Complicated Hydatid Cysts of the Lung: Clinical and Therapeutic Issues

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Background. The clinical presentation and the preoperative and postoperative complications associated with pulmonary hydatid cysts depend on whether the cyst is intact or ruptured. The aim of this study was to review the problems encountered in treating ruptured pulmonary hydatid cysts and to highlight the risks associated with chemotherapy and the delay of surgical treatment in pulmonary hydatid disease.

Methods. The medical records for 67 patients of pulmonary hydatidosis were retrospectively investigated. The patients were divided into two groups based on whether the pulmonary cyst was intact (group 1, n = 34) or complicated (group 2, n = 33). A complicated cyst was defined as one that had ruptured into a bronchus or into the pleural cavity. All patients were treated surgically. Data related to symptoms, preoperative complications, surgical procedures performed, postoperative morbidity, hospitalization time, and cyst recurrence were collected from each individual's records, and the group findings were compared.

Results. In most cases of intact pulmonary hydatid cysts, the lesions were either incidental findings or the patient had presented with cough, dyspnea and chest pain. In addition to these symptoms, the patients with complicated cyst had presented with problems such as expectoration of cystic contents, repetitive hemoptysis, productive sputum, and fever. The differences between the groups with respect to the rates of preoperative complications and postoperative morbidity, frequency of decortication, and hospital stay were statistically significant ($p < 0.05$).

Conclusions. Surgery is the primary mode of treatment for patients with pulmonary hydatid disease. Complicated cases have higher rates of preoperative and postoperative complications and require longer hospitalization time and more extensive surgical procedures than uncomplicated cases. This underlines the need for immediate surgery in any patient who is diagnosed with pulmonary hydatidosis.

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Hydatidosis remains endemic to many parts of the world, most notably the Mediterranean region, Australia, New Zealand, the Middle East, and South America. In areas where hydatid disease is endemic, it is still a major public health problem. Most individuals who contract this parasite are young, and the majority of patients are less than 40 years of age [1–4].

All hydatid cysts carry the risk of rupture. Pulmonary hydatid cysts usually remain asymptomatic until the time of rupture, and the clinical presentation in these patients is directly related to intact or ruptured cyst status. Rupture may occur during anthelmintic therapy or percutaneous aspiration and can lead to severe complications, such as massive hemoptysis and tension pneumothorax. Surgery remains the treatment of choice for hydatid cysts of the lung. However, pulmonary hydatidosis primarily affects children of young adults who may be infected again, and usually they have great lung capacity for expansion. Thus it is important to always use the most conservative surgical methods possible. More radical surgical procedures may be needed in complicated cases.

In this study we retrospectively assessed a series of patients with pulmonary hydatidosis that were treated at our center, and we compared the clinical presentation, surgical treatment, and postoperative outcome in patients with intact cysts and corresponding findings in patients with ruptured cysts. The aim was to review the problems encountered in treating ruptured pulmonary hydatid cysts and to highlight the risks associated with anthelmintic therapy and delayed surgical therapy in pulmonary hydatid disease.

Material and Methods

The medical records of 67 patients with pulmonary hydatid cysts who had operations for the same at our center between January 1998 and December 2002 were reviewed. The series included 33 males and 34 females with a mean age 34 years (range, 4 to 75 years). Approximately 80% of the patients lived in rural areas. The follow-up times for all 67 patients ranged from 5 months to 5 years.

The pulmonary cysts were diagnosed by various combinations of chest roentgenogram, thoracic ultrasonogra-

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Table 1. Surgical Approaches and Procedures Performed in 67 Patients

Surgical Aproaches	Procedures			No. of Patients	No. of Operations
	Cystotomy With Capitonnage	Cystectomy	Wedge Resection		
Posterolateral thoracotomy	27	2	1	30	30
Bilateral posterolateral thoracotomies	16	...	...	16	32
Right posterolateral thoracotomy with phrenotomy	6	1	...	7	7
Posterolateral thoracotomy and laparotomy	11	1	...	12	24
Median sternotomy	1	...	1	2	2
Total	61	4	2	67	95

phy, and thoracic computed tomography. Each patient also underwent abdominal ultrasonography to assess for concomitant hepatic or splenic cysts. Skin and serologic testing were not part of the routine diagnostic workup.

Any cyst that had ruptured into the bronchus or pleural cavity, with or without infection, was defined as complicated. The patients were divided into two groups based on whether the cyst lesions were intact (group 1, n = 34) or complicated (group 2, n = 33). A ruptured cyst was considered to be infected when the patient exhibited accompanying problems of purulent sputum, leukocytosis, fever, and pericystic pneumonitis with or without lobar and segmental pneumonia.

A total of 95 surgeries were performed on the 67 patients (Table 1). Of the 95 operations performed, there were 81 posterolateral thoracotomies, 12 laparotomies, and 2 median sternotomies. A thoracolaparotomy incision was never used and bilateral thoracotomy was not performed in the same session as the previous procedures. Twenty-eight individuals required two surgeries; the second operation was performed at least 21 days after the first operation. For the period between the first and second operation, each patient was informed about possible complications and warned that a second surgery may be needed sooner than planned if the cyst ruptured.

In regard to the hydatid cysts in the liver, only liver-dome cysts were treated in our department. All other hepatic cysts were treated in the general surgery department. The individuals with liver-dome cysts underwent thoracophrenotomy.

Seven of the 67 patients presented with pneumothorax and empyema. These patients all required closed-chest tube drainage initially, and further surgical procedures were performed after their general condition had stabilized.

One individual presented to the emergency service in septic shock due to empyema and multiple infected hydatid cysts of the liver and lung. A chest tube was placed, and biliary drainage through this tube (200 to 500 mL/day) indicated a biliopleural fistula. The patient went into cardiac arrest and was placed on mechanical ventilation. The patient was initially managed with nasobiliary drainage in addition to chest tube drainage and antibiotic treatment, and when he showed good response to this and to supportive treatment, he was taken off the ventilator. Later, laparotomy and thoracotomy were performed on this patient in separate sessions.

In 61 of 67 patients (34 uncomplicated and 27 complicated), the specific surgical treatment for pulmonary hydatid cysts was cystotomy with capitonnage. Unlike excision, capitonnage refers to emptying the cyst and then closing it by applying sutures so that they approximate the opposing surfaces of the cavity. For this procedure the edges of the surgical wound and the lung surface were protected with wet packs soaked in diluted (10%) povidone-iodine solution. Due to risks of leakage into the bronchial system and postoperative tracheobronchial irritation or pulmonary edema, no scolicidal agent was injected into the cyst. The cyst contents were evacuated by needle aspiration, and the cystic membrane was removed with ring forceps. The remaining cavity was irrigated with saline solution and cleaned with sponges containing diluted povidone-iodine (10%). Bronchial openings in the cavity were stitched up with absorbable sutures in a figure-8 suture. After the bronchial openings were closed, the residual cavity was obliterated with separate purse-string sutures that were placed into the cavity from the deepest level to the surface (capitonnage). In the complicated cysts that exhibited a thickened or liquified pericyst layer and had damaged the adjacent parenchyma, the closure of bronchial fistulas was done more carefully using deeper sutures and placing them closer together, or in 6 patients with complicated cysts (18.2%), a pericystectomy or wedge resection was performed. Decortication was performed in 2 patients (5.9%) in group 1 and 8 patients (24.2%) in group 2. None of the patients in group 1 and group 2 required lobectomy or pneumonectomy.

All patients who had pneumonia (n = 8), infected cysts (n = 4), empyema (n = 4), and sepsis (n = 1) received preoperative antibiotic therapy. The course of treatment was a minimum of 7 days (mean, 11 days; range, 7 to 21 days). Third-generation cephalosporins or β-lactamase antibiotics were the usual drugs of choice for empirical therapy. First-generation cephalosporins were administered as postoperative prophylactic treatment with a duration ranging from 4 to 30 days (mean, 6 days). Efficacy of antibiotic therapy was assessed on the basis of cultures of sputum and pleural fluid and blood and was assessed according to clinical response. After surgery, all patients who had complicated or multiple hydatid cysts, or both, were placed on albendazole anthelmintic therapy (800 mg/d in adults, 10 mg/kg/d in children) for at least 2 months. Only 1 of 67 patients received preopera-

Table 2. Clinical Manifestations of Pulmonary Hydatid Disease in 67 Patients

Clinical Manifestations	Group 1 (n = 34)		Group 2 (n = 33)		p Value
Asymptomatic	9	26.5%	1	3%	$p < 0.001$
Chest pain	17	50%	16	48.5%	$p > 0.05$
Dyspnea	6	17.6%	14	42.4%	$p > 0.05$
Cough	9	26.5%	15	45.5%	$p > 0.05$
Hemoptysis	2	5.9%	11	33.3%	$p < 0.05$
Expectoration of cystic contents	. . .	. . .	5	15.2%	. . .
Sputum production	3	8.8%	11	33.3%	$p < 0.05$
Fever	4	11.8%	12	36.4%	$p < 0.05$
Weight loss	2	5.9%	4	12.1%	$p > 0.05$
Biloptysis	. . .	. . .	1	3%	. . .

Table 3. Preoperative Complications in the Series

Preoperative Complications	Group 1 (n = 34)		Group 2 (n = 33)	
Pneumonia	1	2.9%	6	18.2%
Simple pneumothorax	. . .	. . .	2	6.1%
Tension pneumothorax	. . .	. . .	1	3%
Empyema	. . .	. . .	4	12.1%
Bronchobiliary fistula and pneumonia	. . .	. . .	1	3%
Pleurobiliary fistula and septic shock	. . .	. . .	1	3%
Infected cyst	. . .	. . .	3	9.1%
Infected cyst and pneumonia	. . .	. . .	1	3%
Allergic episode	. . .	. . .	1	3%

tive anthelmintic therapy, and this patient was on albendazole when she was referred to our center for a ruptured infected cyst.

Data related to symptoms, preoperative complications, surgical procedures performed, postoperative morbidity, hospitalization time, and cyst recurrence were collected from each patient's records, and the group findings were compared.

Statistical Analysis

The distribution of hospital stays in groups 1 and 2 was examined using the Kolmogorov-Smirnow test. This showed that the data from both groups were normally distributed. Once this was confirmed, the mean hospitalization times for groups 1 and 2 were compared using the unpaired t test. Other results were compared using the χ^2 test and Fisher's exact test as appropriate. The p values less than 0.05 were considered to indicate statistical significance.

Results

Assessment of clinical findings, chest roentgenograms, thoracic computed tomography, and thoracic ultrasonography led to the correct preoperative diagnosis of pulmonary hydatid disease in 63 patients (94%). The other 4 patients were diagnosed intraoperatively. In 3 of these patients, the respective misdiagnoses before surgery were cavitary tuberculosis with associated fungus ball, malignant tumor, and solitary pulmonary nodule. In the other patient, a suppurative cyst attached to the posterior wall of the thorax resembled a neurogenic tumor on chest roentgenogram and computed tomography.

Thirty-one patients (46.3%) had solitary pulmonary cysts and 36 (53.7%) had multiple lesions. Eighteen patients with multiple cysts (26.9%) had bilateral pulmonary hydatidosis, 5 (7.5%) had unilateral multiple pulmonary hydatidosis, and 13 (19.4%) had unilateral lung lesions and concomitant liver hydatidosis. Of the 18 patients with bilateral pulmonary hydatid cysts, 6 also had cysts in the liver. Twenty patients in group 2 had multiple cysts and 13 patients had a single lesion. When the patients were grouped according to their type of hydatid disease (multiple vs solitary), there was no significant difference in the frequencies of cyst rupture ($p > 0.05$).

As previously noted, 33 of 67 patients (49.3%) had complicated cysts (group 2). Cyst rupture into the bronchial system occurred in 24 patients (35.8% of the total 67 patients; 72.7% of the complicated cases). Cyst rupture into the pleural space occurred in 9 patients (13.4% of the total patients; 27.3% of the complicated cases).

In both groups 1 and 2, the most common symptom was chest pain, followed by cough and dyspnea, respectively (Table 2). There were no differences between the groups with respect to the rates of these symptoms ($p > 0.05$). Hemoptysis, sputum, and fever were significantly more frequent in group 2 than group 1 ($p < 0.05$).

Twenty patients (60.6%) in group 2 had associated parenchymal and pleural complications before surgery (Table 3). The overall frequency of preoperative complications in group 2 was significantly higher than that in group 1 ($p < 0.01$). In concern with surgical procedures, decortication was required in many more patients in group 2, and this difference between the groups was statistically significant ($p = 0.0371$).

Three patients (8.8%) in group 1 and 13 patients (39.4%) in group 2 had postoperative complications develop ($p = 0.008$) (Table 4). There was no perioperative or postoperative mortality.

The hospitalization times in group 1 ranged from 7 to 19 days (mean, 10 days), whereas those in group 2 ranged

Table 4. Postoperative Morbidity

Causes	Group 1 (n = 34)		Group 2 (n = 33)	
Prolonged air leak (>7)	1	2.9%	5	15.2%
Empyema	. . .	. . .	3	9.1%
Pneumonia	. . .	. . .	1	3%
Atelectasis	. . .	. . .	3	9.1%
Hemoptysis	. . .	. . .	1	3%
Pleural effusion	1	2.9%	. . .	. . .
Wound infection	1	2.9%	. . .	. . .

from 7 to 74 days (mean, 21 days) ($p < 0.05$). Two patients in group 2 had recurrence develop during follow-up. There was no recurrence in group 1.

Comment

In most uncomplicated cases of pulmonary hydatid disease, lung cysts are either an incidental finding or the patient presents with cough, dyspnea, and chest pain. With complicated cysts, the clinical picture is variable and depends on the nature of the perforation. Often the cyst ruptures into a bronchus. In most cases, solid remnants of the collapsed parasitic membrane are left in the cavity as a source of recurrent infection [1, 5–7]. Such patients present with all the previously described symptoms and may also exhibit vomit-like expectoration of hydatid fluid and remnants of parasitic membrane, recurrent hemoptysis, purulent sputum, or fever, or a combination of some or all of these symptoms. Expectoration of cystic contents can lead to severe complications, such as acute respiratory failure, massive hemoptysis, and anaphylactic shock [4, 6]. Purulent sputum and fever are strong indicators of pneumonia or infected cyst, situations which may result in sepsis. One of our patients developed sepsis in this way.

In contrast to perforation into a bronchus, rupture of a hydatid cyst into the pleural cavity usually causes pneumothorax, pleural effusion, or empyema. Cyst rupture into the pleural cavity can also result in tension pneumothorax [8], which occurred in 1 of our patients. The documented rates of simple pneumothorax in patients with pulmonary hydatidosis ranged from 2.4% to 6.2% [4, 9, 10]. Empyema is reported to occur in 7.6% of patients with hydatid disease of the lung [9]. In our series, pneumothorax occurred preoperatively in 4.5% of all patients (9.1% of complicated cases), and empyema occurred preoperatively in 6% of all patients (12.1% of complicated cases). It has been proposed that rupture of a cyst into the pleural cavity or rupture into the bronchial tree may also lead to secondary larval spread or to allergic and anaphylactic reaction [6]. One of the patients in our series had an allergic episode.

Some authors contend that the treatment of hydatid disease regimens with oral mebendazole or albendazole are effective against cysts [11, 12]. Research has shown that 73% to 75% of patients respond to medical management to some degree; however, the reported cure rates are only 25% to 30% [13, 11]. This strategy is generally not considered a reliable way of eradicating this parasite [5] and it is a long and tedious process that carries considerable risk [7]. Anthelmintics weaken the cyst wall, thus increasing the likelihood of cyst rupture. Wen and Yang [13] found a 77.3% incidence of cyst rupture in 21 patients with hydatid disease who were treated with albendazole. Hepatic hydatid cysts have a relatively low complication rate, so anthelmintic therapy may be appropriate for patients whose lesions are limited to the liver [12]. In contrast, results with medical therapy for pulmonary hydatidosis have been discouraging. Reports in the literature of treatment with mebendazole or albendazole have been documented as requiring urgent surgery due to massive hemoptysis [14] and severe hypersensitivity reactions [15]. One of the 67 pulmonary hydatidosis patients in our series presented with infected hydatid cysts and cyst rupture after albendazole therapy. In addition to the previously mentioned issues, it is important to note that close follow-up is usually not possible in cases of hydatidosis, because these patients tend to be from rural areas where medical care is often inadequate. Our series was not randomized with respect to surgery versus medical treatment, but we believe that due to the possibility of cyst rupture, medical therapy should be used sparingly in patients with pulmonary hydatidosis. This approach should only be used for patients who are high surgical risks or for preventing recurrence.

Lung-conserving procedures are optimal with pulmonary hydatidosis, but video-assisted thoracic surgery is suggested for selected patients [16]. It is possible to remove the cystic membrane thoracoscopically, but uncontrolled spillage of cyst contents may cause anaphylaxis, pleural hydatidosis (if the cyst is intact), or pleural bacterial spread (if the cyst is infected). We believe that the most appropriate procedures for pulmonary hydatidosis are open surgery involving removal of the cyst membrane, closure of the bronchial openings, and capitonnage. However, complicated hydatid cysts tend to cause significant pleural thickening and parenchymal destruction; therefore, more radical surgical procedures, such as decortication, segmentectomy, and lobectomy may be required in these patients [2, 4, 14, 17]. Reports in the literature note resection rates of 0% to 7% for uncomplicated pulmonary hydatid cysts, whereas the corresponding figures for complicated pulmonary hydatid cysts are considerably higher at 19% to 32% [2, 4, 17]. As previously mentioned, marked pleural thickening due to cyst rupture into the pleural cavity is an important problem. In a study of 43 patients with pulmonary hydatid cysts and associated pleural complications, Aribas and colleagues [9] found that decortication was needed in 30 patients (69.8%) and pulmonary resection was needed in 6 patients (14%). In our series, 24.2% of the patients with complicated cysts required decortication. None of the complicated cysts in our study was associated with severe hemorrhage, serious parenchymal destruction, or bronchiectasis; therefore no lobectomy or pneumonectomy procedures were required.

Complicated pulmonary hydatid cysts are associated with higher postoperative morbidity and mortality than uncomplicated cysts [9, 14, 17]. In complicated cases, infection and inflammation of the adjacent lung parenchyma may affect wound healing and lead to postoperative complications such as prolonged air leakage, empyema, and pneumonia. In addition, many patients with complicated pulmonary hydatid cysts require preoperative antibiotic therapy and supportive treatment. These are the main reasons why complicated cases tend to have a higher morbidity and mortality rate and require longer hospitalization than uncomplicated cases. There was no mortality in either of our patient groups, and we attribute this to successful management of infectious complica-

tions. However, the patients with complicated cysts had higher morbidity and a longer hospital stay. Safioleas and colleagues [10] reported the same hospitalization trend in 42 patients with pulmonary hydatidosis (ie, a 12-day median stay for uncomplicated cases vs a 21-day median stay for complicated cases).

In conclusion, surgery is the safest curative treatment for pulmonary hydatid cysts. Although surgical treatment is effective in patients with complicated as well as uncomplicated cysts for treatment of pulmonary hydatidosis, surgical intervention before rupture of the cysts is essential. Experience with medical therapy in these cases is limited, but it is clear that these drugs can cause the cyst membrane to perforate, which can have life-threatening consequences. Ruptured cysts are associated with increased morbidity, more extensive surgery, and longer hospital stays. Regardless of whether symptoms are present, all pulmonary hydatid cysts should be surgically treated as soon as they are diagnosed in order to avoid complications.

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