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Research Article / Araştırma

Surgical Treatment Methods and Results in Complicated Pulmonary Hydatid Cyst

Komplike Akciğer Hidatik Kistlerinde Cerrahi Tedavi Yöntemleri ve Sonuçları

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ABSTRACT

Objectives: Pulmonary cystic hydatidosis (PCH) is a parasitic infestation caused by *Echinococcus granulosis*. Despite all advanced diagnosis and treatment methods, morbidity is between 3.5 and 18% and mortality is between 0 and 2%. We aimed to contribute to the reduction of mortality and morbidity by discussing the relationship between cyst rupture and pre-operative/post-operative complications and treatment methods in complicated pulmonary hydatid disease with the knowledge gained in the following years.

Methods: The files of 101 patients were evaluated retrospectively. Patients' age, gender, number of hydatid diseases, rupture, symptoms, pre-operative and post-operative complications, type of operation, length of hospital stay, follow-up periods, and mortality rates were analyzed.

Results: The mean age was 39.2±18.2. There were pre-operative complications in 35 (34.6%) of the patients. Cyst rupture was detected in 54 (53.5%) of the cases. The presence of rupture was found to be highly correlated with pre-operative/post-operative complications, symptoms, preference for resection, and length of stay. The most common pre-operative complication was lung abscess in 18 (17.8%) cases. Post-operative complications were observed in 30 (29.7%) of the patients. No relapse was detected. Mortality occurred in 2 (1.9%) cases. Lobectomy was found to be associated with pre-operative complications. Complicated hydatid cysts were associated with high morbidity and prolonged hospital stay.

Conclusion: The curative treatment of pulmonary hydatid disease is surgery. The need for major surgery is higher in complicated PCH. Cyst rupture is associated with pre-operative/post-operative complications. We hope that treating PCH before complications develop will reduce morbidity, length of hospital stay, and therefore the cost of treatment.

Keywords: Capitonnage, cystic echinococcosis, cystotomy, hydatid disease, pulmonary hydatid cyst

ÖZ

Amaç: Pulmoner kist hidatik, *Echinococcus granulosis*'in neden olduğu parazitik bir enfestasyondur. Tüm ileri tanı ve tedavi yöntemlerine rağmen morbidite %3,5–18, mortalite %0–2 arasındadır. Bu çalışmada, komplike pulmoner kist hidatik hastalığında kist rüptürü ile preoperatif/postoperatif komplikasyonlar arasındaki ilişkiyi ve tedavi yöntemlerini tartışarak mortalite ve morbiditenin azaltılmasına ilerleyen yıllardaki bilgi birikimi eşliğinde katkı sağlanması amaçlandı.

Yöntem: Çalışmada, 101 hastanın dosyaları geriye dönük olarak değerlendirildi. Hastaların yaşı, cinsiyeti, kist hidatik sayısı, rüptürü, semptomları, preoperatif ve postoperatif komplikasyonları, ameliyat tipi, hastanede kalış süreleri, takip süreleri ve mortalite oranları analiz edildi.

Bulgular: Ortalama yaş 39,2±18,2 idi. Hastaların 35'inde (%34,6) preoperatif komplikasyon gelişti. Olguların 54'ünde (%53,5) kist rüptürü saptandı. Rüptür varlığının preoperatif/postoperatif komplikasyonlar, semptomlar, rezeksiyon tercihi ve hastanede kalış süresi ile yüksek oranda ilişkili olduğu bulundu. Ameliyat öncesi en sık komplikasyon 18 (%17,8) olguda akciğer apsesi idi. Hastaların 30'unda (%29,7) ameliyat sonrası komplikasyon görüldü. Relaps tespit edilmedi. Mortalite 2 (%1,9) olguda meydana geldi. Lobektominin preoperatif komplikasyonlarla ilişkili olduğu bulundu. Komplike hidatik kistler yüksek morbidite ve uzun hastanede kalış süresi ile ilişkilendirilmiştir.

Sonuç: Pulmoner kist hidatik hastalığının küratif tedavisi cerrahidir. Komplike pulmoner kist hidatikte majör cerrahi ihtiyacı daha yüksektir. Kist rüptürü preoperatif/postoperatif komplikasyonlarla ilişkilidir. Komplike pulmoner kist hidatiği en erken zamanda, komplikasyonlar gelişmeden önce tedavi etmenin morbiditeyi, hastanede kalış süresini ve tedavi maliyetini azaltacağını umuyoruz.

Anahtar sözcükler: Kapitonaj, kistik ekinokokkoz, kistotomi, hidatik hastalık, pulmoner kist hidatik.

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Introduction

Hydatid disease is a parasitic infestation caused by *Echinococcus granulosis*.[1] Lungs are the second most frequent in which hydatid disease found after the liver. In the lung, it often prefers the right lung and lower lobes.[2]

Infestation is common in geographic areas where there is constant contact with fatlings such as domestic carnivorous animals, cattle, and sheep.[3] According to the World Health Organization data, hydatid disease has a worldwide geographic distribution and can occur worldwide.

Despite all advanced diagnosis and treatment methods, morbidity is between 3.5 and 18%, and mortality is between 0 and 2%.[4] Some cases may apply to the hospital with the development of the complication. These patients are more challenging for clinicians and are more challenging to treat.

We aimed to contribute to the reduction of mortality and morbidity by discussing the relationship between cyst rupture and pre-operative/post-operative complications and treatment methods in complicated pulmonary hydatid disease with the knowledge gained in the following years.

Methods

Our study was carried out in accordance with the Declaration of Helsinki, and approval was obtained from the Kahramanmaraş Sütçü İmam Üniversitesi Clinical Research Ethics Committee (Ethics committee date 19.10.2021/ decision no: 02).

The files of 101 patients over the age of 18 who were operated on for pulmonary hydatid diseases in Kahramanmaraş Sütçü İmam University Faculty of Medicine Department of Thoracic Surgery and Necip Fazıl City Hospital between January 2011 and January 2021 were retrospectively analyzed. Patients' age, gender, number of cysts, laterality, affected lobe, cyst rupture, other organ involvement, symptoms, pre-operative and post-operative complications, type of surgery, length of hospital stay, follow-up time, and mortality rates were recorded. Patients under 18, who refused to be operated on, could not be operated on for medical reasons, and whose pathology results were not compatible with hydatid disease were excluded from the study. Detailed information about the patients was obtained from the electronic database of both hospitals. Methods such as Casoni intradermal skin test, Weinberg complement fixation test, and eosinophil count were not used because of low diagnostic values due to cross-reactions.

Lung abscess, pleural effusion, empyema, chest wall invasion, and allergy or anaphylaxis, present at the time of admission, were considered preoperative complications. Empyema, atelectasis, expansion defect, prolonged air leak, wound infection, residual cavity, and hemorrhage developed during the treatment were post-operative complications. Hydatid cyst rupture was evaluated as a different parameter.

Double lumen intubation was performed in all cases, and a posterolateral thoracotomy incision was performed. When the cystic parenchyma area was reached, compresses were made with sponges moistened with the Povidone-iodine solution to prevent contamination of the surrounding tissue. Cystotomy-capitonnage operations were the preferred method for small, uncomplicated, and intact cysts. In cystotomy-capitonnage operations, after the cyst content was aspirated, cystotomy was performed and the germinative membrane was removed. The cavity was washed with a Povidone-iodine solution. Hypertonic NaCl was not used in any of our operations. After the open bronchial openings in the cavity were closed with absorbable suture materials, the cavity was also quilted with absorbable suture materials.

Other preferred modes of operation were wedge resection or lobectomy operations in ruptured, complicated, or multiple hydatid diseases. We did not have a case who underwent liver surgery in the same session. Sequential posterolateral thoracotomy was performed at appropriate time intervals in patients with bilateral hydatid diseases. No sternotomy was performed in any of the cases.

Medical treatment was not applied to any of the cases before the operation. After post-operative 3 weeks, albendazole treatment at a dose of 10 mg/kg/day was administered to our patients for 8 weeks. Liver enzymes (ALT/AST) and hemogram were checked at 3-week intervals. Albendazole treatment was discontinued in cases with elevated liver enzymes or neutropenia.

Statistical Analysis

Mean, standard deviation, median lowest, highest, frequency, and ratio values were used in the descriptive statistics of the data. The Kolmogorov-Smirnov test measured the distribution of the variables. Mann-Whitney U test was used in the analysis of independent quantitative data. The Chi-square test was used to analyze independent qualitative data, and the Fischer test was used when Chi-square test conditions were not met. SPSS 27.0 program was used in the analysis.

Results

Forty-nine (48.5%) of the 101 cases included in the study were female, and 52 (51.4%) were male (M/F=1.06). In this 10-year retrospective study, 75 (74.3%) cases were identified in the first 5 years and 26 (25.7%) cases in the past 5 years. The mean age was 39.2±18.2, the youngest 18 and the oldest 88 years old. The most common reason for ad-

mission to our clinic was cough in 30 (29.8%) cases. Pulmonary cystic hydatidosis (PCH) was localized on the right in 71 (70.3%) cases, on the left in 26 (25.7%), and bilaterally in 4 (4%). The rupture was detected in 54 (53.5%) cases. There were pre-operative complications in 35 (34.6%) of the patients. In the pre-operative period, the most common complication was lung abscess in 18 (17.8%) cases, and the second most common complication was pleural effusion in 13 (12.9%) cases.

The most frequent type of surgery was cystotomy+capitonnage in 67 (66%) cases, and the second frequent type was wedge resection in 22 (21%) cases.

Post-operative complications were seen in 30 (29.7%) cases. The most frequent post-operative complication was empyema and atelectasis in 9 (8.9%) cases, and the second frequent complication was expansion defect in 4 (3.9%) cases. Five (4.9%) of the patients were operated on due to relapse. The cases with recurrence were those who had been operated on in other centers and were referred to our clinic due to the development of recurrence. There was no case of relapse in our clinic. Mortality occurred in 2 (1.9%) patients. Age, gender, pre-operative and post-operative complications, laterality, lobes, type of surgery, relapse, and mortality rates are given in Table 1.

Cyst rupture was statistically significantly associated with pre-operative and post-operative complications ($p<0.001$). Drooling ($p<0.001$), cough ($p=0.028$) lobectomy ($p=0.011$), wedge resection ($p=0.022$), and duration of hospital stay ($p<0.001$) were significantly higher in the ruptured group than in the non-ruptured group ($p<0.05$). The rate of cystotomy was significantly lower in the ruptured group ($p<0.001$). The recurrence rate did not differ significantly between the ruptured and non-ruptured groups ($p>0.05$).

Cyst rupture ($p<0.001$), preference for lobectomy ($p<0.001$), post-operative complication ($p<0.001$), and length of hospital stay ($p<0.001$) were significantly higher in the group with pre-operative complications compared to the group without complications. On the other hand, the rate of cystotomy was significantly lower in the group with pre-operative complications ($p=0.003$). The rate of lobectomy in the group with pre-operative complications was significantly ($p<0.05$) higher than the group without pre-operative complications. Factors associated with pre-operative complications are presented in Table 1.

Drooling ($p=0.048$) and dyspnea ($p=0.021$) were significantly higher in the group with post-operative complications compared to the group without complications. There was no significant relationship between postoperative complications and surgical method, recurrence rate, and mortality ($p>0.05$).

Discussion

The most common sites of hydatid disease are the liver and lungs, respectively.[5] Embryos often reach the thorax after passing through the liver through the portal system. Rarely, it may cause lung infestations by inhalation from the lymphatic or bronchial system.[6]

The rate of co-incidence of lung and liver cysts is between 10 and 20%.[7] In our series, the hydatid disease incidence in the liver with a rate of 16% was consistent with the literature. In our study, similar to the literature, the most common localization was the right in 73 cases (72.3%) and the lower lobes in 69 (68.3%) cases.[8,9]

According to the current literature, cough is the most common symptom in PCH (52–62%) and occurs due to bronchial irritation or compression of the cyst.[10] The most common symptom in our study was cough. However, it was seen in 30 (29.8%) cases and was proportionally lower than in the literature. Drooling (hydatoptysis) suggests that the cyst has ruptured, and the cyst fluid is draining from the mouth. Pathognomonic for PCH. The second most common reason for admission in our study, higher than the literature, was profuse drooling.[11] We attribute this to late admission to the hospital in our region. Cases come to the hospital after rupture develops and complications begin.

Complicated PCH may be associated with or lead to bronchiolitis obliterans or organizing pneumonia.[12] In our study, similar to the literature, parenchymal infection was observed in the pre-operative period. In our study, the most common paramkinal infection seen in the pre-operative period was lung abscess in 18 (18%) cases. Lung abscess developed due to contamination of the cavity left behind by ruptured hydatid diseases with infectious agents. A definitive diagnosis was made by thoracotomy.

Echinococcal cysts always riska rupture with anaphylactic potential.[13] In our study, allergic and anaphylactic reactions were observed in 3% of rupture cases and were brought under control with appropriate interventions. In some studies, rupture has been reported in 34–64% of patients with hydatid diseases.[14,15] Our analysis detected a rupture rate of 53.5%, which is consistent with the literature. There was a statistically significant correlation between the rate of cases presenting with the symptom of drooling and rupture ($p<0.05$). In addition, a statistically significant correlation was found between rupture and pre-operative complications (such as pleural effusion, and lung abscess) in our series ($p<0.05$). The most frequent complications due to spontaneous or traumatic rupture of the cyst were reported as pleural effusion and empyema with 7.6%.[11,16] An abscess may develop in the cyst cavity after rupture.[16] In our study, the most common complication associated with rupture was pleural effusion.

Table 1. Demographic characteristics of the cases, the relationship between the characteristics of hydatid diseases and pre-operative complications

	Pre-operative complication (–)				Pre-operative complication (+)				p
	Mean±SD	n	%	Median	Mean±SD	n	%	Median	
Age	37.1±16.7			33.0	41.0±19.6			40.5	0.439*
Gender									
Female		34	51.5			15	42.9		0.407$^{X^2}$
Male		32	48.5			20	57.1		
Number of intrathoracic cysts	1.7±1.0			1.0	2.2±2.7			1.0	0.702*
Symptom									
(–)		9	13.6			1	2.9		0.084$^{X^2}$
(+)		57	86.4			34	97.1		
Rupture									
(–)		45	68.2			2	5.7		<0.001
(+)		21	31.8			33	94.3		
Side									
Right		48	72.7			25	71.4		0.890$^{X^2}$
Left		18	27.3			10	28.6		
Lobes									
Upper		46	69.7			23	65.7		0.864$^{X^2}$
Middle		4	6.1			2	5.7		
Lower		16	24.2			10	28.6		
Surgery types									
Cystotomy+capitonnage		51	773			16	45.7		0.003$^{X^2}$
Lobectomy		1	1.5			11	31.4		<0.001$^{X^2}$
Wedge resection		14	21.2			8	22.9		0.095$^{X^2}$
Albendazole									
(–)		5		7.6		7	0.0		0.066$^{X^2}$
(+)		61		92.4		28	80.0		
Associated other organ									
(–)		57	86.4			27	77.1		0.239$^{X^2}$
(+)		9	13.6			8	22.9		
Liver		8	88.9			8	100.0		
Skin		1	11.1			0	0.0		
Post-operative Complication									
(–)		56	84.8			15	42.9		<0.001
(+)		10	15.2			20	57.1		
Empyema		2	20.0			7	35.0		
Atelectasis		3	30.0			6	30.0		
Expansion defect		1	10.0			3	15		
Persistent air leak		1	10.0			2	10.0		
Wound infection		0	0.0			2	10.0		
Residual cavity		2	20.0			0	0.0		
Hemorrhage		1	10.0			0	0.0		
Recurrence									
(–)		63	95.5			33	94.3		1.000$^{X^2}$
(+)		3	4.5			2	5.77		
Mortality									
(–)		66	100.0			33	94.3		0.118$^{X^2}$
(+)		0	0.0			2	5.7		
Hospitalization duration	7.8±2.9			7.0	12.3±4.7			10.0	<0.001*
Follow duration (year)	7.1±3.0			8.0	6.3±3.1			6.0	0.299*

*: Mann-Whitney u test; $^{X^2}$: Ki-kare test (Fischer test).

In treating PCH, surgical treatment was performed to all cases. Albendazole was not administered in the pre-operative periods due to the low cure rate and increased complications such as rupture and hemoptysis as a result of its application in the preoperative period.[16] While choosing surgery, the primary goals were to provide the cure, preserve as much intact parenchyma tissue as possible, and prevent relapse and contamination.[11,17] For this reason, the most frequently preferred surgical method was cystotomy+capitonnage in 66 cases (66.3%).

In the literature, the rate of resection surgery for intact cysts is approximately 0–7% and 19–32% for complicated cysts.[16,17] The most important factor that changed our operation preference was lung abscess, one of the preoperative complications. Lobectomy was performed in 11 (10%) of 18 cases who developed lung abscesses due to severe destruction of the lung parenchyma. In one of our cases with giant hydatid disease, lobectomy had to be performed due to uncontrolled bleeding during capitonnage. Our lobectomy was rate is 11.9%, which is consistent with the literature.

The cases in which we preferred wedge resection were 22% higher than the literature data.[17] The reason for this was that the cystic cavity could not be quilted due to advanced calcification in 12 cases, that the cavity was necrotic due to cystic lung abscess in seven patients, that the hydatid disease could not be detected radiologically, and it needed to be removed by wedge resection and sent to frozen section examination in three cases. Pre-operative complications have changed our surgical choice. More major surgical methods such as wedge resection and lobectomy were needed in complicated hydatid cysts. Pre-operative complications reduce the patient's physical performance and increase major surgery, thereby increasing postoperative complications. In complicated hydatid disease, the hospitalization period was higher due to the high rate of major surgery and the high post-operative complication rate.

The complication rate after surgical treatment in pulmonary hydatid diseases has been reported as 1.4–19.1% and the mortality rate as 0.6–4.2%.[16,18] In our series, post-operative complications were seen with a rate of 29.7%, which is higher than the literature data. The most frequent complications were empyema and expansion defect. We attributed this to the high rate of complications in the pre-operative period. We think that performing high-risk resections due to pre-operative complications increases the rate of post-operative complications.

Our mortality was 2%, consistent with the literature.[3,17] Both deceased cases were in advanced ages (88 and 72) and had multiple comorbidities. One of the cases died due to massive pulmonary thromboembolism on the 1st post-operative day, and the other died due to septicemia in the late period. Although our post-operative complication was is higher than the literature data, we attribute that our mortality was not high, both to the success of the surgery and to our advanced post-operative care conditions.

Our cases were followed for 1–10 years (mean 6.9±2.9 years). Relapse did not occur in any patient operated in our clinic. However, the rate of patients who had their first operation in other centers and applied to our clinic with relapse is 5%. Relapse rates have been reported as 2–25% in the literature.[16,19] We attribute our successful results in relapse rates to our surgical techniques and surgical experience. Albendazole treatment could not be continued in 12% of cases due to various complications, and relapse did not occur.

In our study, the mean hospitalization duration was 7.4±2.4 days, consistent with the literature.[6] Hospitalization duration was statistically associated with pre-operative complications, post-operative complications, and rupture ($p<0.05$). Hospital stay was longer in those with preoperative complications. In our study, a significant decrease was observed in the number of cases in the last 5 years. There were 75 (74.3%) cases in the first 5 years and 26 (25.7%) cases in the past 5 years. We attribute this to advances in animal care.

Conclusion

The curative treatment of PCH is surgery. In surgery, cystotomy+capitonnage, in which the cyst content is evacuated, and the germinative membrane is removed, is usually sufficient. Cyst rupture is associated with preoperative complications. Complicated hydatid disease is associated with major surgery, high morbidity, and prolonged hospital stay. Therefore, early treatment of hydatid disease will reduce morbidity and prevent economic loss. Our high surgical success prevented recurrences.

Disclosures

Ethics Committee Approval: The study was approved by The Kahramanmaraş Sütçü İmam University Non-interventional Clinical Research Ethics Committee (Date: 19/10/2021, No: 02).

Informed Consent: Written informed consent was obtained from all patients.

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