

## Surgical treatment of 599 patients with hydatid cysts in the liver and lungs



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### Abstract

**Introduction:** Hydatid cyst disease is characterized by the occurrence of cysts as a result of the transmission of *Tenia echinococcus* larvae to humans. The most frequent location is the liver (60–70%). The larvae which fail to attach to the liver pass through the lungs (20–25%), and can be located in all organs and tissues connected to the systemic circulation (10%). They create hydatid cysts in the organs in which they are located. In our multi-center study, we aimed to present different types of surgical treatment, its results, and the associated complications on the basis of a wide series of cases.

**Material and methods:** 599 patients who received surgical treatment were included in our study. The patients' data from archive records were examined retrospectively. Patients with unusual locations of the cysts (without liver or lung involvement) were excluded from the study.

**Results:** 381 of the patients were women and 218 were men. It was established that the disease was more frequent among women than among men, which was in accordance with the literature [9]. The ratio of women to men was 1.747. The average age was 35.6; the youngest patient was 3, while the oldest was 83. When the patients were evaluated with regard to organ involvement, liver cysts were found in 425 patients, while lung cysts occurred in 236 patients. Both liver and lung involvement was revealed in 62 patients.

**Conclusions:** In the case of lung cysts, surgical treatment is preferred to medical treatment if there are no serious contraindications for operation. Even if only single cysts are found in the liver, lungs, or both of these organs, the stomach and the thoracic space must also undergo radiological screening.

### Streszczenie

**Wstęp:** Bąblowica objawia się występowaniem torbieli, powstających w wyniku przedostania się larw *Tenia echinococcus* do organizmu człowieka. Najczęstszym miejscem występowania torbieli jest wątroba (60–70%). Larwy, którym nie udało się przytwierdzić do wątroby, przemieszczają się do płuc (20–25%) lub innych organów i tkanek połączonych z układem krążenia ogólnego (10%). W organach, do których się przytwierdzą, tworzą się torbiele bąblowca. Celem wielośrodkowego badania autorów było zaprezentowanie różnych typów leczenia operacyjnego, ich wyników, jak również związanych z nimi komplikacji, na podstawie dużej liczby przypadków.

**Materiał i metody:** W badaniu uwzględniono 599 leczonych operacyjnie pacjentów. Użyte dane pacjentów były danymi archiwalnymi. Pacjenci, u których torbiele znajdowały się w innych miejscach niż wątroba i płuca, zostali wyłączeni z badania.

**Wyniki:** W badaniu wzięło udział 381 mężczyzn i 218 kobiet. Ustalono, że omawiana choroba częściej występuje u kobiet niż u mężczyzn. Dane te były zgodne z danymi dostępnymi w piśmiennictwie [9]. Stosunek kobiet do mężczyzn wynosił 1,747. Średnia wieku pacjentów wynosiła 35,6 roku; najmłodszy pacjent miał 3 lata, najstarszy 83 lata. Rozmieszczenie torbieli było następujące: 425 pacjentów miało torbiele wątroby, natomiast 236 pacjentów torbiele płuc. Zajęcie zarówno płuc, jak i wątroby zaobserwowano w 62 przypadkach.

**Wnioski:** W przypadku torbieli płuc, jeżeli nie występują istotne przeciwwskazania, preferuje się leczenie operacyjne. Nawet w przypadku stwierdzenia pojedynczych cyst w płucach, wątrobie lub obydwu tych organach konieczne jest przeprowadzenie badania radiologicznego jamy brzusznej i klatki pier-

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One of the main concerns during surgical treatment, in the case of both lung and liver cysts, is protecting the organ parenchyma and removing the cyst without damaging organ function.

**Key words:** hydatid cyst, liver and lungs, surgical treatment.

## Introduction

Hydatid cyst disease is characterized by the occurrence of cysts as a result of the transmission of *Tenia echinococcus* larvae to humans. There are four different forms of echinococcosis. The most frequent ones are *Echinococcus granulosus*, causing cystic echinococcosis, and *E. multilocularis*, causing alveolar echinococcosis [1]. The disease has been known since the time of Galen and Hippocrates. It was first described by Thebesius in the 17<sup>th</sup> century, while the term 'hydatid cyst' was used for the first time by Rudolphi in 1808 [1-3]. It is asserted that the disease was carried to Europe by the dogs of Icelandic whale hunters. Human infestation is common in those geographical regions in which people are in constant contact with domestic flesh-eating animals or livestock, such as sheep [4]. The disease is endemic to Southern Europe, North Africa, Asia, South America, Australia, and New Zealand. It occurs sporadically in regions of North America other than Alaska and North Western Canada. Dogs, foxes, wolves, jackals, deer, and sheep are included in the natural cycle of the parasite. The prevalence of hydatid cyst disease in Turkey is estimated at 50 out of 100 000 and the incidence is nearly 2-6 out of 100 000 [5]. The parasite egg hatches in the small intestine, releasing the embryo. The latter reaches the liver through the wall of the intestine, and from there it travels to the hepatic portal vein using its hooks. This is why the liver is the most frequent location for the cysts (60-70%). The embryos which cannot hold on to the liver pass to the lungs (20-25%); they can be found in all tissues and organs in the systemic circulation (10%), including bone joints. They create hydatid cysts in the organs that they attach to [6, 7]. In the presence of contraindications, or if the patient refuses to undergo surgical treatment, medical therapy is used, even though surgery is the first-choice option in the case of this disease. The optimal anti-parasite effect is achieved by administering albendazole in 3 cycles per day. In our multicenter study, we aimed to present different types of surgical treatment, its results, and the associated complications on the basis of a wide series of cases.

## Material and methods

599 patients who received surgical treatment were included in our study. The patients' data were examined retrospectively using archive records. Patients without liver or lung involvement were excluded from the study. In addition, patients who were treated medically instead of surgically were excluded as well. Tomography was performed on all the patients. Abdominal ultrasonography or tomography was

siowej. Podczas leczenia operacyjnego, zarówno w przypadku torbieli płuc, jak i wątroby, niezwykle ważną kwestią pozostaje ochrona miększu organu i usunięcie torbieli w taki sposób, aby nie zakłócić jego prawidłowej pracy.

**Słowa kluczowe:** torbiel bąblowca, wątroba i płuca, leczenie operacyjne.

performed as follow-up, depending on the place of involvement. During the preoperative period, the Casoni intradermal test and the Weinberg complement fixation test were not performed because of their low diagnostic value [8]. Bronchoscopy was only conducted in 7 patients, hospitalized due to other thoracic diseases, as part of the differential diagnosis. Bronchoscopy was not utilized in any other patients.

## Results

381 of the patients were men and 218 were women. It was established that the disease was more frequent among women than among men, which was in accordance with the literature [9]. The ratio of women to men was 1.747. The average age was 35.6; the youngest patient was 3 and the oldest was 83. The most frequent symptoms were stomach ache, cough, chest pain, shortness of breath, fever, and expectoration of hydatid cyst fluid. With regard to organ involvement, 425 patients had liver cysts and 236 patients had lung cysts. In 62 patients, both liver and lung involvement was revealed. Among the 425 patients with liver cysts, the number of patients with liver involvement only was 338, 62 patients had cysts in both the liver and lungs, while 25 had cysts in both the liver and other organs, excluding the lungs (Fig. 1).

Among the patients with lung cysts, isolated lung involvement occurred in 166 patients. 62 had both lung and liver cysts, while 8 patients had cysts in both the lungs and other organs. 510 cysts in total were found in 425 patients with liver involvement; 341 of them were in the right lobe, 164 in the left lobe, while 5 were in the caudate lobe. 298 cysts were found in 236 patients with lung involvement; 190 of them were located in the right lung, while 108 were located in the left lung (Table I).

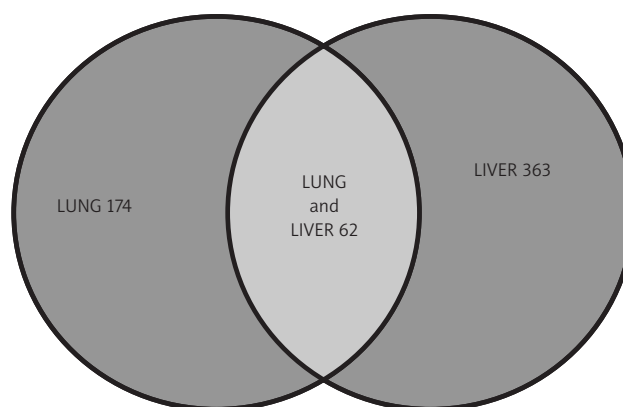


Fig. 1. Location of cysts

Tab. I. Distribution of cysts

Distribution of cysts	Liver cysts			Lung cysts				
	Right lobe	Left lobe	Caudate lobe	Upper lobe of right lung	Middle lobe of right lung	Lower lobe of right lung	Upper lobe of left lung	Lower lobe of left lung
	341	164	5	50	35	105	41	67
<b>808</b>		<b>510</b>				<b>298</b>		

Tab. II. Postoperative complications

Liver	Fistulization	14
	Biloma	3
	Pleural effusion	3
	Pancreatitis	1
	Intestinal bleeding	1
	Liver abscess	3
Lung	Anaphylaxis	1
	Atelectasis	7
	Empyema	3
	Prolonged air leakage	3
	Incisional infections	12
	Pneumothorax	2
<b>Total</b>		<b>53</b>

Tab. III. Procedure applied

Procedure	Total
Laparotomy	356
Laparoscopy	7
Thoracotomy + laparotomy	23
Thoraco-phreno-laparotomy	39
Bilateral thoracotomy	11
Thoracotomy	163

The cysts were more frequently observed in the right lung and on the right side of the liver. In children, lung involvement was more common than liver involvement. With regard to lung involvement, the cysts appeared in the lower lobes more frequently than in the upper lobes. Three patients died. Recurrence was observed in 13 patients. Moderate complications occurred in 43 patients in total (Table II).

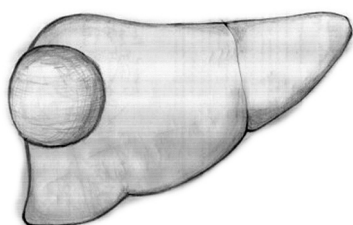
Endoscopic retrograde cholangiopancreatography (ERCP) was applied in 20 cases with liver cysts, 19 patients un-

derwent cholecystectomy, T-tube drainage was applied in 18 cases, omentoplasty was used in 15 cases, splenectomy was employed in 3 cases, and 2 patients received percutaneous drainage. With regard to the performed surgical interventions, thoracotomy was conducted in 163 patients, thoraco-phreno-laparotomy in 39 patients, thoracotomy plus laparotomy in 23 patients, bilateral thoracotomy in 11 patients, laparotomy in 356 patients, and laparoscopy in 7 patients (Table III).

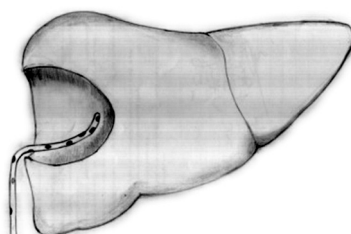
Cystotomy-capitonnage was applied to 256 out of 298 lung cysts, wedge resection to 36 cysts, segmentectomy to 5, while lobectomy was used in one case only. Partial cystectomy was applied to 283 out of 510 cysts, cystectomy to 50, cystotomy and drainage to 177 (Table IV) (Figs. 2-4).

Tab. IV. Surgical method applied

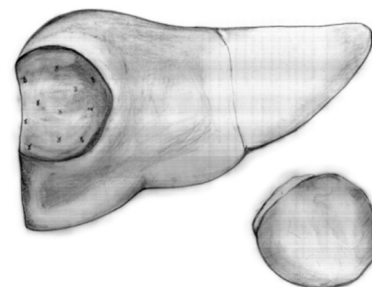
Surgical method	Liver cysts				Lung cysts		
	Partial cystectomy	Cystotomy drainage	Cystectomy	Cystotomy capitonnage	Wedge resection	Segmentectomy	Lobectomy
808	283	177	50	238	56	3	1



Hydatid cyst in liver right lobe

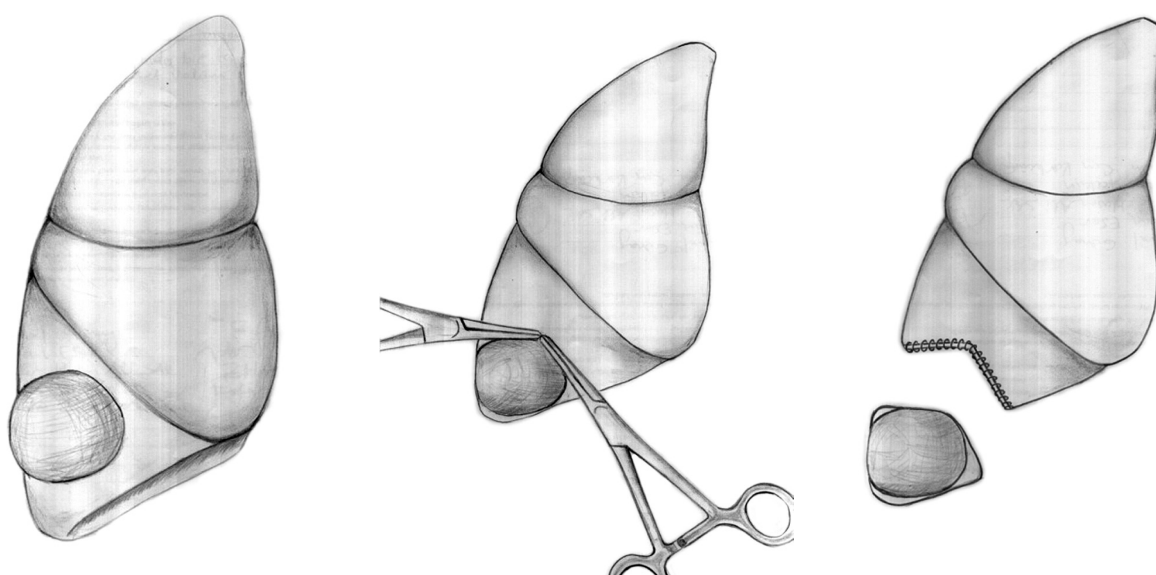


Partial cystectomy + drainage: After 5 minutes 3% NaCl was injected into the cyst, and the free part of the cyst wall was excreted. Drain was placed in cyst pouch



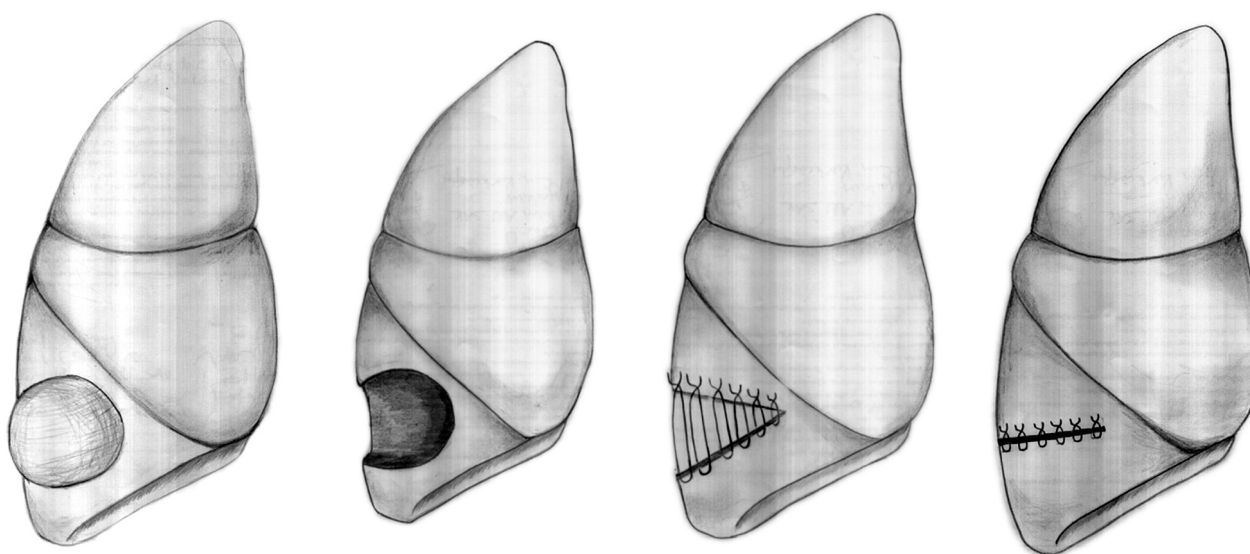
Pericystectomy: Complete resection is made with the pericystic layer around the cyst. The veins and bile ducts present are tied

Fig. 2. Partial liver cystectomy – drainage and pericystectomy



After the two clamps have been placed in the border of the cyst in the right lung lobe as a wedge, the cyst is removed by cutting. Then it is sewn continuously

**Fig. 3.** Lung cystectomy



5 minutes after the injection of 3% NaCl into the hydatid cyst in the right lung lobe, the content is excreted, opening the free wall of the cyst. After the cyst base has been cleaned it is closed sewing from inside to outside as a fish mouth without any loss of parenchyma

**Fig. 4.** Lung cystotomy – capitonnage

## Discussion

Hydatid cyst disease is as old as the history of medicine, dating back to the times of Hippocrates. In general, hydatid cysts can be observed in every part of the body, but occur mainly in the liver and lungs. Approximately, every two cysts out of three are located in the liver and one out of five in the lungs. The remaining cysts can be found in any part of the body [10]. This is why abdominal and thoracic scans must be performed when searching for hydatid cysts. Especially in the regions that the disease is endemic to, this still poses a serious problem. The primary treatment

for the disease is surgery. In the presence of contraindications for surgical intervention, or if the patient refuses to undergo surgery, medical treatment is used. Medical treatment is also employed in order to prevent recurrence. Because of the capsule structure of the cysts, symptoms occur only when pressure is exerted on healthy tissue or when the cyst is ruptured. Rupture risk increases in direct proportion to the size of the cyst. Rupture after trauma is more frequent in the case of liver cysts. Different broad case studies reported that free cyst rupture occurred in the intraperitoneal space in 1.7% or 8.6% of all liver hydatid

cyst cases [11, 12]. Its most serious complication is anaphylactic shock. However, depending on the organ the cyst is located in, as well as the cyst's size and exact place on the organ, the following complications may develop as well: stomach ache, fever, cough, shortness of breath, nausea, vomiting, hepatitis, allergic reactions, and anaphylaxis [13, 14]. The mortality and morbidity caused by hydatid cysts is dependent on the organ in which they are located. Thus, the main goal of surgical treatment is to remove the cysts with minimum damage to the involved organs. Methods sparing the parenchyma should be preferred. Parenchymal sparing surgery was prioritized in our study. In the past years, parenchymal resections were avoided as much as possible; not a single hepatic parenchymal resection was performed. For hydatid cysts located in the lungs, only one patient underwent lobectomy, while segmentectomy was performed in 3 cases. Lung parenchyma was protected as much as possible with the use of wedge resections. Peripherally located small hydatid cysts were removed using a stapler. During the operation, the area containing the cyst was left uncovered, while the surrounding area was covered with gauze bandage saturated with povidone-iodine (polyiodine). Afterwards, a 20 gauge injector with a 50 ml bulb was inserted into the cyst, and 50 ml of hydatid fluid was aspirated. 10% povidone-iodine was then injected into the cyst in the amount equal to the amount of the aspirated fluid. The cyst was opened with cystotomy, and the germinal membrane was excised. In this way, surrounding tissue contamination and cyst recurrence were prevented. Recurrence took place in 7 patients (1.16%). Three patients (0.5%) died: one of them had the hydatid cyst located in the lung, while the remaining two had both liver and lung cysts. In the literature, the mortality rates are 0.6-4.1% and recurrence rates are 0.6-1.7% [1, 4]. Bile leakage occurred in 18 out of 425 patients with liver cysts, who received T-tube drainage. Different rates were reported for bile leakage and bile fistula, ranging from 2.6% to 9.3% [15-17]. Laparoscopic surgery was performed in 7 patients with liver cysts. This relatively new method helps improve morbidity rates. None of the eleven patients with bilateral lung involvement underwent median sternotomy or concurrent bilateral thoracotomy. Two separate thoracotomies were performed with an interval of 6 weeks. In 39 out of 62 patients with liver and lung involvement, both liver and lung cysts were removed during a single thoraco-phreno-laparotomy, because the lung cysts were located in the right lung. The tissue surrounding liver cysts is more solid than that of lung cysts, and there are no structures through which the hydatid fluid could spread, such as the bronchi in the lungs. Therefore, the risk of cyst rupture, resulting from the thinning of the external layer of the cyst by medical treatment, is lower. In lung cysts, the wall of a cyst which was thinned by drug treatment is likely to burst and spread larvae to other parts of the lungs. Hence, medical treatment provides better results in the case of liver cysts than in the case of lung cysts.

## Conclusions

As a result, if there are no serious contraindications, surgical treatment should be chosen over medical treatment for lung cysts. Even if the liver or lungs appear to be the only organs involved, the abdomen and thorax must also be scanned. The main aim of surgical treatment for both lung and liver cysts must be to protect the organ parenchyma as much as possible, as well as to make the cyst inactive without harming the function of the organ involved.

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