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INTRODUCTION

Hydatid cyst is one of the most common parasitic diseases caused by *Echinococcus granulosus*. Although it can be seen worldwide, it is a significant health problem in developing countries such as Türkiye. The incidence of the disease in humans is generally low, and a significant portion of human cases cannot be diagnosed. The World Health Organization (WHO) estimates that more than one million people worldwide are infected with *Echinococcus* each year (1). The incidence frequency varies based on the endemic nature of different regions. Notably, high endemic areas include a number of regions such as numerous African countries, the Middle East, Mediterranean countries, Central Asia, South

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Abstract

Aim: Hydatid cyst is one of the most common parasitic diseases caused by *Echinococcus granulosus*. Although it can be seen worldwide, it is a significant health problem in developing countries such as Türkiye. In this study, we reviewed the epidemiological and clinical data of cystic echinococcosis cases in a tertiary hospital and reviewed current trends in the diagnosis and treatment of the disease.

Materials and Methods: A total of 41 patients were included in the study between January 1, 2008, and December 31, 2022.

Results: In the retrospectively planned study, 16 (39%) of the patients were female, and the mean age was determined to be 47.4±11.7 years. Twenty-seven (65.9%) of the patients were living in rural areas. The most affected organ was the liver (48.8%). Multiple organ involvement was observed in 2 cases. The diagnosis of most patients was made using ultrasound (73.2%). Anemia (22%) was the most frequently detected finding in the laboratory tests performed before treatment. Other laboratory findings were ranked as eosinophilia and leukocytosis. The treatment of the majority of patients was done both medically and surgically (75.6%). Thirty-four patients (82.9%) did not have a recurrence. Among the 7 patients who had a recurrence, the first site of recurrence was observed in the liver in 3 of them.

Conclusion: Hydatid cyst is still an important disease in our country due to its impact on both human and animal health and serious economic losses in areas engaged in animal husbandry.

Keywords: *Echinococcus granulosus*, cystic echinococcosis, hydatid cysts, parasitic disease

America, and parts of Western China (2). In Türkiye, the true prevalence is not fully known due to the extremely limited prevalence of research studies. However, the HERACLES project supported by the European Union 7th Framework Program revealed a prevalence rate of 6% (3).

Humans are only accidental intermediate hosts. The disease is transmitted by eating infected unwashed vegetables and undercooked meat, and by swallowing the parasite's eggs (4). After oral intake, the embryos pass through the intestinal mucosa and reach the liver through the portal vein. They then metastasize and spread to neighboring areas such as the abdomen, retroperitoneum, or distant organs such as the lungs, brain, and

bones, forming cystic lesions in many anatomical regions. The liver and lungs are the most well-known sites of involvement (5).

Diagnosis mainly relies on the clinical presentation and symptoms. Ultrasonography is particularly important in the classification of hydatid cysts. If there is involvement in an area where ultrasound cannot be performed, Computerized Tomography (CT) or Magnetic Resonance Imaging (MRI) is used. In addition, the detection of specific antibodies plays an important role in the diagnosis of cystic echinococcosis. The guidelines prepared WHO working group aim to facilitate patient management by classifying patients (6,7).

Although some types can be successfully treated with percutaneous aspiration, injection, and re-aspiration, the surgical approach remains the preferred treatment. Albendazole and mebendazole are recommended for medical treatment (8-10).

MATERIALS AND METHODS

Our hospital is a heart, lung, and liver transplant center. Patients are admitted to cardiovascular surgery, chest surgery, and gastroenterology surgery clinics from many health centers.

In this study, we reviewed the epidemiological and clinical data of cystic echinococcosis cases in a tertiary hospital between January 1, 2008, and December 31, 2022, and reviewed current trends in the diagnosis and treatment of the disease. In this retrospective study, the diagnostic aspect encompassed various techniques including imaging modalities such as ultrasound, CT, and MRI, as well as serological assessments employing the indirect hemagglutination test. For medical intervention, treatment options involved the administration of oral mebendazole or albendazole.

Radical surgical procedures for liver cysts include total cystectomy (pericystectomy), liver resections, or lobectomies. Conservative surgical methods applied include partial cystectomy, external drainage, omentoplasty, unroofing, introflexion, capitonage, capsule repair, and marsupialization.

For cysts in the heart, standard cardiopulmonary bypass with bicaval venous cannulation was initiated with mild hypothermia following median sternotomy and potassium crystalloid cardioplegia was used to induce cardiac arrest and protect the myocardium. The surgical approach varies according to the location of the cyst. Cysts in the atria and interatrial septum were reached by right atriotomy. A ventriculotomy was performed at the apex to reach the cysts in the interventricular septum. Superficial cysts on the right or left ventricle were treated with a direct approach. After reaching the cyst, the surrounding tissues were protected with a gauze soaked in 30% hypertonic NaCl solution, and the same solution was injected into the cyst. After the cyst content was aspirated with a needle, the cyst was opened, the germinal membrane and remaining cyst material were completely removed, and the cavity was washed for the last time with hypertonic solution. For cysts located in the myocardium, the cavity was closed by passing individual U-sutures with Teflon

strip support through the myocardium and inner cavity wall. In cysts located in the interatrial septum, the defect was repaired with a pericardial patch after removal of the cyst.

For lung cysts, a complete surgical treatment consisting of cystectomy, wide resection, and reconstruction of pericystic, and adjacent tissues was applied.

This study received approval from the Ethics Committee of the University of Health Sciences Kartal Koşuyolu Research and Training Hospital, adhering to the principles outlined in the Declaration of Helsinki (1964). Ethical consent was obtained from the Non-Interventional Clinical Research Ethics Committee of Koşuyolu Heart High Research and Training Hospital, denoted by Approval Decision No. 2023-03-13, dated January 31, 2023.

Statistical Analysis

Regarding the statistical methodology, the descriptive analysis of the data involved metrics such as mean, standard deviation, median, minimum, maximum, frequency, and ratio values. The analysis itself was conducted employing the SPSS 28.0 software program.

RESULTS

A total of 41 patients were included in the study 16 (39%) of the patients were female, and the mean age was determined to be 47.4±11.7 years (minimum-maximum 24-73). Twenty-seven (65.9%) of the patients were living in rural areas. The most affected organ was the liver (n=20, 48.8%), heart (n=10, 24.4%), lungs (n=6, 14.6%), spleen (n=2, 4.9%), Heart-Liver simultaneously (n=1, 2.4%), Liver-Spleen (n=1, 2.4%), Pleura (n=1, 2.4%) (Table 1). The investigation identified 11 distinct sites of heart involvement. Specifically, these encompassed 2 instances within the interventricular septum, 2 cases affecting the right atrium, 2 occurrences in the interatrial septum, 1 case related to the right ventricle, and 3 instances observed within the left ventricle. Furthermore, a patient with a liver hydatid cyst experienced the occurrence of a biliary fistula. Subsequent to surgical intervention, 8 patients received treatment within the post-operative phase in the intensive care unit, ultimately resulting in their discharge.

In the subsequent follow-ups, no complications developed in the patients with two heart involvements, liver involvement, and lung involvement, who were lost for various reasons. Multiple organ involvement was observed in 2 cases. The diagnosis of most patients was made using ultrasound (73.2%), and CT (19.5%). Anemia (n=9, 22%) was the most frequently detected finding in the laboratory tests performed before treatment. Other laboratory findings were ranked as eosinophilia (n=5, 12%) and leukocytosis (n=7, 17%). The treatment of the majority of patients was done both medically and surgically (75.6%). Thirty-four patients (82.9%) did not have a recurrence. Among the 7 patients who had a recurrence, the first site of recurrence was observed in the liver in 3 of them (Table 1).

Table 1: Demographic Information (n=41)					
		Min-Max	Median	Mean±sd	n-%
Age		24.0-73.0	48.0	47.2±11.7	
Gender	Female			16	39.0%
	Male			25	61.0%
Residence	Rural			27	65.9%
	Urban			14	34.1%
Affected organ	Liver			20	48.8%
	Heart			10	24.4%
	Lungs			6	14.6%
	Spleen			2	4.9%
	Heart-liver simultaneously			1	2.4%
	Liver-spleen			1	2.4%
Radiology	Pleura			1	2.4%
	CT			8	19.5%
	CT Thorax			1	2.4%
	ECHO			1	2.4%
	MRI			1	2.4%
	USG			30	73.2%
WBC (x10³)		3300-21700	8100	9094±3023	
HB (g/dl)		4.2-15.7	12.6	12.2±2.2	
Eosinophil (x10³)		0.0-5700	300.0	591.5±1055.7	
Sedimentation (mm/hour)		5.0-97.0	21.0	32.4±23.4	
CRP mg/dL		0.3-13.0	6.0	5.6±4.1	
Monocyte (x10³)		20.0-1350.0	90.0	286.0±338.0	
Neutrophil (x10³)		830.0-14300.0	5350.0	5548.8±2581.9	
Treatment	Surgical+medical			31	75.6%
	Medical			10	24.4%
Operation	Absent			11	26.8%
	Present			30	73.2%
Recurrence	Absent			34	82.9%
	Present			7	17.1%
Site of recurrence	Heart first			1	2.4%
	Liver first			3	7.3%
	Unknown			3	7.3%
Results	Discharge			41	100.0%

CT: computed tomography, ECHO: extracorporeal membrane oxygenation, MRI: magnetic resonance imaging, USG: ultrasonography

DISCUSSION

Cystic echinococcosis remains a serious public health problem worldwide, especially in Mediterranean countries including Türkiye where the disease is endemic. In a multicenter study conducted in our country, the mean age was shown to be 45.4 ± 17.4 (4), while in another study it was found to be 53.5 ± 19.4 (10), and in another study that included 448 people, it was 40.6 ± 20.58 (11). In our study, the mean age presented was 47.2 ± 11.7 , which is close to the values of other studies. Similarly, the disease is thought to be more prevalent in the middle age group due to the long asymptomatic period after contact with the parasite.

Studies conducted in Türkiye have also shown a higher proportion of females among those who tested positive for the disease. In a study conducted in Erzincan, of the positive cases included in the study between 2010-2018, 99 were male (42%) and 137 were female (58%) (12). In the study by Bağcı et al., seropositivity was found to be 14.81% in males and 13.65% in females, and the difference was not statistically significant (13). Similarly, the HERACLES study reported no significant difference in the incidence of the disease between men and women in Türkiye (13). In our study, the gender distribution was different, with 61% of cases being male and 39% female. It appears that positivity is higher among males.

When examining the organs affected by the disease, the liver and lungs were found to be the most affected, although it has been shown that many other organs and tissues can be affected as well. In hydatid cysts, tissues outside the liver and lungs are considered unusual locations. In a study conducted in Iran, unusual cystic echinococcosis locations were investigated, and it was observed that the most common organ affected was the spleen (30.4%). This rate was calculated as 2.8% of all cystic Echinococcosis cases (14,15). Cardiac involvement was seen in 8.7% of patients (16). In our study, while the liver (48.8%) was found to be the most affected organ, the heart (24.4%), which is among the unusual locations, was found to be the second most common location of involvement. The lung was in second place, followed by the spleen (4.9%) among the unusual locations. The fact that cardiac involvement was second in all cases can be explained by our center being a hospital specializing in cardiovascular diseases. It should also be kept in mind that hydatid cyst disease can affect the chambers of the heart, regardless of whether it involves the liver or lungs (17).

Non-specific laboratory abnormalities may be observed with Echinococcus infection. In the study conducted by Akkaya et al., the most common laboratory finding in cases was anemia (25.3%), while eosinophilia was seen in 19% of cases (18). Anemia (n=9, %) was the most observed laboratory finding before treatment. Other laboratory findings were eosinophilia (n=5, %) and leukocytosis (n=7, %). In a retrospective study conducted in Greece, which included 287 patients, eosinophilia was observed in 35% of patients, and abnormal liver function was found in 43% of patients. However, whether the patients

were complicated or uncomplicated was not specified (19). In a study from Türkiye, eosinophils were found to be significantly higher in patients with complicated cysts (rupture or fistula) (35.3%) (20). In our study, eosinophilia (absolute eosinophil count >500 /cubic millimeter) was observed in 11 (26%) patients, with a maximum absolute eosinophil count of 5700/cubic millimeter. Another study investigating the relationship between eosinophilia and cystic echinococcosis found similar results to ours, with eosinophilia detected in 24.8% and 25% of patients (20,21). In asymptomatic patients, the hemogram may be an indicator for active screening of Echinococcus. However, in another study conducted in Nepal on cases of hepatic hydatid cysts, only 13.2% of patients presented with eosinophilia. This data suggests that eosinophilia is not mandatory for the diagnosis of Echinococcus infection, contrary to other studies (22,23).

According to the articles, ultrasound, which is the gold standard test for diagnosing cystic echinococcosis in humans, can also detect the location, number, and size of cysts, as well as their prevalence (24,25). Additionally, compared to serologic tests, ultrasound is a preferred method for screening for cystic Echinococcosis due to its higher specificity and sensitivity (26).

CT has also been reported to provide more accurate information by identifying small cysts and providing a detailed view of lesion characteristics, intra- and extra-hepatic vascular and ductal structures, and anatomical relationships including extrahepatic lesions (27). In an 88-patient series evaluating diagnosis and treatment, all patients underwent ultrasound, and only 28.86% required additional CT imaging (28). The patients in our study were generally diagnosed and referred to our hospital for surgical procedures. Due to its accessibility and ease, we also diagnosed 73.2% of cases in our study with ultrasound. The second most common method for diagnosis was CT, with a rate of 15.5% in accordance with the literature.

In a study conducted in 2020, a model was developed and analyzed for transmission dynamics. It was observed that the most sensitive parameter with a significant impact on disease transmission was the rate of Echinococcus eggs spreading from the environment to sheep. Thus, an effective control strategy that reduces the rate of Echinococcus eggs spreading from the environment to sheep was highlighted as crucial for controlling disease transmission (29).

CONCLUSION

In conclusion, the hydatid cyst is still an important disease in our country due to its impact on both human and animal health and serious economic losses in areas engaged in animal husbandry. Recognition of the in-depth evaluation of cystic echinococcosis cases in humans may be valuable in developing appropriate epidemiological and control programs to reduce parasite transmission to humans and planning randomized prospective studies. The main treatment for hydatid disease is surgical removal of the cysts. Conservative surgical approaches are safe methods, and for most patients it is effective.

Conflict of Interests

The authors declare that there is no conflict of interest in the study.

Financial Disclosure

The authors declare that they have received no financial support for the study.

Ethical Approval

Ethical consent was obtained from the Non-Interventional Clinical Research Ethics Committee of Koşuyolu Heart High Research and Training Hospital, denoted by Approval Decision No. 2023-03-13, dated January 31, 2023.

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