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Complicated Pulmonary Hydatid Disease

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Abstract

Background: Echinococcus is widely distributed parasite through temperate and subtropical region where sheeps are extensively raised. In Iraq, the disease is endemic and is considered to be one of the most serious helminthic diseases in the country.

Patients and Methods: 170 surgically treated patients with pulmonary hydrated disease. They were admitted to the thoracic and cardiovascular department at Ibn Al Nafees teaching hospital from the 1st of January 1998 to the first of October 1999.

Results: Eighty patients were only presented with different complications, forty-three females and thirty-seven males with a F:M ratio of 1.16:1. The mean age value was 26.7 year more than one third of patients were in the third decade. Sixty six percent of patients were from rural population. Regarding the occupation of patients, 47.5% of the patients were housewives whereas farmers constitute 15% only. Five patients gave history of previous pulmonary hydatid disease (two in the Right, two in the Left, and one bilateral). Analysis of the presenting symptomatology revealed hemoptysis in 68.7%. In order of frequency, right lower lobe was more frequently involved followed by left upper lobe, left lower lobe, right upper lobe, and right middle lobe. The diagnosis in the vast majority of patients was based on plain chest x-ray only. Fifty-five patients (68.7%) had a smooth postoperative period while twenty-five patients (31.25%) developed different complications. The overall mortality rate in this study was 2.5% (two patients), one of them died from irreversible shock at the time of re-exploration for bleeding, and the other patient died suddenly in the ward two weeks after surgery as a result of long immobilization following operation and clinically diagnosed as pulmonary embolism.

Conclusion: Early diagnosis of the disease by early request of chest x-ray for patients with chest symptoms and advice early operation to decrease complication rate.

Keywords: Hydatid cyst, thoracotomy, VATS, Echinococcosis

Introduction : Hydatid disease has a very long history; the word "hydatid" is Hellenic in origin and means a cyst full of water.[1,2] Hippocrates mentioned it in his Aphorisms and Al-Razi

referred to it in describing disease of the liver.[3] It's Rudolphi who gave it the name of vesicular Echinococcosis in 1808.[4]

Echinococcus is widely distributed parasite through temperate and subtropical region where sheeps are extensively raised. In Iraq, the disease is endemic and is considered to be one of the most serious helminthic diseases in the country.

Pulmonary hydatid cyst may be asymptomatic found incidentally on a routine chest radiograph.[2,5,11,14] mild to moderate symptoms may range from mild pain , cough , and hemoptysis to acute onset of rigor , fever , productive cough with purulent sputum or fragments of the membrane or daughter cysts described by the patient as grape skin if the cyst rupture into bronchial tree .[5,15] while severe symptoms may include dyspnea , hydro pneumothorax if rupture occurred into pleural cavity , anaphylaxis . Patients may also present with symptoms due to concomitant extra pulmonary hydatidosis.[5,14] The diagnostic workup of a patient with pulmonary hydatid disease may include one or more of : plain chest x-ray , ultrasonic study , CT-scan , MRI, Bronchoscopy , Serological tests , Blood tests , Parasitological tests . the usual radiological findings include: well defined circular homogenous opacity of simple intact HC, Signet ring sign, Double arc sign, Water lily or Camalote sign, Rising sun sign, Cavity with remnants, empty cavity, egg shell sign and air-fluid level or Hydropneumothorax in suppuration and abscess formation. Bronchoscopy may be performed for unclear consolidated shadow in chest x-ray, and may reveal a HC wall at the end of bronchial tree .[2,5,7,13,14]

immunological tests include: Casoni intradermal test, Weinberg complement fixation test, Latex slide agglutination test, Indirect hemagglutination test and immunoelectrophoresis . Eosinophilia is a common finding in peripheral blood smear. Finding of scolices, brood capsules, and daughter cysts in the sputum or pleural aspirate are used as parasitological tests.

In Iraq, the finding of a solitary opacity of homogenous density and clear-cut borders on a chest x-ray was a HC in 90% of patients .and it was a major cause of lung abscess in the country, still must be differentiated from other causes of abscess .[5]

Patients and Methods: A study was conducted in a group of 170 surgically treated patients with pulmonary hydatid disease. They were admitted to the thoracic and cardiovascular department at Ibn Al Nafees teaching hospital from the 1$_{st}$ of January 1998 to the first of October 1999. Eighty patients were only presented with different complications of pulmonary hydatid disease which were included in this study. Information and medical history were taken from each patient directly. These were the age, gender, occupation, residency, history of contact with animals, and history of previous history for hydatid disease.

Patients were clinically examined concentrating particularly on chest and abdomen.

Palin chest x-ray examination was taken for all patients, in both views, posteroanterior and lateral views and were utilized as a main diagnostic tool in all patients.

Investigations including complete blood picture, erythrocyte sedimentation rate, blood tests for sugar, urea, and blood grouping were done.

Some patients underwent pulmonary function tests and 60 patients underwent abdominal ultrasound study to exclude hydatid disease of the abdominal organs.

Bronchoscopy was performed for some patients with suspicion of tumor (4 patients), TB (one), and FB inhalation (one). One diagnostic bronchoscopy had showed shreds of a cyst in bronchus which was then confirmed by cytology and histopathology to be a HC wall.

Results: In this study there were eighty patients with complicated pulmonary hydatid disease, forty-three females and thirty-seven males with a F:M ratio of 1.16:1.

The mean age value was 26.7 year ranging from six years to sixty-five years. more than one third of patients were in the third decade and 4% of patients were in the first decade.

Sixty six percent of patients were from rural population, whereas the remaining were urban.

Complications included the followings: Ruptured HC, infected HC (abscess), empyema, broncho biliary fistula, hydropneumothorax, anaphylactic shock, and rupture into the bronchus as shown below.

Table (1): Types of Complications.

No.	Complications	Number	%
1	Ruptured Cyst	61	76.25
A. With Pleural Effusion (44) B. Without Pleural Effusion (17)			
2	Lung Abscess	5	6.25
3	Empyema	4	5
4	Broncho biliary Fistula	3	3.75
5	Hydropneumothorax	2	2.5
6	Spontaneous Pneumothorax	2	2.5
7	Anaphylactic Shock	2	2.5
8	Rupture Into Bronchus	1	1.25

Regarding the occupation of patients, 47.5% of the patients were housewives whereas farmers constitute 15% only, as shown in table (2).

Table (2)

Occupation	No.	%
Housewives	38	47.5
Students	12	15
Farmers	12	15
Workers	10	12.5
Employees	3	3.75
Non Employees	3	3.75
Children	2	2.5
Total	80	100

Five patients gave history of previous pulmonary hydatid disease (two in the Right, two in the Left, and one bilateral).

Sixteen patients gave history of previous pulmonary hydatid disease, nine in the liver, four in the spleen, two in the skin, and one in the kidney. The cyst was solitary in forty-four patients, and multiple in thirty-six patients.

Analysis of the presenting symptomatology revealed hemoptysis in 68.7%, cough 50%, sputum 47.5%, fever 43.7%, SOB 37.5%. other symptoms stated in Table (3).

Table (3):

Presentation	No.	%
Hemoptysis	55	68.7
Cough	40	50
Sputum	38	47.5
Fever	35	43.7
Dyspnea	30	37.5
Chest pain	25	31.2
Empyema	4	5
Cough bile	3	3.7
Hydropneumothorax	2	2.5
Spontaneous pneumothorax	2	2.5
Anaphylaxis	2	2.5
Skin cyst	2	2.5
Coughing grape skin like material	1	1.2

The time of presentation of our patients from the time of the starting of their symptoms was variable, from one week to ninety-six weeks with a mean of 13.8 weeks as shown in Table (4).

Table (4)

Period/weeks	No.	%
1	10	12.5
2	10	12.5
3	2	2.5
4	14	17.5
8	13	16.25
12	15	18.75
16	3	3.75
20	1	1.25
28	2	2.5
32	2	2.5
48	2	2.5
72	5	6.25
96	1	1.25
	80	100

The right lung was more frequently involved than the left, the right lung involved in forty-seven patients (58.7%), twenty-six in the left (32.5%), and seven patients has had a bilateral involvement (8.75%).

In order of frequency, right lower lobe was more frequently involved followed by left upper lobe, left lower lobe, right upper lobe, and right middle lobe.

The radiological appearance shown in table (5) and water lily sign which was pathognomonic of ruptured hydatid cyst was the most frequent radiological finding and had been seen in fifty-six patients followed by bilateral ruptured cysts which were seen in seven patients.

Table (5)

Radiological appearance	No	%
Water- Lilly	56	70
Bilateral ruptured	7	8.75
Abscess	5	6.25
Effusion	4	5
Peri vesicular Pneumocyst	4	5
Hydropneumothorax	2	2.5
Pneumothorax	2	2.5
Total	80	100

The diagnosis in the vast majority of patients was based on plain chest x-ray only. Blood tests showed the following: Hb was estimated in all patients, anemia was noticed in fifty patients (62.5%) and the leukocyte count was elevated in ten patients only. Casoni and Weinberg tests were not available.

Ultrasound study of the abdomen was performed in sixty patients and it has shown associated liver hydatid in nine patients (one of them was associated with kidney hydatid) , splenic hydatid in four patients and kidney hydatid in one patient .

Bronchoscopy was done as a diagnostic measure in seven patients due to aforementioned causes, otherwise bronchoscopy is not a routine investigation.

Pulmonary function tests were performed for nearly all elderly patients and patients having big lesions, when lung resections were anticipated.

Blood was prepared before surgery for all patients who were covered with antibiotics before surgery. Surgical procedures varied with the type of lesion and the level of complication. Conservative or limited procedures were performed for fifty-four patients in which cystectomy was performed for forty-seven patients and seven patients needed decortication as well. Resection was performed in twenty-four patients where lobectomy was done in eighteen patients, three patients needed segmentectomy and three others required pneumonectomy as shown in table (6).

Table (6):

Procedure	No.	%
Conservative operations	54	67.5
Cystectomy	47	58.75
Cystectomy + Decortication	7	8.75
Resection	24	30
Lobectomy	18	22.5
Pneumonectomy	3	3.75
Segmentectomy	3	3.75
Thoracoabdominal approach	1	1.25
Simultaneous removal of subdiaphragmatic HC during Rught thoracotomy	1	1.25
Total	80	100

Subdiaphragmatic liver hydatid cyst was dealt with at the same time of right thoracotomy in one patient. An extended thoracoabdominal approach for simultaneous removal hydatid cyst in right lung and liver was done in one patient only.

Hospitalization period was ranging from 3-23 days with a mean value of 7.8 days as shown in table (7).

Table (7)

Hospitalization	No	%
1-5	20	25
5-10	40	50
10-15	17	21.25
15-20		
>20	3	3.75
Total	80	100

Fifty five patients (68.7%) had a smooth postoperative period while twenty five patients (31.25%) developed different complications. These included persistent air leak , and collapse of affected lung (six patients) which necessitate keeping chest tube with controlled suction and physiotherapy .

Empyema occurred in six patients which mandates chest tube insertion and antibiotic cover and physiotherapy and it was enough in three patients to clear infection. The remaining three patients in whom conservative measured were not sufficient to resolve the problem until Elloeser flap was done. Wound infections occurred in five patients who were treated by frequent dressing and antibiotic cover.

Residual space occurred in three patients which was left untreated because it presented no respiratory embarrassment and followed with frequent chest x-ray and gradually the space cleared.

Pneumonia was developed in two patients and was treated by antibiotic and physiotherapy.

Bleeding occurred in two patients and was severe enough to undergo re-exploration, no active bleeding was seen in one of them, and only oozing from raw infected surface was seen. The other has died during re-exploration before identifying the bleeding source.

Pulmonary embolism occurred in one patient after two weeks.

The overall mortality rate in this study was 2.5% (two patients), one of them died from irreversible shock at the time of re-exploration for bleeding, and the other patient died suddenly in the ward two weeks after surgery as a result of long immobilization following operation and clinically diagnosed as pulmonary embolism as shown in table (8).

Table (8)

Post-operative Chest x-ray	No	%
Persistent air leak	6	7.5
Empyema	6	7.5
Wound infection	5	6.25
Residual space	3	3.75
Pneumonia	2	2.5
Bleeding	2	2.5
PE	1	1.25
Total	25	31.25

Discussion: Hydatid disease is a major health problem in the middle ease as well as in many other countries. In Iraq the disease is still endemic.[1,9]

The focus of this study was on the complicated cases which include the ruptured cysts with or without pleural effusion, lung abscess, empyema, broncho biliary fistula, hydropneumothorax, spontaneous pneumothorax, anaphylactic shock and rupture to the bronchus. In contrast to other study[20] where the criteria for pulmonary hydatid disease were as follow: 1. Huge cyst in size occupying 2/3 or more in one side of the chest .2. Ruptured hydatid cyst or cysts, with or without secondary infection.3. Multiple cysts involving both lungs, sometimes combined with liver involvement.4. The presence of hepatopleural and/or Hepatobiliary fistula.

In our study, out of the total number (170 patient) eighty were found to have complicated hydatid disease (47%) which was just exactly similar to other study.[20] However, a lower ratio was found in another studies .[21,22] The ratio of unruptured/ruptured cyst was 3:1 (20%) respectively.

The causes of increasing number of complicated cases in our study can be explained by: late seeking medical advice and neglection of patients to some complaints like cough and dyspnea, which are mostly treated as simple bronchitis, without achieving Chest- x-ray. Other explanation is that the disease is still endemic in rural areas where the people are of low socio-economic status and limited medical facilities. Last but not least, decreased number of mass radiography which was used before for screening of tuberculosis.

The youngest and the oldest patients in our study were 6 years and 65 years respectively ,[18,22] and when we compare the former with other studies we will find that they have a younger age 1.5 years and 2.5 years respectively , this may be attributable to the possibility of dealing with complicated cases and many of them lately presented.

We found in our study that 62.5% of patients were in their second and third decades of their lives (11-30years) with a mean of 26.7 year which is similar to others (60%) .[18,22,23] and this may be explained on the bases that the increase in size of the cyst and thus it ruptures easily in younger age .[27]

No significant difference in gender distribution was found in this study, the F:M ratio was 1.16:1 (43F, 37M) and this is comparable to other studies ,[11,22] however, another study showed a slight predominance of male to female ratio (1.3:1).[18]

In our study 66.2% of patients were coming from rural areas and the remaining were urban which is relatively lower than other study .[22] where 77% of patients were found in the rural areas while only 23% were found in urban districts, and this explained by the fact that the important three parameters of the disease is more often available in rural areas (dogs, sheep, and farms) in addition of low standards of living.

In our study, complicated cases were found to be more common in housewives and students (62.5%) which is comparable to other study .[22]

It's obvious that the disease affected people in the reproductive age group, thus it isn't only a major health problem, but an economic one as well.[5]

Hemoptysis was the most frequent complaint in this study (68.7%). This high percentage was found because our study focused on the complicated cysts only, however, hemoptysis can occur in both rupture and intact cysts, whereas in other study ,[22] cough and chest pain were the most frequent symptoms followed by expectoration of mucoid sputum, dyspnea and hemoptysis.

Cough, chest pain, and dyspnea were common in ruptured cyst, but they can also accompany intact cyst. Dyspnea was produced by moderate to large size cysts.[15] The cause of chest pain is not very clear, it might be related to extension of the lung cyst to the pleural surface causing reaction in the sensitive parietal pleura ,[20] when the cyst is close to the chest wall, it can cause dull pain due to pressure on the roots of intercostal nerves .[15] the causes of hemoptysis are also not very clear. The bleeding comes from adjacent bronchial mucosa and not from the peri cyst

layer. The most likely explanation is rupture of the small bronchial arteries, as occurs in bronchiectasis. High fever indicates an onset of infection or rupture of a cyst .[15]

Occasionally, spontaneous pneumothorax may be the presenting symptom in patient with ruptured HC into the pleura and it is due to intrapleural rupture of the cyst .[13]

Anaphylaxis is a rare presentation due to high antigenicity of hydatid fluid. Some patients presented with symptoms due to concomitant extra pulmonary HC.[7]

Five patients gave history of previous pulmonary hydatid and this not absolutely clarifies recurrent cysts, in fact, it might be a primary one, resulting either from a failure to detect them at previous thoracotomy or from reinfestation, or secondary as a result of contamination during previous surgery.[5]

The cysts were solitary in 60 patients (75%), and multiple in the remaining and this was consistent with other study and in general the percentage of multiplicity as reported in more than one series were between 14-22% .[11,22]

Seven patients presented with bilateral HC (8.75%), which appears slightly less than that in other series 9.1% and 16% respectively .[11,12]

The right lung was involved more than the left (58.7%,32.5%) respectively. In order of frequency the right lower lobe was more frequently involved followed by left upper , left lower , right upper , right middle lobes .this is due to greater circulation of right lung as compared with the left lung and many series showed comparable results.[1,2,7,13,15]

In our study, extra pulmonary hydatid disease was found in 20% which is a little bit higher than other series 12%, 15.1%.

Plain chest x-ray is diagnostic in 90% of cases. [2] In this study it is 91.2% with a similar figure in other studies, however, chest radiograph gave a correct diagnosis in 99% of patients in another study .[10,17,20,21]

in our study complicated cases presented in the following radiological appearance: water lily 70%, bilateral ruptured 8.72%, lung abscess 6.25%, empyema 5%, peri vesicular pneumocyst 5%, hydropneumothorax 2.5%, spontaneous pneumothorax 1.25%. the last two radiological appearances constitute the least common pictures when they are compared for example to waterlily owing to the fact that rupture of the cyst to the pleural space is much less common than to the bronchus.[23]

Bronchoscopy was done to seven patients (8.75%) in four of them , bronchogenic carcinoma was suspected , TB was the provisional diagnosis in one case and FB inhalation in the other and the diagnosis of these six cases were not settled until thoracotomy was done. Bronchoscopy helped in the diagnosis of one case only. The finding of shreds of a cyst which was whitish yellow membrane and histopathological examination of the material confirmed the diagnosis.

Anemia was found in fifty patients (62.5%) this is probably due to long standing history of hemoptysis which was the presenting symptom in 68.2% of the patients . our figure was much higher than other study which was 24.3% probably due to long standing infection of the cyst and hemoptysis.

Eosinophil count was found to be elevated in 10% of patients only, however, must authors agree that eosinophilia is of little diagnostic value since multiple parasitic infestations are common in the middle east including our country.[2,17]

Ultrasonography of the abdomen was performed in 60 patients (75%), and in 14 patients (17.5%) it showed abdominal HC, nine of them were occupying the liver, four in the spleen , and one in the kidney. This is comparable to that reported by other study (15.4%) with the majority in the liver. However other series showed higher percentage (32.5%).[7,22]

Medical treatment is of unproven value in controlling the disease like mebendazole and albendazole, praziquantel .[10] In our study, no single case was treated medically. But all cases were submitted to surgery which was the treatment of choice for pulmonary hydatid disease. Routine prophylactic antibiotic therapy just before surgery was given, in addition many patients received a course of postoperative antibiotics.

Median sternotomy wasn't utilized in any of our patients whether the disease was unilateral or bilateral. Instead, simple cystectomy and conservative surgery was performed in fifty-five patients (68.75%) including cystectomy alone in 47 patients (58.7%) and cystectomy/decortication in 8 patients (10%). Resectional surgery (as expected was higher than what was found in other studies in which resection was done in 20% of cases while other study showed lower figures (8.5%). This is probably because our series is concerned with complicated cases only, and it's done in 24 cases 30%. Lobectomy was the most common pulmonary resection performed and it constitute 22.5% (18 patients). Other forms of pulmonary resection, segmentectomy, and pneumonectomy were performed in three patients (3.7%) for each.[3,5]

Decision for resection (lobectomy) was taken for large ruptured cyst causing infection and permanent damage to the lung parenchyma and atelectasis of the lung. This was found in fourteen cases. resection was done for lung abscess and empyema in seven cases.

Pneumonectomy for broncho biliary fistula extending from liver hydatid through diaphragm and involving the right lung was done in three cases.

The period of hospitalization varies between 3-23 days with a mean of 7.8 days and this is consistent with other study.[18]

Fifty-five patients (68.7%) had an uneventful postoperative period with no complications. In the remaining 25 patients, complications were developed. Most of the complications were treated conservatively by the means of antibiotic cover, chest physiotherapy and keeping the chest tube according to the situation for a longer period of time or reinsertion of chest tube. this high rate of

complications in our study may be attributed to the fact that we are dealing with already infected and complicated HC.

Conclusion and Recommendations:

The disease is more common in rural area where standards for living are low and poor medical facilities. The disease regarded to be a major health and economic problem, being affecting people in their reproductive life.

There's an increasing number of complicated cases due to neglection of some chest symptoms and treated as bronchitis , which need proper assessment and evaluation especially in endemic area.

The increased number of pulmonary resections morbidity and mortality rates were due to already complicated cases.

Establishment of an educational program in sheep raising areas hopefully to break the life cycle of the parasite, and strict observation of the slaughter houses.

Early diagnosis of the disease by early request of chest x-ray for patients with chest symptoms and advice early operation to decrease complication rate.

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