Surgical Treatment of Pulmonary Hydatid Cysts, which Perforated to the Pleura

Plevral Perforasyonlu Akciğer Kist Hidatığında Cerrahi Tedavi

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Abstract

Objective: Hydatid cyst disease, endemic in Eastern region of Turkey, is a significant parasitic public health problem. In this study, pleural complications of hydatid cysts were presented in 76 cases.

Materials and Methods: In our study, of the 412 pulmonary hydatid cyst cases operated on between 2003 and 2011, 76 cases had ruptured into the pleura for various reasons, and the different clinicoradiological presentations were evaluated retrospectively. The age distribution of the cases was between 7 and 56 years, and the mean age was 26.20±13.04.

Results: The most frequent symptom due to pleural rupture in patients was dyspnea (44 cases, 57.8%). Etiologically, iatrogenic perforation was detected in four cases and thoracic trauma in nine cases (six car accidents and three falls from a height). An anthelmintic drug use history was found in three cases of ruptured pleura. Spontaneous perforation was detected in the other 60 (78.9%) cases. Two cases that were admitted to the emergency unit and were immediately administered a tube thoracostomy developed tension pneumothorax. In addition, 21 cases had hydro pneumothorax, 17 had pneumothorax, and 36 had pleurisy. Morbidity was observed in 30 cases (39.4%). Atelectasis was the most frequent cause of morbidity in these patients (10 cases). The mean duration of hospitalization was determined to be 12.26±2.90 days.

Conclusion: This disease can progress asymptptomatically for a long time and can sometimes lead to life-threatening emergency situations, such as tension pneumothorax. The delayed admission of a patient to a physician causes the disease to become more complicated and to increase the morbidity and mortality rates. Treatment of the disease is in the form of surgery. Possible parenchymal protection should be applied in surgical treatment, and anatomic resection should not be performed unless necessary.

Key Words: Empyema, Capitonnage, Pleura, Rupture

Introduction

The hydatid cyst, which is a parasitic disease formed by the larval forms of Echinococcus granulosus, is endemic in Turkey. It occurs more commonly in agriculture and livestock in rural life. The incidence rate is 1 in 2000, and lung involvement has been reported to be 35% [1]. According to data from the Ministry of Health of Turkey, the incidence is reported to be 14 per 100,000 [2].

Pleural involvement of the hydatid cyst can be in the form of a primary infestation [3, 4]. Secondarily, hydatid cysts invasion of the pleura can be the result of a tube thoracostomy.
given to a patient who previously had hydatid cyst surgery or the treatment of thoracic hydatid cyst [5, 6].

Pleural manifestation may develop as a complication of pulmonary hydatid disease, which does not involve a parasitic infestation [6-9]. The rupture of a hydatid cyst in the pleura can cause a wide range of clinical and radiographic symptoms, such as pneumothorax, hydropneumothorax, empyema, abscess formation, pleural thickening, and tension pneumothorax, to develop in the pleural area. The diagnosis is sometimes very difficult and even confused with malignancies. Perforated hydatid cysts emerge as a distinct entity that should especially be considered in differential diagnoses. Opening of the hydatid cyst to the pleura can be spontaneous and can also develop as a result of trauma or iatrogenically.

In this study, we aimed to present the pleural complications of this disease, which is frequently encountered in our region.

Materials and Methods

Of the 412 pulmonary hydatid cyst cases operated on between 2003 and 2011, 76 cases that had ruptured into the pleura for various reasons and with different clinico-radiological presentations were evaluated retrospectively. Forty-two of the cases were male, and thirty-four were female. The age distribution of the cases was 7-56 years, and the mean age was 26.20±13.04. The vast majority of the treated patients were living in rural areas (62 cases, 81.5%).

Because the cases were consulted on for complicated clinical and radiological symptoms, a wide range of diagnostic methods were utilized for the diagnosis. A clinical and radiological evaluation was performed initially. Routine plain and lateral chest radiography, computed thorax tomography (CT), abdominal tomography and abdominal ultrasonography, fiber optic bronchoscopy (FOB), and, in some cases, magnetic resonance imaging (MRI) were used. Routine hematomatological and biochemical analyses were performed on the cases. Because serological tests are not routinely used in our clinic, the indirect hemagglutination test (IHA) was used for a limited number of cases that were difficult to differentially diagnosis. In the evaluation of the perforated cyst, the cyst was considered to be a complicated cyst regardless of whether it was infected. Pleural abnormalities were categorized as pleural thickening, free fluid, pneumothorax, and hydropneumothorax radiographically. In those cases with pleural fluid, biochemical and microbiological evaluations were performed. In cases in which the pleural fluid was purulent, the empyema evaluation was assessed by considering the findings of the microbiological examinations.

Perforation was diagnosed by clinical and radiological symptoms and intraoperative symptoms. The cases were analyzed in terms of their clinico-radiological status upon admission, pleural complications, causes of pleural rupture, preoperative and postoperative complications, surgical procedures, and hospital treatment procedures.

Surgical technique

In all cases, a double-lumen endotracheal intubation was performed by the anesthesia clinic. The thorax was incised with a posterolateral thoracotomy from fifth or sixth intercostal spaces. The thorax was incised extrapleurally in those patients who had chronic pleural adhesions and pachypleuritis. The pachypleuritis, which emerged locally or throughout the entire pleural space, was decorticated, and the pleural space was visualized. In patients with a bilateral pathology, the lateral lung was incised after a three-week period. After the lung was relieved, the area of the cystic lesion was determined. Although these cases were considered perforated hydatid cysts, betadine packs and compresses were used to protect potential non-perforated hydatid cysts and vesicles in the pouch around the pet. Then, a cystectomy was performed. The cyst pouch was irrigated with gas, and the pets were impregnated with betadine after the germinative membrane inside the pouch was removed. Bronchial leaks were closed with absorbable suture material (Vicryl Ethicon, Inc, USA), and the pouch was obliterated. The lung was pumped by working in coordination with the anesthesia clinic, and those lung fields that were suffocated and destroyed were excised. Surgical intervention continued while maintaining the parenchyma where possible. Resection was performed after the surgeon determined the expanse state of the lung and in situations in which a significant proportion of the lung was destroyed and not functioning. Although wedge resections were performed many times, anatomic resection was avoided as much as possible, and a lobectomy was performed in only one patient. All patients were given antibiotics during the preoperative and postoperative periods, and cases that developed infection-empyema were given appropriate antibiotics based on the results of microbiological evaluations. All patients were treated with andazol in the postoperative period, which was continued for 45 days over two periods. While being given andazol, the patients were called to the clinic at regular intervals to check their blood biochemistry.

Results

In terms of the distribution of the lesions, the hydatid cyst localized in the right lung in 47 cases and in the left lung in 39 cases. The lesion was detected in the right lower lobe in 26 cases, the right middle lobe in 9, and the right upper lobe in 12. The location of the cysts in the left lung was the upper lobe in 21 cases and the lower lobe in 18. Bilateral hydatid cysts were identified in 13 cases. While a single hydatid cyst lesion was observed in 58 patients, multiple hydatid cyst lesions in either the right or left lung were detected in 18 cases.
Due to pleural rupture, the most frequent symptom was dyspnea in patients (44 cases, 57.8%). Other symptoms included cough (42 cases, 55.2%), chest pain (39 cases, 51.3%), fatigue (32 cases, 42.1%), and fever (28 cases, 36.8%) (Table 1). When the etiopathology was investigated by diagnostic thoracentesis, the perforated hydatid cyst was a result of iatrogenic perforation in four cases and thoracic trauma in nine (six car accidents and three falls from a height). In three cases with pleural rupture, the patients had already been taking anthelmintic drugs. The remaining 60 (78.9%) cases were found to have spontaneous perforation (Table 2).

Of the cases admitted to the emergency unit and immediately administered a tube thoracostomy, tension pneumothorax was observed in two, hydropneumothorax in 21, pneumothorax in 17, and pleurisy in 36. Although five cases were treated with an emergency tube thoracostomy due to a persistent, massive air leak despite tube thoracostomy, all other cases were treated with elective thoracotomy. In all of these cases, the pleural rupture of the hydatid cyst and subsequent complications were detected.

Although a hydatid cyst could be diagnosed in 61 patients during the preoperative period with radiological diagnostic tools, serological methods, and FOB, other patients could not be diagnosed until the operative period. Only two of the seven cases administered FOB could be diagnosed. Positivity was detected in four of the six cases considered to be hydatid cysts in the differential diagnosis and administered the indirect hemagglutination test. Most cases were evaluated as nonspecific pleuritis and pleurisy in the preoperative period because of intensive pleural thickening and pleural complications. In these patients, a thoracotomy was performed for various reasons, such as an on-going air leak despite the tube thoracostomy and pleural thickening, and a diagnosis was then established in the operative period.

Thirty-six patients were admitted for pleural effusion, and twenty-one for hydropneumothorax. In the analysis of the pleural fluids in these cases, 32 cases of nonspecific exudative character and 25 cases of empyema were detected.

In those patients with a pulmonary hydatid cyst with pleural perforation, the hydatid cyst was detected in the liver in 11 cases and in the spleen in one case.

All patients underwent a tube thoracostomy before treatment. Five cases underwent thoracotomy urgently due to a massive air leak, despite thoracostomy. Other cases were operated on under elective conditions. Capitonnage plus cystectomy was the most frequent surgical procedure (52 cases, 68.4%). Eighteen cases were administered partial or total decortication with extrapleural surgery because of intense pleural adhesions and the formation of pachypleuritis (Table 3).

Morbidity was observed in 30 cases (39.4%). Atelectasis was the most frequent cause of morbidity in patients (10 cases). Other complications included wound infection (9 cases), a prolonged air leak lasting more than 10 days (5 cases), empyema (3 cases), and pneumonia (3 cases) (Table 4). Pneumonia and sepsis appeared in one of the pleural perforation hydatid cyst cases.

The mean duration of hospitalization was 12.26±2.90 days. During the first six months of the postoperative period, the patients were called for monthly checks. After six postoperative months, they were followed-up every three months. Recurrence was detected in three of the patients within the two-year follow-up postoperative period.

### Discussion

Pulmonary hydatid cysts and their complications have been known to exist since the time of Hippocrates [10]. A pulmonary hydatid cyst does not form symptoms unless it reaches a large size and shows indications of lung compression or becomes complicated. When a pulmonary hydatid cyst becomes complicated, it can result in serious clinical outcomes. Cysts that open to the pleura or bronchia and that become infected develop a number of pleural and parenchymal complications, a condition known as a complicated cyst. Complicated cysts are difficult to diagnose and differentially diagnose. In perforated cysts, the clinical situation varies depending on the characteristics of the perforation and whether the cyst ruptures into the pleura or the bronchia. Perforation of the cyst into the bronchia is a common occurrence. In Qian’s study, 356 of 842 patients (42.3%) were reported as having complicated hydatid cysts [11]. In the 412 hydatid cyst patients seen in our clinic between 2003 and 2011, 176 (42.71%) had complicated hydatid cysts. Of these cases, 100 (56.8%) had cysts that ruptured into the bronchial tree, and 76 (43.2%) had cysts that ruptured into the pleura.

### Table 1. Distribution of the patients according to symptoms

<table>
<thead>
<tr>
<th>Symptoms</th>
<th>Number and % of patients</th>
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<tbody>
<tr>
<td>Dyspnea</td>
<td>44 (57.8%)</td>
</tr>
<tr>
<td>Cough</td>
<td>42 (55.2%)</td>
</tr>
<tr>
<td>Chest pain</td>
<td>39 (51.3%)</td>
</tr>
<tr>
<td>Fatigue</td>
<td>32 (42.1%)</td>
</tr>
<tr>
<td>Fever</td>
<td>28 (36.8%)</td>
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### Table 2. Causes of pleural perforation

<table>
<thead>
<tr>
<th>Causes</th>
<th>Number and %</th>
</tr>
</thead>
<tbody>
<tr>
<td>Iatrogenic</td>
<td>4 (5.2%)</td>
</tr>
<tr>
<td>Trauma</td>
<td></td>
</tr>
<tr>
<td>Motor vehicle accident</td>
<td>6 (7.8%)</td>
</tr>
<tr>
<td>Falling from a height</td>
<td>3 (3.9%)</td>
</tr>
<tr>
<td>Intake of anthelmintic drug</td>
<td>3 (3.9%)</td>
</tr>
<tr>
<td>Spontaneous perforation</td>
<td>60 (78.9%)</td>
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</table>
As a result of the pulmonary hydatid cyst perforating the pleura, symptoms such as chest pain, coughing, cyanosis, fever, and shortness of breath develop. As a result of the rupture of the cyst, the cyst contents pour into the pleura, and the ruptured germinative membrane collapses. Therefore, signs of compression occur in the latent lung parenchyma. Severe clinical manifestations, such as tension pneumothorax, can sometimes occur as a result of a bronchopleural fistula. In addition, air-fluid leveling within the pleural space occurs. Cernay identified tension pneumothorax in five of 336 patients in a case series (1.3%) [12]. In our study, tension pneumothorax occurred in two cases, depending on the pleural perforation. Severe air hunger, tachycardia, agitation, and severe chest pain were observed in the tension pneumothorax patients. These patients underwent thoracotomy immediately after a tube thoracostomy. In most of the cases with pneumothorax and hydropneumothorax, there was also significant mediastinal compression and a mediastinal shift. These patients underwent surgical treatment under elective conditions after the tube thoracostomy.

A hydatid cyst is diagnosed with radiology (X-ray, computed tomography, MRI, ultrasound), serological investigations, and pathological examinations [13]. Thoracic hydatid cysts are diagnosed on the basis of radiological findings [14, 15]. As a result of pleural complications, the diagnosis could be incorrect, and eliminating hydatid cyst as a diagnosis may be difficult. Indeed, in 61 patients, a hydatid cyst was diagnosed during the preoperative period, but the other cases were diagnosed using diagnostic thoracotomy. Radiological images can reveal symptoms varying from locular hydropneumothorax to non-locular partial, complete or tension hydropneumothorax, which occur during an acute phase in which the cyst ruptures the pleura. In cysts that have ruptured the pleura, air-liquid leveling also emerges. Most cases of pleural rupture (61 cases, 80.2%) were diagnosed with radiological methods, FOB, and serology. For the other 15 (19.7%), there were difficulties in the differential diagnosis due in particular to benign tumors, inflammatory masses, other pathologies that generate an air-fluid level, infectious processes, metastatic disease, and malignant masses. The diagnosis of a hydatid cyst was confirmed with thoracotomy in all patients.

The rupture of a hydatid cyst might be spontaneous, traumatic, or iatrogenic and may also occur with cough or a growth in the cystic diameter. When the etiopathology was investigated by diagnostic thoracentesis in the pleural rupture cases, four had iatrogenic perforation, and nine had thoracic trauma (six car accidents and three falls from a height). Three pleural rupture cases had already been using anthelmintic drugs. Spontaneous perforation was detected in the other 60 (78.9%) cases.

Due to the elasticity of the lung, a hydatid cyst may not be perforated even though it sometimes reaches a large size. This situation is more common among children and young adults [16]. An extremely large hydatid cyst, in excess of 10 cm in diameter, was detected in 41 (23.29%) cases of our hydatid cyst series (412 cases).

Bacterial infection is one of the most serious complications of a perforated cyst. Empyema was detected in 25 cases in our series. Therefore, there were a high number of cases (18 cases, 23.6%) in which decortication was performed because of pleural contamination and pleural thickening.

Surgical treatment remains the most valid method of treatment for a pulmonary hydatid cyst, regardless of whether it is symptomatic [17]. Treatment was in the form of surgery for all our cases. The standard posterolateral thoracotomy incision was used. In the case of bilateral hydatid cysts, the latent part was operated on after a three-week period. In the cases of a complicated cyst in the right lung, which was accompanied by a liver dome cyst, an operation was performed using a single-stage, right thoracophrenotomy incision. Surgical treatment was performed by preserving the lung parenchyma where possible. Resection should not be avoided if complications, depending on significant infection, develop or if there is bronchiectasis. In the literature, the rate of resective surgery is approximately 0-7% for intact cyst surgery and 19-32% for complicated cysts [18]. Although wedge resection was performed on five of the cases with pleural perforation, a lobectomy was performed in one patient whose upper right lobe was almost completely destroyed.
There are various opinions regarding the medical treatment of hydatid cysts, but the cure rates of anthelmintic treatment are not greater than 30-35% [18]. In addition, anthelmintic treatment causes serious complications requiring urgent intervention, such as pneumothorax and massive hemoptysis, as we observed in three cases in our study. Therefore, drug therapy should be performed only in inoperable patients, and we do not recommend the use of anthelmintic drugs during the preoperative period.

The surgical treatment of a hydatid cyst is usually satisfactory. Recurrence rates were reported as 2-25% in the literature, and mortality rates associated with the operation were reported as 0.5-4% [19, 20]. In complicated cysts rupturing into the pleura or bronchia, higher rates of morbidity and mortality have been reported [21]. The risks of empyema, prolonged air leak, pneumonia, pneumothorax, hydro pneumothorax, hemoptysis, dyspnea, allergic reactions and anaphylaxis are higher in perforated cysts. Thus, the duration of the patient’s response to treatment, healing process and discharge is extended. The need for antibiotics and supportive treatment increases in these patients. Morbidity was observed in 30 cases (39.4%). Atelectasis was the most frequent cause of morbidity in these patients (10 cases). Other complications included wound infection (9 cases), a prolonged air leak lasting more than 10 days (5 cases), empyema (3 cases) and pneumonia (3 cases). Pneumonia and sepsis appeared in one of the pleural perforation hydatid cyst cases.

The mean duration of hospitalization was 12.26±2.90/ days. In the first six months of the postoperative period, patients were followed-up monthly, and after six postoperative months, they were followed-up every three months. Recurrence was detected in three of the patients within a two-year follow-up postoperative period.

In conclusion, hydatid cyst disease, which is particularly endemic in our region, is a public health problem. The disease can progress asymptptomatically for a long time and can occur alongside various other diseases, such as pneumonia, empyema, pneumothorax, pyopneumothorax or tension pneumothorax. Invasive procedures, such as thoracotomy or the use of anthelmintic drugs, can lead to complicated hydatid disease, and trauma can also be a cause of the pleural perforation of the cyst. Pleural perforation can sometimes lead to life-threatening emergency situations, such as tension pneumothorax. The delayed referral of patients to physicians causes the disease to become more complicated and leads to an increase in morbidity and mortality. The treatment of the disease is in the form of surgery. Possible parenchymal protection should be applied during the surgical treatment, and anatomic resection should not be performed unless necessary.

Conflict of interest statement: The authors declare that they have no conflict of interest to the publication of this article.

References