

Evaluation of Hydatid Cyst Cases: A Single-center Retrospective Study

Kist Hidatik Olgularının Değerlendirilmesi: Tek Merkezli Retrospektif Bir Çalışma

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Cite this article as: Şahin S, Kaya B. Evaluation of Hydatid Cyst Cases: A Single-center Retrospective Study. Türkiye Parazitolojisi Dergisi. 2024;48(4):222-7.

ABSTRACT

Objective: Cystic echinococcosis (CE) is a zoonotic condition that can be encountered, particularly in developing countries, and leads to significant economic losses. This study was planned to observe the treatment options, complications, in the patients we followed.

Methods: Patients aged 18 and over who were diagnosed with hydatid cyst and followed in our hospital between January 2018 and December 2023 were included in the study. Data were obtained from the hospital's record system. The patients with CE were retrospectively evaluated in terms of age, gender, cyst location, treatment method applied, presence of relapse, and complications.

Results: A total of 30 patients, with a mean age of 42.8 years (range: 19-68), were included in the study; 13 (43.3%) were male and 17 (56.7%) were female. The most common presenting complaint was abdominal pain (n=14, 46.7%), and 6 patients (20.0%) were asymptomatic. Sixteen patients had multiple cysts in the same region, and 6 patients had cysts in different regions. The most common site of involvement was the liver (n=21, 70.0%), followed by the lungs in 4 patients (13.3%). Single cases of brain, spinal cord, spleen, kidney, and bone involvement were observed. Diagnosis was made by ultrasonography in 16 patients (53.3%), magnetic resonance imaging in 8 patients (26.7%), and computed tomography in 6 patients (20.0%). Surgical intervention was performed in 20 patients (66.7%), and percutaneous drainage in 3 patients (10.0%). All patients received albendazole treatment. Complications included intra-abdominal abscess in three patients (10.0%) and rupture in one patient. One patient with intracranial involvement died.

Conclusion: Although observed worldwide, CE maintain their importance in terms of morbidity and mortality, particularly in developing countries.

Keywords: Hydatid cyst, cystic echinococcosis, zoonotic disease, *Echinococcus granulosus*

ÖZ

Amaç: Kistik ekinokokoz (KE), dünya üzerinde özellikle gelişmekte olan ülkelerde karşılaşılabilen, ciddi ekonomik kayıplara yol açan zoonotik karakterli bir hastalıktır. Takip ettiğimiz hastalardaki tedavi seçenekleri, relaps ve komplikasyonları görebilmek için bu çalışmanın yapılması planlandı.

Yöntemler: Ocak 2018-Aralık 2023 yılları arasında hastanemizde takip edilen ve kist hidatik tanısı alan 18 yaş ve üstü hastalar çalışmaya dahil edildi. Veriler hastane kayıt sisteminden elde edildi. Kist hidatik tanısı alan hastalar yaş, cinsiyet, kistin yerleşim yeri, uygulanan tedavi yöntemi, komplikasyonlar açısından retrospektif olarak değerlendirildi.

Bulgular: Yaş ortalaması 42,8 (19-68), 13'ü (%43,3) ve erkek ve 17'si (%56,7) kadın toplam 30 hasta çalışmaya dahil edildi. En sık başvuru şikayeti (n=14, %46,7) karın ağrısıydı, 6 (%20,0) hastada herhangi bir şikayet yoktu. On altı hastada aynı bölgede birden fazla ve 6 hastada da farklı bir bölgede de kist mevcuttu. En sık tutulum bölgesi (n=21, %70,0) karaciğerdi, 4 (%13,3) hastada akciğer tutulumu görüldü. Bunu birer hasta ile beyin, omurilik, dalak, böbrek ve kemik tutulumu izledi. On altı (%53,3) hastada tanı ultrasonografi, 8 (%26,7) hastada manyetik rezonans görüntüleme ve 6 (%20,0) hastada bilgisayarlı tomografi ile kondu. Yirmi hastaya cerrahi (%66,7) ve 3 (%10,0) hastaya perkütan drenaj uygulandı. Tüm hastalara albendazol tedavisi verildi. Üç (%10,0) hastada komplikasyon olarak batın içi apse ve bir hastada rüptür gelişti. İntrakraniyal tutulumu olan bir hasta öldü.

Sonuç: Dünyanın her yerinde görülmekle birlikte özellikle gelişmekte olan ülkelerde kist hidatik morbidite ve mortalite açısından önemini korumaktadır.

Anahtar Kelimeler: Kist hidatik, kistik ekinokokoz, zoonotik hastalık, *Echinococcus granulosus*



Received/Geliş Tarihi: 09.07.2024 Accepted/Kabul Tarihi: 13.12.2024 Publication Date/Yayınlanma Tarihi: 22.01.2025

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INTRODUCTION

Cystic echinococcosis (CE) is a zoonotic disease that, while more concentrated in certain regions globally, can be found in every country. It causes significant public health issues and serious economic losses (1). The common causative agents of the disease are *Echinococcus granulosus* and *E. multilocularis*. In Türkiye, it is quite prevalent as a zoonosis, with most cases caused by *E. granulosus* (2,3). Although there are case series in our country, a study has found that the number of publications on CE has decreased in recent years (4). Liver involvement is the most common manifestation, it can affect various parts of the body including the heart, eyes, brain, kidneys, and bones (5,6).

Transmission to humans occurs through ingestion of eggs from contaminated hands, water, and food, typically via the feces of infected animals. The eggs reach various organs either through direct inoculation or secondary dissemination. While liver involvement is the most common, the disease can manifest anywhere in the body (7,8). Symptoms are not observed in the initial stages of infection, and it is almost always asymptomatic. Clinical findings vary depending on the affected organ and the size of the cyst. If the cyst is small and the cyst wall is calcified, it may not cause symptoms (8). In particular, liver hydatid cysts may not show symptoms until they reach a size of 10 cm (7-9). If a vital organ is affected, complications can arise, including mass effect, edema due to blood or lymphatic flow obstruction, systemic symptoms in case of rupture, and sometimes death or secondary bacterial infection (7,8).

At the time of diagnosis, most patients are asymptomatic (8). Diagnosis is established through serological tests such as indirect hemagglutination (IHA) and enzyme-linked immunosorbent assay (ELISA), along with radiological methods such as ultrasound (USG), magnetic resonance imaging (MRI), and computed tomography (CT) (9). Routine laboratory tests are generally normal unless a complication is present. When a cyst is associated with bile ducts, elevated liver function tests and cholestasis enzymes may occur; leukocytosis can be seen if there is an infected cyst. In 25% of patients, eosinophilia may develop following cyst rupture or leakage of cyst fluid. Serological tests have a sensitivity of 90%. However, serological tests can produce false positives, and a negative result does not definitively exclude the diagnosis of CE (2,9).

The classification of CE based on their morphology has been established according to the World Health Organization and Gharbi criteria (10,11). According to this classification, Type I cysts are unilocular and consist solely of fluid; Type II cysts have a multiloculated wall; Type III cysts contain daughter cysts; Type IV cysts have a heterogeneous echo; and Type V cysts have a calcified inactive wall (10,11). Alveolar echinococcosis (AE) is caused by the larval stage of the fox tapeworm (*Echinococcus multilocularis*) and is often diagnosed as a space-occupying lesion in the liver. The PNM classification system is used for AE because the growth pattern resembles that of a malignant tumor (P = parasitic mass in the liver, N = involvement of adjacent organs, and M = metastasis) (12). The management of CE includes medical therapy, percutaneous therapy, and surgical intervention. Surgery is the treatment of choice for pulmonary hydatid cysts. A total splenectomy is the treatment of choice for splenic CE. Differential diagnoses should include simple liver cysts, hemangiomas,

hepatocellular carcinoma, liver and lung abscesses, Caroli disease, hemangioendotheliomas, and mesenchymal hamartomas (13).

METHODS

The study was planned as a retrospective, cross-sectional, descriptive study in a hospital located in northwestern Türkiye. Patients over the age of 18 who were followed up in our hospital between January 2018 and December 2023 and diagnosed with CE were included in the study. The patients were reached using the ICD-10 diagnosis codes B67.8 (hepatic echinococcosis, unspecified) and B 67.9 (echinococcosis, other and unspecified). The information was obtained from the hospital registry system. The study was conducted in accordance with the principles of the Declaration of Helsinki. The study was approved by the Clinical Research Ethics Committee of University of Health Sciences Türkiye, Kartal Dr. Lütfi Kırdar City Hospital with the decision numbered 2024/010.99/2/47 on 27.03.2024.

Patients were retrospectively evaluated in terms of age, gender, cyst location, imaging methods used for diagnosis, laboratory tests used for CE diagnosis, treatment applied, presence of relapse, and complications. Ultrasound, CT or MRI, depending on the location of the cyst, were used as imaging methods, and *Echinococcus* IHA was used as laboratory test.

Statistical Analysis

The statistical analysis of the data was performed using the Statistical Package for the Social Sciences (SPSS) for Windows version 25.0. Numerical variables were presented as mean \pm standard deviation and minimum-maximum values. Continuous variables were expressed as medians, while categorical variables were expressed as frequencies and percentages.

RESULTS

The study included a total of 30 patients, aged between 19 and 68 years with a mean age of 42.8 years. Among these patients, 13 (43.3%) were male and 17 (56.7%) were female. Twenty-one (70.0%) of the patients had no underlying disease. In 6 (20.0%) patients who had no complaints at the time of admission, cysts were detected in the liver during imaging performed for other complaints. The most common presenting complaint among the other patients was abdominal pain (n=14, 46.7%). Other presenting complaints included cough, itching, seizures, and back pain (Table 1). Four (13.3%) of these patients had a prior history of CE.

Of the patients, 16 (53.3%) had multiple cysts in the same organ. In six patients (20.0%), CE were simultaneously detected in another organ outside the primary region. The liver (n=21, 70.0%) was the most commonly affected organ, and lung involvement was observed in 4 (13.3%) patients. This was followed by involvement of the brain, spinal cord, spleen, kidney, and bone in one patient each (Table 2). The smallest cyst, measuring 20x20 mm, was located in the liver, while the largest cyst, measuring 170x110 mm, was located in the kidney. Of the liver hydatid cysts, 15 (50.0%) were in segment 8, and 7 (23.3%) were in segment 7, predominantly in the right hepatic regions. In 11 (36.7%) patients, cysts were present in more than one segment. All lung cysts were located in the left lung.

The most commonly used imaging method for diagnosis was USG, employed in 16 patients (53.3%). Depending on the

Table 1. General characteristics of the patients

Demographic characteristics		Application assessment	
Gender	n (%)	Complaint	n (%)
Female/male	17 (56.7)/13 (43.3)	Abdominal pain	14 (46.7)
Age distribution		Cough	4 (13.3)
18-20	2 (6.7)	Side pain	2 (6.7)
21-30	6 (20.0)	Hip pain	1 (3.3)
31-40	5 (16.7)	Seizures	1 (3.3)
41-50	6 (20.0)	Itching	1 (3.3)
51-60	5 (16.7)	Swelling in the back	1 (3.3)
>60	6 (20.0)	No complaints	6 (20.0)
-	-	-	30 (100.0)

Table 2. Cyst characteristics

Location		Dimensions		Classification	
Site	n (%)	Millimeter (mm)	n (%)	Stage [†] (WHO-IWGE)	n (%)
Liver	21 (70.0)	0-10	-	1	3 (10.0)
Lung	4 (13.3)	11-30	6 (20.0)	2	4 (13.3)
CNS ^{**}	2 (6.7)	31-50	6 (20.0)	3	1 (3.3)
Spleen	1 (3.3)	51-100	11 (36.7)	4	4 (13.3)
Kidney	1 (3.3)	>100	7 (23.3)	5	2 (6.7)
Bone	1 (3.3)	-	-	-	-
	30 (100.0)	-	30 (100.0)	-	14 (46.7)

[†]: The World Health Organization Informal Working Group on Echinococcosis, ^{**}: Central nervous system

location, MR was used in 8 patients (26.7%), and CT was used in 6 patients (20.0%). *Echinococcus* indirect hemagglutination assay (IHA) was used as a biochemical test for cyst diagnosis. Among the 21 patients tested, 11 (36.7%) had an *Echinococcus* IHA titer >2560. Cysts of two patients with positive *Echinococcus* IHA are shown as examples in Figure 1. *Echinococcus* IHA test results were negative in 4 (13.3%) patients. No significant difference was found between *Echinococcus* IHA titers in hepatic and extrahepatic CE (p=1.00). There was also no significant difference in terms of CE localization site in patients with negative *Echinococcus* IHA (p=1.00). Cysts were demonstrated in these patients by imaging methods (Figure 2). No examination was requested for 5 (16.7%) patients whose CE diagnosis was confirmed by MRI in 2 patients and by CT imaging in 3 patients. Initial laboratory examinations revealed eosinophilia (eosinophil percentage >4%) in 8 patients (26.7%), elevated gamma-glutamyl transferase (GGT) levels in 9 patients (30.0%), elevated alkaline phosphatase levels in 5 patients (16.7%), and elevated aspartate aminotransferase and alanine aminotransferase levels in 3 patients (Table 3).

Surgery was used in the treatment of 20 patients (66.7%), while cyst fluid drainage was applied in 3 patients (10.0%). Relapse occurred in 5 (16.7%) patients, 4 (20.0%) of the patients who underwent surgery and 1 patient who underwent drainage. In addition to surgical and drainage treatments, all patients were given albendazole therapy for a minimum of 3 months. Three patients (10.0%) developed intra-abdominal abscesses, and one patient experienced a rupture. One patient with intracranial involvement died.

DISCUSSION

CE remains a significant and preventable zoonotic condition worldwide, particularly in developing countries (14). Many cases can progress asymptotically without proper medical diagnosis and treatment, but it can also present with various symptoms and findings depending on the organ in which the cyst is located (9,15). In our study, alongside asymptomatic cases, the majority of symptoms included abdominal pain, and less frequently, symptoms such as cough, itching, seizures, and back pain were observed.

The disease is observed in 50-70% of cases in the liver and 20-30% in the lungs, with other organs being affected less frequently. In adults, two-thirds of patients have liver involvement, whereas pulmonary involvement is higher in pediatric patients. Although typically only one organ is affected, concurrent cysts can be seen in other organs (9,16,17). Similar to the literature, in our cases, 70.0% presented with liver involvement (particularly the right liver), and 13.3% presented with lung involvement. Among the patients with CE in the liver, cysts were also detected in the lungs in 2 patients (6.7%) and in the pancreas in one patient (3.3%), albeit with smaller diameters. Additionally, there were cases of cyst involvement in the brain, spinal cord, spleen, kidneys, and bones.

Although radiological examinations are mostly sufficient for diagnosis, it is recommended that serological methods be used concurrently. USG is the first choice for diagnosis, differential diagnosis, staging, guiding interventional treatment, and follow-up. In cases where USG is insufficient, such as for lung or brain hydatid cysts, or in obese patients, CT can be used.

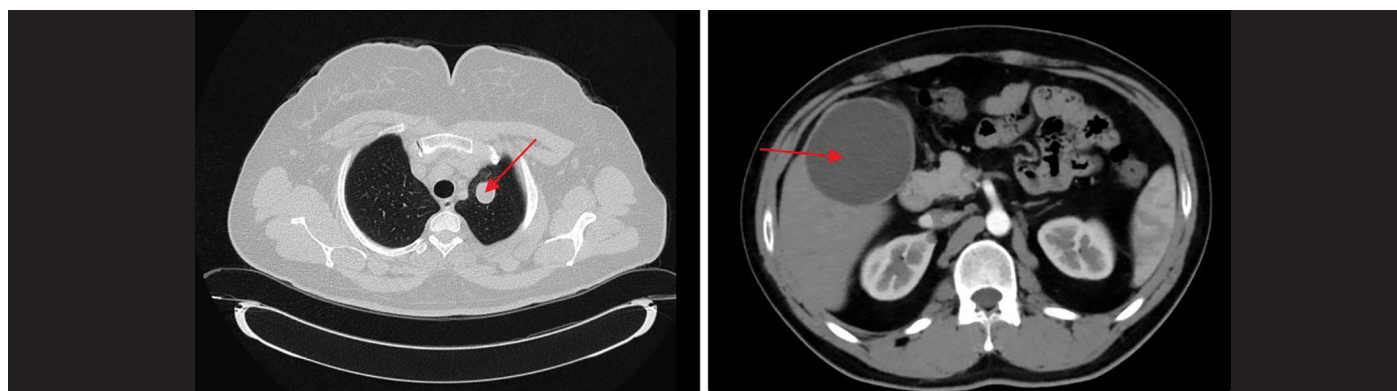


Figure 1. Cyst appearances on lung CT and abdominal CT in 2 different patients with positive *Echinococcus* IHA
 CT: Computed tomography, IHA: Indirect hemagglutination

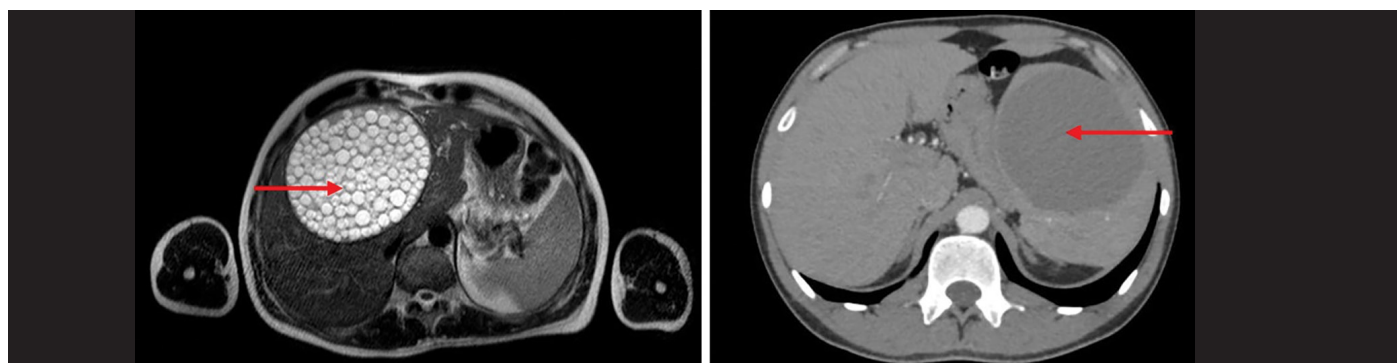


Figure 2. Cyst appearances on abdominal MRI and abdominal CT in 2 different patients with negative *Echinococcus* IHA
 MRI: Magnetic resonance imaging, CT: Computed tomography, IHA: Indirect hemagglutination

Table 3. Diagnostic methods and laboratory results

Imaging method		Indirect hemagglutination		Laboratory findings		
Method	n (%)	Result	n (%)	Parameter	n (%)	Location
USG	16 (53.3)	Negative	4 (13.3)	Eosinophilia (>4%)	8 (26.7)	Liver, lung, kidney
MRI	8 (26.7)	No test requested	5 (16.7)	ALT, AST elevation (>35 U/L)	3 (10.0)	Liver, CNS
CT	6 (20.0)	1/320-1/1280	10 (33.3)	ALP elevation (>98 U/L)	5 (16.7)	Liver, CNS
-	-	>2560	11 (36.7)	GGT elevation (>38 U/L)	9 (30.0)	Liver, lung, CNS

USG: Ultrasound, MRI: Magnetic resonance imaging, CT: Computed tomography, ALT: Alanine aminotransferase, AST: Aspartate aminotransferase, ALP: Alkaline phosphatase, GGT: Gama-glutamyl transferase, CNS: Central nervous system

MRI may also be necessary to show cyst wall defects and neural involvement (9,13). In our patients, the most frequently used imaging method, especially for liver cysts, was USG (n=16, 53.3%). CT (n=6, 20.0%) was used for lung CE and in obese patients, while MRI was particularly used for bone, brain, and spinal cord involvement. The sensitivity of serological tests varies depending on the characteristics of the case; for liver cysts, sensitivity ranges from 80% to 100% and specificity from 88% to 96%, whereas it is lower for lung infections (50-56%) or other organ involvements (9). A negative serological test does not rule out CE. In 4 (13.3%) of our patients, *Echinococcus* IHA was found negative. Among 21 patients with a titer >160, in 11 (36.7%) cases the *Echinococcus* IHA titer was >2560. IHA positivity in liver CE was detected at 81.0% (n=17). Serological testing was not requested for 5 (16.7%) patients whose CE diagnosis was confirmed by imaging methods.

Routine laboratory tests for cases are usually normal; however, leukocytosis and eosinophilia can be observed in 25% of cases with infected CE (18). The incidence of eosinophilia in our cases (n=8, 26.7%) was similar to the literature. Leukocytosis was present in two patients who developed intra-abdominal abscesses and in one patient with cyst rupture. Elevated GGT levels were detected in nine patients (30.0%) and elevated ALP levels in five patients (16.7%), while three patients exhibited elevated liver function tests.

The type of lesion is important in treatment planning. Types I, II, and III, as well as Type IV cysts containing fluid, are considered active and require treatment (10). Surgery, percutaneous drainage, and concurrent administration of albendazole or mebendazole may be employed (9,10). Among patients who undergo surgery, the average mortality rate is 2.2%, with a recurrence rate of approximately 6.5% (14). Surgical treatment was applied to

66.7% (n=14) of patients with liver cysts. Percutaneous drainage was performed on a total of three patients: two with liver cysts and one with a spleen cyst. All patients with lung or other organ involvement underwent surgical treatment. Additionally, all patients received albendazole treatment for at least three months. Although percutaneous drainage is often preferred due to shorter procedure times and reduced hospitalization, it does not show a significant difference in relapse rates compared to surgical treatment. In fact, some studies report higher relapse rates in patients who underwent drainage (19,20). In our study, relapse was observed in one patient with intracranial involvement and three patients with liver involvement. All patients who experienced relapse had previously undergone surgical treatment. Patients with small cysts, who did not receive surgical treatment or drainage, were treated with albendazole for a minimum of six months.

The incidence of complications in patients is approximately 10%. Depending on the cyst's location in relation to the bile ducts, it may open into the bile ducts or a bronchus. The cyst contents can cause luminal obstruction or post-obstructive bacterial infection. One significant complication is the secondary spread of daughter cysts to other parts of the body, leading to the development of new foci in different organs (9). In our study, intra-abdominal abscesses developed in three patients (10.0%) as complications, and one patient experienced a rupture. One patient with intracranial involvement was died.

Türkiye's rural areas are particularly endemic for CE. Globally, it is estimated that over one million people are infected (21). Preventing unsanitary slaughtering practices, ensuring proper collection and disposal of waste, maintaining adequate environmental hygiene, and informing the public about transmission routes will reduce the emergence of new cases (22). Since dogs play a crucial role in transmission, vaccination programs for infected animals will help reduce the spread of the disease (23).

Study Limitations

Due to the retrospective nature of the study, accessing patient information through medical records and the small number of cases were limitations of the research.

CONCLUSION

In conclusion, CE is a preventable public health issue for our country, and the expenses related to its diagnosis and treatment hold significant importance for the national economy. Individuals presenting at hospitals with various complaints and symptoms in endemic and rural areas should be evaluated for CE. Prevention and control programs are crucial to reducing the incidence of CE. Although there are case series in our country, the number of publications on CE has been decreasing in recent years. More comprehensive, prospective, multicenter studies are needed on this disease, which is endemic in our country.

*Ethics

Ethics Committee Approval: The study was conducted in accordance with the principles of the Declaration of Helsinki. The study was approved by the Clinical Research Ethics Committee of University of Health Sciences Türkiye, Kartal Dr. Lütfi Kırdar City Hospital with the decision numbered 2024/010.99/2/47 on 27.03.2024.

Informed Consent: Retrospective study.

Footnotes

*Authorship Contributions

Concept: S.Ş., B.K., Design: S.Ş., B.K., Data Collection or Processing: S.Ş., B.K., Analysis or Interpretation: S.Ş., B.K., Literature Search: S.Ş., B.K., Writing: S.Ş., B.K.

Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study received no financial support.

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