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Original Article

Capitonnage Method for Surgical Management of Pulmonary Hydatid Cysts: A Case Series

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Abstract

Introduction

Hydatid disease of the lung, which is caused by *Echinococcus granulosus*, is commonly found in Mediterranean countries. Surgery is the primary mode of treatment for patients with pulmonary hydatid disease. This study aims to assess the efficacy of capitonnage in the management of the condition.

Methods

This retrospective study was done between February 2017 and December 2019, in Al-Jumhoory Teaching Hospital, Al-Khansaa Teaching Hospital, and Al-Rabee Private Hospital in Mosul, Iraq. Patients with different presentations of pulmonary hydatid disease were treated surgically with cyst enucleation and capitonnage technique.

Results

A total of 118 patients with pulmonary hydatid cyst cases were included in our study. There were 73 males and 45 females, with a mean age of 30 years. More than one-third (45 patients) were asymptomatic, and 73 complained of chest pain, cough, and/or respiratory distress. Other organs were involved in 27 patients, 22 in the liver, 3 in the spleen, and 2 in both. Forty-nine patients had intact cysts that were extracted intact, while the remaining 69 had ruptured cysts, and 6 patients of the latter group presented with pneumothorax. A small number of these patients developed prolonged air leakage, and the majority stayed in the hospital for one day. No death occurred in our study.

Conclusion

We concluded that when the capitonnage technique is used carefully on the foldings of pericystic tissue, the rate of atelectasis and distortion is very low and may be beneficial in reducing the risk of postoperative air leak.

1. Introduction

Pulmonary hydatid cyst is a zoonotic disease that can be seen worldwide, with the highest incidence reported in Australia, New Zealand, South Africa, South America, Turkey, and Mediterranean countries. Hydatid disease is a major public health problem in locations where it is endemic. Most individuals who contract this parasite are young, with most patients being under the age of 40 [1,2]. It is common in sheep- and cattle-raising areas around the world. Humans serve as an accidental intermediate host for cysts, most commonly found in

the liver and lungs; however, they can also be found in nearly any organ [3].

In humans, the lungs are the second most common lodgment site of the parasite (18-35%), with liver involvement occurring in 50 to 60% of cases [4]. Conventional radiography is commonly used to diagnose the disease. A CT scan of the chest and upper abdomen may be useful for an accurate diagnosis of complicated cysts [5].

The most common surgical procedures for hydatid disease are cystotomy, capitonnage, enucleation, and pericystectomy. Capitonnage has long been used to prevent postoperative air leak and empyema formation [6]. However, some researchers have recently suggested that capitonnage provides no additional benefit, as well as only a cystotomy with bronchial closure will be sufficient for treatment [2]. This has prompted us to review our experience of 118 consecutive patients to further clarify the effectiveness of capitonnage in managing pulmonary hydatid disease.

2. Methods

2.1. Study design

This study is a multi-centered retrospective case series.

2.2. Setting

Between February 2017 and December 2019, 118 patients were collected in Al-Jumhoory Teaching Hospital, Al-Khansaa Teaching Hospital, and Al-Rabee private hospital in Mosul, Iraq.

2.3. Eligibility criteria

This study included pulmonary hydatid cyst patients of all ages who were surgically managed via the capitonnage method. Patients with co-morbid diseases like acute myocardial infraction and severe diabetes mellitus (where surgical intervention was prolonged) were excluded.

2.4. Diagnostic workup

Clinical workup included history of petting dogs, chest x-ray (showing rounded homogeneous opacity in intact cysts and water lily sign present in perforated cysts), computed tomography scan in special cases, indirect hemagglutination assay (IHA) for echinococcosis. Additionally, complete blood count (CBC), blood sugar, liver and renal function tests were done.

2.5. Surgical technique

All the patients were carefully assessed preoperatively. Routine blood tests, chest radiograph, electrocardiogram, and random blood sugar was done. Blood transfusion and nutritional supplements were given when required. Prophylactic antibiotic was started at the time of induction of anesthesia and continued postoperatively for 10 days. During this procedure, the patient was put on a pulse oximeter, a non-invasive blood pressure monitor, and a good intravenous line. After induction of anesthesia, single lung ventilation was maintained through a double-lumen endobronchial tube and, in some cases, single-lumen. A bi-channel foley urinary catheter was introduced, and the patient was positioned on a lateral decubitus position. Standard muscle-sparing posterolateral thoracotomy was done through the 5th or 6th ribbed. The lung was freed from any pleural adhesions. The cyst was covered with a water-soaked mob to prevent implantation of daughter cysts in the event of rupture. The adventitia was incised carefully to avoid perforation of the cyst. The incision was enlarged in two directions using blunt scissors. The edge of the opening in pericyst was grasped with two fine forceps placed next to each other. The opening of the pericyst was enlarged to its maximum. When the cyst was exposed from one-third to one-half in that way, the hand was gently pushed beneath the cyst, and the remaining part was separated from the pericyst by careful finger dissection until the cyst lied in the palm of the hand. It was then placed in a basin. The enucleation of the cyst was not easy when the cyst was large and under tension. In such cases, the cystic fluid was aspirated with a large-sized needle, then the cyst was opened with a 1 cm incision, and a suction tip was entered into the cystic cavity to evacuate the remaining fluid. The germinal membrane was removed with forceps. After flushing the cystic cavity with saline solution, the cavity was obliterated by capitonnage, a suture approximation of the pericystic tissue. Pulmonary resection, like lobectomy and segmentectomy, was not needed in our patients, which may be required if the cyst had occupied the whole lobe or had destroyed the adjacent lung. Decortication was performed in patients with pleural complications. In all patients, one chest drainage catheter was positioned posteriorly or anteriorly in respective order and was connected with an underwater seal drainage system. Patients were given Albendazole in doses of l0 mg/kg body weight as a postoperative prophylactic to prevent recurrence. Postoperative patients were managed accordingly, and operative outcomes were observed for a few months. Patients with satisfactory outcomes were discharged on the 1st - 7th postoperative day, and stitches were removed after 10 days.

2.7. Follow-up

All patients were followed up for three months at monthly intervals and after one year. In every follow-up, the patient was evaluated clinically and radiologically.

2.7. Data collection and analysis

The data was extracted from the database of the center. Microsoft Excel 2019 was used for the arrangement of the data. The Statistical Package for the Social Sciences (SPSS) Version 25 was used for coding and analyzing the data.

3. Results

Among the 118 cases, 73 (61.9%) were male and 45 (38.1%) were female. Eight (6.8%) patients were between 3-5 years of age (pre-school), 42 (35.6%) were 6-12 years (school age), 15

Table 1. Air-leakage and hospital stay of patients with ruptured and non-ruptured cysts

Air-leakage and hospital stay of ruptured and non-ruptured cysts										
	Air-leakage		Hospital stays per day							
			1 day n (%)		2 days n (%)		3 days n (%)		4 days n (%)	
Ruptured n (%)	6	5%	69	58.4%	9	7.6%	0	0	1	0.8%
Non-ruptured n (%)	4	3.4%	36	30.5%	6	5%	1	0.8%	0	0

(12.7%) were 13-18 years (adolescent), 40 were (33.9%) 19-40 years (young adult), 12 (10.1%) 41-65 years (middle age), and one patient (0.8%) was above 65 years (old adult). The mean age was 30 years, ranging from 3 to 65 years. (45 patients) (38.1%) were asymptomatic, and 73 (61.9%) of them were symptomatic (chest pain, cough, and\or respiratory distress). Forty-nine patients (41.5%) had right lung involvement, 55 (46.6%) had left lung involvement, and the rest 14 (11.9%) with both lung involvement. Other organs were involved in 27 patients, 22 in the liver, 3 in the spleen, and 2 in both.

Intact cysts were extracted from 49 (41.5%) patients, and the remaining 69 (58.5%) exhibited ruptured cysts, 6 patients of the latter group presented with pneumothorax. During surgery, the size of cysts was different; 18 (15%) were 5-10 cm, 11 (9.3%) were more than 10 cm, and 7 (5.9%) were presented with giant cysts.

All patients with different presentations underwent surgery with enucleation of the cysts and capitonnage. Out of 69 patients of ruptured hydatid cysts 6 patients (5%) developed prolonged air leakage, 69 (58.4%) had to stay in the hospital for one day, 9 (7.6%) for two days, and one (0.8%) for four days. Moreover, out of 49 patients (of non-ruptured hydatid cysts), 4 (3.4%) developed prolonged air leakage, 36 (30.5%) had to stay in the hospital for one day, 6 (5%) for two days, and 1 (0.8%) for three days (Table 1) (Figure 1).

The majority of patients 86 (72.9%) had their chest tubes removed on the third postoperative day, 17 (14.4%) patients on the fifth day, 8 (6.8%) on the fourth day, 4 (3.4%) on the second day, and 3 (2.5%) on the seventh day (Table 2).

Table 2. Chest tube removal.

Postoperative day	N. patients (%)
2nd day	4 (3.4%)
3rd day	86 (72.9%)
4th day	8 (6.8%)
5th day	17 (14.4%)
6th day	0 (0.0%)
7th day	3 (2.5%)

Regarding the development of postoperative complications, there were ten patients with air leakage, eight with atelectasis, three with pneumothorax, three with wound infection, two with hemoptysis, and one patient had a collapsed lung (Table 3).

4. Discussion

Echinococcosis, also known as hydatid disease, is a parasitic infection caused by larvae, which is the metacestode stage of the tapeworm. Four species are recognized and belong to the family *Taeniidae*; *Echinococcus granulosus* is responsible for the vast majority of human infestations. It is the causative agent of cystic echinococcosis, which has a global distribution and is most prevalent in sheep-raising areas [4].

Table 3. Early postoperative complications.

Characteristic	Frequency
Air leak in ruptured cyst	6
Air leak in non-ruptured cyst	4
Atelectasis	8
Rim of pneumothorax	3
Wound infection	3
Hemoptysis	2
Collapsed lung	1

Hydatid disease can affect people of any age or sex, but it is more common in people between the ages of 20 and 40 [4]. The mean age group in the current study was 30, which is consistent with the findings of other studies. Turna et al., reviewed 75 patients and showed an average age of 30.2 ± 17.4 years [2]. The male predominance of hydatid cysts has been documented. This male predominance was observed in nearly all lung hydatid cysts and may be explained by boys' earlier and frequent contact with dogs [7]. In this study, there were 73 (61.9%) males and 45 (38.1%) females, as opposed to the study by Goni MO et al., in which 65% of the patients were female and 35% were male. While Ahsan et al., reviewed 137 patients, they discovered that male incidence was 57% and female incidence was 43% [8].

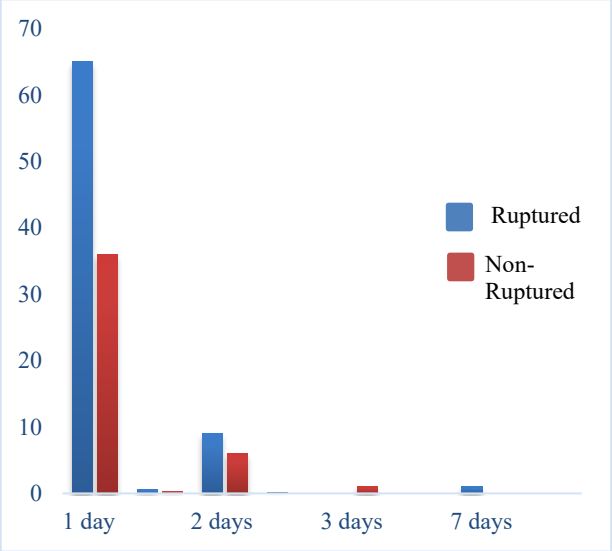

Figure 1. Hospital stays of patients with ruptured and non-raptured cysts

Pulmonary hydatid cysts can occur in any pulmonary lobe, but the right and lower lobes are the most commonly affected lobes; 60% of cases are in the lower lobes of the right lung, 30% have multiple pulmonary cysts, and 20% have bilateral cysts [9, 4]. In The current study, the right lung was affected in 39.8% of the cases, the left lung was involved in 44.9%, and both (right and left) lungs were involved in 11.8% of the patients. The majority of patients are asymptomatic; however, some may occasionally expectorate cyst contents or develop symptoms associated with compression of the surrounding structures [10]. In our study, more than one-third of the patients (38.1%) were asymptomatic, while 73 (61.9%) had common symptoms of echinococcosis, such as chest pain, cough, and\or respiratory distress.

Clinical findings, chest radiographs, thoracic CT scans, and echinococcus serologic studies all help to confirm the correct preoperative diagnosis of pulmonary hydatid disease [2]. On a chest X-ray, an uncomplicated hydatid cyst appears as a well-defined homogenous radio-opacity; a water lily or signet ring sign is a characteristic of a perforated cyst. On CT scan, pulmonary hydatid cyst can be identified by the inverted crescent sign, signet ring sign, high CT density, and thick wall [5].

The definitive treatment of pulmonary hydatid cysts is complete excision of the cyst. Various surgical procedures have been described in the literature, such as resection by means of enucleation without needle aspiration, removal of the intact cyst after needle aspiration, pericystectomy (Perez-Fontana), wedge resection, segmentectomy, and lobectomy [9]. In this study, all patients with different presentations underwent surgery with enucleation of the cysts and capitonnage.

Regarding the residual cavity management study, all the patients had the cyst enucleated with closure of the bronchial opening capitonnage. Almost the same group of surgeons carried out the operations. Nearly all of the patients were cured immediately, without any mortality. In the Goni MO et al., study, 53.4% of the cysts were enucleated with closure of the bronchial opening without capitonnage, while the remaining 46.6% were enucleated with capitonnage [8]. A French surgeon first described capitonnage, the suturing of the pericystic cavity, as a surgical technique in intact or complicated pulmonary hydatid disease [9].

Many published authors (Kuzucu et al., 2003, Hacubrachimoglu et al., 2003, and Kanat et al., 2004) favored the capitonnage method, in which the residual cavity was obliterated with separate purse string sutures placed into the cavity from the deepest level to the surface, as it reduced postoperative chest tube drainage time and morbidity [1, 11, 12]. Similarly, Sonmez and colleagues concluded that capitonnage reduces morbidity rates during the postoperative chest tube drainage period [9]. While the obliteration of the post-cyst resection cavity (capitonnage) is controversial, according to Halezeroglu and associates [13].

Kosar et al., cleared that capitonnage provides a complete cavity closure and acts as a second barrier against prolonged air leak, and reported a 5.4% frequency of prolonged air leakage in the capitonnage group, while the non-capitonnage group had a frequency of 4 (30.4%). As a result, many reports have advocated using capitonnage as part of the procedure to reduce the risk of prolonged air leak, the development of empyema and residual space, and, thus, reduced hospital stay [9].

Turna et al. reported 71 patients, 39 of whom underwent enucleation of the cyst with capitonnage, and five developed air leakage [2]. In this series, we had 49 (41.5%) intact cysts and 69 (58.5%) ruptured cysts, with air leakage occurring in 5% of ruptured cysts and 3.4% of non-ruptured cysts. Chest tubes were removed from day 2 to day 7 postoperatively, with the majority (86) being removed on the 3rd postoperatively.

A study by Goni MO et al. showed that air leakage was more common in patients who had cyst enucleation with capitonnage. They argued against capitonnage because they believed that it provided no benefit and increased complications, such as atelectasis and distortion, only it can be omitted to shorten the operative time [8]. Likewise, Ahsan et al. and Turna et al. reported that the best operative method is cyst enucleation without capitonnage [2, 8]. According to Turna et al., it is not required to approximate and suture the cavity edges because the pulmonary parenchyma obliterates the space. The surface of the lung at the site of cavity is covered by pleura. Without capitonnage, the wall of the pericystic cavity is expected to be covered by epithelial cells for an unknown period [2]. Yaldiz et al. employed capitonnage in 271 patients (92.2%); atelectasis was found in only four of them (1.3%) [14]. In our series, eight patients developed atelectasis that resolved within two weeks. Our strategies were asking the anesthetists to hyperinflate the lung after capitonnage to minimize atelectasis.

According to Eren et al., the cystic cavity completely disappeared on radiography two weeks after the operation without capitonnage due to the ability of the lung to expand [15]. Similarly, Turna et al. showed no significant difference in postoperative chest radiographs between patients with and without capitonnage [2]. While, Yaldiz et al. revealed that the most common complications without capitonnage were lung

collapse, empyema, repeat thoracotomy, and sepsis. They also reported that radiological improvement took more than two weeks, similar to chest radiographs of patients undergoing capitonnage in the early postoperative period. Following that, radiological follow-up after 2 months revealed normal radiological findings or a thin fibrotic band, indicating that the cavity had been completely obliterated [14].

5. Conclusion

Surgical treatment for pulmonary hydatidosis is effective in both ruptured and intact cysts. Capitonnage for residual cavity obliteration is commonly recommended after the removal of a hydatid cyst. If the capitonnage technique is applied carefully on the foldings of pericystic tissue, it results in a very low rate of atelectasis and distortion.

Declarations

Conflicts of interest: The author(s) have no conflicts of interest to disclose.

Ethical approval: Not applicable.

Patient consent (participation and publication)**:** Consent has been taken from the patients and the family of the patients.

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Authors' contributions: OFA, OMH and MHN were the surgeons who performed the operation. BAA were RQS were a major contributor to the conception of the study, as well as in the literature search for related studies. DMH and FHK were involved in the literature review, the writing of the manuscript, and data analysis and interpretation. SHK and BHA were involved in the literature review, the design of the study, revision of the manuscript and in the processing of the figures. SHM and FM confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Data availability statement: All data and materials are kept by the first and corresponding authors.

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