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Management of Pulmonary Hydatid Disease: Review of 66 Cases from Iraq*

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Abstract

Background: Hydatid disease is a serious health hazard and a major problem to the community in Iraq. The disease is still endemic as witnessed in everyday surgical practice. The aim of this prospective study was to review the management of pulmonary hydatid disease (PHD) in two major thoracic surgical centers (Ibn-Alnafis and Medical City Teaching Hospitals), Baghdad, Iraq over one year period. Materials and Methods: Sixty six patients (38 females and 28 males) with PHD admitted and treated surgically in the Departments of Thoracic Surgery in the aforementioned hospitals were studied. Demographic and clinical features were obtained by direct patients' interviews and thorough physical examination. The diagnosis in the vast majority of patients was based on plain chest radiography while few had bronchoscopy. All patients had posterolateral thoracotomy for removal of pulmonary hydatid cysts (PHC). The clinical and radiographic findings as well as operative procedures and postoperative complications were reviewed. Results: The age ranged from two and a half years to 60 years with a mean of 22.3. Sixty percent of patients were in second and third decades. Most patients (n = 51, 77%) lived in rural areas. Housewives and students predominated. A positive family history was obtained in 5. Cough, chest pain, dyspnoea and haemoptysis were the main symptoms whereas 15 were acutely ill. Three patients presented with pathognomonic expectoration of laminated membrane and 2 had intra-pleural cyst rupture. The total number of cysts was 99 (61 intact, 50 solitary, and 55 unilateral). The right lung was more frequently involved (n = 64) and right lower lobe was on the top. Cyst size ranged from 3 to 25 cm with a mean of 8.5. The main radiographic appearance was the "full moon against dark sky" visible in 61.6%. Abdominal ultrasound was carried out in 40 patients who revealed 12 hepatic and one splenic HCs. Lung pre-

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serving surgery was done in 76 cysts (91.5%) while lobectomy was necessary in 7. Capitonnage was used in 16 cases only (19.2%). Two patients had lung decortication and four had trans-diaphragmatic removal of liver HCs. Few complications developed mostly managed conservatively. Reoperation was necessary in 4 patients (prolonged air leak, n = 2 and bleeding, n = 2). Two patients died (3%). Conclusion: PHC is endemic in Iraq, mainly diagnosed by plain chest radiography, and can be safely managed by lung preserving surgery with minimal morbidity and mortality.

Keywords

Pulmonary Hydatid Disease, Cyst, Echinococcosis, Iraq

1. Introduction

Hydatid disease (echinococcosis) is a worldwide zoonosis which is produced by the larval stage of the echinococcus (E) tapeworm [1] [2]. Human echinococcosis was first described in ancient times by Hippocrates as "cysts full of water" in a human liver [1]. Infection with *Echinococcus granulosus* is the most common form of echinococcal infection in humans [1] [3]. Though hydatid disease is endemic in many parts of the world [2], unfortunately it is classified among the most neglected parasitic diseases nowadays [1]. The parasite involves dogs (the definitive host) and sheep (the intermediate host) [1]. Man is an occasional intermediate host [1]-[3]. Ingesting embryonated eggs through hands, food, drinks or materials contaminated with parasite eggs infects humans; the larvae reach the blood and lymphatic circulation and reach the liver, lungs and other organs [4]. Cystic hydatid disease may develop in almost any part of the body [2], although the most commonly involved organs are the liver (60%) and the lung (30%) [3]. The two organs can be affected simultaneously in about 5% - 13% of the cases [2]. In Iraq, the disease is still endemic, although there are no accurate statistics. There are many national studies reviewing the surgical aspects of pulmonary hydatidosis [5]-[14]. While most authors agree upon the importance of plain chest radiography in the diagnosis, there is an ongoing debate on the role of CT scan and bronchoscopy [12]. There is also a controversy about the best method of management of the residual cavity after cyst removal [10]. There is a general agreement about the central role of surgery in the management of pulmonary hydatidosis and the adjuvant value of medical therapy [13]. Percutaneous aspiration has not been accepted or reported as a therapeutic option for pulmonary hydatidosis in Iraq, however, thoracoscopic removal of few PHCs has been reported once [14]. The aim of this study was to review our experience in the management of pulmonary echinococcosis in two major thoracic surgical hospitals, Baghdad, Iraq in view of the current literature.

2. Materials and Methods

Sixty six patients (38 females and 28 males) with PHD admitted and surgically treated in Medical City and Ibn-Alnafis Teaching Hospitals, Baghdad, Iraq over 1 year period were reviewed. Full history was taken particularly documenting age, sex, occupation, residence, contact with pet animals, previous surgery for and family history of hydatid disease and the presenting symptoms. Each patient was thoroughly examined physically noting abnormalities on chest examination as well as evidence of extra-pulmonary hydatidosis such as abdominal mass or organomegally. The diagnostic work-up consisted of plain chest radiography (PA and lateral views), complete blood picture, blood film morphology and an erythrocyte sedimentation rate (ESR). Abdominal ultrasonography was requested in patients with right lung hydatid cysts. Bronchoscopy was arranged for haemoptysis or suspicion of tumour. Serological tests were ordered infrequently. All patients were operated upon via posterolateral thoracotomy. A double-lumen endobronchial tube was used in all adult patients. Intact hydatid cysts were removed mostly by aspiration/evacuation technique after surrounding the cyst with Povidone-iodine (PVP-I) soaked packs taking care to avoid spillage of fluid into the operative field while ruptured cysts were removed by evacuation technique. Enucleation was occasionally used for small to medium-sized cysts. Excision was used for few small peripheral cysts. Bronchial fistulae were closed by non-absorbable cross-sutures. Most cystic cavities were left open. The standard policy was to preserve the lung parenchyma. Lung resection was done infrequently for certain indications. The postoperative morbidity and mortality were studied. Patients' informed consents and ap-

proval of the hospital Ethics Committee were obtained.

In this study, simple refers to intact, complicated refers to injured, ruptured or infected HCs. Evacuation refers to removal of the parasite by sponge forceps through adequate adventitial incision. Enucleation refers to incising the adventitia and removal of the cyst intact aided by lung inflation by the anesthesiologist. Aspiration/evacuation referred to preliminary decompression of the cyst followed by incision and evacuation of the contents of intact HC. Excision is used to describe removal of small peripheral HCs by excising the lung parenchyma together with the cyst. Capitonnage refers to obliteration of the cyst cavity after removing its contents and closure of bronchial fistulae. Prolonged air leak refers to persistent drainage of air via the chest tube after one postoperative week.

Statistical analysis was performed using z-test for 2 population proportions.

3. Results

The age ranged from two and a half year to 60 years with a mean of 22.3. Sixty percent of patients were in second and third decades as shown in **Table 1**. Most patients (n = 51, 77%) lived in rural areas. Housewives (n = 30) and students (n = 22) predominated while there were only 2 farmers. A positive family history was obtained in 5.

Table 2 displays the clinical presentations. Cough, chest pain, fever, shortness of breath and haemoptysis were common symptoms.

All patients had plain chest radiography. It was the main diagnostic tool. The radiological appearances are shown in **Table 3**; the commonest sign in intact cysts was rounded-oval opacity (n = 61) whilst water-lily was the commonest appearance in ruptured cysts (n = 18).

Table 1. Age and sex distribution.

Age (years)	Male, n	Female, n	Total, n (%)
2 - 5	4	1	5 (7.5)
6 - 10	5	2	7 (10.6)
11 - 20	11	10	21 (31.8)
21 - 30	6	13	19 (28.7)
31 - 40	2	7	9 (13.6)
41 - 50	0	2	2 (3)
51 - 60	0	3	3 (4.5)
Total	28	38	66 (100)

Table 2. Clinical presentations.

Presentation	Patients' n	Presentation	Patients' n
Cough	50	Anorexia	3
Chest pain	49	Loss of weight	3
Fever	37	Hoarseness of voice	1
Shortness of breath	35	Scars of previous operations	5
Haemoptysis	32	Hydropneumothorax	2
Signs and symptoms of toxicity	15	Bronchopleural fistula	1
Expectoration of grape skin-like material	3	Aspiration of hydatid fluid due to misdiagnosis of empyaema	1
Hepatomegally	7		

Table 3. Radiological appearances.

Radiological appearance	Patients' n	Radiological appearance	Patients' n
Homogenous rounded-oval opacity	61	Hydropneumothorax	2
Water-lily sign	18	Pericystic inflammatory reaction	4
Perivesicular pneumocyst	4	Mediastinal shift	2
Lung abscess	9	Elevated right hemidiaphragm	3
Residual cavity	4	Pleural thickening	1
Bilateral opacities	11		

The total number of cysts in this study was 99; 83 (46 intact and 37 ruptured) cysts were surgically removed. The remaining 16 cysts were not removed as the patients had bilateral disease with surgery done on one side but did not return for operation on the contra-lateral side.

The cysts ranged in size from 3 to 25 cm with a mean of 8.5. The cysts were solitary in 50 patients (75.8%) whilst the remaining 16 patients (24.2%) had multiple cysts (2-8 cysts per patient).

Table 4 reveals the lobar distribution of the cysts; the most frequently involved lobe was the right lower whereas the middle lobe was the least. Almost two thirds (64.5%) of the cysts were located in the right lung (statistically significant at $p < 0.05$).

Ultrasonic examination of the abdomen was done in 40 patients; it was normal in 27 cases while it revealed 12 liver HC and one cyst in the spleen.

Bronchoscopy was part of the diagnostic work-up of 4 patients only. All of them had haemoptysis and suspicion of tumour. It was diagnostic in a case of middle lobe HC by seeing a laminated membrane protruding through the offended bronchus.

Haematological tests revealed anaemia, elevated ESR and esinophilia in some patients; all were non-specific and insufficient to make a diagnosis.

The operative procedures used in this study are summarized in **Table 5**. Conservative surgery was used for most cysts (n = 76, 91.5%).

The postoperative complications vs. type of cyst and type of surgery are shown in **Table 6** and **Table 7** respectively. It is evident that the complications were higher after surgery for ruptured than for intact HCs but were fewer following lung resection than conservative surgery. By statistical analysis, empyema rate was higher after surgery for ruptured cysts whereas bleeding rate was higher following resection vs. conservative surgery.

The overall mortality was 2 patients (3%); one intra-operative cardiac arrest and one postoperative death due to septicaemia.

4. Discussion

4.1. Age and Sex

Most patients become infected with *E. granulosus* in childhood [7], as children are in an intimate contact with infected animals at play [5] [7] [15]. The cyst grows very slowly, so symptoms may not appear and the disease may not be diagnosed until many years later [7]. Five to twenty years may elapse before the cysts enlarge sufficiently to cause symptoms [2]. Hydatid disease is seen in subjects of any age and sex, although it is more common in those aged 20 - 40 yrs [16]. Sixty percent of our patients were in the second and third decades; this finding was similarly reported by Sarsam [8]. The youngest patient with PHD was one year old reported by Kavukcu *et al*. [17]. Our youngest patient was two and a half years old.

Fifty seven percent of our patients were females. Al-Mukhtar, from Iraq [18] and Yaghan *et al*. from Jordan [19] got a similar finding. A higher prevalence among females has also been reported in some other communities such as East Africa and Ethiopia [19]. In contrast, some authors reported slightly higher prevalence among males [17] [20]. "Women in rural areas are closely associated with domestic and farm duties, such as milking animals and cultivating crops while most men are military or government personnel" [19]. Moreover, Rokni [4] and Yaghan *et al*. [19] blame geophagia in poor children and pregnant women to be another source of infection.

Table 4. Lobar distribution of 99 cysts.

Lobe	Right, n (%)	Left, n (%)	Total, n (%)
Upper	21 (21.2)	14 (14.2)	35 (35.4)
Middle	9 (9)	-	9 (9)
Lower	34 (34.3)	21 (21.2)	55 (55.6)
Total	64 (64.5)*	35 (35.4)*	99 (100)

*The z-Score is 4.1219. The p-value is 0. The result is significant at $p < 0.05$.

Table 5. Operative procedures.

Conservative operation	Cysts' n	Operation	Cysts' n
1. Evacuation of ruptured cysts	29	Capitonnage	16
2. Aspiration/evacuation	31	Decortications	2
3. Enucleation	13	Exploratory thoracotomy without removal of the cyst*	1
4. Excision	3	Bilateral thoracotomies (Re-thoracotomy on the contralateral side after one month interval)	2
Resection (lobectomy)	7	Trans-diaphragmatic removal of hepatic HC	4

*One cyst was explored but not removed as it was adherent to the arch of the aorta simulating an aneurysm. The patient had no preoperative aortography to clarify the diagnosis.

Table 6. Postoperative complications vs. type of cyst.

Complication	In ruptured cysts, n = 37 (%)	In intact cysts, n = 46 (%)	Total, n = 83 (%)	$p < 0.05$
Prolonged air leak*	4 (10.8)	3 (6.5)	7 (8.4)	Not significant
Empyaema	6 (16.2)	0 (0)	6 (7.2)	Significant
Wound infection	4 (10.8)	1 (2.1)	5 (6)	Not significant
Atelectasis	2 (5.4)	1 (2.1)	3 (3.6)	Not significant
Postoperative bleeding*	2 (5.4)	0 (0)	2 (2.4)	Not significant

*Two patients with prolonged air leak as well as two patients with postoperative bleeding were managed surgically whereas others were managed conservatively.

Table 7. Postoperative complications vs. type of surgery.

Complication	Conservative, n = 76 (%)	Resection, n = 7 (%)	$p < 0.05$
Prolonged air leak	7 (9.2)	0 (0)	Not significant
Empyaema	6 (7.8)	0 (0)	Not significant
Wound infection	5 (6.5)	0 (0)	Not significant
Atelectasis	2 (2.6)	1 (14.2)	Not significant
Postoperative bleeding	1 (1.3)	1 (14.2)	Significant

4.2. Occupation

Forty five percent (n = 30) of our patients were housewives. The rate was even higher in studies from Iran [4] in which housewives had the highest rate of infection (51.3% - 75%). "Housewives, especially in rural areas, where the most infected cases can be found, have the highest chance of contact with the sources of infection.

Contact with contaminated vegetables, cleaning the house containing the dog faeces, and desire to eat soil (geophagy) as longing in pregnancy embrace the etiological issues" [4].

4.3. Clinical Features

The symptoms depend on size and site of the lesion. Slowly growing echinococcal cysts generally remain asymptomatic until their expanding size and their space occupying effect in the lung elicits symptoms [2]. The common symptoms in our study as well as in other studies were cough, chest pain and breathlessness [1]-[4] [6] [13] [17]. There are many case reports of chronic cough and progressive dyspnoea due to PHC misdiagnosed as bronchial asthma [1] [15]. Therefore, physicians working in endemic areas or treating patients from endemic areas should be aware of hydatid disease [1] [19]. "It is important to remember that HC may have very unusual presentations" [19]. Haemoptysis is associated with both simple and complicated cysts, although it is more common in the latter [8]. High fever characterizes the onset of infection or rupture of a cyst [5] [6] [15]. Symptoms of intoxication are common in suppurative cysts with fetid contents [6] [7] [15]. In the present study, 15 patients were acutely ill. Three of our patients had expectoration of grape skin-like material that was pathognomonic of intra-bronchial rupture of PHC [15] while two had tension pneumothorax due to the rare intra-pleural HC rupture [1] [9] [11]. Hoarseness of voice was observed once due to a big HC compressing the left recurrent laryngeal nerve. Anorexia and weight loss each observed in 3 patients were most likely related to low economic class of most patients particularly those living in rural areas (77%). Inadvertent aspiration of hydatid fluid was diagnostic in one case.

4.4. Diagnosis

The diagnosis is based on 1) history and geography, 2) imaging, 3) serology [21] and 4) bronchoscopy. Routine haematological and biochemical tests do not help in the diagnosis of hydatid disease [2].

4.5. Imaging

Hydatid disease is a dynamic entity with varying imaging appearances. Familiarity with imaging findings, especially in patients living in countries where this disease is endemic, provides important advantages in making the diagnosis. Hydatid cyst should be kept in mind when a cystic lesion is encountered anywhere in the body [22].

Plain chest radiography is the main diagnostic tool. Plain films will usually define the pulmonary cysts as rounded, irregular masses of uniform density, but they may miss the cysts in other organs [2].

PHCs may vary from 1 to 20 cm in diameter. Because of their compressibility, the lungs are the only organ in which HCs can grow so large. The high prevalence of PHCs in childhood can also be attributed for this feature [22].

There are 3 radiologic signs considered diagnostic of ruptured pulmonary hydatid cyst (PHC): perivesicular pneumocyst, double-domed arch, and water lily. Apart from these, every localized radiologic density seen in any patient more than 3 years of age in an endemic area should be looked on as a possible ruptured hydatid cyst [12].

In our study, plain chest radiography was the main diagnostic tool. None of the patients required a CT scan of the chest; however, CT scan was sometimes necessary in cases of complicated hydatid cysts or suspicion of tumours.

4.6. Bronchoscopy

Bronchoscopy is unnecessary in cases of ruptured PHC with a pathognomonic clinical picture, radiologic picture, or both [12]. However, it is indispensable when there is suspicion of tumor or when the radiologic picture is atypical. The source of hemoptysis (if present) can be traced [12]. The finding of laminated membrane in a bronchus draining the cyst cavity (be it lobar, segmental, or subsegmental) is diagnostic. A piece of the membrane can be obtained and examined with a hand lens or microscopically to confirm its laminated nature. The bronchial aspirate can be examined for the presence of hooklets [12].

In a huge intact PHC, bronchoscopy is hazardous as it can induce rupture of the cyst due to the effect of cough that usually follows bronchoscopy; moreover, the findings are usually not conclusive (mucosal erythema, external compression or normal bronchoscopy).

In patients with pulmonary opacities simulating tumours, bronchoscopy is necessary for diagnosis.

In our study, bronchoscopy was part of the diagnostic work-up of 4 patients only. All of them had haemoptysis and suspicion of tumour. It was diagnostic in a case of middle lobe HC by seeing a laminated membrane protruding through the offended bronchus.

4.7. FNAC

Although Das *et al*. from India had diagnosed a PHC and mediastinal HC by ultrasound-guided fine needle aspiration cytology [23], we believe that such a maneuver should not be attempted whenever there is suspicion of PHC as it causes cyst rupture with its dangerous sequels.

4.8. Surgery

The aim of surgery in pulmonary hydatid cyst is to remove the cyst completely while preserving the lung tissue as much as possible. Lung resection is performed only if there is an irreversible and disseminated pulmonary destruction [24]. Lung preserving surgery was done in 76 cysts (91.5%) while lobectomy was necessary in 7.

There are two methods for management of the residual cyst cavity: capitonnage that is closure of the cavity after removing its contents with a series of purse string sutures starting from the bottom outwards and cystotomy and closure of bronchial orifices leaving the cavity open [10]. Which option is better? This is a very controversial issue [10]. We share many authors their opinion that capitonnage is unnecessary [8] [10] [13]; we used it in only 16 of our cases (19.2%). Sarcasm [8] and Elhassani [6] recommended leaving the residual sac open to obliterate spontaneously after closure of bronchial fistulae and emphasized that no attempt should be made to suture it. Moreover, Sarsam believed that obliteration of the cavities of multiple cysts, particularly when large, may convert the remaining lung tissue into a collapsed and distorted mass, prone to infection and other complications [8]. Shehatha *et al*. (2008) studied 763 cases of thoracic hydatidosis managed by leaving all the cavities open and obtained good results [13]. In contrast, Al-Ali and Baram claimed that low complication rate was achieved following capitonnage in 72 cases of PHC [cited in 10].The study, however, had several limitations particularly the lack of a control group and deficient follow-up [10].

Not infrequently, thoracic surgeons are asked for the management of hydatid cysts located at the upper part (subdiaphragmatic location) of the liver. A thoracotomy provides better exploration and access to the cyst located in this area when compared to laparotomy [24]. Four patients were managed by this approach in our study.

4.9. Postoperative Complications

In this study, we observed that complications such as prolonged air leak, wound infection, empyema, atelectasis and bleeding seemed to be higher following surgery for ruptured than intact cysts. However, only empyema was statistically significant. Ruptured cysts are usually already infected and this may explain this finding.

Regarding complications vs. type of surgery (lung preservation or resection), apparently they were fewer after resection. However, the differences were not statistically significant except for bleeding which was higher after resection (This may be due to small sample size).

4.10. Other Therapies

There is a general agreement about the central role of surgery in management of pulmonary hydatidosis and the adjuvant value of medical therapy [13]. Percutaneous aspiration has not been accepted or reported as a therapeutic option for pulmonary hydatidosis in Iraq, however, thoracoscopic removal of few PHCs has been reported once [14].

Conflict of Interests

None declared.

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