

Lobectomy for pulmonary hydatid cyst

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Aim: Although parenchymal sparing surgery is the most appropriate for the treatment of hydatid cysts, rarely lobectomy is needed. We aimed to retrospectively evaluate the cases in which we had to perform lobectomy due to hydatid cyst.

Materials and methods: A total of 36 (1.88%) patients that underwent lobectomy (n = 35) and pneumonectomy (n = 1) from among 1909 patients operated on for hydatid cyst in our hospital between 1992 and 2012 were retrospectively evaluated.

Results: Among the patients, 35 underwent lobectomy and 1 right pneumonectomy. The mean age of the patients was 11.1 ± 3.4 years (6–16 years) among children and 43.2 ± 13.8 years (20–78 years) among adults. The most common indications for lobectomy were destroyed parenchyma in 22 (61.1%), hemoptysis in 17 (47.2%), and parenchymal loss due to giant cyst in 12 (33.3%) cases. Postoperative morbidity was observed in 9 (25%) patients, and no mortality was observed.

Conclusion: The effective treatment for hydatid cyst is parenchymal sparing surgery. However, lobectomy is an inevitable result with indications such as destroyed parenchyma, hemoptysis, parenchymal loss due to giant cyst, lobe bronchus ruptured into the cystic cavity, broncho-pleural fistula, suspected malignancy, broncho-bilier fistula, and pulmonary abscess.

Key words: Hydatid cyst, lobectomy, pulmonary

1. Introduction

Hydatid disease, known in the times of Galen, was described by Thebesius in the 17th century (1). Hydatid cyst is the most common parasitic disease of the lung and is often caused by *Echinococcus granulosus* and rarely *Echinococcus multilocularis*. Carnivora such as dogs and wolves are the primary host in the life cycle of echinococcosis, while humans, sheep, and cattle are the intermediate hosts. In humans, hydatid cyst disease is frequently seen in the liver and lungs, but considering that the scolices are carried through blood, the disease can be seen everywhere the blood reaches (2–4).

Echinococcosis is endemic in many regions such as the Middle East, the Mediterranean, South America, Australia, New Zealand, Alaska, and Canada (1–5). The disease is currently endemic in Turkey, and according to the Turkish Ministry of Health in Turkey the incidence rate of the disease is 14 in 100,000 (6).

Because hydatid cyst is a parasitic infection, when faced with the agent in endemic areas, recurrence of the disease is always possible. The treatment for lung hydatid cyst is surgery. Parenchymal sparing surgery is the most

appropriate and preferable form of treatment (2,3,7,8). In the endemic areas, a surgical procedure that always results in parenchymal loss is not acceptable. Lobectomy and anatomical resections must be avoided in these cases as much as possible (7,8). However, lobectomy is inevitable in some cases (5). In this article we discuss 36 cases and lobectomy indications in the cases in which we had to perform lobectomy in the last 20 years.

2. Patients and methods

A total of 1909 patients were operated on due to hydatid cyst in the Atatürk Chest Diseases and Thoracic Surgery Training and Research Hospital between 1992 and 2012. Of these patients, 35 underwent lobectomy and 1 patient underwent pneumonectomy. Thirty-six (1.88%) patients who required anatomic resection were examined from hospital records and patient files and evaluated for demographic, clinical, and radiological data, and the indications for lobectomy were discussed.

Descriptive features of our patients were expressed as mean ± standard deviation. Since the data showed a nonparametric distribution, statistical analysis was

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performed using the Mann–Whitney U-test. P values of less than 0.05 were considered significant.

Five patients (13.8%) were asymptomatic, and the cyst was discovered incidentally upon plain chest X-ray for other causes. The remaining patients had one or more symptoms, which are summarized in Table 1.

In the pre-operative evaluation chest X-ray, thorax CT, bronchoscopy, respiratory function tests, abdominal USG, and abdominal CT were used when necessary. Patients with neurologic symptoms were evaluated with cerebral CT. Biochemical tests (e.g., Casoni skin test, Weinberg complement fixation, and recording the number of eosinophils) have not been used recently due to their low diagnostic value.

Of the patients, 10 (27.8%) were children, and 26 were (72.2%) adults. Twelve (33.3%) patients had intact cysts, 23 (63.9%) had ruptured hydatid cysts, and 1 (2.8%) had *Echinococcus multilocularis*. Two of our patients were complicated cases who had had previous operations due to hydatid cysts. All the patients in our series were treated surgically. Out of the patients with massive hemoptysis (n = 2), the decision for lobectomy and pneumonectomy was made according to the intraoperative findings.

The standard approach consisted of posterolateral thoracotomy through the fifth or sixth intercostal space under general anesthesia with a double lumen endotracheal tube, except in small pediatric patients, who received a single lumen tube.

Seventeen patients with hemoptysis underwent pre-operative rigid bronchoscopy to localize the hemoptysis focus. Following thoracotomy, in order to avoid contamination in our intact cyst cases, the parenchymal area outside the hydatid cyst site was wrapped with a 10% povidone-iodine-soaked compress. Hydatid fluid was aspirated from the top of the cyst with a 16-G needle to reduce the intracystic pressure. Then cystotomy was performed, and remaining fluid was completely aspirated with a suction system, and germinative membranes were evacuated with ring forceps. The cystic space was irrigated

with 10% povidone-iodine. Bronchus orifices opened to the cyst cavity were closed using absorbable sutures, and pericystectomy was performed. Then the lung was ventilated, and the remaining parenchyma was evaluated. Of our 12 intact cases in the preoperative evaluation, 8 (22.2%) had giant cysts (>10 cm). The majority of these cases (n = 6) were in children. In the intact giant cyst cases, the cysts were seen to invade the lobe completely or nearly completely. For patients with total invasion, lobectomy was planned. Following pericystectomy in the cases without total invasion, the remaining parenchyma was seen to be destroyed and not ventilated, and lobectomy was planned for these patients. Lobectomy was performed in 3 of our intact cyst patients due to hemoptysis and in 1 patient due to multiple cysts localized at the same lobe.

In our series, ruptured hydatid cysts accounted for the majority (n = 23, 63.9%) of the cases. Six of these patients presented with pleural complications. Empyema was detected in all 6 patients. Two of these patients were followed-up due to the rupture of the capitonnage following previous cyst operations and 2 patients due to a broncho-pleural fistula that developed due to a cyst that ruptured into the pleural cavity. The remaining 2 patients who were followed-up because of pleural complications underwent lobectomy due to pulmonary abscess. In the patients with broncho-pleural fistula and empyema, a chest tube was inserted, empyema was drained, antibiotherapy was administered, intrapleural irrigation was carried out, and thoracotomy was planned after the parameters of infection (e.g., CRP, culture) were regressed. Decortication was performed following the thoracotomy in these patients with pleural complications. Cystotomy was performed in all the ruptured cyst cases in which the cyst pouch could be reached; the infected cyst pouch was irrigated with 10% povidone-iodine, the germinative membrane was removed, the lung was ventilated following closure of the bronchus orifices with suture, and the cases were evaluated for lobectomy.

In the ruptured cyst cases the most common indications we encountered for lobectomy were hemoptysis in which the parenchyma was unventilated with irreversible parenchymal destruction and the lobe bronchus was ruptured into the cyst cavity.

3. Results

Of the patients, 22 (61.1%) were males and 14 (38.9%) were females, with a mean age of 34.3 ± 18.8 (6–78). Twenty-six patients were adults and 10 were children. The mean age of the patients was 11.1 ± 3.4 years (6–16 years) among children and 43.2 ± 13.8 years (20–78 years) among adults. Right lower lobectomy was the most common type performed (n = 11, 30.5%). The lobectomies performed and localizations are given in Table 2.

Table 1. Symptoms of pulmonary hydatidosis in this series.

Cough	n = 26	72.2%
Chest pain	n = 23	63.8%
Sputum	n = 21	58.3%
Hemoptysis	n = 17	47.2%
Dyspnea	n = 14	38.8%
Fever	n = 11	30.5%
Membrane expectoration	n = 6	16.6%
Asymptomatic	n = 5	13.8%
Vomiting	n = 3	8.3%
Biliptysis	n = 1	2.7%

Table 2. Lobectomy localizations for hydatid cyst.

Right lower lobectomy	n = 11	30.5%
Medium lobectomy	n = 8	22.2%
Left lower lobectomy	n = 7	19.4%
Right upper lobectomy	n = 6	16.7%
Left upper lobectomy	n = 2	5.5%
Bilobectomy inferior	n = 1	2.8%
Right pneumonectomy	n = 1	2.8%

Indications for lobectomy resulted from one or more factors. The most common cause was parenchymal loss due to giant cyst in the intact cases, while destroyed parenchyma was the most common indication in the ruptured cyst cases. At the evaluation carried out for all patients, destroyed parenchyma was the most common indication for lobectomy (n = 22, 61.1%). Destroyed parenchyma cases were those most frequently accompanied by hemoptysis. There was destroyed lobe in all patients in whom the lobe bronchus ruptured into the cyst cavity. Hemoptysis was the second most frequent indication (n = 17, 47.2%) with ruptured hydatid cysts (n = 14) accounting for the majority of the cases in which hemoptysis played a role in the indication for lobectomy. Giant cyst cases (n = 12, 33.3%) that led to complete or near complete parenchymal loss were mostly intact cysts (n = 8) and pediatric cases (n = 8). There was an association between hemoptysis and destruction of the remaining parenchyma in these patients. All the patients in which the lobe bronchus ruptured into the cyst cavity (n = 6, 16.6%) had ruptured cysts, and the lobe parenchyma was completely destroyed in all these patients, with hemoptysis accompanying it in 2 cases. Two of the broncho-pleural fistula patients (n = 4, 11.1%) had rupture of the capitonage due to previous cyst operations, and 2 patients had cysts rupture into the pleural cavity. Empyema and destroyed parenchyma were observed in all these patients. Lobectomy was performed in 2 patients (5.5%) due to pulmonary abscess, in 1 patient (2.7%) due to suspicion of malignancy, in 1 patient (2.7%) due to multiple hydatid cysts in the same lobe with diameters between 1 and 5 cm, and in 1 patient (2.7%) due to a liver dome cyst accompanied by a ruptured cyst in the lower lobe and bronchobiliary fistula. Right pneumonectomy was carried out in 1 patient (2.7%) due to *E. multilocularis*. Lobectomy indications and frequencies are summarized in Table 3.

In addition to the 4 patients with broncho-pleural fistulas, chest tubes were inserted in 2 other patients with pulmonary abscesses due to empyema, empyema therapy was administered, and a thoracotomy was performed after the infection symptoms regressed.

Table 3. Lobectomy indications for pulmonary hydatid cysts.

Destroyed parenchyma	n = 22	61.1%
Hemoptysis	n = 17	47.2%
Parenchymal loss due to giant cyst	n = 12	33.3%
Rupture of the lobe bronchus into cyst cavity	n = 6	16.6%
Bronchopleural fistula	n = 4	11.1%
Pulmonary abscess	n = 2	5.5%
Suspicion of malignancy	n = 1	2.7%
Multiple cyst in the same lobe	n = 1	2.7%
Bronchobiliary fistula	n = 1	2.7%
<i>E. multilocularis</i>	n = 1	2.7%

Postoperative morbidity was observed in 9 patients (25.0%). The association of one or more complications was revealed in these patients. Eight of these patients had ruptured hydatid cysts. Evaluation of the complications is given in Table 4.

Among the total 12 giant cyst patients (>10 cm) 8 had intact cysts. Mean cyst diameter was 15.58 ± 6.1 cm (max.: 22 cm), while children were dominant among the giant cyst patients (n = 8, 66.6%). In the giant cyst patients we encountered complete or near complete parenchymal loss as the most common indication for lobectomy. Irreversible alterations in the remaining parenchyma were the second most common indication for lobectomy.

When ruptured cyst patients (n = 23) were compared with the intact cyst patients in our series in terms of hospitalization duration and postoperative complications, ruptured cyst patients stayed significantly longer in the hospital and had more morbidity (P = 0.003). Mean hospitalization time was 15.53 ± 15.8 days for all patients. This duration was 19.2 ± 18.2 (7–75) days in the ruptured cyst patients and 8.17 ± 3.7 (5–17) days in the intact cyst patients. No mortality was observed. Liver functions of all the patients were followed-up for 2 months after discharge from the hospital, and the patients were administered albendazole 10 mg/kg/day.

Table 4. Complications.

Wound site infection	n = 7	19.4%
Pneumonia	n = 4	11.1%
Atelectasia	n = 3	8.3%
Empyema	n = 3	8.3%
Prolonged air leakage	n = 2	5.5%
Drainage requiring revision	n = 2	5.5%
Aseptic space	n = 2	5.5%

4. Discussion

Echinococcus embryos taken in through the digestive system may reach the liver and from here travel to the lungs via the vena cava inferior. However, while less common, the lymphatic system or bronchi can also serve as the pathway for infestation (1,9). Cysts surviving the filter in the lungs may reach the left heart and enter systemic circulation. This means every point that the systemic circulation reaches may be infected with hydatid cysts. In this pathway followed by the cyst embryo, the most frequently infected organ is the liver, followed by the lungs (2,3,9).

Because of their elastic tissue compared to the other organs hydatid cysts of the lungs may reach a giant size and can invade large part of a lobe (10,11). Giant pulmonary cysts occur more often in children than in adults (12). In addition, lung hydatid cysts may rupture with spontaneous iatrogenic and traumatic causes at any stage of their life cycle (13). Ruptured lung hydatid cysts can become easily infected because the tracheobronchial system is open to the external environment (13,14). Delay in the diagnosis and treatment may lead to parenchymal destruction in patients with hydatid cysts (1-3,13,14). The standard treatment modality is parenchyma-sparing surgery for lung hydatid cysts. Cystotomy plus capitonnage or cystotomy without capitonnage are accepted surgical procedures; lobectomy or pneumonectomy are undesirable surgical procedures in cyst surgery (7,10,11).

However, when the English literature published in the last 2 decades was searched, rates of lobectomy were found to vary between 0% and 52.1%, while the rates of pneumonectomy were between 0% and 11.4% in the same series published around the world (1,5,14-18). In this review for a series of lung hydatid cysts that had been operated on, the most outstanding study was published by Vaquez et al. in 2009 (5). In that study, 115 hydatid cyst

cases were surgically treated with lobectomy performed in 60 (52.1%) and pneumonectomy in 3 (2.6%) patients. The authors stated that they preferred cystectomy for the hydatid cyst surgery and performed lobectomy in cases where the hydatid cyst invaded more than 50% of the lobe and in the presence of multiple cysts at the same lobe. We think this surgical technique increases the rates of lobectomy. In addition, rates of lobectomy in the cyst series from different countries around the world are summarized in Table 5. We use cystotomy plus capitonnage in our surgical practice. After closure of the bronchial leaks following cystotomy with absorbable suture, we perform capitonnage if the remaining parenchyma is ventilated regardless of the extent of the invasion by the cyst, and we are contented with closure of the bronchial leaks in the event that the capitonnage interferes with the ventilation of the remaining parenchyma. We also apply these rules for multiple cysts at the same lobe.

When our series and the other series were analyzed destruction and nonventilation of the remaining lobe parenchyma and irreversible alterations were the most common indications for lobectomy (1-5,14-19). All the authors emphasized that lobectomy was necessary in cases of irreversible alterations in the lung parenchyma (1,5,8,10,11,14,16,18-20). The decision for lobectomy was made in the presence of the destroyed parenchyma during the evaluation made after cyst fluid and germinative membrane in the intact cysts and infected material in the ruptured cysts were removed.

Hemoptysis is a rarely seen symptom for hydatid cysts (13,18). It may present with various clinical manifestations from mild to massive hemoptysis. In intact cysts, hemoptysis occurs as a result of pneumonia developed in atelectatic ground, cyst eroding the bronchus, rupture of the cyst, or erosion in the vascular structures. Hemoptysis is often seen with rupture of the cysts. In different cyst

Table 5. Rates of lobectomy and pneumonectomy in the series of cyst hydatid published in various years.

Author	Year	Country	Case number	Lobectomy	Pneumonectomy
Burgos	1991 (1)	Spain	240	2.5%	0%
Athanassiadi	1998 (15)	Greece	83	3.6%	0%
Salih	1998 (16)	Turkey	405	1.2%	0.2%
Amez	2002 (17)	Peru	88	28.4%	11.4%
Kuzucu	2004 (14)	Turkey	67	0%	0%
Dinçer	2006 (18)	Turkey	301	3.6%	0%
Vasquez	2009 (5)	Peru	115	52.1%	2.6%
This series	2012	Turkey	1909	1.88%	0.05%

series, hemoptysis was reported at between 1.8% and 8% (13,16,18,21,22). In the case of close neighborhoods with large vascular structures, cysts localized in the lung caused erosion of the vascular wall, resulting in massive hemoptysis and mortality (21,22). In our series, hemoptysis was the second most common indication for lobectomy. The majority of hemoptysis cases ($n = 17$) consisted of ruptured cysts ($n = 13$). While lobectomy was performed in 2 patients due to massive hemoptysis, the operation was completed with lobectomy in 2 patients because of the persistence hemoptysis following cystotomy plus capitonnage. Although hemoptysis was often observed in association with destroyed parenchyma, other causes (e.g., pulmonary abscess, bronchobiliary fistula) were also accompanied by hemoptysis.

Hydatid cysts with the largest diameter (>10 cm) were defined as giant cysts (10,11). These are more common in children than in adults (18,23,24). In various giant cyst series, rate of lobectomy was reported to be higher than in series of normal cysts (6.3%–54.5%) (10,11,18,19). Various series of giant cysts and rates of lobectomy are given in Table 6. There were 12 cases of giant cysts in our series

with 8 pediatric cases (Figure 1). In all these patients, there was complete or near complete invasion of the lobe due to cysts (Figure 2). Many authors recommended lobectomy in cases when parenchymal loss of a lobe from invasion by hydatid cysts was about 50%–60% (1,5,19,25). As a surgical procedure, we perform capitonnage if the parenchyma is ventilated regardless of the extent of invasion; however, if capitonnage is not suitable we close the bronchial leakages and terminate the surgery, avoiding resection. However, this was impossible in the giant cyst cases in our series.

In 6 patients in our series, after the cyst cavity was discharged lobe bronchus was seen to have ruptured into the cavity. Lobectomy was inevitable in these patients. All these patients had ruptured, infected, and destroyed parenchyma. Hemoptysis accompanied in 2 patients. Decision for lobectomy should be easily made in the cases where the lobe bronchus has ruptured into the cyst cavity at a preoperative evaluation with bronchoscopy.

Complicated conditions of hydatid cyst, such as in broncho-pleural fistula and pulmonary abscess, mostly define the delayed cases (1,2,9,13). As in our series, an association between destroyed parenchyma and hemoptysis

Table 6. Giant cyst series and lobectomy rates.

Author	Year	Country	Case number	Lobectomy/pneumonectomy
Halezaroğlu	1997 (11)	Turkey	47	3 (6.3%)
Karaoglanoglu	2001 (10)	Turkey	67	9 (13.4%)
Dincer	2006 (18)	Turkey	11	6 (54.5%)
Arroud	2009 (19)	Morocco	32	5 (15.6%)



Figure 1. Giant cyst is seen to completely invade right lower lobe.



Figure 2. Appearance of the pericyst after the germinative membrane was removed following cystotomy. Giant cyst is seen to completely invade the lobe.

was seen in most of these cases. Presence of empyema in the patients with pleural complications prolonged the treatment duration and required decortication during the operation.

Lobectomy was performed due to bronchobiliary fistula in 1 patient with a ruptured infected liver dome cyst accompanied by right lower lobe cyst, in 1 patient with ruptured cyst who had a mass image mimicking malignancy due to suspected histopathological findings at the preoperative evaluation, and in 1 patient because of multiple cysts at the lower lobe with diameters between 1 and 5 cm. Right pneumonectomy was performed in 1

patient in having *E. multilocularis* as the agent and multiple cystic foci, with hemoptysis, which we thought resulted from a hilus-localized lesion.

Community-based health policy should be developed to eradicate hydatid cyst, which is a preventable disease. Timely surgical treatment should be administered before the cysts become complicated and reach giant sizes, and this will reduce hospital costs and decrease morbidity and mortality. Parenchymal-sparing surgery must always be the surgical procedure of choice, but lobectomy is an effective treatment method when its indications are present.

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