced sensitivity for lung cysts.[15,18] The decrease in the antibody titre is more rapid in patients with lung HCs than in those with liver HCs.[19] In this study, the most negative IHA test results were seen in patients with a ruptured lung HC.

In total, 72 (92.3%) patients were pre-diagnosed with HC disease radiologically. However, 6 (7.7%) patients (two in the liver, two in the lung, one in the omentum, and one on the anterior abdominal wall) were not pre-diagnosed with HC disease by radiological imaging. Serological tests are needed in radiologically suspicious cases, although this is controversial.[6,20] In this study, we performed IHA tests in all patients, except for the six with no pre-diagnosis of HC disease.

The main treatment is surgery. However, according to cyst location, size and number, PAIR, or medical treatment (albendazole or mebendazole) can be applied in addition to surgery.[1] Endoscopic procedures should be used sparingly in children due to the sparsity of data on anesthesia, contamination, and anaphylaxis[21-23]; however, such procedures are minimally invasive, require a shorter duration of hospitalization and are associated with greater patient comfort.[24] Initiating medical treatment before surgery is important to prevent contamination during the operation.[21] We performed open surgical procedures in all patients, except the six who underwent PAIR for a liver HC.

The most important complication of HC disease is a fistula (5–17%).[25] The main treatment is surgery to relieve portal vein pressure or close the fistulae. Infection of the cyst, atrophy of the lobes due to pressure in the biliary tract and portal vein, hypertrophy on the opposite side, and opening of the cyst to the biliary system can also occur.[2,6,11] In this study, bile leakage was found in 9.4% (n = 5) of the patients. ERCP is important in the treatment of patients with bile leakage and biliary fistulae.[26] Decreasing the pressure in the biliary system by endoscopic sphincterotomy may help to close a biliary fistula in the postoperative period.[27] Early endoscopic sphincterotomy in patients with bile leakage may reduce the rate of complications and shorten the hospitalization duration. The complications of lung HCs are pneumothorax, pleural effusion, empyema, abscess, fluid and/or membrane fragments (e.g. rock juice from the mouth), anaphylaxis, and death.[9] A bronchoalveolar fistula developed in 13 (44.8%) patients in this study.

There was no significant difference in HC size or the rate of eosinophilia between patients with and without complications [Tables 2 and 4]. During and after surgery, the most important complication of HCs in the lung and liver was a fistula. During surgical removal of a cyst, the fistula should be considered, and potential complications should be evaluated in advance.

The recurrence rate of HCs differs depending on the surgical method and follow-up period (0–25%).[28-30] Serological tests alone are not sufficient to identify recurrence and must be supported by US or CT.[30] The follow-up period should be at least 3 years after surgery. Epidemiological characteristics, including employment in agriculture or animal husbandry, were not related to recurrence, but cyst contamination was a risk factor.[28] Liver HCs, difficult surgery, multiple intraabdominal cysts, and laparoscopic treatment were nonsignificantly associated with recurrence compared to open surgery.[29,31] The recurrence rate was reduced by the medical treatment of patients who underwent surgery.[31] No recurrence was observed in this study, possibly due to the use of open surgery and initiation of medical treatment before surgery and its continuation for 3–6 months thereafter.

In conclusion, HCs cannot be detected until they reach a certain size and/or rupture. Because of the high incidence of multiple organ involvement, other organs should be examined in patients with HC disease. Due to the nonspecific physical examination results, an absence of specific laboratory findings and low sensitivity of serological tests, HC disease should be considered as a diagnosis for patients with radiologically defined cysts, particularly in endemic regions. Importantly, IHA tests yielded negative results in cases of ruptured HCs. There was no significant difference between cyst rupture and cyst size. Open surgery and medical treatment can reduce the recurrence of HCs.

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Conflicts of interest

There are no conflicts of interest.

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