

## Original Article

# Therapeutic strategies for complications secondary to hydatid cyst rupture

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**Abstract:** Objective: Clinical approach and therapeutic methods are important in cases with complicated hydatid cysts of the lung. This study was designed to retrospectively investigate cases with hydatid cysts, thereby discussing diagnostic methods, treatment modalities, and rates of morbidity and mortality in line with the literature. Methods: 176 cases with perforated hydatid cysts, who presented to our clinic and underwent surgery between 2003 and 2011, were included in the study. There were 71 (40.34%) females and 105 males (59.66%) with a mean age of 27.80±14.07. The most common symptom was dyspnea (44.31%) and the most common radiological finding was the water lily sign (21.02%). 88.06% of the cases were surgically treated by Cystotomy+closure of bronchial opening+capitonnage, 3.97% by wedge resection, 4.54% by segmentectomy and 3.40% by lobectomy. Results: The cysts exhibited multiple localization in 24 cases (13.63%), bilateral localization in 14 cases (7.95%), with the most common localization (43.75%) being the right lower lobe. While the hydatid cyst rupture occurred due to delivery in three (1.70%), trauma in 11 (6.25%), and iatrogenic causes in seven (3.97%) cases, it occurred spontaneously in the rest of the cases (88.08%). Fourteen of the cases with spontaneously occurring rupture (7.95%) were detected to have received anthelmintic treatment for hydatid cyst during the preoperative period (albendazole). The rate of morbidity was 27.27% and the rate of mortality was 1.13% in our study. Two cases recurred during a one-year follow-up (1.13%). Conclusion: Hydatid cyst rupture should be considered in the differential diagnosis of cases with pleural effusion, empyema, pneumothorax and pneumonia occurring in endemic regions. Symptoms occurring during and after perforation lead to errors in differential diagnosis. Performing the surgery without delay favorably affects postoperative morbidity and mortality. While parenchyma-preserving surgery is preferential, there is a need for resection in perforated hydatid cysts.

**Keywords:** Hydatid cyst, rupture, management

## Introduction

The fact that Hydatid cyst is a parasitosis occurring secondary to *Echinococcus granulosus* has been known since Hippocratic times [1]. This parasite remains a significant healthcare issue in endemic regions such as the Mediterranean. Turkey is such an endemic region with a hydatid disease incidence of 18-20 cases per 100,000 people [2, 3]. Echinococcal cysts always pose a risk of rupture along with anaphylactic potential. This risk also involves the spread of the agent to the healthy organs. Factors such as trauma or cough that increase intrathoracic pressure and facilitate enlargement of the cyst diameter are the

factors known to be responsible for rupture; however, no correlation has been detected between cyst diameter and intra-cyst pressure [4]. When the cyst is ruptured up to the bronchus, a part or all of the symptoms, including fever and hemoptysis, may occur as well as hydatid fluid and parasitic membrane expectoration [5]. Also, the rupture of the hydatid cyst up to the pleural space commonly results in pneumothorax, pleural effusion or empyema [6].

## Materials and methods

In this study, we retrospectively evaluated cases diagnosed with hydatid cyst presenting to and

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undergoing surgery at our clinic between 2003 and 2011. In total, there were 412 cases diagnosed with hydatid cyst within this period, among which 716 with ruptured hydatid cysts (42.71%) were included in the study.

There were 71 (40.34%) females and 105 males (59.66%) with a mean age of  $27.80 \pm 14.07$ . Most of the patients were from rural areas (87%). The time between the onset of the symptoms and presentation to the hospital was  $16.78 \pm 6.41$  days (9-32 days).

For the diagnosis, clinical examination, routine investigations, pulmonary x-ray, CT, USG and fiber optic bronchoscopy (FOB) and MRI, when necessary, were used. Due to their low diagnostic value, serologic tests (Casoni, Weinberg etc) were not used on a routine basis. Blood tests showed eosinophilia in most of the patients. Perforation was described as the impaired integrity of the germinative membrane of the cyst. The diagnosis of perforation was established by clinical manifestations, findings, radiologic data and the intraoperative assessments of the patient. The ruptures to the pleural space or bronchus were also considered to represent perforation. The hydatid cysts with diameters  $\geq 10$  cm were considered to be giant hydatid cysts.

The most common symptom was dyspnea occurring in 78 cases (44.31%); the other findings were cough-sputum [n:54 (30.68%)], chest pain [n:37 (21.02%)], fever [n:47 (26.70%)], hemoptysis [n:12 (6.81%)], hydroptysis [n:19 (10.79%)], lack of appetite [n:61 (34.65%)] and fatigue [n:69 (34.65%)]. Eleven cases (6.25%) were asymptomatic.

While the most common radiologic finding was the water lily sign in 37 cases (21.02%), the other findings included hydropneumothorax [n:21 (11.93%)], air-fluid level mimicking abscess [n:19 (10.79%)], total pneumothorax [n:17 (9.65%)], localize empyema [n:21 (11.93%)], pleural thickening [n:19 (10.79%)], pleural fluid [n:36 (20.45%)], double-dome arc sign [n:11 (6.25%)], pneumonia [n:14 (7.93%)], tension pneumothorax [n:2 (1.13%)] and mediastinal shift [n:2 (1.13%)].

The cases were assessed with respect to the site of localization, the number and size of the cysts, the cause of the cyst rupture,

preoperative and post-operative complications, surgical procedure performed, hospitalization period, and mortality rates.

### *Surgical procedure*

All patients were operated on via thoracotomy (the thorax was opened from the 5-6<sup>th</sup> intercostal space at the lateral decubitus position by thoracotomy). In cases with contralateral lesions, the operation was performed on the opposite side, two weeks after the initial thoracotomy. Median sternotomy could not be used to due to infection risk. All procedures were performed using a double-lumen endotracheal tube under general anesthesia. After the lungs were completely freed following thoracotomy, the site of the cyst/cysts was detected. To avoid the flow of the cyst fluid into the thoracic cavity, the operation site was covered with sponge or compresses wet with 10% Savlon (1.5% *chlorhexidine* gluconate + 15% cetrimide) at the time of access to the thorax. Subsequently, the content of the cyst was aspirated using a thick needle. Following a minor incision in the pericystic layer to open it, the germinative membranes in the cyst space and the daughter vesicles were transferred into a container full of 10% Savlon using an over clamp. After cleaning the cyst space with diluted Savlon, the cyst space was rinsed with isotonic fluid to avoid the irritating effect of Savlon. During this time, the tracheobronchial system was continuously aspirated by the anesthetic staff. To avoid air leakage, the bronchial openings in the cyst cavity were sutured with 3/0 or 2/0 Vicryl (Polyglactin 910 synthetic absorbable suture) one by one. Capitonage was performed in multiple stages starting from the lowest part of the cavity, and surrounding all walls. Capitonage was conducted, taking into consideration the anatomic integrity of the lungs. The lung tissue adjacent to the cyst was assessed with respect to participation in the pulmonary ventilation. If the adjacent parenchyma was hepatized and could not be ventilated by insufflations applied by the anesthetist, resection was considered indicated. The width of the resection was determined based on the intraoperative findings and the experience of the surgeon. If the lesion was larger than 2/3 of a lobe, resection was completed to lobectomy. Decortication was performed in patients with pleural cortex

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**Table 1.** Operative methods for Perforated Hydatid Cysts

Procedure	Cases (n)	%
Thoracentesis	43	24.43
Tube thoracostomy + drainage	76	43.18
Decortication	22	12.5
Cystotomy +closure of bronchial opening+ capitonnage	155	88.06
Wedge resection	7	3.97
Segmentectomy	8	4.54
Lobectomy	6	3.40

**Table 2.** Hydatid cyst localization observed

	Cases with multiple hydatid cysts (n:24)						Cases with a single hydatid cyst (n:152)			Total number of cases (n:176)	%
	Bilateral localization (n:14)			Lateral hemithorax localization (n:10)			Upper lobe	Middle lobe	Lower lobe		
	Upper lobe	Middle lobe	Lower lobe	Upper lobe	Middle lobe	Lower lobe					
Right lung	5	4	7	4	5	3	21	10	67	126	71.59
Left lung	8		8	4		4	18		36	78	44.31

formation secondary to empyema (**Table 1**). All patients received antibiotic treatment before and after the surgery. If there was no specific culture finding, first- and second- generation cephalosporins were used empirically. Unless contraindicated, albendazole was initiated in all cases (10-14 mg/kg/day) in the postoperative period. Albendazole was not used in pregnant women and patients with high liver enzymes.

### Results

Twenty-four of the cases (13.63%) had multiple cysts, 14 had (7.95%) bilateral cysts, and ten had (5.68%) cysts located in the lateral hemithorax. While the hydatid cysts were located in the right upper lobe, middle lobe and right lower lobe in 30 (17.04%), 19 (10.79%) and 77 (43.75%) cases respectively, 48 cases had cysts in the left lower lobe (27.27%) and 30 had cysts in the left upper lobe (17.04%). A total of 41 cases (23.29%) had a giant cyst with a diameter larger than 10 cm (**Table 2**).

The pulmonary hydatid cysts were accompanied by hepatic, spleen and renal cysts in 26 (14.77%), two (1.13%) and one case (0.56%), respectively.

While hydatid cyst rupture occurred due to delivery and trauma in three (1.70%) and 11 (6.25%) cases respectively, it occurred due to iatrogenic causes in seven cases (transthoracic fine needle aspiration biopsy in four cases, thoracentesis in three cases) (3.97%) and spontaneously in the rest of the cases (88.08%). Fourteen of the cases with spontaneously occurring rupture (7.95%) were detected to have received anthelmintic treatment for hydatid cyst during the preoperative period (mebendazole or albendazole).

The most common complication occurring in the preoperative period was pleural perforation of the cyst (**Table 3**).

The morbidity rate was detected to be 27.27% with 48 cases. The most common postoperative complication was atelectasia (6.25%) (**Table 4**). Two (1.13%) cases resulted in mortality due to pneumonia and sepsis.

The mean duration of hospitalization was  $10.95 \pm 3.50$ /day and the mean follow up duration was  $19.7 \pm 13$  months (range: 10-35 months). During the one-year follow-up, two

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**Table 3.** Preoperative complications

Complication	Number of cases (%)
Hydroptisis	19 (10.79)
Perforation of the pleura	76 (43.18)
Tension pneumothorax (2)	
Hydropneumothorax (21)	
Total pneumothorax (17)	
Pleural effusion (36)	
Infection	72 (40.90)
Abscess ( 23)	
Empyema ( 35)	
Pneumonia (14)	
Allergic complications	14 (7.95)
Bronchial perforation	37 (21.02)
Urticaria -erythema (6)	
Asthma bronchial provocation (3)	
Cramp (2)	
Diarrhea (3)	
Pulmonary hemorrhage -hemoptysis	12 (6.81)

**Table 4.** Postoperative complications and the causes of mortality

Postoperative complication	Number of cases (%)
Pneumonia	9 (5.11)
Empyema	3 (1.70)
Bronchopleural fistula	2 (1.13)
Atelectasia	11 (6.25)
Prolonged air leakage (>10 days)	7 (3.97)
Lesion site infection	8 (4.54)
Post cystectomy residual cavity	6 (3.40)
Post lobectomy residual cavity	1 (0.56)
Thrombophlebitis	1 (0.56)

cases were detected to have (1.13%) recurrence (one on the operated side, the other on the contralateral lung).

### Discussion

Patients with thoracic hydatid cyst with pleural involvement of the lung and/or those associated with infection are included in a special clinical entity called complicated hydatid cyst [7].

The diagnosis of hydatid cyst is established by anamnesis, x-ray and other radiologic imaging techniques (chest x-ray, computed tomography, magnetic resonance imaging), serologic tests and microscopic examination [8].

Non-perforated pulmonary cysts may be asymptomatic or may manifest with various

clinical findings associated with the compression of the pulmonary tissue [9]. The clinical appearance is variable for the perforated hydatid cysts and dependent on the characteristic of the perforation. Frequently, the cyst is ruptured up to the bronchus. In most of the cases, the solid residues in the collapsed parasitic membrane cavity are the source of the recurrent infection [10]. Cough-sputum, pain, hemoptysis, dyspnea, and fever are the most common pulmonary hydatid cyst symptoms [11]. Since there were symptoms such as dyspnea, cough-sputum, chest pain, fever and hemoptysis that lacked a specific diagnostic value, radiographic imaging methods were used to establish diagnosis.

Radiologic methods establish diagnosis in 89% of cases. The differential diagnosis includes benign tumours, inflammatory masses,

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metastases, solid or fluid-filled cysts and carcinomas [12].

The intact cyst is imaged as a homogenous round radio-opacity with clear borders on radiography. In complicated cysts, the radiological findings may include water lily sign, hydroaeric level, meniscus signs, pneumonic filtration around the cyst, and pleural fluid due to pleural opening [13]. Complicated pulmonary hydatid cysts may be associated with, or lead to, benign conditions such as bronchiolitis obliterans organizing pneumonia [14]. While the most common radiologic finding was the water lily sign (21.02%), other findings included hydropneumothorax, air-fluid level mimicking abscess, total pneumothorax, localized empyema, pleural thickening, pleural fluid, and double-dome arc sign, and 14 cases had pneumonia (7.93%).

Serologic tests are used in the diagnosis and follow-up of patients with hydatid cysts in the endemic regions due to their low cost and convenient administration [15]. Echinococcus IHA, ELISA, IgG, immunoelectrophoresis, and indirect fluorescent antibody tests are the serologic tests that can be used in the diagnosis and follow-up of hydatid cysts. However, they have a low diagnostic sensitivity, particularly to pulmonary hydatid cysts. Due to their low diagnostic value, serologic tests (Casoni, Weinberg etc) are not used on a routine basis.

Since the lung is an elastic organ, the hydatid cyst can reach large sizes [16]. This enlargement is more common in children and young people [17]. A total of 41 of our cases (23.29%) had a giant cyst with a diameter larger than 10 cm.

In cases with hydatid cysts, the increase in the cyst size, factors such as trauma and cough that cause an increase in the intrathoracic pressure may predispose patients to cyst rupture formation [18]. Intra-cyst pressure is proportional to the cube of the cyst diameter. Therefore, even a small increase in the cyst diameter results in a large rupture by severely increasing the intra-cyst pressure. In addition, one should note that invasive diagnostic procedures would increase the risk of perforation. While the hydatid cyst rupture occurred due to delivery and trauma in three (1.70%) and 11 (6.25%) cases respectively, it

occurred due to iatrogenic causes in seven cases (3.97%), and spontaneously in the rest of the cases (88.08%).

The fact that the time between the onset of symptoms and the presentation to the hospital was  $16.78 \pm 6.41$  days could suggest that the disappearance of the compression on the surrounding tissue by the perforated cyst after expectoration could be associated with the reduction in the symptoms secondary to compression, and also that the symptoms occurring during perforation could cause errors in the differential diagnosis; literature screening revealed no such studies.

Whether the cyst is symptomatic or asymptomatic, surgical treatment is still a valid option in the treatment of pulmonary hydatid cyst due to its potential complications [5, 19, 20]. Lung-preserving, and single-step surgery, if applicable, are the recommended modality of surgery [19]. As a rule, the pulmonary parenchyma should be preserved and the radical procedures should be avoided. In the presence of severe infection and bronchiectasis, the affected part of the lung should be resected; if very large or multiple cysts have damaged the parenchyma, lobectomy or pneumonectomy may be performed [5].

Based on the literature reports, radical methods may be required at a rate of 0-7% in intact hydatid cysts and at a rate of 19-32% in complicated hydatid cysts [21]. In giant pulmonary hydatid cysts, a resection rate of 6.6-13% was reported [16, 17]. Resection should be avoided in children because damaged pulmonary parenchyma has a high capacity to heal [5]. While various approaches are reported in the literature, cystotomy with capitonnage by thoracotomy is the standard procedure [19, 22]. In cases of pulmonary hydatidosis, video-assisted thoracic surgery can only be performed in selected patients. However, while excising the cyst, one should consider the risk of anaphylaxis, pleural hydatidosis and if the cyst is complicated, the potential for the infection to spread should also be kept in mind [21]. In our study, 88.06% of the cases were surgically treated by Cystotomy + closure of bronchial opening+ capitonnage, and 21 cases (11.93%) underwent wedge resection, segmentectomy and lobectomy.

Another treatment modality is medical treatment in hydatid cyst disorder. Data in the WHO guidelines reveal that cases followed for 12 months on medical treatment achieved a complete cure of 30%, cyst degeneration and/or severe size reduction of 30-50%, and no improvement at a rate of 20-40% [20]. The studies show that cases respond to medical treatment at varying degrees between 73 and 75%; however, cure rates are only 30 to 35% [21]. This is not adequate for the eradication of the cyst. In addition, the literature reports show that cases with hydatid cysts may require urgent surgery secondary to severe hemoptysis and hypersensitivity reactions with preoperative albendazole use [20, 21]. Dogan et al [5] administered mebendazole treatment to 28 cases in the preoperative period. During this treatment, four cases were urgently operated on due to massive hemoptysis and six cases were reported to have cyst rupture into the bronchus. Only three cases showed regression of the cyst. Based on these results, Dogan et al [5] recommend drug treatment only for inoperable cases and recurrent cases with high surgical morbidity and mortality.

In our study, 14 of the cases (7.95%) with spontaneously occurring rupture (7.95%) were detected to have received anthelmintic treatment for hydatid cyst during the preoperative period (albendazole or mebendazole) and unless contraindicated, albendazole treatment (10-14 mg/kg/day) was started for all cases in the postoperative period. Albendazole was not used in pregnant women and patients with high liver enzymes.

In general, hydatid cyst has a good prognosis. Depending on the localization of the cyst and the experience of the surgeon, the rates of recurrence vary between 2 and 25%. Operation-associated mortality rates vary between 0.5 and 4% due to the same reasons [23, 24]. Perforated cysts involve higher postoperative morbidity and mortality relative to non-perforated pulmonary cysts [22]. In perforated cases, infection and inflammation of the adjacent pulmonary parenchyma may affect wound healing and lead to postoperative complications, including prolonged air leakage, empyema and pneumonia. Additionally, perforated pulmonary hydatid cyst patients require preoperative antibiotic and supportive treatment. These are the main reasons why

perforated cases have high mortality and morbidity. Perforated cases need longer hospitalization compared to non-perforated cases. The rate of morbidity was 27.27% in our study. The most common postoperative complication was atelectasis (6.25%) (Table 4). Two (1.13%) cases resulted in mortality due to pneumonia and sepsis. The mean duration of hospitalization was  $10.95 \pm 3.50$ /day and the mean follow up duration was  $19.7 \pm 13$  months (range: 10-35 months). During a one-year follow-up, two cases (1.13%) were detected to have recurrence (one on the operated side, the other on the contralateral lung).

In conclusion, hydatid cyst rupture should be considered in the differential diagnosis of cases with pleural effusion, empyema, pneumothorax and pneumonia occurring in endemic regions such as our country. In addition to the other factors, preoperative albendazole may increase the risk of perforation in cases of pulmonary hydatid cysts, and in pulmonary hydatid cyst disorder, cyst perforation may occur due to invasive diagnostic procedures. In case of pulmonary hydatid cyst, patients may present relatively late to the hospital due to disappearance of the compression on the surrounding tissue by the perforated cyst after expectoration with the resulting reduction in the symptoms secondary to compression. The symptoms occurring during and after the perforation lead to errors in the differential diagnosis. Performing the surgery without delay favorably affects postoperative morbidity and mortality. While parenchyma-preserving surgery is preferential, there is a need for resection in perforated hydatid cysts.

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