

Evaluation of pulmonary hydatid cysts in children: 5-year data from a single center

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ABSTRACT

Aims: The aim of this study was to evaluate the demographic characteristics, clinical symptoms, complications, and treatment options of patients with pulmonary hydatid cysts.

Methods: A retrospective chart review of all pediatric patients (age <18 years) diagnosed with pulmonary hydatid cysts between January 1, 2019 and June 30, 2024 was performed. Patients were divided into two groups: patients with ruptured cyst and patients with unruptured cyst. All data were compared between the two groups.

Results: A total of 44 patients with a mean age of 116.01±44.41 months were included in the study. The most common symptom was cough (n=38, 86.4%), followed by chest pain (n=22, 50%). The most common complication was cyst rupture (n=10, 22.7%), and followed by pleural effusion (n=9, 20.5%). Forty-two (95.5%) patients underwent surgery and 14 (31.8%) patients required admission to the intensive care unit admission after surgery. Hemoptysis, cyst diameter, intensive care unit admission, length of stay, and recurrence were significantly more associated with cyst rupture.

Conclusion: Pulmonary hydatid cysts should be considered in the differential diagnosis of children presenting with lower respiratory tract symptoms such as fever, cough, hemoptysis, and chest pain, especially in regions where echinococcosis is endemic. Pulmonary hydatid cysts can lead to life-threatening complications such as bronchial rupture. Hemoptysis and larger cyst diameter are associated with bronchial rupture.

Keywords: Childhood, cystic echinococcosis, pulmonary hydatid cyst, rupture

INTRODUCTION

Cystic echinococcosis, or hydatid disease, is a zoonosis caused by the larval stage of the cestode *Echinococcus granulosus*.¹ The life cycle of *Echinococcus granulosus* typically involves dogs as the definitive host and ruminants (e.g., sheep, goats) as intermediate hosts. Humans serve as incidental intermediate hosts by acquiring infection through ingestion of food contaminated with dog-laid eggs.^{2,3} It is endemic in South America, East Africa, Australia and Mediterranean countries and still poses a health problem especially in the eastern and southeastern parts of Türkiye.^{4,5}

In contrast to adults, the lung is the most common localizing organ in children. Hydatid cysts may rarely occur in organs other than the lung and liver, such as the spleen, kidney, brain, and soft tissues.⁶ Hydatid cysts grow more rapidly in the lung than in the liver. This is due to the elastic structure of the lung and the negative intrathoracic pressure.⁷ Rupture is the main

complication of cysts and may be related to the presence of symptoms, and to morbidity and mortality. The diameter and the location of the cyst has been proposed as risk factors for cyst rupture. Pulmonary hydatid cysts (PHCs) are more likely to rupture than hepatic hydatid cysts.⁸ In children, PHCs often cause respiratory symptoms such as cough, hemoptysis, chest pain, shortness of breath, and fever.¹

The aim of this study was to evaluate the epidemiologic characteristics, clinical symptoms, complications, and treatment options of patients with PHCs.

METHODS

Ethical approval was obtained from the Institutional Ethics Committee of Necmettin Erbakan University Faculty of Medicine (Date: 18.10.2024, Decision No: 2024/5260). All procedures were carried out in accordance with the ethical rules and the principles of the Declaration of Helsinki.

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A retrospective chart review of all pediatric patients (age <18 years) diagnosed with PHC at Necmettin Erbakan University Faculty of Medicine, Pediatric Pulmonology Clinic between January 1, 2019 and June 30, 2024 was performed. Patients with (1) compatible histopathologic findings, (2) positive serology with compatible clinical presentation, or (3) a clinical presentation, epidemiology, and imaging compatible with PHC were included in the study. Patient demographics, presenting symptoms, clinical findings, computed tomography (CT) findings (cyst location, diameter), treatment modalities (surgical, medical), complications, length of hospital stay, history of recurrence and intensive care unit (ICU) admission were recorded.

Statistical analyses

The Kolmogorov-Smirnov test was used to determine the normality of the distribution. Continuous data were presented as mean±standard deviation for variables with a normal distribution and as median and interquartile range (IQR) for variables with a non-normal distribution. Categorical data were presented as frequencies and percentages. The patients were divided into two groups as patients with ruptured cyst and patients with unruptured cyst. Student's t-test was used to compare two groups of continuous data and chi-squared test was used to compare categorical data. MedCalc statistical software (MedCalc Software Ltd., Ostend, Belgium; <https://www.medcalc.org>), version 20.110 was used for all analyses. A p-value of <0.05 was considered statistically significant.

RESULTS

A total of 44 patients diagnosed with PHC were included in the study. The mean age was 116.01±44.41 months. Twenty-four (54.5%) patients were male and 20 (45.5%) were female. Twenty-eight (63.6%) patients were from rural areas and 16 (36.4%) were from urban areas.

Two (4.5%) patients were asymptomatic. The most common symptom was cough (n=38, 86.4%), followed by chest pain (n=22, 50%). There were 8 (18.2%) patients who had two cysts. The most common lung involvement was found in right lower lobe with 13 (29.5%) patients. Twenty (45.5%) patients had liver involvement and 3 (6.8%) patients had other organ involvement. The most common complication was cyst rupture (n=10, 22.7%), and followed by pleural effusion (n=9, 20.5%). All of the patients received albendazole as a first-line treatment. Forty-two (95.5%) patients underwent surgery and 14 (31.8%) patients required admission to the ICU admission after surgery. **Table 1** summarizes the detailed clinical findings of the patients.

When comparing patients with a ruptured cyst to patients without a ruptured cyst, hemoptysis was significantly more common in patients with a ruptured cyst. In addition, cyst diameter, ICU admission, length of stay, and recurrence were significantly more associated with cyst rupture. **Table 2** summarizes the comparison of patients with and without cyst rupture.

DISCUSSION

This infection is more common in areas where livestock markets are common. Humans become infected through food

Table 1. Clinical characteristics of the study population

Symptoms, n (%)	
Asymptomatic	2 (4.5)
Cough	38 (86.4)
Weakness	12 (27.3)
Sputum	20 (45.5)
Chest pain	22 (50)
Fever	19 (43.2)
Hemoptysis	3 (6.8)
Abdominal pain	5 (11.4)
Nausea-vomiting	2 (4.5)
Dyspnea	6 (13.6)
Night sweats	2 (4.5)
Weight loss	3 (6.8)
Anaphylaxis	1 (2.3)
Localization, n (%)	
Right	
Upper lobe	5 (11.4)
Middle lobe	5 (11.4)
Lower lobe	13 (29.5)
Left	
Upper lobe	7 (15.9)
Middle lobe	3 (6.8)
Lower lobe	11 (25)
Hepato-pulmonary involvement	20 (45.5)
Spleen	3 (6.8)
Patients with two cysts, n (%)	8 (18.2)
Diameter of the cyst, mm, mean±SD	64.02±29.02
Complications, n (%)	
Rupture	10 (22.7)
Bronchus	7 (15.9)
Parenchyma	3 (6.8)
Pleural effusion	9 (20.5)
Atelectasis	6 (13.6)
Pneumothorax	1 (2.3)
Surgery, n (%)	42 (95.5)
Intensive care unit admission, n (%)	14 (1.8)
Length of stay in hospital, days, mean±SD	7.68 ± 4.27
Recurrence, n (%)	6 (13.5)
SD: Standard deviation	

contaminated with eggs in the feces of the primary host. In the gastrointestinal tract, the larvae penetrate the intestinal mucosa and travel via the porto-caval anastomosis to the liver or via the general circulation to the lungs or other organs.⁹ While it is an endemic disease in South America, East Africa, Australia, and Mediterranean countries, it is also a growing thoracic pathology in non-endemic countries due to increased global travel and immigration.^{5,10} In a study conducted in Istanbul, which is an industrialized city, Arinc et al.⁴ reported that 54.1% of pulmonary HC patients came from rural areas, and this finding shows the effect of increased migration and travel. Our hospital is located in an Anatolian city where livestock markets are common and an important source of livelihood even in the city center, and our results showed that 63.6% of the admissions were from rural areas. Although most of the patients from rural areas, rupture was more common in urban patients even no statistical difference.

Once cysts enter the body and mature in the resident organ, they may remain latent for a long time and symptoms may not appear until they reach a certain size or complications arise. Only 10-19% of patients are reported to be asymptomatic. The most common symptoms reported are cough, chest pain, and sputum production.¹¹ In this study, only 2 (4.5%) patients

Table 2. Comparison of patients with ruptured and unruptured cysts

	Ruptured cyst (n=10)	Unruptured cyst (n=34)	p value
Age, months, mean±SD			
Gender, n (%)			
Male	3 (30)	21 (61.8)	0.147
Female	7 (70)	13 (38.2)	
Sociogeographic status, n (%)			
Rural areas	5 (50)	23 (67.6)	0.456
Urban areas	5 (50)	11 (32.4)	
Symptoms, n (%)			
Cough	10 (100)	28 (82.4)	0.310
Weakness	1 (10)	11 (32.4)	0.241
Sputum	7 (70)	13 (38.2)	0.147
Chest pain	6 (60)	16 (47.1)	0.472
Fever	4 (40)	15 (44.1)	0.817
Hemoptysis	3 (30)	0 (0)	0.009
Abdominal pain	0 (0)	5 (14.7)	0.198
Nausea-vomiting	1 (10)	1 (2.9)	0.346
Dyspnea	2 (20)	4 (11.8)	0.505
Night sweats	1 (10)	1 (2.9)	0.346
Weight loss	1 (10)	2 (5.9)	0.548
Anaphylaxis	1 (10)	0 (0)	0.227
Localization, n (%)			0.481
Right	4 (40)	19 (55.9)	
Upper lobe	0 (0)	5 (14.7)	
Middle lobe	1 (10)	4 (11.8)	
Lower lobe	3 (30)	10 (29.4)	
Left	6 (60)	15 (44.1)	
Upper lobe	2 (20)	5 (14.7)	
Middle lobe	1 (10)	2 (5.9)	
Lower lobe	3 (30)	8 (23.5)	
Hepato-pulmonary involvement	2 (20)	18 (52.9)	0.083
Spleen	0 (0)	3 (8.8)	0.331
Diameter of the cyst, mm, mean±SD	84.40±30.77	58.02±26.00	0.010
Intensive care unit admission, n (%)	8 (80)	6 (17.6)	0.001
Length of stay in hospital, days, mean±SD	10.40±4.47	6.88±3.92	0.020
Recurrence, n (%)	4 (40)	2 (5.9)	0.018

SD: Standard deviation

were asymptomatic and had uncomplicated PHC, which is lower than the literature. Among the symptomatic 42 (95.5%) patients, the most common symptoms were cough and chest pain, as expected.

While hydatid cysts are more common in the liver in adults, they are more common in the lungs in children.¹² PHCs grow more rapidly and are more likely to become symptomatic due to the compressibility of lung tissue, high vascularity, and low negative pressure.^{6,13} Faster growth leads to more complications. While there are some studies reporting that the rupture rate increases with increasing cyst size, there are also studies reporting the opposite situation.¹⁴⁻¹⁸ Previous studies have reported rupture rates ranging from 21.1% to 53.1%.^{18,19} In this study, there were 10 (22.7%) patients who experienced cyst rupture, which is consistent with the literature. All 3 patients with hemoptysis were in the ruptured group. The mean diameter of the cysts was higher in the ruptured group than in the unruptured group with a statistical difference. In addition, the length of stay, ICU admission, and recurrence

were more common in ruptured cysts, as expected. Although there was no statistical difference, the left-sided cysts were more likely to be rupture. This may be due to the lower compressibility of the lung tissue on the left side compared to the right side as a result of the left-located heart.

Conventional surgery is the main treatment for PHC. The goal of this surgery is to remove all parasitic material and repair the bronchial fistulas while preserving as much lung tissue as possible, especially in children. Kocaman et al.²⁰ reported that 93.2% of their patients underwent surgery. In this study, 42 (95.5%) patients underwent surgery, which is consistent with the literature. One patient with anaphylaxis underwent emergency surgery. Two patients did not require surgery and were treated with albendazole alone.

Limitations

The major limitations of our study were the retrospective collection of data from a single center and the small number of patients. Cystic echinococcosis, especially PHC, is difficult to diagnose and requires a high index of suspicion, especially in children and centers from non-endemic regions. Furthermore, due to the retrospective nature of the study, we were unable to assess and evaluate the surgical data and its impact on recurrence.

CONCLUSION

In conclusion, hydatid cysts are still a health problem, especially in urban areas. PHC can be life-threatening, leading to anaphylaxis and bronchial rupture. PHC should be considered in the differential diagnosis of children presenting with lower respiratory tract symptoms such as fever, cough, hemoptysis, and chest pain, especially in regions where echinococcosis is endemic.

ETHICAL DECLARATIONS

Ethics Committee Approval

Ethical approval was obtained from the Institutional Ethics Committee of Necmettin Erbakan University Faculty of Medicine (Date: 18.10.2024, Decision No:h 2024/5260).

Informed Consent

Because the study was designed retrospectively, no written informed consent form was obtained from patients.

Referee Evaluation Process

Externally peer-reviewed.

Conflict of Interest Statement

The authors have no conflicts of interest to declare.

Financial Disclosure

The authors declared that this study has received no financial support.

Author Contributions

All of the authors declare that they have all participated in the design, execution, and analysis of the paper, and that they have approved the final version.

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