



Evaluation of Clinical and Epidemiological Characteristics and Treatment Methods of Pediatric Hydatid Cyst Cases

Pediyatrik Kist Hidatik Olgularının Epidemiyolojik ve Klinik Özelliklerinin ve Tedavi Rejimlerinin Değerlendirilmesi

Yalçın Kara¹(ID), Mahmut Can Kızıl¹(ID), Merve İşeri Nepesov¹(ID), Çiğdem Öztunalı²(ID), Mehmet Surhan Arda³(ID), Hüseyin İlhan³(ID), Nihal Doğan⁴(ID), Ömer Kılıç¹(ID), Ener Çağrı Dinleyici⁵(ID)

¹ Department of Pediatric Infectious Diseases, Eskişehir Osmangazi University Faculty of Medicine, Eskişehir, Türkiye

² Department of Radiology, Eskişehir Osmangazi University Faculty of Medicine, Eskişehir, Türkiye

³ Department of Pediatric Surgery, Eskişehir Osmangazi University Faculty of Medicine, Eskişehir, Türkiye

⁴ Department of Microbiology, Eskişehir Osmangazi University Faculty of Medicine, Eskişehir, Türkiye

⁵ Division of Pediatric Intensive Care, Eskişehir Osmangazi University Faculty of Medicine, Eskişehir, Türkiye

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Abstract

Objective: Hydatid cyst is a common zoonotic disease caused by echinococcal cestodes. The diagnosis of hydatid cysts is based on clinical symptoms, serology, and radiological findings. There are treatment options such as medical and surgery. In this study, we evaluated the clinical, epidemiological, laboratory, and radiographic findings of pediatric hydatid cysts.

Material and Methods: Eighty-four pediatric cases followed between 2012-2022 in our hospital were included. Epidemiological and clinical features of the cases were evaluated.

Results: Fifty (59.5%) of the 84 patients were males, and mean age was 137 months. Forty-eight (57.1%) of the patients were from rural areas, and 38 (45.2%) had animal contact. Indirect hemagglutination test was positive in 54.8% of the cases, and 48.8% had eosinophilia. Lung cysts were found in 41.7% of the patients, liver cysts in 32.1%, lung and liver cysts in 23.8% of the patients. Surgery and albendazole treatment were applied in 71.4% of patients, only medical therapy in 25%, and PAIR in one case. While the risk of rupture was higher in patients with lung cysts, IHA positivity was higher in patients with liver cysts.

Conclusion: Hydatid cyst continues to be a public health problem for the whole world. Educating the society and taking measures to prevent the spread and transmission are the most important methods of protection. Although it is not a standard treatment regimen, cyst size, location and number are important in the decision of surgery or medical treatment.

Keywords: Hydatid cyst, child, treatment

Öz

Giriş: Hidatik kist, ekinokokal sestodların neden olduğu yaygın zoonotik bir hastalıktır. Hidatik kistlerin tanısı klinik, seroloji ve radyolojik bulgulara dayanır. Medikal ve cerrahi tedavi seçenekleri mevcuttur. Bu çalışmada pediyatrik hidatik kist olgularının klinik, epidemiyolojik özelliklerini, laboratuvar ve radyografik bulgularını değerlendirmeyi amaçladık.

Gereç ve Yöntemler: 2012-2022 yılları arasında hastanemizde takip edilen 84 pediyatrik kist hidatik olgusu çalışmaya dahil edildi. Olguların epidemiyolojik ve klinik özellikleri değerlendirildi.

Bulgular: Seksen dört hastanın 50 (%59.5)'si erkekti ve yaş ortalaması 137 aydı. Hastaların 48 (%57.1)'i kırsal kesimden ve 38 (%45.2)'inin hayvan teması vardı. İndirekt hemagglütinasyon testi olguların %54.8'inde pozitif ve %48.8'inde eozinofili vardı. Hastaların %41.7'sinde akciğer kisti, %32.1'inde karaciğer kisti, %23.8'inde akciğer ve karaciğer kisti saptandı. Hastaların %71.4'üne cerrahi ve albendazol tedavisi, %25'ine sadece medikal tedavi ve bir olguya PAIR uygulandı. Akciğer kisti olan hastalarda rüptür riski daha yüksek bulunurken, karaciğer kisti olan hastalarda İHA pozitifliği daha yüksekti.

Sonuç: Hidatik kist tüm dünya için bir halk sağlığı sorunu olmaya devam etmektedir. Toplumun eğitimi, bulaşmayı ve yayılımı önleyici tedbirlerin alınması, kist hidatikten en önemli korunma yöntemidir. Standart bir tedavi rejimi olmamakla birlikte cerrahi veya medikal tedavi kararında kistin boyutu, yeri ve sayısı önemlidir.

Anahtar Kelimeler: Kist hidatik, çocuk, tedavi

Correspondence Address/Yazışma Adresi

Yalçın Kara

Eskişehir Osmangazi Üniversitesi Tıp Fakültesi,
Çocuk Enfeksiyon Hastalıkları Anabilim Dalı,
Eskişehir, Türkiye

E-mail: dryalcinkara@hotmail.com

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Introduction

Hydatid cyst is a common zoonotic disease caused by echinococcal cestodes. It is one of the 17 neglected tropical illnesses listed by the World Health Organization (WHO), impacting more than one million people and generating a financial loss of more than three billion dollars annually (1,2). Our country is one of the high endemic regions where more than 50-400 patients per hundred thousand are seen each year. There are four types: *E. granulosus*, *E. multilocularis*, *E. vogeli*, and *E. oligarthus*. *E. granulosus* is the most common species in our country and worldwide. People get it when they eat echinococcal eggs spread by the feces of infected dogs (3). Hydatid cysts are mostly asymptomatic; however, depending on their location in the body, they may present clinical symptoms. Findings often happen as a result of the cyst's rising pressure. It occurs in 60% of the liver, 30% of the lungs, and 10% of other organs (4). Cysts can be found in all organs where echinococcal larvae settle (5). According to the research, liver hydatid cysts are more prevalent in adult patients, but lung hydatid cysts are more common in pediatric patients. The diagnosis of hydatid cysts is based on clinical symptoms, serology, and radiological imaging. Radiological methods play a significant role in disease diagnosis, classification, treatment, and follow-up (6). Due to its low cost, high sensitivity, and high specificity, ultrasonography is the most critical imaging method utilized in patients, community screenings, and the classification of liver hydatid cysts (7). Serological tests diagnose, follow up after surgery or medical therapy, and evaluate the prognosis of hydatid cyst disease (8-10). There are treatment options such as medical, percutaneous, surgical, and follow-up, and the most appropriate therapy is decided by the cyst's anatomical location, size, and classification (11,12). In this study, we evaluated the clinical, epidemiological, laboratory, and radiographic findings and treatment regimens of pediatric hydatid cysts.

Materials and Methods

The study included pediatric patients with hydatid cysts who were followed up by the pediatrics clinic, and pediatric surgery clinic between 2012 and 2022. The cases' epidemiological and clinical characteristics, laboratory and radiological results, and treatment regimens were reviewed retrospectively. Age, sex, complaints, physical examination findings, and laboratory findings were studied for the diagnosis of the cases; hemogram, eosinophil, indirect hemagglutinin values, ultrasonography, computerized tomography, and magnetic resonance results from radiological diagnostic tests, and treatment modalities were retrospectively reviewed from the hospital information system. Eosinophil count $>500/\mu\text{L}$ was accepted as eosinophilia. Anemia was defined as hemoglobin below -2SD of normal for age according to WHO

criteria. Indirect hemagglutination (IHA) test was positive if the titer was more than 1:160. Gharbi score was used to classify liver hydatid cysts (Type-1: Simple fluid collection, Type-2: Appearance showing separation of membranes, Type-3: Cystic appearance with daughter vesicles, Type-4: Heterogeneous echo structure and solid appearance, Type-5: Thick-walled calcified cyst appearance). Anatomical location, size, number, and a class of the cysts according to the Gharbi score, and treatment regimens were compared (2,6). The study was started after the approval of University Ethics Committee (Decision no: 1322, Date: 06.01.2023).

Statistical Analysis

Statistical Package for Social Sciences (SPSS) version 18.0 for Windows (SPSS, Chicago, IL, USA) was used for statistical analysis. Continuous variables were expressed as median (minimum-maximum). After assessing normal distribution with the Kolmogorov-Smirnov test, parameters were compared between the groups with the Mann-Whitney U test for continuous variables and a chi-square test for categorical variables. p values of <0.05 were considered statistically significant.

Results

Fifty (59.5%) of the 84 patients were males, and mean age was 137 (15-216) months. Forty-eight (57.1%) of the patients were from rural areas, and 38 (45.2%) had animal contact. The most common symptoms at presentation were cough (58.3%), abdominal pain (40.5%), dyspnea (26.2%), chest pain (17.9%), fever (17.9%), hemoptysis (13.1%) and vomiting (13.1%). There was a history of anaphylaxis in 16.7% of the cases and cyst rupture in 17.9%. In laboratory examinations, mean hemoglobin (hb) was 12.6 mg/dL (4-17.2), leukocytes was $9434 \pm 336/\text{mm}^3$, and eosinophile was $630/\text{mm}^3$ (0-3600). Indirect hemagglutination test was positive in 54.8% of the cases, 14% had anemia, and 48.8% had eosinophilia. Lung cysts were found in 41.7% of the patients, liver cysts in 32.1%, lung and liver cysts in 23.8%, bladder cysts in one patient, ovarian cysts in one patient, and heart cysts in one patient. While 74.5% of the cases had a single cyst, 25.5% had multiple cysts. Of the patients with lung cysts, 58.2% had right lung cysts, 30.9% had left lung cysts, and 10.9% had bilateral cysts. Ultrasonography was performed in 95.2% of the cases, computed tomography was performed in 66.7%, and magnetic resonance was performed in 10.7% of the cases. According to the Gharbi scoring system, 44.7% of the hydatid cysts were Type-3, 27.7% were Type-2, 10.6% were Type-1, 10.6% were Type-5, and 6.4% were Type-4. Cyst sizes were >5 cm in 75% of cases with lung cysts and <5 cm in 42%. 59.3% of the cases with liver cysts were >5 cm, and 40.7% were <5 cm. Surgery and albendazole treatment were applied in 71.4% of patients, only medical therapy in 25%, and PAIR in one case. Mean duration of medical treatment was 7.3 months (Table 1). In the surgical treatment group, compared

Table 1. Clinical and epidemiological characteristics of the cases

	n= 84 (%)		n= 84 (%)
Sex		Residence	
Boy	50 (59.5)	City	36 (42.9)
Girl	44 (40.5)	Rural	48 (57.1)
Age (month)	137 (15-216)	Animal contact	38 (45.5)
Symptoms-Signs		Size	
Cough	49 (58.3)	Liver	27
Abdominal pain	34 (40.5)	<5cm	11 (40.7)
Dyspnea	22 (26.2)	>5cm	16 (59.3)
Chest Pain	15 (17.9)	Lung	32
Fever	15 (17.9)	<5cm	8 (25)
Hemoptysis	11 (13.1)	>5cm	24 (75)
Vomiting	11 (13.1)	Liver + Lung	20
Hepatomegaly	25 (29.8)	<5cm	14 (70)
Splenomegaly	3 (3.6)	>5cm	6 (30)
Anaphylaxis	14 (16.7)	USG	80 (95.2)
		CT	56 (66.7)
		MRI	9 (10.7)
Rupture	15 (17.9)	Treatment	
Lung	12 (14.3)	Medical	21 (25)
Liver	3 (3.6)	Surgery + Medical	60 (71.4)
		PAIR	1 (1.2)
Laboratory		Gharbi classification (liver)	
Hemoglobin	12.6 ± 0.17	Type-1	47
Anemia	12 (14)	Type-2	5 (10.6)
Leukocyte	9434 ± 336	Type-3	13 (27.7)
Eosinophil	390 (0-3600)	Type-4	21 (44.7)
Eosinophilia	41 (48.8)	Type-5	3 (6.4)
IHA positivity	46 (54.8)		5 (10.6)
Location		Cyst number	
Lung	35 (41.7)	Liver	
Liver	27 (32.1)	Single	34 (72.3)
Lung + Liver	20 (23.8)	Multiple	13 (27.7)
Other	2 (2.4)	Lung	
		Single	41 (74.5)
		Multiple	14 (25.5)
Symptom duration (month)	2.3 (1-24)	Medical treatment duration (month)	6 (1-24)

IHA: Indirect hemagglutination, USG: Ultrasonography, CT: Computed tomography, MRI: Magnetic resonance imaging, PAIR: Puncture, aspiration, injection, reaspiration.

to the medical treatment group, the rate of eosinophilia was higher, and the cyst size was larger ($p= 0.008$, $p= 0.01$) (Table 2). While cough, dyspnea, and chest pain were more common in patients with lung cysts, abdominal pain was more common in people with liver cysts. While the risk of rupture was higher in patients with lung cysts, IHA positivity was higher in patients with liver cysts ($p= 0.045$, $p= 0.025$) (Table 3).

Discussion

Hydatid cyst has been accepted as one of the 17 neglected diseases by WHO, and it continues to be seen as endemic in many countries, including our country (2). In this study, we aimed to evaluate the epidemiological, clinical features and treatment regimens of hydatid cyst cases followed in our clinic. In our study, 57.1% of hydatid cysts lived in rural areas, and 45.2% had contact with animals. The prevalence of

hydatid cysts was found to be 0.59% in the HERACLES study, which was conducted in Bulgaria, Romania, and Türkiye (13). Another study showed that hydatid cysts are 2-6 times more common in rural areas than in urban areas. Another study with children and adults in Bulgaria showed that hydatid cyst cases were more common in rural areas, and animal contact was more prevalent (14,15). This can be explained by the fact that people living in rural areas have a higher risk of animal contact than in urban areas. There must be more than precautions for storing and disposing of slaughtered animal waste; dogs consume animal offal, and hygiene measures are less. Tamarozzi et al. have noted in their multicenter study that hydatid disease is not only a food-borne infestation but also a public health problem that is spread by soil and through household contact (15). Altıntaş et al. have focused on social education and distinction in hydatid cyst prevention and

Table 2. Comparison of the cases according to treatment types

	Surgery, n= 62 (%)	Medical, n= 21 (%)	
Sex			0.32
Girl	24 (38.7)	10 (47.6)	
Boy	38 (61.3)	11 (52.4)	
IHA	36 (60)	30 (42)	0.15
Eosinophilia	36 (60)	4 (23.5)	0.008
Anatomy			0.38
Lung	29 (46.8)	6 (28.6)	
Liver	17 (27.4)	9 (42.9)	
Lung + Liver	15 (24.2)	5 (23.8)	
Other	1 (1.6)	1 (4.8)	
Gharbi classification	32	14	0.61
Type-1	2 (6.3)	3 (21.4)	
Type-2	10 (31.3)	3 (21.4)	
Type-3	14 (43.8)	6 (42.9)	
Type-4	2 (6.3)	1 (7.1)	
Type-5	4 (12.5)	1 (7.1)	
Number			0.21
Single	45 (72.6)	12 (60)	
Multiple	17 (27.4)	8 (40)	
Size			0.1
<5 cm	17 (28.8)	17 (81)	
>5 cm	42 (71.2)	4 (19)	

IHA: Indirect hemagglutination.

reported that educating the public, teachers, health workers, and school children is the most critical step, especially in rural areas (16). In our study, in parallel with the literature, there was a high rate of rural life and animal contact. This emphasizes the importance of hygiene measures and of education.

In our study, the most common hospitalization symptoms were cough, abdominal pain, and respiratory distress. Median duration from first symptom onset to hospital admission was 2.3 (1-24) months. Tuz et al. reported that the most common symptoms during the presentation were abdominal pain, cough, vomiting, and chest pain (17). Another study by Jia et al. showed that abdominal pain was the most prevalent presenting symptom (18). The symptoms of a hydatid cyst vary depending on the location of the cyst, its size, and the pressure on the surrounding tissues and organs. Our study's higher incidence of cough is due to the high prevalence of pulmonary cyst hydatid disease. Mean duration of hospital admission for the patients was 2.3 months, which resulted in the cysts being big and widespread due to late entry, which increased the effects of rupture and compression.

Clinical, radiographic, and serological tests are used in diagnosing hydatid cysts. In our study, IHA positive was 54% higher in patients with liver involvement than lung involvement. There are very different results about the sensitivity of IHA in the literature, and the sensitivity of the test varies between 60-100%. In a study of pediatric patients, Oral et al. showed that IHA positive was 75% (19). Koca et al. found

that 54% of cases of pediatric hydatid cysts were IHA positive (20). Some studies show an increase in the number and size of cysts and IHA positive, but no association was detected in our study (8). Higher IHA positivity has been reported in liver cysts than in lung and other organ cysts (8,21). In our study, like in the literature, patients with liver cysts had a higher IHA positivity. This was explained by the fact that liver tissue had a stronger immune response to echinococci. Eosinophilia was found in 48.8% of the cases in our study. Various studies have shown that the rate of eosinophils in cases of hydatid cysts is between 20-34%. However, its diagnostic value is not very significant and is extremely high in ruptured cyst fluid (22,23). In our study, we found a higher rate of eosinophils than the literature, which was related to a higher rate of lung and liver cyst rupture.

In our analysis, 41.7% of the patients had lung cysts, 32.1% had liver cysts, 23.8% had lung and liver cysts, there was bladder cyst in one patient, ovary in one patient, and cardiac cyst in one patient. While 74% of the cases had a single cyst, 26% had multiple cysts. According to the literature, 60% of hydatid cysts are found in the liver, 30% in the lung, 10% in other organs, and 10-20% involve more than one organ. While the liver is the most commonly involved organ in adults, the lung is more commonly involved in children (5,24). Jordanova et al. reported that the most common organ involvement was the liver (14). Although hydatid cysts are most commonly diagnosed as single in children, multiple cysts have been reported to occur at

Table 3. Characteristics of liver and lung hydatid cysts

	Lung, n= 35 (%)	Liver, n= 27 (%)	p
Sex			0.480
Girl	13 (37.1)	11 (40.7)	
Boy	22 (62.9)	16 (59.3)	
Residence			0.130
City	19 (54.3)	10 (37)	
Rural	16 (45.7)	17 (63)	
Animal contact	14 (40)	13 (48.1)	0.350
Symptom-Sign			
Cough	31 (88.6)	1 (3.7)	0.001
Abdominal pain	1 (2.9)	25 (92.6)	0.001
Dyspnea	16 (45.7)	0 (0)	0.001
Chest pain	10 (76.9)	0 (0)	0.015
Fever	8 (22.9)	3 (11.1)	0.195
Hemoptysis	6 (17.1)	0 (0)	0.026
Vomiting	3 (8.6)	4 (14.8)	0.350
Anaphylaxis	6 (17.1)	4 (14.8)	0.540
Rupture	6 (17.1)	3 (11.1)	0.045
IHA	13 (38.2)	18 (66.7)	0.025
Eosinophilia	14 (42.4)	16 (59.3)	0.086
Size			0.156
>5 cm	24 (75)	16 (59.3)	
<5 cm	8 (25)	11 (40.7)	
Number			0.605
Single	26 (74.3)	20 (74.1)	
Multiple	9 (25.7)	7 (25.9)	
Treatment			0.100
Medical	6 (17.1)	9 (33)	
Surgery	29 (82.9)	17 (65.4)	

IHA: Indirect hemagglutination.

a rate of 9-30%. Hallaç Keser et al. reported that 15.6% of cystic hydatid cases had organ involvement other than liver and lung (25). In our study, as in the literature, the most common organ involvement was lung and liver, respectively, with just three cases having involvement outside of these two organs (heart, bladder, ovary) and 74% having a single cyst.

There is no standard therapy for hydatid cysts in children; however, there are medical, surgical, and percutaneous treatment regimens depending on the cyst's location, size, and number (26,27). In our study, only medical treatment was given to 25% of the cases, surgical and medical treatment was applied to 71.4%, and PAIR was applied to one case. The main treatment approach in lung hydatid cysts is surgery, and multicenter studies by the World Health Organization (WHO) investigating the effects of drugs have reported conflicting results about the effectiveness of medical treatment. According to Sarkar et al., medical therapy decreased cyst size and pressure and was successful in 70% of cases (28). Publications show that medical therapy damages the cyst membrane and raises the risk of lung cyst rupture (29,30). In our study, as in the literature, 82.9% of lung hydatid cysts

had surgical and medical treatment, whereas 17.1% received only medical treatment. In liver hydatid cysts, treatment regimen varies according to the size and number of the cyst. According to the WHO classification, percutaneous treatment is suggested for CE1 and CE3a cysts, surgical treatment is advised for CE2 and CE3b cysts, and treatment is not recommended for CE4 and CE5 cysts. According to the Gharbi score, PAIR therapy is advised for Type-1 and Type-2 cysts, and surgical treatment is indicated for Type-2-Type-3 cysts. Treatment is not recommended for Type-4 and Type-5 cysts because they are inactive (2,6). The Gharbi score method was also used in our study, and 44.7% of the cases were classified as Type-3, 27.7% as Type-2, and 10.6% as Type-1 and Type-5 hydatid cysts. Surgical treatment was applied to 65.4% of liver hydatid cysts, medical treatment was applied to 33%, and PAIR was applied to one case. Surgical treatment was applied, and 43.8% of the cases were Gharbi-3, and 31.3% were Gharbi-2, unlike the literature, PAIR treatment was applied only in one case. This can be explained by the high number of cases of lung hydatid cysts and the high rate of cases with cyst sizes more than 5 cm.

The use of benzimidazoles known as albendazole and mebendazole is recommended in the medical treatment of hydatid cysts. However, there is no consensus on the optimal duration of their use. There are few studies in the literature on the appropriate indication, duration, and safety of these drugs in children. In a multicenter study conducted in Greece, it was recommended to administer albendazole treatment for one to four weeks prior to surgery and to continue medical treatment for one to three months after surgery (25). Another study reported that albendazole treatment should be continued for 3-6 months, and up to two years for pulmonary hydatid cysts, depending on the type of cyst (6,17). In our study, the median duration of albendazole use was six months, and there was no significant difference in the duration of use between liver and lung cysts. Similarly, there was no significant difference in the duration of albendazole use between patients who underwent surgery and those who received only medical treatment. These findings highlight the need for large-scale, multicenter studies to determine the optimal duration of albendazole use.

Our study's main limitations are that it was retrospective, covered ten years, some laboratory and radiological data could not be accessed, and it was a single centre.

In conclusion, hydatid cyst still continues to be a public health problem for our country and many other countries. Especially in pediatric cases with liver and lung cysts, the history of rural life and animal contact should be questioned and hydatid cyst should be kept in mind in its etiology. Although the most important factors in determining the treatment of hydatid cyst are the anatomical location of the cyst, its size and compression symptoms, unfortunately there is no standard treatment regimen for hydatid cysts can be used worldwide. Multicenter, multinational, and big participation trials are required.

Ethics Committee Approval: This study was approved by Eskişehir Osmangazi University Non-Invasive Clinical Research Ethics Committee (Decision no: 30, Date: 20.12.2022).

Informed Consent: Patient consent was obtained.

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