Surgical treatment of echinococcosis by a transthoracic approach: a review of 85 cases

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Abstract

Objective: Human echinococcosis remains a serious health problem for the Mediterranean countries, among them Greece. As there is no effective medical therapy, surgery is still the treatment of choice.

Material and methods: We present our experience in the surgical management of hydatidosis by a transthoracic approach, based on 85 patients (49 male, 36 female, aged 4–86 years) treated during 1986–1996.

Results: Twenty-one patients (26.3%) appeared with complications as: hydatidemesis (n = 5), hydropneumothorax (n = 3), cyst infection (n = 3), empyema thoracis (n = 8), cholebronchial (n = 3), and cholebronchopleural fistula (n = 1). The location of the cysts was: 61 in the lungs (right, 29; left, 24; bilateral, eight), 31 on the liver dome, six in the pleural cavity, two in the mediastinum, and one in each of pericardium, chest wall, and right pararenal space. Surgical approach involved a thoracotomy or median sternotomy in all cases. Pulmonary endocystectomy and capitonnage was the procedure of choice in the surgical management. Hepatic cysts were approached through a right thoracophrenotomy and were managed with evacuation of the main and daughter cysts, suture of the diaphragm to the margins of the cyst, and drainage of the cystic and pleural cavities. There was no in-hospital mortality. Major postoperative complications were: empyema thoracis (n = 3), biliary fistula (n = 2), and bronchopleural fistula (n = 1). Five patients presented later with seven recurrences of the disease.

Conclusion: Transthoracic approach is a good and safe choice in surgical treatment of both the intrathoracic and the (concomitant or not) hydatid cysts on the upper surface of the liver.

Keywords: Echinococcosis; Lung echinococcosis; Liver echinococcosis; Surgical management

1. Introduction

Echinococcosis has been known since the time of Hippocrates, who first introduced methods of treatment. Even today it remains a significant health problem in endemic areas, such as the Mediterranean countries, the Middle East, Central Asia, China, Australia and New Zealand, East Africa, South America, northwestern Canada and Alaska, where sheep- and cattle-raising is a mainstay of existence [1–3].

The lung is the second most commonly affected organ after the liver (10–40%) [3,4].

In this study, we present the experience of a Thoracic Surgery Department in the surgical management of lung and liver hydatidosis by a transthoracic approach.

2. Patients and methods

We retrospectively studied the records and films of 85 patients with hydatid disease, who were treated in the Department of Thoracic and Vascular Surgery at 'Evangelismos' General Hospital in Athens from September 1986 to December 1996. Information on history, clinical picture, radiological findings, methods of surgical management, and results of surgery were analyzed and classified. Fol-
low-up was based on personal or telephone contact with the patients. There were 49 male (57.6%) and 36 female (42.4%), aged 4–86 (mean: 44) years. All were Greek citizens. Most of them (64/85, 77.3%) were living in rural areas, being farmers or working in contact with animals and vegetables. All were infected by Echinococcus granulosus.

The methods of surgical management are outlined as follows.

### 2.1 Thoracic cysts

Surgical approach and management depend on the anatomic and clinical features of the hydatid cyst(s). A standard posterolateral thoracotomy is employed in most cases. After entering the hemithorax, the cyst is identified as a grey-white swelling on the surface of the affected lobe. The operative field is isolated with saline pads for protection from spillage of cystic fluid. In non-complicated hydatid cysts with no great tension, the ‘intact endocystectomy’ (Ugon’s technique [3]) is attempted [1,3,6]. The pericyst is incised with a fine scalpel until the right plane of the non-adherent tissue between endocyst and pericyst is found and the whitish endocyst appears. As the outer pressure decreases abruptly, rupture of the endocyst is most likely to occur at that point. The flaps of the pericyst are lifted up with fine forceps and the pericyst is carefully separated from the endocyst. When most of the endocyst has appeared, the anesthesiologist is asked to increase the intrapulmonary pressure and the endocyst can be delivered intact into a basin. During ‘delivery’ of the cyst, one or two tubes are ready to suction in case of sudden rupture. The free flaps of the pericyst are excised. The residual cavity of the cyst is carefully inspected and the bronchial leaks found are closed individually with fine sutures. The cavity is obliterated with pursestring suture of fine absorbable material starting from the bottom (capitonnage). The concept of capitonnage is to avoid abscess formation in the residual cavity. After full expansion of the lung is achieved, the thoracic cavity is closed with two drainage tubes.

An alternative to intact endocystectomy, first described by Barrett in 1947 [3], is aspiration of the endocyst prior to its removal, and infusion of hypertonic saline solution (NaCl 15%) into the cyst. Then, the excision of the crumpled cyst is easier.

### 2.2 Hepatic cysts

Surgical approach is achieved via a right posterolateral low thoracotomy. After entering the hemithorax through the 7th or 8th intercostal space, the cyst is palpated and the overlying diaphragm is incised, as well as the pericyst and the main cyst. The main and the daughter cysts are removed by suction. The residual cavity is sterilized using hypertonic saline solution. Any bile duct leak is closed with fine suture material. The margins of the diaphragmatic incision are sutured to the margins of the residual cavity with a running non-absorbable suture, preventing spillage of the contents into the abdomen. The remnants of the incised diaphragm and the pericyst are left within the cavity, which is drained with a mushroom catheter brought out through the 9th intercostal space. The pleural space is drained in the usual manner [6].

In infected hepatic hydatid cysts with diaphragmatic and pleural or pulmonary involvement, the attempt was to dissect the cyst adherences to the lung and preserve as much as possible, to remove the purulent content of the cystic and the pleural cavity, and to establish adequate drainage of both cavities.

### 3. Results

The total number of hydatid cysts diagnosed was 111. In

<table>
<thead>
<tr>
<th>Location</th>
<th>RL</th>
<th>LL</th>
<th>Liver</th>
<th>Pleura</th>
<th>Mediastinum</th>
<th>RPRS</th>
<th>Total</th>
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<tr>
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<td>Mediastinum</td>
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<tr>
<td>Chest wall</td>
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<td>Pericardium</td>
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<tr>
<td>Total</td>
<td>37</td>
<td>32</td>
<td>31</td>
<td>6</td>
<td>2</td>
<td>1</td>
<td>111</td>
</tr>
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RL, right lung; LL, left lung; RPRS, right pararenal space; RUL, right upper lobe; RML, right middle lobe; RLL, right lower lobe; LUL, left upper lobe; LLL, left lower lobe.
26 patients (30.7%) multiple cyst locations were identified (Table 1).

3.1. Clinical picture

Clinical manifestations varied widely depending on the status of the hydatid cyst. Patients with intact cysts (62/85, 73%) were asymptomatic (38/85, 44%) or reported minimal and non-specific symptoms, such as chest pain, cough, mild dyspnea, for several months before seeking medical advice (24/85, 29%). In the majority of them diagnosis was made accidentally on a routine chest roentgenogram. Twenty-three patients (27%) presented with complicated cysts (Table 2).

3.2. Radiological findings

Chest roentgenogram (posteroanterior and lateral views) was the most helpful diagnostic tool providing useful information on the anatomic location and pathologic condition of the cyst. A well-defined spherical or oval opacity within the lung towards its periphery was found in 41 cases of intact simple hydatid cysts. In nine cases the outline of the shadow was blurred due to pericystic inflammatory reaction of the lung parenchyma. The ‘camalote’ or ‘water-lily’ or ‘iceberg’ sign (rupture of the cyst into the bronchus and partial evacuation of its fluid results to a cavity with a undulating fluid level due to the floating echinococcus) was present in three cases (Fig. 1). Perivesicular pneumocyst or ‘detachment’ or ‘meniscus’ or ‘double arch’ sign, that is a round shape with a superior small crescent of air, like a new moon, due to partial separation of the endocyst from the pericyst by the entrance of air (pathognomonic sign for echinococcus), was observed in two cases. The picture of the incarcerated membrane, produced after rupture of the cyst into a bronchus and closure of the draining bronchus due to inflammatory reaction resulting to imprisionment of the hydatid membrane, was seen in two cases (Fig. 2). Hydropneumothorax existed in three cases.

The radiological image of a lung abscess was present in three cases (Fig. 3), with concomitant pleural effusion in one case. In four of the six cases with empyema thoracis, the hydatid cyst was not apparent on plain chest film.

In the two patients with hydatid cysts of the posterior mediastinum, the round/oval opacity of the cyst was partially covered by the cardiovascular shadow, so it was difficult to differentiate it from neoplasm, despite the well-defined outline of the lesion. In one of them the coexistence of a pulmonary hydatid cyst was helpful to diagnosis, while in the other one computed tomography (CT) scanning was necessary.

Plain tomography was in use in the first 2–3 years of our study, but then it was completely replaced by ultrasonography (u/s) and CT. Both the latter imaging techniques are most sensitive in defining the size, number, and position of the cysts in either the lung parenchyma (CT) or the hepatic tissue (u/s, CT).

![Fig. 1. Bilateral pulmonary hydatid cysts. RUL, intact cyst; LUL, cyst ruptured to the pleura (waterlily sign).](https://academic.oup.com/ejcts/article-abstract/14/2/134/459155/136)
Bronchoscopy was performed in two patients with hydatidemesis and in one with infected cysts, and clear hydatid fluid and purulent secretions were aspirated, respectively. No scolices were isolated.

The Casoni skin test and the Weinberg reaction test have been abandoned in the diagnostic approach of hydatid disease in our Department during the recent decade, due to their low diagnostic value.

3.3. Surgical treatment

Eighty-three patients were surgically treated. Two aged patients (the oldest male and female in our series) presented with hydro pneumothorax and empyema thoracis, respectively, and were treated successfully with tube thoracostomy and drainage of the pleural cavity. The one refused operation, and the other was denied an operation due to her bad general condition. They left hospital been improved, and at follow-up 12 and 14 months after their discharge no related to the disease problem was revealed. A transthoracic approach was employed in all surgically treated patients through an ipsilateral thoracotomy ($n = 75$), or bilateral (submammary) thoracotomy ($n = 3$), or median sternotomy ($n = 5$). Table 3 summarizes the procedures employed in the management of hydatid cysts.

The intact endocystectomy technique was attempted in 44 pulmonary cysts with rupture occurring in eight cases. The cyst aspiration prior to removal was the method of treatment in 19 intact cysts of the lung.

Radical surgical procedures (pulmonary resection) were employed in case of destruction of the surrounding the cyst lung parenchyma due to prolonged compression or inflammation. Five wedge resections were performed in three cases of infected lung cysts and in two cases of uncomplicated huge lung cysts. In 5 cases of empyema thoracis due to lung hydatid cyst, lung decortication completed the surgical procedure.

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The hepatic cysts were treated in the manner previously described. In seven cases of infected hepatic hydatid cysts with diaphragmatic and pleural or pulmonary involvement, the cyst adherences to the lung were dissected, the purulent content of the cystic and the pleural cavity was drained, and adequate drainage of both cavities was established. In the 3 cases of bronchobiliary communication (Figs. 4 and 5), segmental or atypical resection of the affected lung parenchyma adherent to the cyst was performed. Lung decortication seemed necessary in one of the two cases of empyema thoracis due to liver cysts.

In case of concomitant right lung and liver dome hydatid cyst, they are removed at one stage through a right thoracophrenotomy. The choice of the intercostal space for the

<table>
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<th>Table 3 Surgical procedures for hydatid cysts</th>
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<tr>
<td>Procedure</td>
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<tr>
<td>Lung cyst excision</td>
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<td>Wedge resection</td>
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<td>Atypical lobectomy/segmentectomy</td>
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<td>Pleural cyst excision</td>
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<td>Chest wall cyst excision</td>
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<td>Liver cyst excision</td>
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<tr>
<td>Fistula excision</td>
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<tr>
<td>Lung decortication</td>
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<td>Tube thoracostomy alone</td>
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*Two patients had undergone liver hydatid cyst excision elsewhere.
incision depends on the location of the lung cyst. The latter is treated first. The one-stage procedure was employed in 11 patients.

3.4. Medical treatment

The last 16 patients of our study were administered post-operatively chemotherapy (albendazole, 400 mg twice a day for the first 15 days of the month, for 3 months), according to a protocol established in cooperation with the Department for Infectious Diseases of our hospital. Although no recurrence in these patients has been encountered yet, we are unable to come to powerful conclusions at present.

3.5. Mortality and morbidity

There was no in-hospital mortality in our series. The post-operative complications are summarized in Table 4.

3.6. Follow-up

The follow-up span ranged from 10 months to 10 years. Five (5.9%) patients (four male) presented with either a recurrence or a new onset of hydatid disease 9 months to 10 years after surgery (Table 5). Four of them had ruptured cysts pre- or intraoperatively. In two patients with two incidents of echinococcosis each, a new onset of the disease should be considered due to the location of the cysts in the subsequent episodes and the time span. All were treated surgically (cystectomy), without complications.

4. Discussion

Surgery remains the principal mode of treatment of echinococcosis, as chemotherapy with benzimidazole compounds (mebendazole, albendazole) is not yet satisfactory [7]. Concerning E. granulosus, cure can be expected in about 30% of patients and improvement in 30–50% after 12 months follow-up [7].

Methods of surgical management of pulmonary hydatid cysts are divided into two groups [3]. (1) Conservative: the parasite is removed, the bronchial openings are closed, and the residual cavity is obliterated. Two forms of cystectomy are employed: removal of the endocyst (endocystectomy, Ugon’s or Barrett’s technique) or excision of both the endocyst and the pericyst (pericystectomy or Perez-Fontana’s...
Liver RLL 10 years
LLL 1. LUL 8 years
LLL LUL 5 years

Concomitant hepatic hydatid cyst under the diaphragm, hidrosis, has been emphasized in the past [4,20]. In case of echinococci of the liver in every patient with lung hydatidosis surgically treated, published in the recent decade (1988–1997) and covering approximately in 20% of all surgical procedures [1,5,8–12,14,18]. The low proportion of resections in our material (12%) indicates that we always try to preserve as much lung parenchyma as possible. The reason for that is double. First, pericyst is not parasitic, so excision in its entirety is not necessary. The surrounding lung tissue is atelecstatic but often not infected, and after cyst removal, it usually reexpands well. Second, after pulmonary resection, the patient chances of surviving future lung disease are reduced. Hydatid disease may recur as he returns to the endemic area. Five patients in our series had a second incidence of hydatidosis, and two of them a third one.

In case of giant cysts with prolonged compression of a considerable amount of lung parenchyma, the decision on the type of operation is made at the operating theater. If, after cyst removal, the compressed lung portion cannot reexpand, then parenchymal resection is justified. In case of infected cysts with gross parenchymal affection, resection is the procedure of choice.

The overall mortality rate after surgical management of intrathoracic (lung and liver) echinococcosis is very low (0–4%) [1–5,6,8–25]. A review of 14 reports on 4,255 patients with intrathoracic hydatidosis surgically treated, published in the recent decade (1988–1997) and covering the period from the early 70’s till now, revealed a perioperative mortality of 1.45% [1,2,5,8–14,16–18]. The overall postoperative early and late morbidity ranged from near zero to 17% in 3,433 patients [2,5,8–10,12–14,16–18].

Hepatic hydatid cysts coexist with lung cysts in 4–14%, with a mean of 8.34% of all patients with pulmonary cysts [2,4,5,8,12,15,17,18,20,21,23,24]. The need to search for echinococci of the liver in every patient with lung hydatidosis, has been emphasized in the past [4,20]. In case of concomitant hepatic hydatid cyst under the diaphragm, removal of both lung and liver lesions at one stage is a good option [4,6,15,17,19,21,24]. Many times the approach from above the diaphragm is the only option. Our technique in the management of hepatic cysts differs from others described in the literature [2,4]. We do marsupialize and drain the pericystic cavity through the pleura, not through the abdomen. The diaphragm is not repaired. The margins of the phrenotomy are sutured to the margins of the residual cavity after cyst removal. Thus, the peritoneal cavity is isolated and protected from spillage of hydatid contents. Our approach is always transpleural (right low thoracotomy). Others use sometimes thoraco-abdominal incision [19–21,23]. In our series, there was no mortality and morbidity among patients with one-stage operation. The two patients with postoperative cholepleural fistula had only hepatic cysts, and the total morbidity after hepatic hydatidosis surgery was 6.4% (2/31). Our results are comparable with the ones reported in the recent literature. In 120 patients undergone simultaneous excision of lung and liver cysts, mortality was zero, and morbidity was 17.5% (13 empyema, four cholebronchial fistulas, 2 biliocutaneous fistulas, two sub-diaphragmatic abscesses) [2,4,15,17,19–21,23,24].

We feel that simultaneous removal of concomitant lung and liver hydatid cysts is not so widely performed, as the incidence of the entity requires. In 1983, Aubert and Viard [25] reviewed 8384 cases of pleuropulmonary hydatidosis from 69 centers around the Mediterranean basin. They found that 18 surgical teams usually follow the one-stage approach and five perform it occasionally. Since then, we were able to find only seven reports on the simultaneous treatment [2,4,15,19–21,24]. Obviously, it is appropriate to raise the subject again in the thoracic surgical literature in order to find in the every day practice the place it deserves.

In conclusion, we are convinced that transthoracic approach is a good and safe choice in the surgical treatment of both the intrathoracic and the (concomitant or not) hydatid cysts on the upper surface of the liver.

Table 5

<table>
<thead>
<tr>
<th>Cyst location at 1st episode</th>
<th>Cyst location at subsequent episodes</th>
<th>Interval</th>
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<tbody>
<tr>
<td>RLL + R. pleura</td>
<td>R. chest wall</td>
<td>10 months</td>
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<tr>
<td>LLL</td>
<td>LUL</td>
<td>5 years</td>
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<td></td>
<td>1. LUL</td>
<td>8 years</td>
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<tr>
<td></td>
<td>2. LLL</td>
<td>9 months</td>
</tr>
</tbody>
</table>

References

[6] Exarchos N. Experience with surgical treatment of hydatid cysts of...
the lung, heart and diaphragmatic portion of the liver. Presentation at New York Society for Thoracic Surgery, May 24, 1984.


