

A 10-year experience of hydatid cysts of lung in association with other extrathoracic system involvement in a tertiary care hospital in Kolkata, India



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Submission: 25-02-2022

Revision: 02-05-2022

Publication: 01-06-2022

ABSTRACT

Background: The metacystode of tapeworm *Echinococcus* causes hydatid disease. Infection in man is mainly caused by *Echinococcus granulosus*. Surgical intervention to treat complicated hydatid cyst (HC) is different from uncomplicated HC. **Aims and Objectives:** The present study was performed with an aim to review 82 cases of pulmonary HCs and associated HCs in other locations. We analyze and evaluate our experience in the surgical treatment and complication. **Materials and Methods:** In a 10-year period, from August 1, 2005, to July 31, 2015; 82 patients were operated on in our hospital for HCs in pulmonary as well as other associated extrapulmonary locations. The diagnosis was established on the basis of clinical features and imaging studies supplemented by serological test. Surgical approach consisted of a posterolateral thoracotomy in all cases of thoracic HCs. For cysts in extrathoracic locations, either laparotomy or local excisions were performed. **Results:** Radical resections [lobectomy] were done in four cases, and conservative operations in the remaining. Post-operative complications occurred in 12 cases (14.63 %). We had two recurrences, three cases of prolonged air leak with bronchopleural fistula, two had atelectasis, and five wound infections. At 6 months follow-up, 97.56% of our patients were free of any recurrence. **Conclusion:** HCs of the lung are best treated with surgery. Even when associated with HCs in other locations in the body, surgery is considered the treatment of choice. When supplemented with long-term perioperative Albendazole therapy, the chance of recurrence is practically negligible.

Key words: Hydatid cyst; Laparotomy; Lobectomy; Lung

INTRODUCTION

Hydatid cyst (HC) caused by the larval form of *Echinococcus* is a worldwide zoonosis, which is most commonly caused by *Echinococcus granulosus* (EG). The other less common but more aggressive type is caused by *Echinococcus multilocularis*.^{1,2} Although the liver is the most common site for HC, it can involve almost any organ of the body. The lungs are the second most frequent location of hematogenous spread in adults and probably the most common location in children.^{3,4}

While the lung parenchyma is the most common site within the thorax, it may develop in any extrapulmonary region, including the pleural cavity, fissures, mediastinum, heart, vascular structures, chest wall, and diaphragm. Imaging plays a pivotal role not only in the diagnosis of HC but also in the visualization of the extent of involvement and complications.⁵ The metacystode of tapeworm *Echinococcus* causes hydatid disease. Infection in man is mainly caused by EG. This species is common in sheep-rearing areas worldwide. There are three other species – *E. multilocularis*, *Echinococcus vogeli*, and *Echinococcus*

Access this article online

Website:

<http://nepjol.info/index.php/AJMS>

DOI: 10.3126/ajms.v13i6.43431

E-ISSN: 2091-0576

P-ISSN: 2467-9100

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oligarthrus. Of these, *E. multilocularis* is found in cold environment and often associated with wild animals (fox). The treatment of pulmonary HC is surgical and we prefer intact endocystectomy with or without capitonnage (as a parenchyma preserving method). However, if surgical treatment is delayed due to the earlier medical treatment of uncomplicated HC, the cyst may rupture, and suppurate. Complicated pulmonary HCs of this type may resemble many other pulmonary lesions and carcinomas of the lung. A surgical intervention to treat complicated HC is different from uncomplicated HC. In humans, *E. multilocularis* is rare and causes alveolar disease. The remaining two species cause polycystic disease and are extremely rare.⁶

Aims and objectives

The aims of this study were to present our experience in the surgical treatment and complications of pulmonary HCs and associated HCs in other locations in our institution for 10 years.

MATERIALS AND METHODS

In the present interventional study, 82 patients underwent surgery in our department for parenchymal HCs for the duration from August 1, 2005, to July 31, 2015. Data was collected from the patients and written informed consent was taken at the time of data collection. Some of these patients also had associated extrapulmonary HCs, in addition to their lung hydatid. Ethical approval was taken from the Institutional Ethical Committee.

Pre-operative chest radiography and computed tomographic scanning were performed on all patients, as was ultrasonography of the abdomen to exclude HCs elsewhere in abdomen (particularly liver). Percutaneous transthoracic needle aspiration was avoided for diagnosis or treatment. In later years of the study (2011 onward), enzyme-linked immunosorbent assay (ELISA) to detect immunoglobulin (Ig)G echinococcal antibody in serum was performed to supplement clinical and radiological diagnosis. Diagnosis was confirmed in every case by histopathological study of the resected specimen.

All patients underwent a posterolateral thoracotomy for lung HCs. Surgical options for lung hydatid included lobectomy, wedge resection, pericystectomy, as well as intact endocystectomy with and without capitonnage. In the event of bilateral lung HCs, surgery was performed first on the larger cyst followed by the other at a second stage 3 weeks after the first operation. In cases of hydatid cyst elsewhere, the site was addressed 6 weeks after the first operation. Intraoperatively, 10% Betadine was used as scolicidal agent for protection of surrounding structures.

All our patients were started with albendazole 10 mg/kg/day in two divided doses after diagnosis. The same dose was continued till 6 months after operation.

Statistical analysis

The recorded data were compiled and entered in a spreadsheet computer program (Microsoft Excel 2007) and then exported to data editor page of SPSS version 15 (SPSS Inc., Chicago, Illinois, USA). For all tests, confidence level and level of significance were set at 95% and 5%, respectively.

RESULTS

In our patient population, 48 were male and 34 were female with a mean age of 30.26 years, ranging from 10 to 48 years. Most patients belonged to a lower socioeconomic background [80.25%]. The clinical presentation, as summarized in Table 1, shows dry cough [96.34%] and chest discomfort [48.78%] as the main presenting symptoms. Only five patients were asymptomatic. Serological test with ELISA for IgG antibody to echinococcal antigen was done in 30 cases and was positive in 28 cases [93.33%]. The locations of hydatid cysts in our patients, as in Table 2, show that unilateral lung hydatid was the most common [n=71; 86.59%], followed by bilateral lung hydatid [n=11; 13.41%]. The most common extrathoracic location of hydatid cysts was liver [n=9; 10.98%]. Surgical options for lung hydatid included lobectomy, wedge resection, pericystectomy, and intact endocystectomy with and without capitonnage, as shown in Table 3.

Post-operative complications occurred in 12 cases, as shown in Table 4. The most common complication was wound infection in five cases [6.09%], followed by persistent air leak with bronchopleural fistula in three cases [3.66%]. We did not have any mortality.

DISCUSSION

Although commonly found in the 2nd–4th decades of life, hydatid disease of lung can occur in individuals of any age and gender.^{6,7} For the hydatid cyst to occur in lung, the

Table 1: Clinical presentations

Symptoms	Number of cases (n=82)	Percentages
Asymptomatic	5	6.10
Chest discomfort	40	48.78
Dry cough	79	96.34
Expectoration	25	30.49
Hemoptysis	7	8.54
Vomiting	6	7.32
Fever	25	30.49
Dyspnea	10	12.20

Table 2: Various locations of hydatid cysts and their surgical approaches

Locations of hydatid cysts	Surgical approaches	Numbers (n=82)	Percentages
Unilateral lung parenchymal cysts	Posterolateral thoracotomy	71	86.59
Bilateral lung parenchymal cysts	Posterolateral thoracotomy	11	13.41
Associated liver hydatid cysts when the lung cyst was in the left lung	Laparotomy in a separate sitting	6	7.32
Associated liver hydatid cysts when the lung cyst was in the right lung	Liver hydatid removed transdiaphragmatically in the same sitting	3	3.66
Associated splenic hydatid cyst	Laparotomy	1	1.22
Associated pericardial hydatid cyst	Left Posterolateral thoracotomy	1	1.22
Associated parotid hydatid cyst	Superficial parotidectomy	1	1.22
Associated right mammary hydatid cyst (as in Figure 1)	Local excision	1	1.22
Associated chest wall hydatid cyst (as in Figure 2)	Local excision	1	1.22

Table 3: Different types of surgeries performed for lung hydatid cysts

Types of surgeries for lung hydatid	Numbers	Percentages
Lobectomy	4	4.30
Intact endocystectomy with closure of bronchial openings	81	87.10
Intact endocystectomy with capitonnage	8	8.60
Total number of lung cyts addressed=93 (number of patients being 82)		

larvae have to pass the hepatic sinusoids. However, they can also reach lung through lymphatics.^{8,9}

The annual growth rate of a lung HC could be as much as 1–5 cm.¹⁰ HCs mostly involve one organ (85–90% cases). The most common organ being liver, followed by lung. In more than 70% cases, only a single cyst is found. Other sites of affection are spleen, kidneys, orbit, heart, brain, and bone.⁸⁻¹¹ In about 20–40% cases of lung hydatid, liver cysts may coexist.^{12,13} Talking of lung echinococcal cysts, interestingly, 60% occur in right lung, while 20% are found bilaterally. Multiple cysts are found in 30% cases. Again lower lobes of lung harbor the cysts in 60% of the patients.¹⁴

Although an intact lung HC can sometimes cause cough, hemoptysis, or chest pain, mostly they are incidental finding on a chest radiograph.¹⁵ Cyst may rupture as a result of secondary infection, trauma, or even spontaneously and cause symptoms. This rupture may be a communicating rupture, where the contents escape through the bronchioles incorporated in the pericyst and reach the airway. The cyst may directly rupture into the pleura. The rupture may also be a contained one when the endocyst ruptures but the pericyst is intact.¹⁶ When rupture happens, it may

be associated with sudden onset cough and fever. There may also be expectoration of clear HC fluid along with membranes and scolices. It tastes salty or peppery.

HC of heart is a rarity (0.5–2% cases). They are mostly asymptomatic, but sometimes cause chest pain. The most common location of cysts in heart is the left ventricle. Cysts have also been reported in inter ventricular septum and right ventricle. Pericardial and right atrial cysts are extremely rare.^{17,18}

A chest radiograph is invaluable in diagnosing lung HC.^{10,16-20} Unruptured cysts appear as smooth bordered, homogeneous round, or oval masses surrounded by normal lung parenchyma.²¹ Size may be as big as 20 cm.²²⁻²⁴ Contrast-computed tomography (CT) shows a thin enhancing rim with homogeneous density (like water) if cyst is intact.²² Sensitivity of ELISA for IgG echinococcal antibody is about 50% of lung HC and 90% of liver hydatid. Therefore, it is an important screening tool.²⁵

Studies have been conducted to compare perioperative morbidity and complications between open approach and video-assisted thoracoscopic surgery with or without mini thoracotomy.^{26,27} We operated all cases through open thoracotomy. The closure of bronchial openings and management of the residual cavity is important to prevent prolonged post-operative air leak and empyema formation.²⁸ Nabi et al. proposed it is difficult to close the bronchial openings in ruptured or infected cysts and found all of them had post-operative prolonged air leak due to cut-through of sutures leading to empyema formation. The addition of capitonnage had the advantage of securing unidentified air leaks during surgery.²⁹

Surgery, whenever feasible is treatment of choice ensures complete removal of cyst with the parasites. Lobectomy, wedge resection, pericystectomy, intact endocystectomy, and capitonnage are described as surgical options in lung

Table 4: Post-operative complications and treatment offered

Post-operative complications	Numbers (n=82)	Percentages	Treatment offered
Atelectasis which resolved with chest physiotherapy	2	2.44	Antibiotics
Wound infection	5	6.10	Antibiotics
Persistent air leak with bronchopleural fistula	3	3.66	Reintervention [Lobectomy 1 case]. Other 2 healed on conservative management
Recurrence following intraoperative spillage in an attempt at intact endocystectomy	2	2.44	Right middle lobectomy, 6 months after the first surgery in both cases



Figure 1: Infected hydatid cyst of the right mammary (cyst wall can be seen being extracted)



Figure 2: Hydatid cyst of the left chest wall (red arrow)

hydatid.³⁰ Unlike liver hydatid, percutaneous aspiration, instillation of scolical agent, and resection are rarely done for lung echinococcal cysts.^{31,32} Albendazole administered pre- and postoperatively inactivates protoscolices and reduces incidence of recurrence.^{33,34}

Limitations of the study

As the present study was performed in limited sample size, we cannot generalize the results of current study to the whole population.

CONCLUSION

Lung HC, irrespective of its association with HCs elsewhere in the body, is a curable disease. Surgery, when combined with pre- and post-operative Albendazole therapy cures the disease completely with minimal complications or recurrence.

ACKNOWLEDGMENT

We thank all the individuals included in this study.

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SSG- Concept and design of the study, prepared first draft of manuscript; **UC-** Interpreted the results, reviewed the literature, and manuscript preparation; **SD-** Concept, coordination, preparation of manuscript; **MO-** statistical analysis and interpretation.

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Source of Support: Nil, **Conflict of Interest:** None declared.