Surgical Treatment of Pulmonary Hydatid Disease in Kayseri State Hospital, Turkey: Ten Years of Experience

Ali Yeginsu, MD; Recai Buyruk, MD; Mustafa Köseahmetoğlu, MD

Kayseri State Hospital, Deparment of Thoracic and Cardiovascular Surgery, Kayseri, Turkey

Abstract

Background: We reviewed a series of 70 patients with pulmonary hydatid disease treated surgically in our center from 1990 to 2000.

Patients and methods: We retrospectively reviewed the patients' symptomatology, diagnostic studies, treatment options, and morbidity and mortality. Male/female ratio was 38/32 and the mean age was 25.5 years (range:7-70 years).

Results: Radiological studies were the main diagnostic tool. The correct preoperative diagnosis could be made in 71% of the patients by plain chest roentgenogram and in 96% of the patients by chest roentgenogram plus tomography, and in three patients diagnosis were made per or postoperatively. Eighty one operations were performed in 70 patients with 46 intact and 40 ruptured cysts. Sixty seven parenchyma saving operations, 11 pulmonary resections and 7 pleural decortications were performed. Patients with extrathoracic hydatid disease were refferred to the relevant subspecialty clinics and were not operated on by our team. Eighteen complications developed in 8 patients (11%). Prolonged air leak was the most frequent complication (in 7 patients). One patient required resuscitation peroperatively. There was no operative mortality.

Conclusions: Surgery is still the main procedure for treatment of pulmonary hydatid disease. The parenchyma-saving surgical methods sould be preferred.

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Correspondence: *Dr. Ali Yeginsu, MD*
Gaziosmanpaşa Üniversitesi,
Tıp Fakültesi, Göğüs Cerrahisi AD.
60100, Tokat, Türkiye
Tel: +90 (0) 542 252 64 41
e-mail: yeginsu@hotmail.com

Introduction

Hydatid disease is endemic in rural areas in many countries, especially in countries around the Mediterranean and those in the Middle East, Central Asia, South America, New Zealand, Australia, Northwestern Canada, Alaska and Northern Africa (1-6). The incidence of the disease in Turkey is 2/100 000 (approximately 2200 new cases per year) and the disease is still an important public health problem (1,3,4). In this article, we present and discuss our experience of ten years with respect to the treatment of pulmonary hydatid disease.

Patients and Methods

During a period of ten years, from February 1990 to December 2000, our team operated on 70 patients with pulmonary hydatid disease. The male:female ratio of the patients was 38:32 and their mean age was 25.5 (range: 7 to 70 years). Forty seven of the

Table 1. Symptoms

Symptom	Patient (n)	%
Coughing	53	75
Chest pain	50	71
Dyspnea	28	40
Weakness	25	35
Haemoptysis	24	34
Fever	20	28
Sputum	14	20
Expectoration	10	14

Table 2. Radiological findings

Finding	Patient (n)	%
Simple cyst	25	36
Giant cyst (>10cm)	18	26
Diffuse image	17	24
Air-bubble sign	13	18
Air-fluid level	10	14
Camalote sign	7	10
loculation	5	7
Pleural tickening	5	7
Mass lesion	2	3
Pleural effusion	2	3
Peripheral nodule	1	1.5
Calcification	1	1.5
Hydropneomothorax	1	1.5

patients (67%) were from the rural areas of the Central Anatolia.

All patient files were reviewed retrospectively for presenting symptoms, diagnostic studies, operative findings, complications, hospitalization, and recurrences.

Results

Sixty six patients (94%) were symptomatic. Coughing (75%) and chest pain (71%) were the most frequent symptoms at presentation (Table 1). Haemoptysis was seen in 24 patients but all were mild. Fever, purulent sputum, weakness, dyspnea were other frequent symptoms of the cystic disease. No anaphylactic reaction was observed. Four patients were completely asymptomatic and all these patients had intact cysts.

Posteroanterior and lateral chest radiography were performed on all patients. The correct preoperative diagnosis could be made by plain chest radiography in 50 of the patients (71%). Computed tomography of the chest was performed in 48 (68%). All but three patients (96%) could be diagnosed by plain chest radiography plus chest tomography preoperatively. Preoperative radiological findings are listed in Table 2. Eighty two of the cysts (95%) could be visualized radiologically. Except for the 6 patients with suspected disease, 3 of whom had positive results, serology (indirect hemaglutination) was not routinely done. The diagnosis was made intraoperatively in 2 patients and postoperatively in 1.

Table 3. Localization of the cysts

Locus	Patient (n)	%
RLL	18	26
RUL	12	17
LLL	11	16
LUL	11	16
RML	5	7
RUL + RLL	3	4.3
LLL + LUL	3	4.3
RUL + RML	2	3
RML + RLL	1	1.5
RUL +RML + RLL	1	1.5
Bilateral	3	4.3
Total	70	100

RLL=Right lower lobe, RUL=Right upper lobe, LLL=Left lower lobe, LUL=Left upper lobe, RML=Right middle lobe.

Table 4. Surgical treatment

Type of operative procedure	n
Cystotomy and capitonnage (CC)	53
Enucleation and capitonnage (EC)	8
CC and decortication	6
CC and wedge resection	4
EC and wedge resection	2
Lobectomy	2
Lobectomy and decortication	1
Wedge resection	2
Bronchopleural fistula repair (rethoracotomy)	2
Control of bleeding (rethoracotomy)	1
Total	81

The right lower lobe was the site most frequently involved (26%) (Table 3). The disease was unilateral in 67 of the patients (96%) and multiple cysts were present in 10 (14%). Pulmonary and extrathoracic cysts were determined in 8 of the patients (11%). The liver (5/8), the brain (2/8), and the chest wall (1/8) were the most frequently involved extrathoracic sites. Twenty one (30%) of the cysts were infected. Two of the patients were referred to our center with empyema and one with hydropneumothorax. These patients were first subjected to a tube thoracostomy and the operation was performed subsequently.

Table 5. Postoperative complications

Complications	P-S O	Resections	n	%
Prolonged air leak > 7 days	6	1	7	39
Air Space	1	2	3	17
Pneumothorax	-	1	1	5
Bleeding	-	1	1	5
Atelectasis	2	-	2	12
Pleural effusion	1	-	1	5
Empyema	2	-	2	12
Intraoperative resuscitation	1	-	1	5
Total	13	5	18	100

P-S O= Parenchima-saving operations

Fourty six intact and 40 ruptured cysts were determined intraoperatively. Two of the intact and 19 of perforated cysts were infected. A total of 81 operations (70 for primary disease, 3 for rethoracotomy, 3 for contralateral lung disease and 5 for recurrent disease) were performed (Table 4). Operations for bilateral cysts were always performed in two stages via posterolateral thoracotomy. In patients with extrathoracic hydatid disease, operations were not performed simultaneously and all of these pateints were referred to a subspecialist.

Cystotomy capitonnage was the most common procedure (85%). Eleven (15%) lung resections (8 wedge resections and 3 lobectomies) were performed. Our indications for lung resection included multiple cysts in 3 patients, infection in 2, giant cysts in 2 and extensive tissue damage in the distal portion of the lung in 4. In 7 of the patients pleural decortication was performed and all were infected. We have not routinely used albendazole, but only in 24 patients with multiple, ruptured or recurrent cysts.

There was no mortality. One patient required resuscitation intraoperatively due to aspiration of hydatid liquid. Rethoracotomy was required in 3 patients (bronchopleural fistula in 2 patients and bleeding in one). A total of 18 complications were encountered in 8 patients (11%) (Table 5). Prolonged airleak was the most common complication. Mean duration for the operations was 154 minutes (range: 75 to 215 min), mean duration for drainage was 6.8 days (range: 2 to 30 days), mean duration for hospitalization was 16.4 days (range: 7 to 64 days) and mean cost per patient was the equivalent of 870 USD (range: 413 to 1714 USD). Periodic follow-up could not be done because most of the patients were from rural areas, but 5 of the patients (7%) underwent a second operation because of recurrence. Four had ruptured and one had multiple cysts at the initial operation. All of these patients were given albendazole therapy after the operation. All cysts were in the same operated site and all patients were treated with a second thoracotomy and albendazole. No patient was readmitted for recurrence subsequent to this second intervention.

Discussion

Hydatid disease, caused by Echinococcus granulosus, is a parasitic disease and is endemic in many sheep and cattle raising areas around the world. It may occur in all age groups but is most frequent in the third and fourth decades of life (4). Liver is the most frequently involved site (55-70%) and the lung is second in frequency (18-35%) (4). Simultaneous lung and liver cysts are seen less than 10% of the cases (7). Nearly any organ can be involved in hydatid disease. In pulmonary hydatidosis, cysts are predominantly located in the lower lobes and more on the right side than on the left (1-5). Our results are also in accord with these previous reports. In our patients, 26% of the cysts were in the right lower lobe and 7% of the patients had cysts in the liver concomitantly.

The plain chest radiography is the main diagnostic tool in hydatid disease (1-7). It has been shown to be highly sensitive (90%) in several studies (4,8,9). Typically, intact pulmonary cysts are seen as well demarcated, spherical, homogenous and single or multiple lesions (1,3). Some characteristic descriptive images of hydatid cysts such as the water-lily sign, Escudero-Nenerow sign, notch sign, double dome arch sign, meniscus sign, air-bubble sign have been reported (1,2,5,6,9,10). However, plain chest x-ray is not always enough for the diagnosis. Computed tomography of the chest, by revealing the fluid density of a cystic lesion, the air-fluid density of a cavitary lesion, or the solid density of a complicated cyst, may be helpful in establishing the diagnosis. It can also be of value in determining the presence of cysts in areas difficult to visualize with chest x-ray films, especially in the posterior and costophrenic angles (11). Ninety six percent of our cases could be detected preoperatively by chest x-ray plus tomography. We believe that chest tomography increases the diagnosis rate but is not mandatory. Serological studies are highly sensitive (70-100%) in hydatid disease (1,2,3,5,7,10), but false-negative results are reported in 34-42% of the cases (3,4,12). Additionally, false positivity in some parasitic, degenerative, autoimmune, allergic, and neoplastic diseases is reported (10). We used serology not routinely but only when having difficulties in diagnosis.

Surgery is the mainstay of optimal treatment of pulmonary hydatid disease (1-10). The primary aims of surgery are to remove all cysts while preserving as much lung parenchyma as possible, sterilize the resultant cavity, suture air leaks, close the cyst cavity and treat the pleural space. The necessity to close the residual cystic cavity is disputed but most authors accept the importance of closure of patent bronchial openings (10,13). Lung resection should only be resorted to

when the lung tissue has been completely destroyed (1,3). Bilateral cysts are removed by 2-staged posterolateral thoracotomy or a median sternotomy. In intact bilateral cysts, if 2-staged thoracotomy is planned, the operation shoud be performed first on the larger or the multiple cysts (10). And if there is a ruptured cyst on one side and a large intact cyst on the opposite side, first the intact cyst should be operated on, because the intact cyst may rupture during the operation (10). Removal of the cysts located in the left posterior basillary segment may be especially difficult by median sternotomy, but some authors report that this problem can be solved by using video-assisted surgery as an additional method (3). In patients with right lung and liver cysts, simultaneous removal of lung and liver cysts may be possible via a right thoracotomy and phrenotomy (1,3). Bilateral lung cysts may be treated successfully with median sternotomy and right phrenotomy (3). We performed standard posterolateral thoracotomy for all our cases and never performed simultaneous removal of the liver and lung cysts. We preferred to refer these patients to the general surgery department. Lung resection was performed in 15% of cases. Existence of a high number of complicated and/or giant cysts accounts for this high rate of lung resection in our series. However, similar high rates were also reported in other series (5).

Various surgical techniques for removal of hydatid cysts, such as excision of entire cyst by enucleation (Barret technique), excision of pericyst (Perez-Fontana), cystotomy, lung resections, have been described (1,3,10). Choice of surgical method depends on the condition of the cysts. Cystotomy and capitonnage is the most common procedure and is preferred by many teams (1-7). We also preferred cystotomy and capitonnage procedures in 77% (63/81) of our patients. However, in giant cysts, capitonnage of the residual cavity may cause atelectasis by obliterating the bronchus surrounding the cyst. In these cases, extensive resection of damaged superficial pulmonary tissues (partial cystectomy) and closure of bronchial openings may be performed (3). Enucleation was recommended in small peripheral cysts (3), but successful enucleation has been reported for a cyst 18 cm in diameter and 3200 g in weight (2). Video-assisted thoracoscopic and percutaneous aspiration techniques may be performed in selected patients but have not been widely accepted yet (14-16).

Chemotherapy is mainly recommended for inoperable, multiple or complicated cysts to prevent further dissemination or recurrences (1-10). Chemotherapy alone is of no value (1,3 17). Preoperative use of chemotherapeutics is not recommended, because these drugs weaken the wall of the cyst and make rupture of a pulmonary cyst more likely. In addition, there have been reports of toxicity from these drugs, though this seems to be dose-related and more likely to develop in patients with liver failure (1,3,14,18). Recently, chemotherapy combined with surgery for prophylaxis has been recommended (1,3,5-7). Albendazole is currently the drug of choice for perioperative prophylaxis and treatment of inoperable cases of pulmonary hydatid disease in humans (17). With this treatment, the cure rate was only 30% and prolonged, repeated, and high doses were necessary to obtain satisfactory results (17,19). We routinely use albendazole in the following instances:

1 multiple hydatid disease,
2 ruptured pulmonary cysts,
3 recurrent cysts, and
4 inoperable cystic disease.

Surgical treatment of pulmonary hydatid disease carries a morbidity risk of 0 to 17% and a mortality risk of 0 to 5%, while recurrence of cysts varies from 2 to 30% over 5 years even without obvious spillage of cyst contents (1-3,7,19). The most common complication is prolonged air leaks (1-3,6,7). In our series, there was no mortality, but the complication rate was 11%. Prolonged air leak was the most common complication, as was reported in the literature. Mean operation time was 154 minutes, mean duration of drainage was 6.8 days, and mean duration of hospitalization was 16.4 days. An approximate mean value of 800 USD was given as the treatment cost of a patient with hydatid disease (20). Mean cost per patient in our series was estimated as 870 USD. We observed hydatid disease recurrence in 5 patients (7%), all with pulmonary location.

In conclusion, radiological studies are still the main diagnostic tool and surgery is still the optimal treatment procedure in pulmonary hydatid disease. Parenchyma-saving surgical procedures should be preferred when possible. Chemotherapy alone is not effective and its preoperative use is not recommended. Postoperative albendazole may be used for prophylaxis in patient with multiple, inoperable or recurrent cystic disease.

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