



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

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Abstract

There are some controversies on surgical options of pulmonary hydatid cyst; no surgical guideline is available to considering the best option in each various types.

Aim & Objective: We analyzed our experience in intact and infected pulmonary hydatid cysts of >5 cm diameter with focusing on possible preserving the lung tissue, the best surgical option in each type, and compare it with others similarities published studies.

Patients and Methods: This observational cohort analysis was conducted on 138 consecutive patients with pulmonary hydatid cyst during July 2008 to July 2022.

Surgical options were analyzed on cyst >5 cm diameter, classified as infected and intact cyst. The Age, sex, clinical manifestation, recurrent rate, hospital length of stay, postoperative complications and long-term results in each groups were assessed. The American Society of Anesthesiologists Physical Status, Charlson Comorbidity Index (CCI), Complexity of surgery and Clavien-Dindo score also were determined.

Results: Out of 138 patients, between 15 to 79 years old, had 152 cyst >5 cm cyst diameter, 81 case (53.5%) were intact cyst classified as group 1 (G1), and the rest was infected which subdivided in to early infected 43 cases =28.4% as (G2) and cavity suppurated =28 cases (18.4%) as (G3). Cystectomy, closure of major bronchial opening and capitonnage was done in intact and early infected cysts. The result of both was the same, with no considerable major complication.

Options in group G3 required special attention due to pericyst surface was severely inflamed, dirty and had pus, so were subdivided to 3 distinctive group include G3a, undergone bronchial opening closure without capitonnage, G3b group, undergone capitonnage without pericystectomy and G3c group, in addition of bronchial opening closure and capitonnage, pericystectomy also had been performed. Major complication in the subgroups of G3a was 2 patients, G3b 3 patients, but in the subgroup of G3c, not considerable complication was seen.

Conclusion: Capitonnage significantly decreased the complication rate of surgical treatment in of all form of pulmonary hydatid cyst. Optimized approach in both intact and in early infected cysts was: Cystectomy, closure of major bronchial opening and capitonnage, in suppurated cyst, also was bronchial opening closure, Pericystectomy and capitonnage.

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

Introduction

Pulmonary hydatid cyst is a parasitic infection, exists in many endemic areas of worldwide [1]. Due to the elasticity of lung structure, the cyst growths into giant dimensions [2]. The clinical

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manifestation of Pulmonary Hydatid Cysts (PHC), depends on the sizes, location and whether the cyst being intact or ruptured. Surgery is the main stream of treatment, includes: Cystectomy alone, cystectomy and capitonnage, enucleation, and pericystectomy [3]. Despite a numerous studies controversy is still persists on the optional selection, no surgical guidelines is available, and a little consensus is among surgeons for performing pericystectomy and capitonnage in severely infected PHC (suppurated cyst) [4].

Aim & Objective

To share our experience and comparing its outcomes, we doing capitonnage in all form of cyst and Pericystectomy in suppurated cyst. Our plan was Parenchymal saving, unless being confront with situation that lobectomy is indicated such as multiple unilobar cysts, destructed of a lobe or bronchiectasis.

Patients and Methods

In this study, 138 consecutive adult patients with pulmonary hydatid cyst of >5 cm who treated surgically in a teaching Hospital from July 2008 to July 2022 were analyzed. 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 suppurated cyst and the option was cystectomy+ removal of laminated membrane + closure of bronchial opening without capitonnage, because 2 of 5 (40%) patients had been complicated with empyema, were abandoned from our practice, so preferred strategy of management from that time, up to now have been performed obligatory capitonnage in both intact or infected cyst.

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

All informative data were extracted from the Medical records database of our hospital. Analysis were Performed into three groups based on intact, early infected and suppurative. Characteristics of the cysts were obtained from chest computed tomography reports and surgical notes. The American Society of Anesthesiologists Physical Status (ASA-PS). Charlson Comorbidity Index (CCI), [5]. Complexity of surgery and Clavien-Dindoscore (CDC) [6], also were determined.

Definitions

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

Early infected hydatid cysts

Clinical the patients had systemic inflammatory response and radiological finding, which was suggested for hydatid cyst infection, in early infected PHC the patients were not septic [7] and residual cavity surface was not dirty or cleaned by washing.

Severely Infected hydatid cyst or suppurated cavity

The patients were septic, had fever and purulent sputum due to lung abscess. Cyst cavity surface in operation filed was dirty, obvious necrotic tissue and severely were inflamed. The specific CT features of infected pulmonary hydatid cyst had air fluid level, and water lily Sign.

Inclusion criteria

Age of >15 years, cysts >5 cm diameter, complete data and availability for follow up.

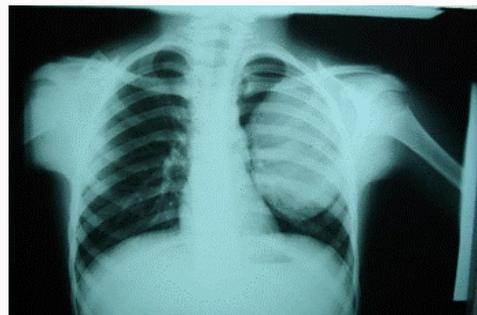


Figure 1: PA chest X-ray a intact giant hydatid cyst in the left upper lobe of 21 years old.

Exclusion criteria

Age less than 15 years, incomplete data in medical record, cysts less than 6 cm.

Surgical Technique

Under general anesthesia, with double-lumen end tracheal tube. A poster lateral, muscle saving thoracotomy through the 5th to 7th intercostal space, depending on the location of the cyst and in bilateral lung cysts, sequential bilateral thoracotomy were performed.

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

In intact cyst after the cyst fluid was suctioned carefully, the laminated membrane, were removed, only major bronchial openings 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 the severely infected cyst due to residual cavity was dirty with obvious necrotic tissue and did not cleaned with normal saline, capitonnage was performed after carefully resection of the pericyst totally or as match as possible, with little parenchymal damage, (Figure 2, 3). Decortication was performed in patients with pleural complications [8].

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.

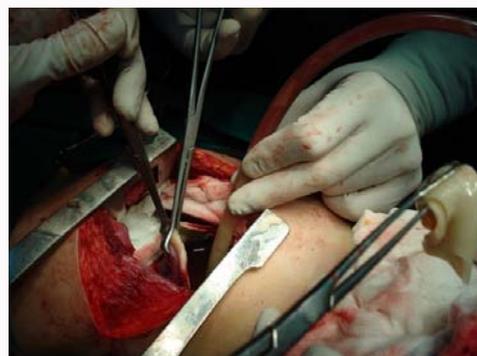


Figure 2: Demonstrate a perforated giant hydatid cyst in the right lower lobe.



Figure 3: Total pericystectomy in the right lower lobe.

Followed up and Outcome

Follow-up was continuing mostly more than five years for possible complications and recurrence rate.

Result

In this study we have 138 Patients, the ages was between 15 to 89 years old, mean age was 41.7 SD 18.8 years, 88 cases (64%) was man the rest 50 (36%) was women, m/w ratio was 1.76/1.81 case (53.%) of cysts were intact, and 71 case (47%) were infected. There was significant difference between non perforated and perforated cyst P-value =0.009 (Table 1).

From 71 cases (47%) of infected cysts, 43 cases (28%) were considered as early infected cyst or G2, from this group, 8 case (5%), had been ruptured into the pleura space, presented as loculated empyema, beside of cystectomy, bronchial opening closure and capitonnage, decortication also was performed.

28 cases (18.4%) of infected cysts were severely infected and considered as G3. This group because of pericyst surface was dirty, severely necrotic, and pus, divided in to 3 sub-groups. From 16 case of G3 who pricystectomy had not been performed, 5 case capitonnage also had not been performed, only bronchial opening closure was performed (no pricystectomy 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) undergone capitonnage, (no pricystectomy but capitonnage and bronchial opening closure was performed) as (G3b), three patients from G3b were complicated (hydro pneumothorax, capitonnage dehiscence and tension pneumothorax).

However in 8 patients post-operative course was uneventful but some of them had prolonged productive sputum. From 12 cases of G3 beside of mentioned procedure, similar as in G1 and G2,

pericystectomy also has been performed; in this subgroup (G3c) we had no found considerable major complication except minor hemorrhage during pericystectomy and about half hour more time consumption. Because of favorites results of this option (G3c), this procedure became routine of our practice and both other sub type of operations (G3a and G3b) in suppurative group were abandoned (Table 2). So we had 5 out of 28 (18%) in severely infected cyst (G3), major complication (2 in G3a and 3 in G3b), none of early and intact cyst or Gc were complicated (Table 2).

The mean hospital stay was 6 days (4 to 8 days) both in G1 and G2 operation but in suppurated operation (G3) 12 days. The most localizations of the cysts were 87 cyst (57%) in the right side (the right lower lobe =56, middle lobe =14 right upper lobe 17 cyst. In left side 65 cyst (43%) lower lobe =36 then upper lobe =16 and middle lobe 13.

The most symptoms were cough in (67%) of patients, productive sputum in 51%, mild hemoptysis 11%. Moderate dyspnea, 20% and fever, 25 %. WBC=10.77 SD 4.23, Hb=12.15 SD 2.29 Eosinophil was between 2 to 9 mean (2.8 SD 0.8) platelet =283950 SD 81.

Simultaneously extra thoracic involvement of liver in 25 case was found, simultaneously lung liver and spleen in 10 cases (7%) was seen Based on radiographic finding diameter of infected cyst (98.5 SD 21.4) mm was slightly greater than intact cyst 74.68 SD 25. Cyst between (6 cm to 10 cm) diameter in intact cyst was more than early infected (56 vs. 25 cyst).

In this study, diameter of cysts between 6 cm to 10 cm were 108 (71%) and 10 cm to 15 cm =44 (29%) 14 cyst (more than 5 cm) were bilaterally (10%) which 5 cyst in intact and 9 in 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 from the capitonnage dehiscence, prolonged sputum and hospital stay among suppurated cyst.

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, had low to moderate risk, ejection fractions between 45% to 50% were reported. No cardiac or respiratory disease after surgery was happened.

In assigned weights for diseases, most of our 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].

Table 1: Demographic data and side of cyst.

Variable	Perforated n=71 (47%)	Non-perforated n=81 (53%)	Total n=152 (100%)	P-value
Age (mean SD)	39.29 ± 17.83	42.92 ± 17.72	41.71 ± 18.08	0.037
Gender (n%)				
Male	34 (60%)	54 (67.0%)	88 (64%)	0.243
Female	23 (40.0%)	27 (33.0%)	50 (36%)	
Side (n%)				
Right	42 (59%)	45 (56%)	87 (57%)	0.009
Left	29 (41%)	36 (44%)	65 (43%)	

Table 2: Surgical results in various options groups.

Operation groups	Diameter	Number	Early morbidity	Late morbidity	Mortality	Mean Hospital stay after operation
systostomy+LBOC +capitonnage in intact cyst. (G1)	6-10 cm=60 (39%)	72	4wound infection	0	0	6 days
	10-15 cm=12 (8%)					
cystectomy + LBOC +capitonnage in early infected cyst (G2)	6-10 cm=24 (16)	38	4wound infection Pneumonia=3			6 days
	10-15 cm 14 (9%)					
cystectomy + LBOC without pericystectomy without capitonnage in severe cavity suppuration. (G3a)	6-10=1 (1%)	5	Reoperation (bronchial fistula and empyema 2 2wound infection Pneumonia=5	2	0	12 days
	10-15 cm=4 (3%)					
cystectomy + LBOC without pericystectomy+capitonnage in severe cavitory suppuration. (G3b)	6-10 cm=8 (5%)	11	empyema copitonnage dehiscence)=3 2 wound infection Pneumonia=4	0	0	12 days
	10-15 cm 3 (2%)					
cystectomy + LBOC with pericystectomy+capitonnage in severe cavitory suppuration. (G3c)	6-10 cm=8 (5%)	12	2 wound infection Pneumonia=3		0	11 days
	10-15 cm=4 (3%)					
bilateral systostomy+large bronchial opening closure +capitonnage in infected cyst	6-10=1 (1%)	5	pneumonia =1	0	0	6 days
	10-15 cm=4 (3%)					
Bilateral systostomy+large bronchial opening closure +capitonnage+intact cyst	6-10=6 (4%)	9	0		0	6 days
	10-15 cm=3 (2%)					
Total=152 (100%)	6-10 cm=108 (71%)	6			0	
	10-15 cm=44 (29%)					
Large Bronchial Opening Closure (LBOC)						

There were no differences regarding laboratory data at admission time in the 2 main groups of intact cyst and infected 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 suppurated group, five major complications were reported in the G3a, 2 complications that need Reoperation for bronchial fistula and empyema, in the G3b, 3 complications including 3 patients with empyema and capitonnage dehiscence (grade 3a CDC), pneumonia 18 case (13%) (grade 2 CDC) and), Surgical Site Infection (SSI) rate was seen in 14 cases (11%), pneumonia 12.7% (grade 2 CDC).

No death from any cause during hospitalization time within 30 days (grade 5 CDC=0). No Clavien grade IV complication occurred during the investigation was occurred, no recurrence of hydatid cyst at least during 5 years of follow up, was occurred in spite of no any anti-parasitic drug has been administered.

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

After operation, the lung parenchyma gradually during 3rd month of follow up was expanded; delayed lung expansion was happen more in infected groups, pericyst cavity also gradually eliminated. No abscess formation in remaining cavity space, air fluid level on chest radiography or any respiratory symptoms was seen after 3rd month of follow up, in intact and early infected cyst and G3c. However the major complication was (uncapitonnage) G3a and un-pericystectomy with capitonnage), G3b. no lobectomy was required except limited number or wedge resection of nonfunctional small cyst, which were accompanied with a giant cyst.

Complications at the Time of Surgery

Transient Hypoxemia during operation occurred in some of G3

patients that managed with tracheal suction and supine positioning.

There was no complication in intact and early infected cyst except the wound site infection rate was mostly in G3 (21%) then G2 (9%) and intact (5%), pneumonia 18 case (13%) 12.7%, but major complication with empyema, Bronchopleural fistula, in 5 cases 4%, of G3 was happened no death from any cause during hospitalization time within 30 days was occurred, no recurrence at least during 5 years of follow up, was occurred in spite of no any anti-parasitic drug has been administered.

Discussion

In this study with impacted on Parenchymal saving treatment, 152 cyst were analyzed, 44 (29%) were giant hydatid cysts and one lobe has been invaded by giant cyst, after surgery the diseased lobe, gradually was expanded anatomically during 3rd month of follow up.

Most surgeon have consensus on preservation of pulmonary parenchyma, however lobectomy may necessary if the invasion is more than 50% of the lobe by cyst or presence of multiple cysts in the same lobe, destructed lobe or sequela of the cyst (bronchiectasis and hemorrhage) [9]. We did not confront with this situation that required lobectomy, except limited number or wedge resection of nonfunctional tissue, which were accompanied with a giant cyst. It seems when lung parenchyma is compressed by giant cyst, the surrounding tissue is not destructed permanently, after decompression, and gradually the majority of lung parenchyma will obtain its anatomical expansion which is confirmed in chest CT scan. However for functional improvement rate, needs more functional studies. In severely suppurated cyst also, with pericystectomy, as our preferred option, since the source of infection is removed, it may potentially helps to preserves the involved lobe.

In a study have suggested, parenchyma saving methods in the

treatment of giant lung hydatid cysts, leads greater length of hospital stay when compared with performing lobectomy 28.21 ± 6.53 vs. 18.41 ± 4.31 days [7]. But recently in a study hospital stay was reported 11.4 ± 5.7 days [9]. In our study, also the length of hospital stay in compare with mentioned lung resection was the same or less.

Seventy-nine of our patients were in ASA-I and CCI scores of 1 to 2 and the rest were in ASA II, with CCI scores of 3 to 4. Surgical site infection rate of 11% is acceptable in our patients may be related to considerable (47%) of our patients had infected cyst that increased the SSI rate. On the basis of the Clavien-Dindo classification, it is compatible with Grade IIIb, with a morbidity rate of in severe cavity abscess as G3 in 28 cases (18.4%), however this complication in G3a was 2(5)=40% and in G3b, 3(11)=27% but in G3c, 0(12)=0%, so patients with higher Clavien scores have severe complications and longer length of hospital stay (12 days in G3 vs. 6 days in G1 and G2).

This study showed, the selected option for parenchymal saving of pulmonary hydatid cyst in intact cyst (G1) and early infected (G2) was the same and both has the same favorable results, there was no different complication rate among medium size cyst (6 cm to 10 cm) and giant cyst, in G1 and G2 option, because in early infected cyst (G2), pericyst when being cleaned carefully with normal saline, it is similar intact cyst, does not increase major complications, Clavien-Dindo scores and hospital stay, so the option of cystectomy + large bronchial opening closure and capitonnage may being considered as choice and optimized conservative surgical treatment of pulmonary hydatid cyst in all size in intact or early infected cyst.

The frequency of rupture of cyst in to bronchial as a main factor of infection, in our study was 71 (47%), this complication in published study were reported as 24.7% to 61% [10,11]. Perforation of the cyst in to thoracic cavity can cause, pneumothorax, hydro pneumothorax, empyema [12,13], Perforation was happened in 8 case (5%) [8], in giant cyst had been ruptured into pleural space and presented initially as thoracic empyema our finding in thoracotomy was perforated PHC, managed with decortication, extraction of laminated membrane, and G2 procedure. So in endemic area, the thoracic empyema may also is due to perforated PHC.

In our experiences the great challenge was seen on severely suppurated cavity (G3). After removal of cyst contain, the cavity surface was suppurated, dirty and necrotic tissue, in this situation for the management of the contaminated cyst cavity, different methods has been used such as: 1) Leaving the cavity open into the pleural space after closure the bronchial openings, 2) capitonnage with or without suturing the openings, and 3) occasionally pericystectomy [14]. In a study has been showed pericystectomy decreased of chances of recurrence rate however it increased risk of air leak and tension pneumothorax in acute cases or broncho-pleural fistula formation so, did not advised the pericystectomy [15], Opponents also believes pericystectomy is not a part of the parasite, and its removal may lead to increased risk of airway leak [16]. Another study also no advocated pericystectomy [17], but some authors recommend partially or resecting the free parts of the pericyst [18]. The proponents of this method believe pericystic cavity may undergo Amyloid degeneration and calcification, with pericystectomy the parasite completely will removed of [19].

Our study showed serious complication was related to only in (G3a) and capitonnage without pericystectomy (G3b). Clavien-Dindo scores and length of hospital also have been increased

significantly, so this options are not recommended, G3c option which included total pericystectomy had no any major complication hence G3c is appropriate and preferred treatment modality in giant cyst of suppurated and cavity abscess cysts, this options currently are routine of our practice and we recommend to performing in endemic areas due to risk of encountering re-infection.

In suppurated cyst due to fragmented tissue and necrosis of pericystectomy, resection is not difficult as seen in the early infected cyst, which is intensive fibrotic adhesion, so this option is safe, feasible and has no any impose of early or late complication, except a half of our time consumption and minimal hemorrhage during pericystectomy.

In all of this procedure only large bronchial opening was closed because small bronchial openings may helps to prevent possible blind cavity space and abscesses formation.

In the severely infected of pericyst due to having necrotic tissue and microbial foci, performing only bronchial openings closure and capitonnage (G3b), may has potential of cavity abscess formation and/or capitonnaged cyst dehiscence, however in some patient who capitonnage was not performed (G3a) the bronchial fistula and empyema also was happened. So we have more stress on doing capitonnage on intact, early infected cyst and in suppurated cyst when pericystectomy had been done.

There are some debates about performing capitonnage among surgeons. Capitonnage has been done since 1952, with the intention that it would reduce post-operative air leak and empyema formation [20]. However, this contention has been challenged recently by various surgical groups [4,21,22]. Some advocate it, as common practice [23] but in another study this option is not recommended due to capitonnage causes a blind contaminated cavity which remains in the lung and its complication can exceed that of an open cavity into the pleural space [24] some suggest that capitonnage is not only unnecessary but also interferes with lung expansion [4,21,23].

In our study performing capitonnage was necessary in all type of PHC and it prevented from major complication, provided additional strength to the lung parenchyma and prevented post-operative air leak and empyema formation with excellent results.

In our study, the most common postoperative complication was atelectasis. Mostly in giant cyst were seen. The wound site infection rate also mostly was seen G3 (21%) then G2 (9%) and intact (5%), pneumonia 10.7%, but major complication with empyema, bronchopleural fistula, in 5 cases 4%, of G3a and G3b was happened no death from any cause during hospitalization time within 30 days was occurred which is in line with the literature [10]. The incidence of bilateral hydatid cyst in our series was 10% however in Literature 17% to 20% was reported [24,25].

In this study, no cyst recurrence was occurred during follow up, in spite of no any anti-parasitic drugs pre or post operation were administered. The cause of it may be careful cyst manipulation during surgery and prevention from any spillage to intra thoracic cavity or incision site. In a study, despite a cyst perforation rate of 29.4%, no recurrences were reported [1]. In another study recurrence occurred in 2 cases in the chest wall. So, the role of the post-surgical use of anti-helminthes for prevention of recurrence is controversial. Since albendazole effects on the germinative layer of the laminated membrane, after properly removal of laminated membrane no need

to albendazole administrated in intact cyst and no any recurrence will be happened. The germinative layer had been inactivated in infect cyst and spillage to intra thoracic cavity also may be harmless.

Conclusion

Capitonnage is essential part of the treatment of all form of PHC cystectomy, cystectomy, closure of major bronchial opening and capitonnage is the best surgical option in both intact and in early infected cysts. Optimal approaching in suppurated cyst is closure of major bronchial opening, pericystectomy and capitonnage. The preservation of pulmonary parenchyma is often feasible and safe.

Imitations

In this study, we could only include those who have no missing data, all must have chest CT scan; other limitations of the study was a retrospective nature, did not bacterial studies in suppurated cyst and functional study after giant cystectomies.

Highlight

Capitonnage is essential part of the treatment of all form of PHC special giant cysts. Optimal surgical approach in suppurated cyst is Pericystectomy. The diagnosis of PHC is made on means of imaging and serology test seems is no require with standard surgery no albendazole administration routinely is recommended before or after surgery.

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