Factors affecting outcome and morbidity in pulmonary hydatid cyst surgeries

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Abstract
Aim: Hydatid cyst disease is an important health problem due to its endemic, progression, serious complications, and rarely spontaneous regression.

Material and Methods: The files of 143 patients who were operated for hydatid cysts in our clinic between January 2000 and December 2010 were analyzed retrospectively. The effects of parameters such as age, gender, symptom, number of cysts, cyst size, whether the cyst is perforated, leukocyte count, and localization of the cyst on mortality and morbidity were investigated.

Results: Morbidity rates were high in perforated cysts. However, gender, cyst size, and localization had no effect on morbidity (p<0.05).

Discussion: We determined that perforated pulmonary hydatid cysts significantly prolonged hospital stay and increased postoperative morbidity. Pulmonary hydatid disease, which is a public health problem in our country, should be treated surgically when diagnosed.

Keywords
Lung, Hydatid cyst, Morbidity, Perforation
Introduction

Hydatid cyst disease, also known as hydatidosis, is a disease characterized by the emergence of cysts as a result of the transmission of Taenia echinococcus larvae to humans. There are four species of Taenia echinococcus, of which Echinococcus granulosus and Echinococcus multilocularis are disease agents in humans [1]. Due to its social conditions and geography, our country is among the places where the disease is common [2]. The prevalence of hydatid cyst disease in Turkey, which is seen in all regions, especially in rural settlements, has been reported to be around 50 in 100,000 and its incidence is around 2 in 100,000 [3]. Following the liver, the organ most affected by the disease, lung involvement is observed in 10-30% of patients [4]. Clinical findings are variable and are mostly related to the growing cyst. As long as it is not perforated, it does not cause a special complaint in the patient. It is usually alone and asymptomatic in humans [5]. Findings are variable, but mostly related to the growing cyst. Rarely, it is related to the rupture of the cyst, the spread of the parasite, and the formation of inflammation and allergic findings [6]. Conventional radiology is mostly used in diagnosis. Sometimes the diagnosis can be made on the basis of anamnesis, while sometimes they can be diagnosed intraoperatively [2]. Radiological methods provide the diagnosis in 98% of cases, but sometimes it can be confused with abscess, malignant tumors or cysts of other origin [7].

Surgical intervention is an effective primary treatment for pulmonary hydatid cyst disease. Perforated cysts should be treated immediately as they can be life-threatening [8]. Anthelmintic therapy is recommended in cases where the cysts are inoperable due to the presence of recurrent hydatid cysts or widespread or difficult localization of the cyst, or when surgery involves a high morbidity and mortality risk [9].

In our study, we investigated some factors that may affect morbidity and mortality in lung hydatid cysts. These include age, gender, location of the cyst, size of the cyst, whether it is perforated, symptom, number of cysts, localization of the cyst, and white blood cell values. We aimed to evaluate the relations of these determined parameters with postoperative complications using appropriate statistical tests.

Material and Methods

After approval of the ethics committee (18.03.2011/58), the files of the patients diagnosed with hydatid cyst who were operated in our clinic between 2000-2010 were evaluated retrospectively. The patients were divided into two groups as those without morbidity (group 1; 106) and those with morbidity (group 2; 37). The age, gender, symptoms of the patients, whether the cyst was perforated, the size of the cysts, the number of cysts, the number of leukocytes, and the treatment methods applied to the patients were recorded. The effects on morbidity and mortality were evaluated.

Statistics

In statistical analysis, continuous variables were expressed as mean± standard deviation, while categorical variables were expressed as number-ratio. The results were evaluated with the Student-t and Chi-square tests, p<0.05 was considered significant.

Results

A total of 143 patients were included in the study. Fifty-three were males and 90 were females. In group 1, 39 patients were male and 67 were female. In group 2, 14 patients were male and 23 were female. The gender difference was not significant in the development of morbidity (p>0.05).

The mean age in Group 1 was 20.84±15.98 years, the mean cyst size of the right lung was 4.23±4.42, the mean cyst size of the left lung was 3.15±4.50, perforation rate was 0.29±0.49, and mean leukocyte was 12.26±6.06. On the other hand, the mean age in Group 2 was 20.46±15.53, mean cyst size of the right lung; 3.32±4.55, the mean cyst size of the left lung was 4.84±5.68, perforation rate was 0.5±0.5, and the mean leukocyte was 12.1±5.10. When compared, a significant difference was found between the perforation rate and morbidity in Group 2 (p<0.05) (Table 1).

In Group 1, 90 patients underwent cystotomy+capitonnage, 4 underwent cystotomy+capitonnage+decortication, 3 underwent cystotomy, 2 had primary closure, 1 underwent wedge resection, 1 underwent decortication, 1 had cystotomy+enucleation, 1 underwent cystotomy+excision, 1 underwent cystotomy+capitonnage+wedge resection, 1 underwent cystotomy+capitonnage+phrenotomy (Table 2).

Table 1. Demographic distribution of the groups and their mean values according to some criteria

<table>
<thead>
<tr>
<th>Criteria</th>
<th>Morbidity (-)</th>
<th>Morbidity (+)</th>
<th>Student-t</th>
<th>P Value</th>
</tr>
</thead>
<tbody>
<tr>
<td>Male</td>
<td>67</td>
<td>23</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Women</td>
<td>39</td>
<td>14</td>
<td>-</td>
<td>-</td>
</tr>
<tr>
<td>Average age</td>
<td>20.84±15.98</td>
<td>20.46±15.53</td>
<td>0.125</td>
<td>0.9</td>
</tr>
<tr>
<td>Average cyst size (right)</td>
<td>4.23±4.42</td>
<td>4.23±4.42</td>
<td>1.06</td>
<td>0.291</td>
</tr>
<tr>
<td>Average cyst size (left)</td>
<td>3.15±4.50</td>
<td>4.84±5.68</td>
<td>-1.82</td>
<td>0.07</td>
</tr>
<tr>
<td>Perforation</td>
<td>0.29±0.497</td>
<td>0.5±0.5</td>
<td>-2.603</td>
<td>0.01</td>
</tr>
<tr>
<td>Leukocyte count</td>
<td>12.26±6.068</td>
<td>12.1±5.10</td>
<td>0.111</td>
<td>0.912</td>
</tr>
</tbody>
</table>

Table 2. Effect of surgical techniques on morbidity

<table>
<thead>
<tr>
<th>Surgical techniques</th>
<th>Morbidity (-)</th>
<th>Morbidity (+)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Cystotomy+Capitonnage</td>
<td>90</td>
<td>21</td>
</tr>
<tr>
<td>Cystotomy+Capitonnage+Decortication</td>
<td>4</td>
<td>9</td>
</tr>
<tr>
<td>Cystotomy</td>
<td>3</td>
<td>0</td>
</tr>
<tr>
<td>Primary Closure</td>
<td>2</td>
<td>0</td>
</tr>
<tr>
<td>Wedge Resection</td>
<td>1</td>
<td>4</td>
</tr>
<tr>
<td>Decortication</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+Enucleation</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+Excision</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+Capitonnage+Wedge Resection</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+Capitonnage+Enukleasyon</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+Wedge Rezaktion+Decortication</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>Cystotomy+ Capitonnage+ Phrenotomy</td>
<td>0</td>
<td>3</td>
</tr>
</tbody>
</table>

Table 3. Localization of cysts in the lung

<table>
<thead>
<tr>
<th>Localization</th>
<th>Morbidity (-)</th>
<th>Morbidity (+)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Extra upper and lower lobe</td>
<td>19</td>
<td>4</td>
</tr>
<tr>
<td>Upper lobe</td>
<td>35</td>
<td>13</td>
</tr>
<tr>
<td>Lower lobe</td>
<td>44</td>
<td>14</td>
</tr>
<tr>
<td>Both upper and lower lobes</td>
<td>8</td>
<td>6</td>
</tr>
<tr>
<td>Total</td>
<td>106</td>
<td>37</td>
</tr>
</tbody>
</table>
capotomy+enucleation, and 1 had cystotomy+wedge resection+decortication. On the other hand, 21 patients from Group 2 underwent cystotomy+capitonnage, 9 underwent cystotomy+capitonnage+enucleation, 4 patients had wedge resection, and 3 had cystotomy+capitonnage+frenotomy. The most common morbidities in Group 2 were atelectasis (n: 18), expansion defect (n: 10), and postoperative air leak (n: 9). However, the difference in surgical techniques in the development of morbidity was not statistically significant (p>0.05) (Table 2).

In Group 1, 44 patients had lower cyst localization, 35 had upper localization, 8 had both upper and lower localization, and 19 had localization outside the lower and upper lobes. In Group 2, 14 had lower localization, 13 had upper localization, 6 had both upper and lower localization, and 4 had localization outside the lower and upper lobes. In Group 2, postoperative atelectasis was more common in cysts located in the lower lobes, while postoperative expansion defect was more common in cysts located in the upper lobes (p<0.05).

When evaluated in terms of the number of cysts, in Group 1, 1 cyst in was found in 81 patients, 2 cysts in 17 patients, 3 cysts in 5 patients, 4 cysts in 1 patient, and 5 cysts in 2 patients were detected. In Group 2 patients, 1 cyst was identified in 28 patients, 2 cysts in 6 patients, 3 cysts in 1 patient, 4 cysts in 1 patient, and 5 cysts in 1 patient. Statistically, there was no significant effect of the number of cysts on morbidity (p>0.05). While the number of giant cysts (>10cm) was 33 in Group 1, it was 15 in Group 2. There was no significant difference in terms of morbidity (p>0.05).

Discussion
While hepatic hydatid cysts are more common in females, pulmonary hydatid cysts are more common in males [10]. In a study with 13000 pulmonary hydatid cysts, 45% of the patients were females and 57% were males [1]. In our study, contrary to the literature, 62% of the patients were females and there was no statistical difference in terms of postoperative morbidity according to gender.

Although hydatid cyst disease can be seen at any age, it is more common in adulthood. In many studies, it has been reported that hydatid cyst disease is more common in the 15-35 age group [12]. In our study, the youngest age was 4 years old, while the oldest was 67 years old, and the mean age was 20.46 years. We found that age did not significantly affect postoperative morbidity.

Although hydatid cysts can be seen in many organs of the body, the most frequently involved organs are liver and lung, respectively [13]. The coexistence of lung and liver involvement is between 20-40% [11]. Cough-sputum, pain, hemoptysis, dyspnea, and fever are the most common symptoms of pulmonary hydatid cyst [14]. The most common symptoms in our study were chest pain (46.5%), shortness of breath (25%), cough (8.3%), and fever (8.3%). We found that these symptoms did not affect postoperative morbidity.

There are no specific clinical and laboratory findings in the diagnosis of hydatid cyst disease. Radiological evaluation is much more important and the first step is direct chest radiography. On the radiograph, intact lung cysts appear as round, homogeneous, and well-circumscribed opacities. However, complicated cysts can mimic many other lung lesions. Computed tomography of the thorax is very successful in imaging both intact and complicated cysts [15]. In our study, the diagnosis was mostly made by direct radiography, computed tomography of the thorax, and ultrasonography.

While no mortality was reported in the hydatid cyst study of 279 cases, mortality was reported in one case in the giant cyst series of 50 cases by the same authors [13]. Karaoglanoglu et al. reported one death due to respiratory failure in their series of 67 cases of giant hydatid cyst [16]. In our study, there was no mortality in 48 patients with giant cysts.

Regardless of whether the pulmonary hydatid cysts are symptomatic or not, the surgical treatment method is still up-to-date [13]. VATS can be applied in selected patients, but it should be kept in mind that control of the cyst content may be difficult in this method, and daughter vesicles may spread to the surrounding tissue [17]. If the cyst fills a lobe, becomes infected, alveolar cyst is present, and the exocyst is calcified, resection is recommended [16]. In their series consisting of 100 cases, Aytaç et al. performed 4% wedge resection, 1% segmentectomy and 11% lobectomy [18]. In our study, 78.3% of patients underwent cystotomy capitonnage, 7.9% had cystotomy+capitonnage+decortication, 3.5% underwent wedge resection, 3.5% had cystotomy, and 3% had enucleation.

Among the postoperative complications, those seen within the first seven days are primarily hemothorax, wound infection, atelectasis, pneumonia and respiratory failure, empyema, sepsis, and thrombophlebitis. Post-cystectomy residual cavity and bronchopleural fistula are more common after the seventh day [10]. In their series of 240 cases with pulmonary hydatid cyst, Raul B. et al. demonstrated pneumothorax in 24 cases (8.8%), pleural effusion in 20 cases (7.4%), wound infection in 18 cases (6.6%), bronchopleural fistula in 12 cases (4.4%), empyema in 10 cases (3.7%), atelectasis in 6 cases (2.2%), phrenic nerve palsy in 2 cases (0.7%), bleeding in 2 cases (0.7%), and deep vein thrombosis in 1 case (0.4%) [19]. In their pulmonary hydatid cyst series of 139 cases, Ulku et al. found atelectasis as the most common postoperative complication [20]. In our study, we found empyema, bronchopleural fistula, atelectasis, prolonged air leakage, expansion defect, and hemothorax as postoperative complications. Consistent with the literature, the most common postoperative complication in our study was atelectasis.

Celik et al. reported that 80% of 13,006 cases had a single cyst, and in half of those with multiple, cysts were bilaterally located [11]. Although pulmonary cysts are found in every lobe of the lung, they are more common in the right hemithorax and lower lobes [21]. When studies on pulmonary hydatid cysts are examined, the rate of localization of pulmonary cysts in the right hemithorax is between 52.7% and 63.2% [20]. Studies have also reported that 50.1% and 76.7% of the cysts are located in the lower lobes [22]. Bilateral lung localization of hydatid cysts ranges from 2% to 14% in patient series [23]. In our study, 51% were localized in the right hemithorax, 43% were in the left hemithorax, and 6% had bilateral localization.

In accordance with the literature, we detected hydatid cysts mostly in the right lower lobe with a rate of 38%. We found
that the number and size of the localization of the cyst did not statistically affect the postoperative morbidity.

In perforated cases, infection and inflammation of the adjacent lung parenchyma may affect wound healing and lead to postoperative complications such as prolonged air leak, empyema, and pneumonia. In addition, patients with perforated pulmonary hydatid cysts require preoperative antibiotic and supportive treatment. These are the main reasons why perforated cases have higher mortality and morbidity. Perforated cases require longer hospital stays than non-perforated cases. In the studies by Kuzucu et al., they found higher morbidity in patients with perforated cysts [24]. In our study, we found that perforated hydatid cysts increased postoperative morbidity and prolonged hospital stay, consistent with the literature.

Conclusion

Hydatid cyst should be considered in undiagnosed lung lesions in areas where Echinococcus granulosus is endemic. Evaluation of CT findings together with serology is useful in confirming the diagnosis. Patients with pulmonary hydatid cysts most frequently present with complaints of shortness of breath, cough and chest pain, and these patients are mostly diagnosed with a single and intact (non-perforated) cyst. Postoperative morbidity is more common in perforated lung hydatid cysts, so the importance of diagnosis and treatment of pulmonary hydatid cysts before perforation increases.

Scientific Responsibility Statement

The authors declare that they are responsible for the article's scientific content including study design, data collection, analysis and interpretation, writing, some of the main line, or all of the preparation and scientific review of the contents and approval of the final version of the article.

Animal and human rights statement

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No animal or human studies were carried out by the authors for this article.

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Conflict of interest

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