

# Optimized Conservative Surgical Treatment of Pulmonary Hydatid Cyst: A Retrospective Observational Cohort Study

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**Background:** There are some controversies on surgical options for pulmonary hydatid cysts. We analyzed our experience in lung tissue preservation during the surgical treatment and optimal surgical options.

**Materials and Methods:** This observational cohort analysis was conducted from July 2008 to July 2022. The Age, sex, clinical manifestation, recurrent rate, hospital length of stay, postoperative complications, and long-term results in each group were assessed. The American Society of Anesthesiologists Physical Status, Charlson Co-Morbidity Index (CCI), Complexity of surgery, and Clavien-Dindo score were also determined.

**Results:** Out of 138 patients, 81case (53.5%) had intact cysts (G1), and the rest were infected cysts which in turn were subdivided into early infected (G2) and cavity suppurated=28 cases (18.4%) as (G3). Group G3 required special attention because the pericyst surface was severely inflamed, dirty, and had pus, so they were subdivided into 3 distinctive groups including G3a, undergone cystectomy, bronchial opening closure alone, G3b group, undergone cystectomy, capitonnage+bronchial opening closure and G3c group, in which pericystectomy also added to previously mentioned procedures. Major complications in the subgroups of G3a were 2 patients and G3b 3 patients, but in the subgroup of G3c, no considerable complications were seen.

Cystectomy, closure of major bronchial opening, and capitonnage were done in intact, and early infected cysts. The results of both were the same, with no considerable major complication.

**Conclusion:** Capitonnage significantly decreased the complication rate. The optimized approach in both G1 and G2 was: cystectomy, closure of major bronchial opening, and capitonnage. In G3, bronchial opening closure, pericystectomy, and capitonnage were the preferred procedures.

**Keywords:** Albendazole; Capitonnage; Endobronchial rupture; Hydatid cyst of lung; Pericystectomy

## INTRODUCTION

Pulmonary hydatid cyst is a parasitic infection that exists in many endemic areas worldwide (1). Due to the elasticity of lung structure, the cyst grows up to giant dimensions (2). The clinical manifestation of pulmonary hydatid cysts (PHC) depends on the size, location, and

whether the cyst is intact or ruptured. Surgery is the mainstream treatment and includes cystectomy alone, cystectomy, bronchial opening closure, capitonnage, enucleation, and pericystectomy (3). Despite numerous studies, controversy persists regarding the optimal approach as no surgical guideline is available, and there is

no consensus among surgeons on performing pericystectomy and capitonnage in severely infected PHC (suppurated cyst) (4).

This study aimed to share our experience and compare outcomes, with other studies. We did capitonnage in all forms of cysts and pericystectomy in suppurated cysts. We planned to save as much lung tissue as possible unless a lobectomy was necessary due to factors such as multiple unilobar cysts, lobe destruction, or bronchiectasis.

## MATERIALS AND METHODS

This study analyzed 138 consecutive adult patients with pulmonary hydatid cysts of more than 5 cm who had been treated surgically in a teaching hospital from July 2008 to July 2022. The diagnosis of patients was carried out by chest X-ray and computed tomography of the chest and abdomen.

In our initial practice, we did not perform pericystectomy and capitonnage in the suppurated cyst and the option was cystectomy+ removal of laminated membrane plus closure of bronchial opening without capitonnage. After 2 of 5 (40%) patients had been complicated with empyema, they abandoned this practice and started to perform obligatory capitonnage in both intact or infected cysts.

Adding pericystectomy in some of the suppurated cysts improved surgical outcomes; however, this option needed to be verified. We did not administer any anti-parasitic drug pre or post-operation.

All data were extracted from the medical records database of our hospital. The cases were divided into intact, early infected, and suppurative categories. Characteristics of the cysts were defined based on chest computed tomography reports and surgical notes. The American Society of Anesthesiologists Physical Status (ASA-PS), Charlson Co-Morbidity Index (CCI) (5), Complexity of surgery, and Clavien-Dindo score (CDC) (6) were also determined.

### Definitions

**Intact cyst:** Patients had no systemic inflammatory response and no abnormality finding in the cyst on chest X-ray (Figure 1).



**Figure 1.** PA chest X-ray of an intact giant hydatid cyst in the left upper lobe of a 21-year-old patient.

**Early infected hydatid cysts:** Clinically, the patients had a systemic inflammatory response, and radiological findings which suggest hydatid cyst infection. In early infected PHC, the patients were not septic (7) and the residual cavity surface was not dirty or cleaned by washing.

**Severely infected hydatid cyst or suppurated cavity:** The patients were septic and had fever and purulent sputum due to lung abscess. The cyst cavity surface in the operation field was dirty, had obvious necrotic tissue, and was severely inflamed. The specific CT features of the infected Pulmonary Hydatid cyst had an air fluid level, and water lily Sign.

**Inclusion criteria:** Age of > 15 years, cysts > 5 cm diameter, complete data, and availability for follow-up.

**Exclusion criteria:** Age less than 15 years, incomplete data in the medical record, cysts less than 6 cm.

### Surgical Technique

Under general anesthesia, with a double-lumen endotracheal tube, a poster lateral muscle saving thoracotomy through the 5<sup>th</sup> to 7<sup>th</sup> intercostal space, depending on the location of the cyst and in bilateral lung cysts, sequential bilateral thoracotomy was performed.

Every effort was made to prevent spillage to the thoracic cavity and incision site with coverage of the adjacent tissues by towels soaked in 10% saline hypertonic sodium chlorides.

In an intact cyst, after the cyst fluid was suctioned carefully, the laminated membrane was removed and only major bronchial openings were closed by non-absorbable fine sutures.

We begin to suture from the deepest part of the cavity with non-absorbable purse string sutures that remain about 1 cm between each layer (capitonnage). In cases of severe cyst infection, when the remaining space is unclean due to evident necrotic tissue and cannot be adequately cleaned with normal saline, capitonnage is carried out after thorough removal of as much of the cyst wall as possible while minimizing damage to the surrounding tissue (Figure 2,3). Decortication was also performed in patients with pleural complications (8).

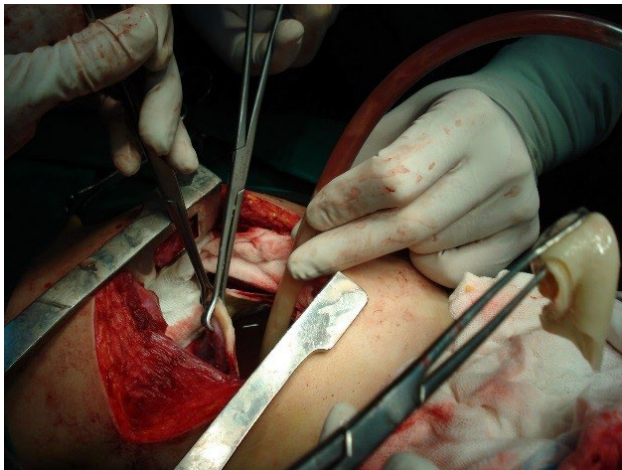


Figure 2 Demonstrate a perforated giant hydatid cyst in the right lower lobe



Figure 3. Total pericystectomy in the right lower lobe

**Data analysis:** All data were analyzed using SPSS16. Results are expressed as means  $\pm$  SD or percentage. P value  $< 0.05$  was considered statistically significant.

**Follow-up and Outcome:** The follow-up process typically lasted for more than five years to monitor for potential complications and assess the rate of recurrence.

## RESULTS

We had 138 consecutive patients between 15-89 years of age (mean: 41.7, SD: 18.8); 88 cases (64%) were male, male/female ratio was 1.76/1. Eighty-one (53%) of cysts were intact, and 71 cases (47%) were infected. There was a significant difference between non-perforated and perforated cysts (P-value=0.009) (Table 1).

Table 1. Demographic data and side of the cyst

Variable	Perforated n=71(47%)	Non-perforated n=81(53.%)	Total n=152(100%)	P-value
Age (mean SD)	39.29 $\pm$ 17.83	42.92 $\pm$ 17.72	41.71 $\pm$ 18.08	0.037
Gender (n%)				
Male	34 (60%)	54 (67.%)	88(64%)	0.243
Female	23(40.%)	27(33.%)	50(36%)	
Side (n%)				
Right	42 (59%)	45(56%)	87 (57%)	0.009
Left	29 (41%)	36(44%)	65 (43%)	

Of 71 cases (47%) of infected cysts, 43 cases (28%) were considered as early infected cysts or G2, 8 of them (5%) being ruptured into the pleura space, presented as loculated empyema. In these cases, decortication was also performed in addition to cystectomy, bronchial opening closure, and capitonnage.

28 cases (18.4%) of infected cysts were severely infected and considered as G3. This group was subdivided into 3 sub-groups based on the status of the pericyst surface (dirty, severe necrosis, pus). From 16 cases of G3 whose pericystectomy had not been performed, 5 cases of capitonnage also had not been performed, only bronchial opening closure was performed (no pericystectomy and no capitonnage (G3a)), 2 patients from G3a due to prolonged bronchial fistula and empyema required reoperation for re closure of bronchial opening and pericystectomy and

capitonnage. The rest (11 patients) underwent capitonnage (no pericystectomy but capitonnage and bronchial opening closure were performed (G3b)). Three patients from G3b were complicated (hydropneumothorax, capitonnage dehiscence, and tension pneumothorax).

In 8 patients, the postoperative course was uneventful, however, some of them had prolonged productive coughs. From 12 cases of G3, besides of mentioned procedure, similar as in G1 and G2, pericystectomy also has been performed in this subgroup (G3c). We did not find considerable major complications except minor hemorrhage during pericystectomy and the procedure took half an hour more surgical time. Because of the favorable results of this option (G3c), this procedure became our routine practice and both other subtypes of operations (G3a and G3b) in the suppurative group were abandoned (Table 2). So, we had 5 out of 28 (18%) severely infected cysts (G3), major complications (2 in G3a and 3 in G3b), and none of the early and intact cysts or Gc were complicated Table 2.

The mean hospital stay was 6 days (4-8 days) both in G1 and G2 operation but 12 days in suppurated operation (G3). The majority of the cysts (87 cysts, 57%) were located on the right side, with 56 (37%) in the right lower lobe, 14 (9%) in the middle lobe, and 17 (11%) in the right upper lobe. On the left side, there were 65 cysts (43%) in total, with 36 (24%) in the lower lobe, 16 (10%) in the upper lobe, and 13 (9%) in the middle lobe.

The most prevalent symptoms were cough in 67% of patients, productive cough in 51%, mild hemoptysis in 11%, and moderate dyspnea in 20%. Fever was reported in 25% of cases. The white blood cell count was 10.77 (SD 4.23), hemoglobin was 12.15 (SD 2.29), and eosinophil levels ranged from 2 to 9 (mean 2.8, SD 0.8). The platelet count was 283950 (SD 81).

The study found that 25 cases had extra-thoracic involvement of the liver. Additionally, in 10 cases (7%), involvement of the lung, liver, and spleen was observed simultaneously.

**Table 2.** Surgical results in various options groups

Operation groups	Diameter	number	Early morbidity	Late morbidity	Mortality	Mean Hospital stay after operation
Cystectomy+ LBOC +capitonnage in intact cyst (G1)	6-10Cm=60 (39%) 10-15cm=12(8%)	72	4wound infection	0	0	6 days
Cystectomy + LBOC + capitonnage in early infected cyst (G2)	6-10Cm=24(16%) 10-15cm14(9%)	38	4wound infection Pneumonia=3			6 days
Cystectomy + LBOC without pericystectomy without capitonnage in severe cavity suppuration. (G3a)	6-10=1(1%) 10-15Cm=4(3%)	5	Reoperation(bronchial fistula and empyema 2 2wound infection Pneumonia=5	2	0	12 days
Cystectomy + LBOC without pericystectomy+ capitonnage in severe cavitory suppuration. (G3b)	6-10Cm=8(5%) 10-15cm3(2%)	11	empyema copitonnage dehiscence)=3 2wound infection Pneumonia=4	0	0	12 days
Cystectomy + LBOC with pericystectomy + capitonnage in severe cavitory suppuration. (G3c)	6-10cmCm=8(5%) 10-15Cm=4(3%)	12	2wound infection Pneumonia=3		0	11 days
Bilateral cystectomy+ large bronchial opening closure + capitonnageininfected cyst	6-10=1(1%) 10-15Cm=4(3%)	5	pneumonia =1	0	0	6 days
Bilateral cystectomy+ large bronchial opening closure + capitonnage + intact cyst	6-10=6(4%) 10-15Cm=3(2%)	9	0		0	6 days
Total==152(100%)	6-10cm=108(71%)	10-15Cm=44(29%)	6		0	

Based on radiographic findings, the diameter of infected cysts ( $98.5 \pm 21.4$  mm) was slightly greater than that of intact cysts ( $74.68 \pm 25$  mm). The diameter of intact cysts (6-10 cm) was greater than that of early infected cysts (56 vs. 25 cysts).

In this study, the diameter of cysts between 6-10 cm was 108 (71%) and 10-15 cm was 44 (29%). Fourteen cysts (more than 5 cm) were bilateral (10%), with 5 cysts in the intact group and 9 in the infected group.

### Outcome

Primary endpoints: capitonnage prevented from the bronchopleural fistula, and prolonged hospital stay in all forms of hydatid cyst.

Secondary endpoints: pericystectomy prevented the capitonnage dehiscence, prolonged sputum, and hospital stay among suppurated cysts.

The majority of our patients (79%) were normal healthy people (ASA I), the rest were in ASA II class, such as controlled diabetes mellitus (DM) or chronic obstructive pulmonary disease (COPD). In the pre-operative pulmonology and cardiology consultation, indicated low to moderate risk, ejection fractions between 45-50% were reported. No cardiac or respiratory disease after surgery happened.

In Assigned Weights for Diseases, most of patients had CCI 1-2; some had CCI 3-4 (e.g., controlled DM and COPD). They had no serious disease, leukemia, or malignancy. CCI was calculated according to the scoring system established by Charlson et al (6). There was no difference regarding laboratory data at admission time between the two main groups of intact and infected cysts; however, in suppurated cysts, patients had leukocytosis and "shift to the left".

Post-operative complications were classified according to the Clavien-Dindo classification (CDC) (7) graded between 1 and 5. In the suppurated group, five major complications were reported. In subgroup G3a, two complications that required reoperation were bronchial fistula and empyema. In subgroup G3b, there were three complications, including three patients with empyema and

copitonnage dehiscence (grade 3a CDC), 18 cases of pneumonia (13%) (grade 2 CDC), and a surgical site infection (SSI) rate of 11% seen in 14 cases. The pneumonia rate was 12.7% (grade 2 CDC).

We had no mortality by any cause during hospitalization time within 30 days (grade 5 CDC=0). No Clavien-Dindo grade IV complication occurred during the investigation, and no recurrence of hydatid cyst at least during 5 years of follow-up occurred despite no anti-parasitic drug having been administered.

The median length of hospitalization for intact and early infected was 6 days and for severely infected was 11-12 days.

After the operation, the lung tissue gradually expanded during the 3rd month of follow-up. Delayed lung expansion was more noticeable in the infected groups, but the pericyst cavity was gradually eliminated. There was no abscess formation in the remaining cavity space, and no air-fluid level on chest radiography or any respiratory symptoms were observed after the 3rd month of follow-up, both in intact and early infected cysts (G3c). However, there were some major complications such as uncapitonnage (G3a) and unpericystectomy with capitonnage (G3b). No lobectomy was required, except for a limited number of wedge resections of nonfunctional small cysts, which were accompanied by a giant cyst.

### Complications at the time of surgery

Transient Hypoxemia during operation occurred in some G3 patients that were managed with tracheal suction and supine positioning.

There was no complication in intact and early infected cysts except the surgical site infection rate was mostly in G3 (21%) then G2 (9%) and intact (5%), pneumonia 15 cases (12%), but major complication such as empyema, bronchopleural fistula in 5 cases (4%), of G3 happened. There was no mortality by any cause during hospitalization time within 30 days, no recurrence at least during 5 years of follow-up, occurred despite no anti-parasitic drug being administered.

## DISCUSSION

In this study on the treatment of impacted parenchymal, 152 cysts were analyzed. Forty-four (29%) of the cysts were giant hydrated cysts, and one lobe was invaded by a giant cyst. Following surgery, the diseased lobe gradually expanded anatomically during the 3rd month of follow-up. Most surgeons agree on the preservation of pulmonary parenchyma. However, lobectomy may be necessary if the invasion is more than 50% of the lobe by a cyst, or in the presence of multiple cysts in the same lobe, a destructed lobe, or complications from the cyst (such as bronchiectasis and hemorrhage) (9). In our series, none of our patients required lobectomy. A limited number underwent wedge resection of nonfunctional tissue, which were accompanied by a giant cyst. It appears that when lung parenchyma is compressed by a giant cyst, the surrounding tissue is not permanently destroyed. After decompression, the majority of lung parenchyma gradually returns to its anatomical expansion, as confirmed in chest CT scans. However, for a functional improvement rate, more functional studies are needed. In severely suppurated cysts, pericystectomy is our preferred option. Since the source of infection is removed, it may potentially help preserve the involved lobe.

A study has suggested that parenchyma-saving methods in the treatment of giant lung hydatid cysts lead to a greater length of hospital stay when compared with performing lobectomy:  $28.21 \pm 6.53$  vs.  $18.41 \pm 4.31$  days (7). However, recently in a study, hospital stay was reported  $11.4 \pm 5.7$  days (9). In our study, the hospital stay after lung resection was either the same or shorter.

Seventy-nine of the patients had ASA-I and CCI scores of 1-2, while the rest had ASA II with CCI scores of 3-4. The 11% surgical site infection rate in our patients may be related to the fact that 47% of them had infected cysts, which increased the SSI rate. According to the Clavien-Dindo classification, it is compatible with Grade IIIb, with a morbidity rate of 18.4% in severe cavity abscesses (G3). However, this complication presented in G3a at a rate of 40%, in G3b at 27%, and in G3c at 0%. Patients with higher

Clavien scores experienced severe complications and had a longer length of hospital stay (12 days in G3 versus 6 days in G1 and G2).

The study found that the preferred treatment option for preserving lung tissue in intact pulmonary hydatid cysts (G1) and early infected cysts (G2) was the same, and both options yielded favorable results. There was no difference in complication rates between medium-sized cysts (6-10cm) and giant cysts in either G1 or G2. In the case of early infected cysts (G2), carefully cleaning the outer layer of the cyst with normal saline yielded similar outcomes to intact cysts and did not lead to major complications, changes in Clavien-Dindo scores, or longer hospital stays. Therefore, the choice of cystectomy, large bronchial opening closure, and capitonnage may be considered as an optimal conservative surgical treatment for pulmonary hydatid cysts of all sizes, whether intact or early infected.

In our study, the frequency of cyst rupture into the bronchial was 71 (47%) as a main factor of infection. This complication is reported in the literature as 24.7%-61% (10,11). Perforation of the cyst into the thoracic cavity can cause pneumothorax, hydropneumothorax, and empyema (12). Perforation occurred in 8 cases (5%) (8). In a giant cyst, the rupture into the pleural space initially presented as thoracic empyema. During the thoracotomy, we found a perforated pleural hydatid cyst (PHC) managed with decortication, extraction of the laminated membrane, and the G2 procedure. Therefore, in endemic areas, one of the causes of thoracic empyema may be due to a perforated PHC.

In our experience, we encountered a significant challenge with a severely suppurated cavity (G3). After removing the cyst, the cavity surface was found to be suppurated, dirty, and containing necrotic tissue. To manage the contaminated cyst cavity, various methods were used, such as draining the cyst, closing all the bronchial openings in the pericyst and leaving the pericyst cavity open into the pleural space (13), performing capitonnage with or without suturing the openings, and occasionally conducting pericystectomy (14).

It has been shown in a study that peri-cystectomy decreases the chances of recurrence rate, but it increases the risk of air leak, tension pneumothorax, and bronchopleural fistula formation in acute cases. Therefore, pericystectomy is not advised (15). Opponents argue that the peri-cyst is not a part of the hydatid cyst itself, and its removal may lead to an increased risk of airway leak (16). Bagheri et al. (17) also did not recommend peri-cystectomy, but some authors recommend partial resection or resection of the free parts of the peri-cyst (18). Those in favor of pericystectomy believe that the cavity may undergo amyloid degeneration and calcification, and therefore, with pericystectomy, the parasite will be completely removed (19).

Our study showed that serious complications were only related to option G3a (capitonnage without pericystectomy) and option G3b (clavien-dindo scores and length of hospital stay also showed significant increases). Therefore, these options are not recommended. On the other hand, option G3c, which includes total pericystectomy, had no major complications. Therefore, G3c is the appropriate and preferred treatment modality for giant cysts and cavity abscess cysts. These options are currently routine in our practice, and we recommend performing them in endemic areas to reduce the risk of encountering re-infection.

In cases of a suppurated cyst resulting from fragmented tissue and necrosis, performing a pericystectomy resection is not difficult, unlike with early infected cysts where there is intensive fibrotic adhesion. This option is considered safe and feasible, with minimal risk of early or late complications. The procedure may take around half an hour and may involve minimal hemorrhage during pericystectomy.

In these procedures, only the large bronchial opening was closed because small bronchial openings may help to prevent the formation of potential blind cavity spaces and abscesses.

In severely infected pericysts with necrotic tissue and microbial foci, performing only bronchial openings closure

and capitonnage (G3b) may have the potential for cavity abscess formation and/or capitonnaged cyst dehiscence. However, in some patients in whom capitonnage was not performed (G3a), bronchial fistula and empyema also occurred. Therefore, there is more emphasis on performing capitonnage on intact, early infected cysts and in suppurated cysts when pericystectomy has been performed.

There is ongoing debate among surgeons about the effectiveness of performing capitonnage. Capitonnage has been a common procedure since 1952, with the goal of reducing post-operative air leaks and preventing empyema formation (20). However, this belief has recently been contested by different surgical groups (4, 21, 22). Some advocate for capitonnage as a standard practice (23), while another study does not recommend it due to the risk of creating a blind contaminated cavity within the lung, with complications that may surpass those of an open cavity in the pleural space (24). Furthermore, some argue that capitonnage is not only unnecessary but also hinders lung expansion (4, 21, 23).

In our study, performing capitonnage was necessary in all types of PHC. It prevented major complications, provided additional strength to the lung parenchyma, and prevented post-operative air leaks and empyema formation with excellent results.

The most common postoperative complication in our study was atelectasis, mostly seen in giant cysts. The wound site infection rate was mostly in G3 (21%), then G2 (9%), and intact (5%). Pneumonia occurred at a rate of 12%. Major complications like empyema and bronchopleural fistula occurred in 4% of G3a and G3b cases. Fortunately, there were no deaths from any cause during the 30-day hospitalization period, which is consistent with the literature.

The incidence of bilateral hydatid cysts in our series was 10%, while the literature reports an incidence of 17-20% (24,25).

The study found that there were no cyst recurrences during the follow-up period, even though no anti-parasitic drugs were given before or after the operation. This could be due to careful handling of the cyst during surgery and preventing any spillage into the chest cavity or incision site. Another study reported no recurrences despite a 29.4% cyst perforation rate (26). However, in a different study, recurrence occurred in 2 cases in the chest wall. Therefore, there is controversy over the use of anti-helminths post-surgery for preventing recurrence (27). After the proper removal of the laminated membrane, albendazole may not be necessary for intact cysts, as its action targets the laminated membrane's terminating layer. This approach resulted in no recurrence for us, as the infected cyst's germinative layer had been inactivated, and any spillage into the chest cavity may not be harmful.

### Limitations

In this study, we could only include those who had no missing data and all must have had a chest CT scan. Other limitations of the study included its retrospective nature, the lack of bacterial studies in supplicated cysts, and the absence of functional studies after giant cystectomies.

### CONCLUSION

Capitonnage is an essential part of the treatment for all forms of PHC (pulmonary hydatid cyst) including cystectomy, and closure of major bronchial openings, and also is the best surgical option for intact and early infected cysts. The optimal approach for a supplicated cyst includes closure of the major bronchial opening, pericystectomy, and capitonnage. Preservation of pulmonary parenchyma is often feasible and safe.

### Data Availability Statement

All informative data were extracted from the medical records database of Shahid Beheshti Teaching General Hospital, Kashan, Iran, and also characteristics of the cysts obtained from chest computed tomography reports, surgical notes, and pathologic results are available on request.

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### Conflicts of interest

The authors declare that they have no competing interests, and no author received research grants, or speaker honorarium from any company.

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