

PULMONARY HYDATID DISEASE: A Retrospective Study

Akciğer hidatik hastalığı: Retrospektif çalışma

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Summary: We have reviewed 197 cases diagnosed preoperatively by chest roentgenograms. Six patients who had false-positive diagnosis (2.9 percent) excluded. The right lung (57.8 %) and the lower lobes were the most frequently affected parts. Preoperatively ruptured cases were diagnosed in 40 patients. Eighteen of them were into the pleural cavity and 22 were into the intrabronchial system. Surgical intervention was carried out in 193 patients. Frequently used surgical techniques were cystectomy plus capitonage (60 %) or only cystectomy. Lobectomy and pneumonectomy were applied in seven cases. In the late period follow-up eight patients who had been operated because of ruptured cyst, was found recurrent hydatid cyst.

Özet: 1978-1990 yılları arasında kliniğimizde tedavi edilmiş 197 pulmoner hidatik kist olgusunu gözden geçirdik. Olguların tümü ameliyat öncesi göğüs röntgenogramlarıyla tanındı. Yanlış pozitif tanısı (% 2.9) olan altı hasta hariç tutuldu. Sağ akciğer (% 57.8) ve alt loplara en sık etkilenen bölgelerdi. Rüptüre olgular ameliyat öncesi 40 hastada tanındı. Bunlardan onsekizi plevral kaviteye ve 22'si intrabronkiyal sisteme rüptüre idi. Cerrahi müdahale 193 hastaya yapıldı. Sıklıkla kullanılan cerrahi teknikler kistektomi ve kapitonaj (% 60) ya da yalnızca kistektomiydi. Lobektomi ve pnömonektomi yedi olguda uygulandı. Rüptüre kist hidatik nedeniyle ameliyat edilmiş olan sekiz hastanın geç dönem takibinde rekürrent hidatik kiste rastlandı.

Key words: Hydatid disease, Pulmonary.

Anahtar kelimeler: Hidatik hastalık, Akciğer

H ydatidosis (hydatid cyst disease), seen in every society, agriculture and cattle-breeding being the main supporting sources but having had inefficient precautions of environmental health and social physicians, is a kind of parasitic disease. It has been known since Hippocrates. In 1965, Hartman described the adult form of echinococcus granulosus in dogs.

Echinococcus has four subgenus, E. granulosus, E. multilocularis, E. vogelli and E. oligarthus (9). A hydatid cyst is the larval form of the parasite E. granulosus. Man as a herbivore is the intermediate host and harbors the larval form only. Man becomes infested with the parasite by eating food contaminated with the ova. Once inside the stomach, the chitinous coat is dissolved by gastric juices, and the hatched embryo passes through the wall of the duodenum into a radical of the portal vein. Some times the hatched embryo passes through the wall of the stomach or the duodenum into lymphatic channel, to the lungs via the thoracic and mediastinal lymph ducts, thus bypassing the liver. This route explains

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those cases of hydatid cyst of the lung in which the liver is free of the disease (2, 15) A pericyst, consisting of host fibrous tissue and compressed lung parenchyma, and forming around the cyst, plays a dual role: (1) pericyst guards the host against dissemination of the echinococcosis; (2) it feeds echinococcus and protects it against mechanical trauma.

Involvement of the various organs by the parasite is different in adults and children. In children, hydatid cyst is found in the lungs in 45-64 % and the liver in 28-35 % of those admitted, whereas, in adults, the lesions are common in the liver than in the chest (17, 22). Though the disease is seen in both sexes, hepatic hydatid cysts in women, pulmonary hydatid cysts in men are more common (4). Hydatid disease shows differences about residence, besides individual specialities such as occupations, ethnic group and religion, traditions and habits, and socioeconomical level. In our country, 60-87 % of cases contain the ones who come from rural area (7, 13).

The cyst may be solitary or multiple, unilateral or bilateral. Solitary cysts are generally primary, whereas multiple cysts may be primary or secondary. Multiple primary hydatid cysts are usually the same size, unless the host was exposed to infestations one more than occasion. The lungs is the second most common focus for this disease, after the liver. In some papers it is asserted that the disease localized more commonly in the lungs (8, 18). Pulmonary hydatid cysts are solitary in 70 percent and multiple in 30 percent. Solitary hydatid cysts prefer right lung and lower lobes (1, 11, 15, 16). Pulmonary hydatid cysts vary greatly in size. The resiliency of the lung tissue may allow the cyst to assume giant proportions without causing symptoms. It is assumed that the average growth of the cysts is probably 1 to 2 cm diameter per year (3).

Roentgenographic signs are usually diagnostic in pulmonary hydatidosis, while the Casoni's intradermal test, Weinberg reaction, and

eosinophilia are non specific and unreliable for diagnosis (1, 11, 15). Currently, computed tomography (CT) scan gives the most accurate finding (14).

Surgical operation, which should be performed early after establishing the diagnosis to prevent complicated cyst rupture or infection, is the principal method of treatment.

We present this paper because (1) pulmonary hydatid cysts are still a major problem in rural areas where cattle breeding and agriculture is the main source of living, and in our department one in every three thoracotomy has been performed for hydatid cyst; (2) hydatid cyst is seen frequently in the "active" age group 20 to 40 (47.7 %); (3) resectional operations such as lobectomy or pneumonectomy has been required in few of our cases (4.6 %) when compared with the others; and (4) surgical management is the sole treatment.

METHODS

Between 1978 to 1990, one hundred and ninety-seven patients with pulmonary hydatidosis were surgically treated in the Department of Thoracic and Cardiovascular Surgery of Erciyes University Medical Faculty. There were 100 (50.8 %) male; the ages ranged from 2.5 to 65 years, as shown in Table I.

Table I. Patients' age.

Age	n	%
2.5-9	24	12.2
10-19	41	20.8
20-29	44	22.3
30-39	50	25.4
40-49	19	9.7
50-59	16	8.1
60-65	3	1.5

n= Number of patients

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Other family members were affected in six cases (% 3); 117 patient were from rural environment (59.4 %); 80 patients (40.6 %) were living in cities; 55 patients (27.9 %) had histories of contact with dogs.

The clinical manifestations of 177 patients are shown in Table II. The remaining 20 patients (10.1 %) were asymptomatic and their diagnosis was purely accidental, and their lesions were noted following a routine chest graphy.

Table II. Clinical manifestations.

Symptoms	n	%
Cough	90	45.7
Chest pain	66	33.5
Dyspnea	32	16.2
Expectoration of cystic fluid	25	12.7
Fever	16	8.1
Anaphylaxis	1	0.5
Vomiting	5	2.8

n= Number of patients

Diagnosis was made routinely by chest roentgenogram, ninety-one of these patients also had ultrasonography and computed tomography (Table III).

For except ten, all patients were treated sur-

gically, apart from two patients who were managed mid-line thoracotomy by means of a posterolateral thoracotomy under general endotracheal anesthesia. In the patients, regardless of the condition of the cyst (ruptured or nonruptured), conservative surgical methods, i.e., cystectomy plus with or without capitonnage, were used. In all patients, the bronchial fistulas were sutured with fine sutures. The remaining cystic cavity was obliterated with absorbable pursestring sutures. We injected 10 % sodium chloride into the cyst and waited 5 to 10 minutes before removal. The surgical techniques used are implicated on Table IV. Operative and postoperative complications evaluated (Table V). Ten patients (5 %), nine of whom refused the operation and one had multiple cysts, were treated by mebendazole, but the long-term follow up results of these patients were not collected.

RESULTS

Twelve patients had admitted to the emergency department with the symptoms caused by perforated cyst. One of them was the patient who came with hydropneumothorax plus anaphylactic shock caused by blunt thoracic trauma. Spontaneous pneumothorax in two cases, hemothorax in one case, hydropneumothorax in four cases, and hydrothorax in four cases were determined. In 140 cases the cysts were intact and uncomplicated. Forty-one cysts were ruptured, 13 were infected

Table III. Diagnostic procedures.

Diagnostic tool	n	%*
X-ray	203	97
Ultrasonography	81	94
Computed axial tomography	10	100
Positive Casoni's skin test	7	85
Positive Weinberg reaction	34	66
Eosinophil cell (> 3 %)	60	67
ESR (> 20 mm/hr)	29	82
Pneumoperitoneum	12	100

* Positive result on diagnosis.

n= Number of patients

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Table IV. Surgical methods used in our patients with hydatid cysts.

Procedure	n	%
Resectional operations*		
Segmental and wedge resections	14	7.3
Lobectomy	6	3.1
Pneumonectomy	1	0.5
Supplementary segmental lobectomy ** (Barret-Thomas' method)	3	1.6
Cystectomy (Hydatidectomy)		
With capitonnage (Ugon's method)***	115	59.9
Without capitonnage	40	2.8
Enucleation of intact endocyst		
With capitonnage	8	4.2
Without capitonnage (Yacoubian-Dajani's method)	3	1.6
Plus pericystectomy (Perez-Fontana's method)	2	1.0

* Five patients, two of them also had a hepatic cyst, who underwent the resectional operations were also performed the pleural decortication at the same time.

** A supplementary segmental lobectomy was required during the operation due to unsatisfactory expansion of one segment of the diseased lobe;

*** Removing the cyst and obliterating the pulmonary cavity by sutures.

n= Number of patients

Table V. Complications.

Complication	n	%
Preoperative		
Ruptured cyst		
intrabronchial	22	11.2
intrapleural	18	9.1
bronchobiliary	1	0.5
Infected cyst	17	8.4
Ruptured plus infected cyst	13	6.4
Calcified cyst	1	0.5
Operative		
Ruptured cyst	2	1.0
Cardiac arrest	1	0.5
Postoperative		
Early		
atelectasis	12	6.0
pneumothorax	4	2.0
pulmonary hematoma	1	0.5
wound infection	2	1.0
Late		
recurrent cyst	8	4.0

n= Number of patients

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and ruptured (Table V).

The right lung was more commonly affected than the left, probably reflecting the relationship of its greatest circulation, and the lower lobe was a more common site than the upper (Table VI). Bilateral cysts were found

the disease was echinococcus granulosus. The findings of the roentgenograms are seen in Table VIII. There was no death in our series, and no instances of postoperative hemorrhage, empyema, or bronchopleural fistula. The mean duration until discharge was 8 days

Table VI. Distribution of hydatid cyst.

Cyst's location	Right	Left	Total
Upper	35	37	72
Middle/lingula	33	-	33
Lower	46*	46	92
Total	114	83	197

* Three of these patients had a cyst both at lower lobe of lung and over the dome of liver.



Figure 1. Solitary cyst in right.

in 5 percent of the cases (Figure 1), and multiple unilateral cysts were found in 12 percent (Figure 2-3). Table VII shows extrapulmonary cysts.

Except for three cases, which were caused by *E. alveolaris*, in all cases the agent causing



Figure 2. Ipsilateral multiple cysts seen in right lung.

for the uncomplicated patients and 15 days for the complicated patients. The immediate postoperative results and complications are summarized in Table V.

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Figure 3. Lateral roentgenographic image of Figure 2

The overall mortality rate was 0 percent in our series. Long-term follow-up information was available in all but 37 patients. There has

been no evidence of recurrence of thoracic hydatid disease in any of the long-term survivor, except eight patients. Three patients with localized cysts on the hepatic dome adjacent to the right hemidiaphragm were managed with thoracotomy transdiaphragmatically. Congenital bronchial cyst in three patients, pericardial cyst in two patients and cystic Schwannoma in one patient had been thought as a hydatid cyst preoperatively. The results of the chest X-ray of all 203 patients were normal postoperatively.

DISCUSSION

In hydatidosis, some serological diagnostic methods have been used to prove the diagnosis, evaluate the follow-up and prognosis after surgery or chemotherapy, study the prevalence, and evaluate the efficacy of the control methods. Counter immunoelectrophoresis (= CIEP), enzyme = linked immunoabsorbant assay (= ELISA), immunoelectrophoresis (= IEP), indirect hemagglutination test, radioimmunoassay and latex agglutination test (= LA) are usually used for diagnosis; complement fixation test, IEP, CIEP, and IgM-ELISA for follow-up; and double diffusion (DD5), CIEP, LA, ELISA for scanning (3). The Casoni's dermal test, the Weinberg reaction, and eosinophilia are not specific for the diagnosis

Table VII. Extrapulmonary cysts.

Location	n	%
Intra- abdominal		
Hepatic	19	9.3
Splenic	3	1.5
Abdominal cavity	1	0.5
Aortic wall	1	0.5
Intrathoracic extrapulmonary		
Pleural	1	0.5
Mediastinal	4	2.0
Pericardial	2	1.0
Left atrial wall	1	0.5
Thoracic wall		

n = Number of patients

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Table VIII. Radiologic findings.

	n	%
Non-complicated cyst		
Simple cyst	133	66.5
Sunset sign	6	3.5
Hydatidocystic respiration sign*	1	0.5
Complicated (ruptured) cyst		
Lotus-on-water sign	21	10.5
Hydro-aeric cyst		
Hydrothorax	18	9.0
Air crescent sign (pneumocyst)	5	2.5
Cumbo's sign (Ivanissevich's double-arch)	1	0.5
Incarceration of germinative layer	1	0.5
Aeric cyst	1	0.5

* This sign, longitudinal elasticity during deep breathing on fluoroscopy, was examined only in two patients.

n = Number of patients

(1, 11, 15). The Casoni's dermal test may also be positive in pulmonary carcinoma and tuberculoma (1, 21). At present these tests are discredited. Therefore, in our series we have not used these tests for diagnosis routinely.

Surgical operation is the principal method of treatment for intrathoracic hydatid cyst. It should be performed early after establishing the diagnosis to prevent complicated cyst rupture or infection, since oral mebendazole, albendazole or praziquantel was found to kill any soem of the scolices in the hydatid cyst and could not inhibit their growth. Surgical goals are: (1) the total eradication of the parasite; (2) the prevention of the cyst's from rupture and recurrency; and (3) the treatment of the residual cavity (2). Three methods are used in the treatment of pulmonary hydatidosis; resectional operations (segmental and wedge resections, lobectomy, pneumonectomy, Barret-Thomas' supplementary segmental lobectomy); hydatidectomy (Ugon's cystectomy with capitonnage, cystectomy without capitonnage); and enucleation of intact endocyst (enucleation with obliteration,

Yacoubian-Dajani's enucleation without capitonnage, and Perez-Fontana's pericystectomy (15, 17, 19). Conservative surgical methods that preserve lung parenchyma are usually preferred. The reasons of recommending conservative surgery rather than resection are as follows: (1) hydatid cyst is an expanding lesion, (2) multiplicity of the lesion is fairly common, and (3) the possibility of the patient developing more cysts which may require further operations in the future should always be kept in mind. Resections such as wedge resection, segmentectomy, lobectomy or pneumonectomy are indicated for some of the ruptured and infected cysts, in the existence of bronchiectatic changes secondary to infection or if the cysts are gaint, and it may be performed in selected patients. Lower lobe exicion is usually indicated in patients with thoracabiliary fistulas (16). Lobectomy is recommended for the followings; (1) if there is a single or multiple cyst filling a lobe, (2) if a cyst or cyst filling at least 95 per cent of the lobe, (3) if there is a serios infection which is not giving any response to the antibiotic therapy, and (4) if diffuse bronchiectasis, pul-

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monary fibrosis or serious hemorrhage happened as the cyst's sequele. Indications for pneumolobectomy included huge hydatid cysts occupying a considerable part of the lung lobe, complicated severe infections with hemorrhage, calcification of ectocyst, and the presence of many alveolar hydatid cyst (9).

As soon as arriving to the cyst, a quantity of cystic fluid is aspirated and the quantity of hypertonic saline (3% NaCl solution) is instilled into the cyst. Some surgeons use 1% formaldehyde, however it has a necrotizing effect on the tissues, particularly on bronchial mucosa (1, 3, 9). The other scolocidal agents are 3 percent hydrogen peroxide, cetrimide iodine and argentinrate of 0.5 percent (3, 9). A waiting period of about 5-10 minutes is necessary for the hypertonic saline solution to deactivate the scolices (1, 3, 15).

Although spilling of hydatid scolices is inevitable in most patients, except those are subjected to enucleation or pulmonary resection, implantation of the scolices in the pleural cavity is extremely uncommon (1, 12, 15). We administered a ten day antibiotic therapy before surgery in 26 case of complicated cysts which had been ruptured into bronchial system. This practice was suited with the literature (20).

Posterolateral thoracotomy is generally carried out rote of operation. Median sternotomy can be made in bilateral hydatid cyst (6). Longitudinal mid line sternotomy was done in 1% of our cases (Figure 4). The simultaneous removal of hydatid cysts of the right lung and liver has been reported (5, 14).

Recurrency changes between 2 to 12 per cent in the literature (2, 10, 21). In order to prevent the recurrencies; (1) antihelminthic therapy is to be begun preoperatively and is to be continued postoperatively; (2) if possible, operation is to be induced when the cyst is dead; and (3) cyst is to be removed without rupturing intraoperatively. Recurrency rate in our series is five percent. The larger hydatid

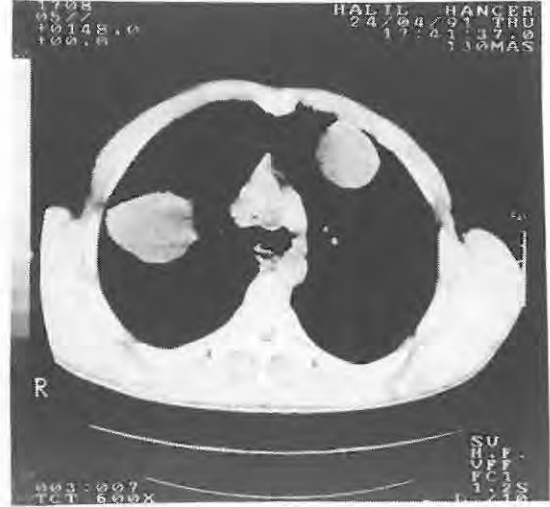


Figure 4. Bilateral cysts in CAT scanning. This patient with bilateral hydatid cyst was treated surgically via median sternotomy.

cyst is, the more rupturing chance. Despite of this, no relation has been found between size of cyst and postoperative recurrency (10). In ruptured cases, mebendazole (50-200 mg/kg each day for at least 3 months) plus cimetidine or albendazole (10-15 mg/kg each day for at least 30 days) are given postoperatively (3, 14).

The mortality rate of hydatidosis is reported as 0.4 to 3 percent in some publications (1, 9, 22). None of our patients died.

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