

# Management Strategies of Pulmonary Hydatid Cyst - A Prospective Observational Study

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## ABSTRACT

**Introduction:** Cystic echinococcosis better known as “hydatid cyst disease”, is caused by the helminth *Echinococcus granulosus*. It has a worldwide distribution and is extremely common in the Indian Subcontinent. Lung is the second most common organ of involvement after liver. In the present study we aimed to clinically evaluate this disease and understand the various treatment modalities available for it.

**Material and methods:** It was a prospective observational study and statistical analysis was done wherever applicable. Present study comprised of 25 patients who attended with pulmonary hydatidosis at our hospital during a span of 18 months.

**Results:** The most common symptoms were cough and chest pain. The mean duration of illness (in years) was  $2.4 \pm 0.91$  (Mean  $\pm$  SD). Computed tomography (CT) scan was done in all patients for confirming the diagnosis. Sixty percent had hydatid cysts located in their right lung and had involvement of the lower lobe. Excision of cyst was done in 20 patients. Lung resection either in the form of segmentectomy or lobectomy was required in 32%. Intercostal chest drains were placed in all the patients during operation. In 10 patients the cumulative daily drainage amount ranged between  $>75$ -100 ml. The difference in proportion of patients having  $<75\%$  collection in drain was not statistically significant. The intercostal chest drains were removed between the 5th -10th postoperative days in 15 patients. As sample size in each individual group was less than 5, Shapiro-Wilk test was applied to assess the distribution pattern of the observation. The P value  $<0.05$  observed in patients without lung resection was statistically significant. The difference in mean time of removal of drain following the different surgical interventions was not statistically significant between the patients who required lung resection and those who did not. The distribution pattern of the observation on duration of hospitalization amongst the patients undergoing different surgical procedures (i.e., those requiring resection of lung & those without any lung resection) were compared. The P value 0.004 observed in patients without lung resection was statistically significant. Patient morbidity and mortality: 16 patients had air leak in their immediate postoperative period, which was the commonest complication. There was no mortality in our study.

**Conclusion:** CT Scan is inevitable for confirmation of diagnosis. Surgery is the treatment modality of choice with pre- and postoperative albendazole therapy. Lung resection may be needed for selected patients and the most common complication following surgery is air leak which can be managed conservatively.

**Keywords:** Hydatid Cyst, Pulmonary Echinococcosis, Thoracic Hydatidosis, *Echinococcus Granulosus*

## INTRODUCTION

Hydatid disease or echinococcosis is a helminthic infestation caused by larvae of the cestode, *Echinococcus*. Of the four recognized species, *E. granulosus* is responsible for most of the infections in humans. It causes cystic echinococcosis which has a worldwide distribution, particularly endemic in regions such as South and Central America, the Mediterranean, the Middle East, sub-Saharan Africa, Australia, New Zealand & Asian countries including China & India. Most cases in the USA and Central Europe occur in immigrants from endemic areas.

Dogs are definitive hosts of the parasite. Humans are accidental hosts and usually get infected by handling an infected dog. The cyst can be found virtually in any organ, but the most frequently involved site is liver. This is followed by lungs, spleen, and kidney. In children, the lungs are often commonly affected.

Hydatid disease may be primary or secondary. Although it is commonly found in patients aged between 20-40 years, yet it may be seen in any individual irrespective of age and sex. Pulmonary echinococcosis is often an incidental finding in a chest radiograph. It may be single or multiple, intact, or ruptured and may involve one or multiple organs. Small cyst is generally asymptomatic, however, enlarged cyst may produce symptoms such as cough, haemoptysis, and chest pain due to compression of adjacent structures. Symptomatic disease is commonly seen in complicated cyst which is defined as one that had ruptured into a bronchus or into the pleural cavity.

Surgery is the treatment of choice for pulmonary hydatid disease and includes pericystectomy, intact endocystectomy with capitonnage as well as lung resection (segmentectomy, lobectomy or rarely pneumonectomy) and decortication. Medical therapy with albendazole has a definitive role in the management of all cases of hydatid diseases.

Study aimed at clinical evaluation of pulmonary hydatid

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disease, diagnosis of pulmonary echinococcosis, i.e., to evaluate the role of – Chest X-ray, CT Scan, & Fiberoptic Bronchoscopy, medical management- to evaluate the role of Albendazole and surgical management in thoracic hydatid cyst- operative details & postoperative management.

## MATERIAL AND METHODS

The present study was a prospective observational study which was carried out at the Department of Cardiothoracic & Vascular Surgery, RG Kar Medical College, Kolkata, during the period January 2014 to June 2015. Twenty-five patients presenting with pulmonary hydatid disease were evaluated. All haemodynamically unstable or any unwilling patients were excluded from this study.

A detailed history including patients' age, sex, body weight, etc. were taken. They were interviewed and clinically examined for any symptoms such as haemoptysis, productive sputum, fever, cough, etc. The role of different investigation modalities such as the routine examination of blood & sputum, chest x-ray, computed tomography, magnetic resonance imaging, fibre-optic bronchoscopy, serology tests like ELISA following were evaluated. Patients were also evaluated on the basis of the available treatment options with detailed clinical notes such as mode of treatment, details of operation done, whether it was an elective or emergency surgery, types of incision made, choice of antibiotic used, drains used or not postoperatively, duration of hospital stay etc.

Standard, appropriate statistical analysis of the observational data (such as history, clinical examination, investigations done, nature of operation/ postoperative details, clinical notes) in the population under study was done. Statistical analysis had been done using the software SPSS Version-20. Approximate statistical technique by using- Shapiro-Wilk Test, Fischer's Exact Test and Mann Whitney U Test were applied to establish the analysis and evaluate the study wherever applicable.

### Life cycle of Echinococcus

The different species of Echinococcus have different geographical distributions and involve different hosts. The definitive host is usually a dog but may be some other carnivore and a variety of species of warm-blooded vertebrates (sheep, cattle, goats, horses, pigs, camels, and humans) are the intermediate host. Humans are accidental hosts and do not play a role in the biological cycle. They may become intermediate hosts through contact with a definitive host (usually a domesticated dog) or ingestion of contaminated water or vegetables. The eggs can also be inhaled causing primary lung disease. The adult worm inhabits the small intestine of the definitive host, is usually 2–7 mm long, is attached to the mucosa by a double row of hooklets contained in its scolex and has at least three proglottids, which contain numerous eggs. The eggs pass out in the faeces of the dog and stick to the animal's fur or to grass. These eggs can survive for at least a year in the outside world, during which time they are widely dispersed. Flies help to spread the eggs, as does the wind. The adult

tapeworms do not make their hosts ill. Intermediate hosts ingest eggs when grazing on contaminated ground. The ovum loses its protective chitinous layer as it is digested in the duodenum. The released hexacanth embryo, or oncosphere, passes through the intestinal wall into the portal circulation and develops into a cyst usually within the liver or lung where they develop into cystic metacestodes. The parasite then grows to form a cyst filled with fluid. The interior of a cyst is filled with protoscolices, each of which can grow into an adult worm when ingested by a canine host. Cysts may contain hundreds or thousands of protoscolices and this tremendous reproductive potential poses a problem in the intermediate host (particularly in humans). Protoscolices can develop into either secondary cysts, known as daughter cysts, or a mature worm. Daughter cysts occur in the organs of the intermediate host, whereas mature worms are found in the definitive host if organs infected with protoscolices are ingested. Development into a mature worm within the intestine of a definitive host occurs over a period of 4–7 weeks and completes the life cycle. Since two mammalian species are required for completion of the life cycle, direct transmission of echinococcosis from human to human does not occur. The life cycle is completed when carnivores ingest the cysts in the viscera of intermediate hosts. Each larval tapeworm can then develop into an adult tapeworm, which eventually produces new eggs and thereby continues the cycle.

## RESULTS

In our study, 3 patients (12%) were in the age group 15-20 years, 6 patients (24%) were in the age group 21- 30 years, 11 patients (44%) were in the age group 31- 45 years, and 5 patients (20%) were above 45 years of age. The mean age was  $35.8 \pm 11.58$  (Mean  $\pm$  SD). Thirteen (52%) were male patients while the remaining were females. The Male: Female Ratio was 1.083: 1.

We found that only 12 patients (48%) were housewives, 10 patients (40%) were laborers, 2 patients (8%) were students and only 1 patient (4%) was a professional skilled worker.

All the patients in our study had complaints of cough and chest pain. Fifteen patients had cough with expectoration. A few of them complained of fever and recurrent lower respiratory tract infection (LRTI) (12 patients each). A couple of patients presented with shortness of breath (SOB). One patient each complained of episodes of haemoptysis and abdominal pain (patient having concomitant abdominal hydatid cyst).

Eleven patients were symptomatic for 3 years or more, 10 patients were symptomatic for 2 years and only 4 patients had their symptoms for about a year. The mean duration of illness (in years) was  $2.4 \pm 0.91$  (Mean  $\pm$  SD).

Computed tomography (CT) scan was done in all patients for confirming their diagnosis. ELISA (enzyme-linked immunosorbent assay) was done in 4 patients & was reported to be positive in all of them. Fiberoptic Bronchoscopy (FOB) was done in 5 patients. A total of 15 patients (60%) had hydatid cysts located in their right lung whereas 6 patients

(24%) had cysts in their left lung. There were 3 patients (12%) who had thoracic as well as abdominal hydatid cysts. Only 1 patient (4%) had hydatid cysts in bilateral lungs. Fifteen patients (i.e., 60%) had involvement of the lower lobe and 4 patients had cyst in the upper lobe of the right lung. Only 1 patient had cyst located in the middle lobe. There were 3 patients (i.e., 12%) who had involvement of the lower lobe of the left lung. Involvement of hydatid cyst was seen in the left lower lobe and lingual in 2 patients each.

In our study, excision of cyst was done in 20 patients. Segmentectomy and lobectomy were done in 5 and 3 patients, respectively. Another 2 patients were also operated for abdominal cysts. A total of 8 patients (32%) required lung resection either in the form of segmentectomy or lobectomy. Lung resection was not required in the remaining 17 patients (68%). The surgical incision was made depending on the location of the cyst. Right posterolateral thoracotomy incision was made in 15 patients whereas left posterolateral thoracotomy incision was made in 6 patients. Bilateral posterolateral incision was made in 1 patient. For the patients who had concomitant abdominal hydatid cyst, right posterolateral thoracotomy incision followed by right phrenotomy was done in 2 patients. In another such patient, a separate lower abdominal midline incision was made.

Intercostal chest drains were placed in all the patients during operation. In 10 patients the cumulative daily drainage amount ranged between >75-100 ml, whereas it ranged between >50-75 ml in another 8 patients. The drainage collection was less than 50 ml per day in 5 patients. The remaining 2 patients had more than 100 ml per day drain

fluid collection postoperatively.

The distribution of cases by amount of collection in drains according to different interventions (i.e., with or without lung resection) was compared statistically (Table-1). As expected, cell values of more than 20% cells were less than 5, the Fisher's Exact Test was considered. The difference in proportion of patients having <75% collection in drain was not statistically significant. The p-value was found to be .411. The intercostal chest drains were removed between the 5<sup>th</sup> -10<sup>th</sup> postoperative days in 15 patients. Another 10 patients had their drains in situ for more than 10 days. Only 1 patient required drain for less than 5 days postoperatively. The distribution pattern of the observation on removal of drain amongst the different surgical procedures performed (i.e., those requiring resection of lung & those without any lung resection) were statistically compared. (Table-2a). As sample size in each individual group was less than 5, Shapiro-Wilk test was applied to assess the distribution pattern of the observation. The P value <0.05 observed in patients without lung resection was statistically significant. Observation on removal of drain in patients undergoing surgical interventions without any lung resection were with skewed distribution pattern. Therefore, Mann-Whitney U Test was applied. (Table-2b). The difference in mean time of removal of drain following the different surgical interventions was not statistically significant between the patients who required lung resection and those who did not. (Table-2c)

Twelve patients were discharged from hospital before the 10<sup>th</sup> postoperative day whereas another 9 patients could only be discharged after the 15<sup>th</sup> postoperative day. The remaining 4 patients were discharged between the 10<sup>th</sup> & 15<sup>th</sup> postoperative

Amount of Collection		Procedure		Total
		With Lung Resection	Without Lung Resection	
< 75 ml/ day	Count	3	10	13
	% within Procedure	37.5	58.8	52.0
>75 ml / day	Count	5	7	12
	% within Procedure	62.5	41.2	48.0
Total	Count	8	17	

Total number of cases in study group: n=25

**Table-1:** Distribution of cases by amount of collection in drains according to different interventions

Variable	Group	P value
Removal of Drain in <10 <sup>th</sup> postoperative day	With lung resection	.17
	Without lung resection	.000

**Table-2a:** Distribution pattern of removal of drain amongst the different surgical interventions

Group	Mean ± SD	Mean Rank	Sum of ranks	Man-Whitney U	P Value
With Lung Resection	14.13 ± 7.492	16.13	129.00	43.0	0.142
Without lung Resection	11.47 ± 9.29	11.53	196.00		

**Table-2b:** Observation on removal of drain in patients undergoing surgical interventions

Variable: Removal of drain (in days)	Range	Mean ± SD	Median (Interquartile range)
In procedures with Lung Resection	7-27	14.13 ± 7.492	12.5
In procedures without Resection	3-32	11.47 ± 9.29	7

**Table-2c:** Mean time of drain removal

Variable	Group	P value
Duration of hospitalization <10 <sup>th</sup> POD	With lung resection	.068
	Without lung resection	.004

**Table-3a:** Distribution pattern of hospitalization amongst the different surgical interventions

Group	Mean ± SD	Mean Rank	Sum of ranks	Man-Whitney U	P Value
With Lung Resection	17.38 ± 9.56	14.81	118.5	53.5	0.394
Without lung Resection	14.41 ± 9.50	12.15	206.5		

**Table-3b:** Observation on duration of hospitalization in patients undergoing surgical interventions

Variable: Duration of hospitalization (in days)	Range	Mean ± SD	Median (Interquartile range)
In procedures with Lung Resection	8-13	17.38 ± 9.56	14.5
In procedures without Resection	5-35	14.41 ± 9.50	9

**Table-3c:** Mean duration of hospitalization

days. The distribution pattern of the observation on duration of hospitalization amongst the patients undergoing different surgical procedures (i.e., those requiring resection of lung & those without any lung resection) were compared. (Table-3a). As sample size in each individual group was less than 5, Shapiro-Wilk test was applied to assess the distribution pattern of the observation. The P value 0.004 observed in patients without lung resection was statistically significant. Observation on duration of hospitalization in patients undergoing surgical interventions without any lung resection were with skewed distribution pattern. Therefore, Mann-Whitney U Test was applied. (Table-3b). The difference in mean time of duration of hospitalization following the different surgical interventions was not statistically significant between the patients who required lung resection and those who did not. (Table-3c)

**Patient morbidity:** A few common postoperative complications were seen. They have been classified under the following groups- Immediate (<24 hours), Early (24 hours – 7 days), and Late (> 7 days).

**Immediate Complications:** As many as 16 patients (64%) had air leak in their immediate postoperative period which was the commonest complication. The next common complication that occurred during this phase was fever which was seen in 4 patients. The other complications seen were bile leak and bleeding which was seen in each of 2 patients.

**Early Complications:** Twelve patients (48%) had postoperative wound infection. Another 5 patients (20%) presented with Broncho-pleural fistula in their early postoperative period.

**Late Complications:** 5 patients (20%) presented with empyema in the late postoperative period. Only 1 patient had wound dehiscence in our study.

**Mortality:** There was no mortality in our study.

## DISCUSSION

In our study, the maximum incidence was found in the age group of 31-45 years, i.e. 44%. The mean age (in years) was 35.80 ± 11.58, the minimum age was 15 years and the

maximum age was 61 years. However, in one of the studies, the mean age has been found to be 34 ± 21 years.<sup>1</sup> Another study has reported the mean age as 36.52 ± 17.13 years.<sup>2</sup> The majority (52%) of our patients were males while the remaining were females. One study<sup>3</sup> has shown the male: female ratio as 7:5. At least three other studies have shown almost equal male: female preponderance.<sup>4,5</sup> Thus no relation could be established considering the above criteria with the incidence of thoracic hydatid cyst.

Forty-eight percent of our patients were housewives, and 40% were laborers. Many clinical studies have found housewives as the commonest occupation in their study group.<sup>2,6</sup> Parija et.al reported maximum patients were labourers.<sup>7</sup> Hence, the observation in our study was comparable to other studies.

Cough (with or without expectoration) and chest pain were the presenting complaints of all our patients. 48% patients had complained of fever and recurrent LRTI. Other complaints included SOB, Haemoptysis, and abdominal pain. Numerous extensive clinical trials have been done over the past many years and it has been reported that cough, chest pain and SOB were the commonest presenting complaints in the symptomatic patients.<sup>5,6,8</sup> Another study reported dyspnoea, fever and haemoptysis as the commonest symptoms.<sup>8</sup> The findings of our study corroborated with the findings of most other studies in literature.

Most of our patients (44%) were symptomatic for over 3 years. Only 16% had symptoms for less than 1 year. The mean duration of illness in our study was 2.4 ± 0.91 (years). It was reported in one study that the duration of symptoms was less than 1 month 35.29% and was more than 1 year in 17.64% of the patients.<sup>6</sup>

CT scan was done in all our patients for confirmation of diagnosis, accurate localization of the cyst & determination of the size and condition of the cyst. ELISA and FOB were not done on a routine basis in our study. ELISA and FOB were done in only 16% and 20% patients in our study group. Many studies have reported in literature that CT Scan is the modality of choice for diagnosing hydatid disease and an indispensable preoperative investigation.<sup>9</sup>

Serological tests are not recommended as a routine investigation for diagnosing hydatid disease by many studies

since they have a high rate of false positive and false negative result.

60% of our patients had disease involving the right lung while 24% had cyst in the left lung. Concomitant association of abdominal cysts were seen in 12% of the patients. Bilateral lung cysts were seen in only 4% patients. The location of thoracic hydatid cyst has been reviewed extensively in literature. It has been concluded that the right lung is the most commonly involved organ in thoracic hydatid disease.<sup>1,4-6,8,10</sup> The incidence of bilateral lung hydatid disease has been reported as 2% by Dogan et al.<sup>4</sup> whereas it was reported as high as 20% in another study.<sup>10</sup> The incidence of hepatic and thoracic hydatid disease occurring together has varied greatly amongst the different studies and has ranged from 1% to 79%.<sup>4</sup> The findings of our study well correlated with that in literature.

We had 60% of patients with involvement of the right lower lobe and 16% had cyst in the right upper lobe. Only 1 patient (4%) had cyst located in the middle lobe. Another 12% had involvement of the lower lobe of the left lung. Cystic involvement was seen in the left lower and lingual lobes in 2 patients (i.e., 8%) each. Studies have also shown that hydatid disease is most prevalent in the lower lobe of lungs, with right lower lobe being more frequently involved than its left counterpart.<sup>1,4,6,10</sup> Thus, the findings in our study is comparable with those reported in literature.

All our patients were operated electively after a 6-week course of oral Albendazole therapy preoperatively. It was found that in 20 patients i.e., 80%, excision of cyst (i.e., cystotomy ± decortication or enucleation) was done. Segmentectomy and lobectomy were required in 20% and 12% of the patients in our group. Concomitant excision of the abdominal cyst was done in 8% of the patients. Similar findings were reported in different literature.<sup>4,8,11</sup> Many studies have concluded that recurrence of hydatid disease was high and was often related to the intraoperative spillage of cyst contents in the operative field. Hence it is recommended that either mebendazole or albendazole be given to reduce the risk of secondary hydatidosis.<sup>11</sup>

Seventeen patients (68%) did not need any form of lung resection. Eight patients (32%) underwent surgery for lung resection (segmentectomy & lobectomy). Surgical management of thoracic hydatid cyst has been studied extensively in literature. It has been suggested that the lung parenchyma should be preserved as far possible, especially in children and endemic areas, where the risk of recurrence is high.<sup>8</sup> Some studies have reported a lung resection rate of around 14%.<sup>12</sup> Another study has reported a lung resection rate of only 6.2%.<sup>8</sup>

Posterolateral thoracotomy was the commonest type of incision used in our study. 60% patients had a right posterolateral thoracotomy approach, 24% patients had a left posterolateral thoracotomy and 4% patients had bilateral posterolateral thoracotomies for bilateral disease. For patients having concomitant abdominal cysts, a right posterolateral approach followed by phrenotomy was used in 8% of patients while separate lower abdominal midline

incision was made in 1 patient (i.e., 4%). Most studies have concluded that the conventional incision i.e., posterolateral thoracotomy approach was commonly used<sup>1</sup>. In literature, it is also reported that bilateral hydatid disease may be managed by one- or two-staged surgery<sup>13</sup> and one-stage bilateral thoracotomy.<sup>3</sup> Many other studies have reported that trans-diaphragmatic approach (e.g. phrenotomy) is a suitable option for concomitant hepatic hydatid cysts<sup>4</sup>.

As previously said chest drains were placed in all the patients during operation. The amount of collection in drain was compared statistically amongst the patients undergoing different surgical techniques (i.e., those who needed lung resection & those who did not. A P-value of 0.411 was obtained which was statistically insignificant. (Table-1).

The mean time of drain removal was  $14.13 \pm 7.492$  in patients requiring resection of lungs, while it was  $11.47 \pm 9.29$  in patients who did not require any resection of lung (Table-2c). The distribution pattern of the observation on removal of drain amongst the different surgical procedures performed (i.e., those requiring resection of lung & those without any lung resection) were compared statistically (Table-2a). The P value  $<0.05$  observed in patients without lung resection was statistically significant. The difference in mean time of removal of drain following the different surgical interventions was not statistically significant between the patients who required lung resection and those who did not. (Table-2b)

The mean duration of hospitalization was  $17.38 \pm 9.56$  days in patients requiring resection of lungs, while it was  $14.41 \pm 9.50$  days in patients who did not require any resection of lung (Table-3c). The distribution pattern of the observation on duration of hospitalization amongst the patients undergoing different surgical procedures (i.e., those requiring resection of lung & those without any lung resection) were compared statistically. (Table-3a). The P value 0.004 observed in patients without lung resection was statistically significant.

The difference in mean time of duration of hospitalization following the different surgical interventions was not statistically significant between the patients who required lung resection and those who did not. (Table-3b)

In literature, it has been shown that postoperative hospital stays ranged between 3-21 days<sup>1,11</sup>. The mean duration of hospitalization was 7.3 days in one study.<sup>11</sup> Another study reported an average hospital stay as  $9.4 \pm 3.8$  days.<sup>1</sup>

#### Patient Morbidity

**Immediate postoperative complications:** 64% of our patients had air leak in their immediate postoperative period which was the commonest complication. This was followed by fever which was seen in 4 patients (16%). The other complications were bile leak and bleeding. In literature, numerous studies have shown that the perioperative morbidity ranged between 0-17%.<sup>4</sup> Studies have also reported that air leak was one of the most common complication<sup>3</sup> and was often seen after procedures such as enucleation<sup>4</sup> and cystotomy with closure of bronchial openings (3%)<sup>14</sup>. Bile leak and bleeding were reported in some

studies.<sup>3,4</sup>

**Early postoperative complications:** Postoperative wound infection was the commonest early complication in our study. This was followed by broncho-pleural fistula. A number of studies have reported in literature that prolonged air leakage or broncho-pleural fistula is a common complication in the early postoperative period.<sup>3-5,15</sup> Surgical procedures such as enucleation<sup>4</sup>, cystotomy<sup>14</sup> and lobectomy<sup>4</sup> has a high incidence of development of broncho-pleural fistula postoperatively. A study<sup>15</sup> has reported that in perforated cases, infection and inflammation of the adjacent pulmonary parenchyma affected wound healing.

**Late postoperative complications:** 5 patients (20%) presented with empyema in the late postoperative period. Only 1 patient had postoperative wound dehiscence in our study. Numerous studies have reported empyema as one of the commonest complication in the late postoperative period.<sup>4,15</sup> Studies have found empyema to be associated with surgical procedures such as pericystectomy, cystotomy<sup>14</sup>, open aspiration by Figuera technique<sup>16</sup> and lobectomy.<sup>4</sup> Wound infection was reported as a late postoperative complication in many studies<sup>5</sup>

**Patient Mortality:** We had no mortality in our study. However, the mortality rate ranged between 0-5% in most studies<sup>3,4,8,11,15</sup> Lobectomy was reported to have a mortality rate of 0-2%<sup>4</sup> The commonly reported causes of death were pneumonia and sepsis<sup>4</sup>

## CONCLUSION

It is evident from this study that thoracic hydatid disease commonly presents with cough and chest pain. CT Scan is an essential tool for diagnosis. Surgery is the treatment of choice with pre- and postoperative albendazole therapy. Lung resection is needed for selected patients and the most common complication following surgery is air leak which can be managed conservatively.

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