Cystectomy With or Without Capitonnage: Analysis of Outcome of 73 Cases in the Management of Hydatid Cyst of Lung

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ABSTRACT

Introduction: Sporadic cases of lung hydatid cyst is common in India apart from its endemicity in Mediterranean regions and other rural areas of developing countries. The patients of Hydatid cyst (HC) of lungs are often diagnosed incidentally. The definitive treatment of such lesions is surgery. Lung preserving surgery in such cases is generally the preferred technique unless there is considerable destruction of pulmonary parenchyma. Cystotomy and capitonnage are the two preferred techniques and generally followed. In this retrospective observational study it was attempted to analyze outcomes of cystotomy and capitonnage with regard to postoperative morbidity and mortality.

Material and Methods: Between 1st May 2014 and 31st May 2019, a total of 73 patients with lung Hydatid cyst were randomly selected from all the hydatid cyst cases operated in a tertiary centre and the results and outcome analyzed. Out of 73 cases 39 (Group A) were operated by Cystotomy and closure of bronchial opening and 34 (Group B) were operated with Cystotomy, closure of bronchial opening and Capitonnage.

Results: The mean chest tube removal time for Group A and B was 5.94 ± 3.17 and 3.41 ± 2.61 days respectively. The mean hospital stay was 10.02 ± 4.53 days for group A and 7.55 ± 2.66 days for group B. Prolonged air leak (>7days) was found in 15 patients in group A, 14 of which required prolonged chest tube drainage and 1 required surgical closure. 4 patients in group B had prolonged air leak and all closed with prolonged chest tube drainage. No abscess formation or mortality was observed in any group in this study. There was a significant difference between group A and group B with regard to chest tube removal time (P=0.000836), hospital stay (P = 0.047585), and all complications (P= 0.008214).

Conclusion: In surgical management of pulmonary Hydatid cyst, capitonnage is superior to cystotomy without capitonnage because it reduces morbidity and hospital stay.

Keywords: Pulmonary Hydatid Cyst, Capitonnage, Cystectomy

INTRODUCTION

Pulmonary hydatidosis is a zoonotic infection of world – wide distribution, caused commonly by the larval stages of tapeworm Echinococcus granulosus. This is endemic in the Indian subcontinent and is prevalent where livestock is raised in association with dogs. Hydatid cysts (HC) are most common in the liver (65%) as it is the site of first filtration of the hexacanth embryo of Echinococcus granulosus. Pulmonary HC’s are found in 25% of cases¹,² and are caused by Echinococcus granulosus. Infestation with Echinococcus multilocularis is also not uncommon in the Indian subcontinent. Alveolar echinococcosis caused by Echinococcus multilocularis usually occurs in a wildlife cycle between foxes, other carnivores and small mammals (mostly rodents). Canines, most commonly the dogs are definitive hosts, which harbour the mature tapeworm in their intestine while sheep and cattle are intermediate hosts. Human beings are accidentally infected by ingesting the parasitic ova from excreta of the infected carnivores in contaminated food and water, and the parasite then develops into larval stages in the viscera. This is endemic in sheep rearing areas of various developing countries and is a major surgical problem.³ The disease often remains asymptomatic or diagnosed accidentally after Chest X-ray examination for unrelated problems. Symptoms may arise when cyst increases in size and erodes into bronchus, compresses surrounding structures, gets infected or ruptures.⁴ Chest X-ray of unruptured Hydatid cyst of lungs may mimic malignancy or tuberculosis. Various radiological signs like crescent sign, inverse crescent sign, water lily sign or camalote sign occurs because of collapse of endocyst and partial evacuation of its fluid. High resolution computerized tomograms (HRCT) is very helpful in locating the lesions in lungs and other organs like pericardium and heart⁵–⁶ and liver and other intra abdominal organs. The detection of Immunoglobulin G (IgG) is more specific and sensitive for confirming diagnosis hydatidosis.⁷ Surgical intervention is the treatment of choice though pharmacotherapy may also be useful in selected patients with poor surgical risk, refusal for surgery and multiorgan disease, patients with contraindication for surgery, multiple cysts, recurrent cysts, and patients with intraoperative spillage of hydatid fluid. Medical therapy of pulmonary hydatid cyst includes benzimidazoles group of drugs, for example, mebendazole (MBZ) or albendazole (ABZ). ABZ

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is preferred, as it has better bioavailability, can be used with lower doses and is more effective than MBZ as shown by Sarkar et al. Albendazole achieves a higher plasma concentration and intracystic drug penetration and its plasma concentration is 10–40 times higher than that of MBZ. However, ABZ requires a minimum contact period of 11 days to have a significant response. The usual recommended dosage of ABZ is 10–15 mg/kg/day, taken in two divided dose daily and that of MBZ is 40–50 mg/kg/day daily. Fat rich meals increase the bioavailability of the drugs. The optimal duration of pharmacotherapy in pulmonary hydatidosis is not known, but it should be given for a period of 6 months and for longer periods in those with inoperable diseases or contraindications to surgery. Current study aimed to analyze outcomes of cystotomy and capitonnage with regard to postoperative morbidity and mortality.

**MATERIAL AND METHODS**

This was a retrospective review of 73 patients with pulmonary hydatid cysts, documented and treated by us in Medical College Hospital, Kolkata, from 1st May 2014 to 31st May 2019. Our patient population as retrieved from the records, were placed into two groups based on the type of surgery performed. Group (A) consists of 39 cases that were treated with cystotomy, closure of bronchial opening and group (B) had 34 cases, which were treated with cystotomy, closure of bronchial opening and capitonnage. The base line information about the demographic profile of the patients, side of HC, state of HC (Ruptured or intact), presenting complaints are noted in Table 1. Type of operation performed, complications, morbidity and mortality are noted in Table-2. This retrospective study was approved by the ethical committee of our institution. The surgical procedures performed using different methods were in accordance with the routine standard of care being followed in our institution during 2014-2019.

**Exclusion criteria:** Multiple hydatid cysts in lungs and concomitant hydatid cysts in heart, pericardium and of liver were excluded from this study.

**Operation**

All procedures were performed under general anesthesia with double-lumen endotracheal tube and single-lumen endotracheal tube for the few younger patients included in our study. The surgical approach was posterolateral or lateral thoracotomy in all the patients depending on the cyst location of the cyst. Serratus anterior was preserved in all the cases. The surgical procedure was cystotomy, removal of germinal epithelium and closure of bronchial opening in 39 patients in group A and cystotomy, removal of germinal epithelium, closure of bronchial opening and capitonnage in 34 patients in group B. When the hydatid cyst was identified, the surgical wound and adjacent tissue were covered with gauges soaked in 10% povidone-iodine so that only the area of the lung containing the cyst was exposed. Cystectomy, that is, excision of the entire intact cyst by enucleation using and closure of bronchial openings were done after identifying by irrigating with hypertonic saline while the anesthesiologist inflated the lung. After removal of intact cyst or ruptured infected cyst, the cystic cavity was thoroughly washed again with saline and 10% povidone iodine and all debris removed. Closure of the small airway openings was done by 4-0 poly dioxanone (PDS) sutures and sometimes with polypropylene sutures with or without pledges according to the bronchial opening size and the surrounding tissue, and then Capitonnage was performed to obliterate the cyst space by imbricating sutures from within the cysts beginning from the depth and gradually proceeding up by using preferably 4-0 PDS sutures. Decortications were performed when required. Chest was closed with apical and basal drains. Albendazole was given in a dose of 10-15 mg/kg/day for 6 months postoperatively in all the patients. Liver function tests were checked at three monthly follow-up. Investigations were made to evaluate the parenchyma and lung functions, preoperatively and at one-year follow-up postoperatively.

**STATISTICAL ANALYSIS**

Data were checked for normality before statistical analysis. Variables were expressed in mean with standard deviation. Differences between study groups were considered statistically significant when p was less than or equal to 0.05.

**RESULTS**

In group A, 25 (64.1%) patients were male and 14 (35.89%) were female and in Group B males and females were 23 (67.6%) and 11 (32.5%) respectively; with mean age of 31.05 ± 11.76 years and 30.11 ± 11.43 years respectively (p= 0.94467). 34 (87.17%) patient presented with intact cyst(s) in group A and 31 (91.17%) in group B; and the others were ruptured cysts. Conservative surgical techniques, such as cystectomy plus closure of bronchial openings and Capitonnage of the residual cystic space, constituted the surgical approach in 34 in Group B. For intact cysts, cystectomy (enucleation) was the operative procedure in 39 patients in Group A. The presentations of the patients are...
### Table-1:

|                  | Group A          | Group B          | P value |
|------------------|------------------|------------------|---------|
|                  | Frequency        | Percentage       | Frequency | Percentage |       |
| **Age**          |                  |                  |          |            |       |
| <20              | 8                | 20.51282         | 9        | 26.47059   | 0.94467 |
| 21-30            | 15               | 38.46154         | 12       | 35.29412   |        |
| 31-40            | 9                | 23.07692         | 7        | 20.58824   |        |
| >40              | 7                | 17.94872         | 6        | 17.64706   |        |
| **Mean ±SD**     | 31.05128 ± 11.76066 | 30.11765 ± 11.43563 |       |
| **Gender**       |                  |                  |          |            | 0.750224 |
| Male             | Male             | 25               | 64.10256 | 23         |        |
| Female           | Female           | 14               | 35.89744 | 11         |        |
| **Presentation** |                  |                  |          |            |       |
| Dyspnoea         | 10               | 25.64103         | 9        | 26.47059   | 0.93871 |
| Chest Pain       | 3                | 7.692308         | 2        | 5.882353   |        |
| Incidental       | 16               | 41.02564         | 12       | 35.29412   |        |
| Empyema          | 6                | 15.38462         | 5        | 14.70588   |        |
| Pneumothorax     | 2                | 5.128205         | 2        | 5.882353   |        |
| Pleural Effusion | 2                | 5.128205         | 3        | 8.823529   |        |
| Tension Pneumothorax | 0              | 0                | 1        | 2.941176   |        |
| **Side of HC**   |                  |                  |          |            |        |
| Left             | 21               | 53.84615         | 19       | 55.88235   | 0.861576 |
| Right            | 18               | 46.15385         | 15       | 44.11765   |        |
| **State of Hydatid Cyst** |      |                  |          |            |        |
| Intact           | 34               | 87.17949         | 31       | 91.17647   | 0.585523 |
| Ruptured         | 5                | 12.82031         | 3        | 8.823529   |        |

### Table-2:

|                  | Group A          | Group B          | P value |
|------------------|------------------|------------------|---------|
|                  | Frequency        | Percentage       | Frequency | Percentage |       |
| **Hospital stay**|                  |                  |          |            |       |
| <8 days          | 19               | 48.71795         | 22       | 64.7059    | 0.047585 |
| 8-11 days        | 8                | 20.51282         | 10       | 29.4118    | 0.294118 |
| 12-14 days       | 7                | 17.94872         | 2        | 0.58824    |        |
| >14 days         | 5                | 12.82051         | 0        | 0          |        |
| **Mean ±SD**     | 10.02564 ± 4.533427 | 7.558824 ± 2.665051 |       |
| **Complications**|                  |                  |          |            |       |
| Air leak         | 15               | 38.46154         | 4        | 11.76471   | 0.008214 |
| Atelectasis      | 6                | 15.38462         | 2        | 5.882353   |        |
| Re-operation     | 1                | 2.564103         | 0        | 0          |        |
| No complications | 17               | 43.58974         | 28       | 82.35294   |        |

Postoperative complications were infrequent and no mortality was seen. Prolonged parenchymal air leak (>7 days) was observed in 15 patients (38.46%) in Group A, one(1) of them had broncho-pleural Fistula (BPF) for which surgical closure was required; rest closed spontaneously. In Group B, only 4 patients (11.76%) had prolonged air leak and all closed spontaneously. The overall complication rate was higher in Group A (p=0.008214). The hospital stay and chest tube removal time were found to be significantly longer in group A (in which no capitonnage was applied) compared with those in group B (in which capitonnage was done). Mean hospital stay in Group A and B were 10.02 ± 4.53 days and 7.55 ± 2.66 days respectively with p values of 0.047585. The mean chest tube removal was done in 5.94 ± 3.17 days in Group A and 3.41 ± 2.60 days in Group B (p= 0.000836). Furthermore, the overall complications occurring in the postoperative period, including prolonged air leak, were higher in group A than in group B. The higher complication rate was statistically significant (P = 0.008214). We consider all complications, especially prolonged air leak, as factors prolonging the chest tube removal time and hospital stay. Air leak was managed by chest physiotherapy and chest tube drainage. Atelectasis developed in 6 cases (15.38%) in Group A and 2 cases (5.88%) in Group B, all resolved after few days of chest physiotherapy.

**DISCUSSION**

The primary cause of Hydatid Cyst in India is E. granulosus and rarely E. multilocularis. E. granulosus conspicuously forms single cyst while E. multilocularis forms multiple
daughter and granddaughter cysts. After ingestion of infective ova through drinking of contaminated water or food the hexacanth embryo enters portal circulation and lodges in liver acting as the first filter. Embryos escaping liver lodges in lungs acting as the second filter. Lung may also get affected when embryos bypass liver through lymphatic channels. Liver is the most frequently affected organ (65%) and lung being the second common organ affected (25%).

Pulmonary Hydatid Cyst requires prompt surgery for its potential complications like rupture into bronchus, pleura or other vital organs or can get compressed and cause symptoms. Most commonly presenting symptoms are cough and haemoptysis.

Lung tissue should be preserved and resection should be avoided irrespective of cyst size. Although we had a parenchyma preserving approach, no recurrence was observed in our series; this was similar to another study. Resection is not recommended unless whole lobe is destroyed. In our practice, conservative surgical techniques, such as cystectomy plus closure of bronchial openings and capitonnage of the residual cystic space, constituted the routine surgical approach. In many cases in our study, a radio-opaque shadow appeared in chest X-ray and chest-CT after the Capitonnage and has also been reported by others; this shadow represents sutured lung tissues; it disappears within 3 weeks to 3 months. Some authors have claimed that capitonnage can cause atelectasis by obliterating the major bronchi surrounding the cyst cavity and present a potential risk of pulmonary parenchymal distortion. However, Erdogan and colleagues have reported that they saw no atelectasis in any of the 44 patients with hydatid cysts who had undergone capitonnage. The main aim of surgery in hydatid cysts is total excision and avoidance of air leaks which was significantly low in group B. Haemoptysis as a complication of capitonnage for management of Pulmonary Hydatid Cyst, as per Kambiz Sheiky et al presence of unabsorbable suture material such as prolene in residual pericyst caused recurrent infection and haemoptysis. However in our study we have not encountered such complication and use of PDS suture should be recommended for closure of bronchus and capitonnage. PDS sutures are delayed absorbable sutures and therefore chances of unabsorbable suture related complications like haemoptysis would be avoided.

Goni MO et al had a different result in their study. They found that capitonnage had no added advantage over removal of germinative layer of Hydatid cyst and closure of Bronchial openings. But Kuzucu et al, Kanat et al recommended capitonnage for its superior results in terms of reduction of hospital stay and air leakage over non-capitonnage group. For prevention of recurrence and minor spillage, which was not uncommon after removal of cyst, we recommend albendazole (10-15 mg/ kg/ day) for its better bioavailability. We started albendazole after diagnosis and continued for six months after surgery with no recurrence. Ozan Usluer et al observed incidence of giant Hydatid cysts (≥ 10 cm) was more common in younger patients and in lower lobes. They also mentioned chance of lobectomy was higher in giant Hydatid cysts even when parenchyma saving operations was performed.

In another study Mohsen Sokouti et al observed capitonnage after cystotomy and closure of air leak in Hydatid cyst of lung significantly reduces post-operative air leak and empyema formation. However, in our study there was no empyema formation in both Groups A and B but capitonnage group had significant reduction of post-operative air leak and hospital stay.

**CONCLUSION**

Both the techniques of lung preserving surgery for pulmonary Hydatid are effective with no recurrence of HC and mortality. But Cystotomy, closure of bronchial openings and capitonnage provides better results in terms of post-operative air leak, early chest tube removal, lesser hospital stay and lower re-intervention rates over cystotomy alone. Bronchial openings, which may get unnoticed during the procedure due to visual obstruction by blood clots and secretion were probably the causative factor for higher incidence of air leak in non-capitonnage group.

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