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ORIGINAL ARTICLE

Clinical Characteristics and Management of Patients with Cystic Echinococcosis in Salmaniya Medical Complex – A Retrospective Descriptive Study

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Abstract

Introduction: Cystic Echinococcosis, commonly known as hydatid cyst disease, is a zoonotic disease caused by *Echinococcus granulosus* and commonly affects hepatic and pulmonary tissues. Although it is prevalent in some countries in the middle east, no epidemiological studies have been conducted to determine the epidemiology of cystic Echinococcosis in the Kingdom of Bahrain.

Methods: This is a retrospective descriptive study of all cystic echinococcosis cases diagnosed in Salmaniya medical complex between 2015 and 2020. Descriptive analysis of clinical, radiological, and histological data using frequencies and percentages was conducted.

Results: A total of 11 patients were identified with Cystic Echinococcosis. Most of them (n=9; 81.82%) reported a travel history to endemic areas and had liver involvement. While most laboratory data were within normal ranges, radiological investigations of hepatic lesions revealed characteristic features of hydatid cysts, including well-defined margins (n=9, 100%), hypoechoic liver cysts (n=9, 100%) and floating lily sign (n=5, 55.56%). All pulmonary (n=6, 100%) and most hepatic cysts (n=7, 77.78%) required surgical interventions. Reoperation rates were 33.33% (n=3) and 16.67% (n=1) for hepatic and pulmonary cysts, respectively.

Conclusion: Hydatid cyst is a rare disease in Bahrain and is seen mainly in patients who travel to endemic areas. Thus, extracting a detailed travel history is essential for the diagnosis of this disease. Clinical presentations are variable due to different tissues involved, natural history of the disease, and patients' characteristics. Although prolonged medical treatment and minimally invasive procedures are of value, surgical interventions are required in most cases.

Keywords: Bahrain, Cysts, Echinococcus granulosus, Humans, Zoonoses

Introduction

Echinococcosis is a zoonotic disease, i.e., transmitted from animals to humans, caused by the larval stage of taeniid cestodes of the genus Echinococcus. There are six recognized species of Echinococcosis. However, four are of public health concern, including *Echinococcus granulosus,* which causes cystic Echinococcosis, *Echinococcus multilocularis,* which causes alveolar Echinococcosis, *Echinococcus vogeli and Echinococcus oligarthrus,* which cause polycystic Echinococcosis. Among these forms, Cystic Echinococcosis (CE) is the most prevalent form.[1-2]

CE, also known as Hydatid cyst disease (HCD), is considered an increasing public health problem in some Middle Eastern and North African countries. In these regions, many studies have suggested that humans typically acquire the infection from sheep or dogs. [3-5]

The pathogenesis of CE is complex as the worm has definitive and intermediate hosts. Adult worms reside in the intestinal tract of the definitive hosts (like dogs and sheep), produce eggs that pass in the faeces to the external environment. Once the egg is ingested by an intermediate host (like humans), it hatches in their intestinal tract, releases oncospheres which move through the portal and lymphatic systems and reach different organs, particularly the liver and lungs, where they usually develop into larvae (also called hydatid cysts or metacestodes). However, other organs such as the abdominal cavity, kidneys, brain parenchyma, spleen, bladder, and bones can be involved.[1-2,6]

Patients with CE can be asymptomatic as echinococcus larva grows slowly (1 cm within the first six months, followed by an annual growth of 1 cm), which indicates that it might take years before developing into a large cyst. Nonetheless, it might lead to a wide range of clinical scenarios depending on the organ involved, size, number, and location of the cyst.[1-2,6]

This study aims to determine the epidemiology, clinical & radiological features, and treatment modalities among patients diagnosed with CE in Salmaniya medical complex (SMC).

Methods

Study setting, Participants and Selection criteria

This is a retrospective descriptive study of all patients who presented to SMC due to HCD between January 2015 and December 2020. Salmaniya medical complex is the primary government hospital in Bahrain that treats patients with communicable and complicated infectious diseases.

All patients with confirmed CE were included and patients with incomplete medical records were excluded. Since all patients with CE were included, sample size calculation was not performed. The Secondary Care Research Committee approved the study protocol at SMC, and written consents were obtained. All data were de-identified during the analysis.

The diagnosis of hydatid cyst was based on the pathological, serological and/or radiological features. A list of the cases with CE was obtained from the hospital's electronic medical records. Then, the patients' medical notes, along with their relevant laboratory and radiological investigation, were retrieved & tabulated. Baseline characteristics including demographics, risk factors, clinical presentations, laboratory and radiological investigations, and treatments were collected.

Radiological and Pathological characteristics

Radiological findings in abdominal ultrasound, chest radiographs and computed tomography (CT) studies, as well as pathological reports from serological and histological tests were included. Features including size (cm), number, involved lobe, presence of septations, regularity of borders, vascularity and calcification of the lesions were retrieved from the radiological reports.

Statistical analysis

Descriptive statistics were reported for all patients in this cohort. Continuous variables were expressed as means and standard deviations, while qualitative variables were presented as frequencies and percentages. Due to the descriptive nature of this study, inferential statistics were not performed.

Results

Baseline Characteristics

Baseline characteristics of the participants are presented in Table 1. A total of 11 patients were identified with HCD in the selected period. Most of them were Bahraini (n=9, 81.82%), females (n=8, 72.73%) and reported a history of travel to endemic areas (n=9, 81.82%). Diabetes Mellitus was the most frequent comorbidity among the patients, followed by Hypertension (36.36% and 27.27%, respectively).

Table 1: Baseline characteristics of the patients

Demographics	n (%)
Age (Mean ± SD)	41.45 ± 16.86
<20 years	1 (9.09)
20-39 years	4 (36.36)
40-59 years	5 (45.46)
60 years and above	1 (9.09)
Sex	
Male	3 (27.27)
Female	8 (72.73)
Nationality	
Bahraini	9 (81.82)
Non-Bahraini	2 (18.18)
Comorbidities	
Diabetes Mellitus	4 (36.36)
Hypertension	3 (27.27)
Hyperlipidaemia	1 (9.09)
Rheumatoid Arthritis	1 (9.09)
Risk factors	
Travel history to an endemic area	9 (81.82)

n = number of subjects; SD, standard deviation.

Clinical Presentation

Most patients presented with clinical features and were admitted through the emergency department (n=10, 90.91%), while only one subject was discovered to have the disease incidentally. Gastrointestinal symptoms such as abdominal pain, nausea and vomiting were more commonly encountered than respiratory symptoms like shortness of breath and cough (72.73% vs 54.55%). As shown in Table 2, fever was seen in two cases only, while an elevated level of inflammatory markers was the most common laboratory finding (n=6, 54.55%).

Table 2: Clinical and laboratory characteristics of the patients

Clinical Presentation	n (%)
Admission type	
Emergency	10 (90.91)
Elective	1 (9.09)
Fever	2 (18.18)
Gastrointestinal symptoms	**8 (72.73)**
Abdominal pain + mass	6 (54.55)
Nausea	2 (18.18)
Diarrhoea	1 (9.09%)
Respiratory symptoms	**6 (54.55)**
Shortness of Breath	4 (36.36)
Chest pain	2 (18.18)
Cough	2 (18.18)
Laboratory Findings	
Leucocytosis (> 12,000)	2 (18.18)
Neutrophilia (> 70%)	2 (18.18)
Eosinophilia (> 500)	0 (0)
Elevated Inflammatory markers (ESR*, CRP**)	6 (54.55)

*n = number of subjects; * ESR; Erythrocyte Sedimentation Rate; ** CRP; C-Reactive Protein*

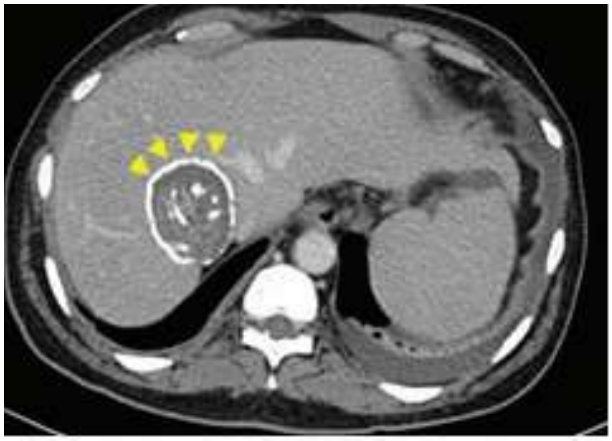
Radiological Features of Hydatid Cysts

Radiological features of hydatid cysts according to the involved organ are summarized in the Table 3. The mean size of hepatic hydatid cysts among the patients was 7.88±2.81 cm and 8.18±2.94 cm in hepatic ultrasound and computed tomography (CT) studies, respectively. All hepatic cysts were hypoechoic, avascular and had well-defined margins (n=9, 100%). Other standard features included septations and the classical floating lily sign (n=6, 55.56%). All pulmonary lesions were clearly defined and cystic (n=6, 100%) (Figure 1).

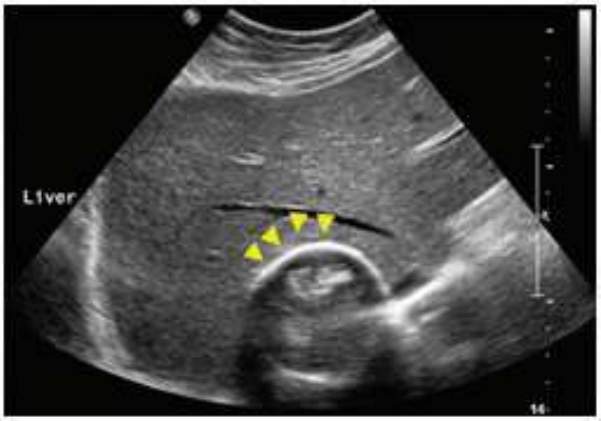
Table 3: Hepatic ultrasound and computed topography characteristics

Liver Ultrasound Findings (n=9, %)		Liver CT Findings (n=9, %)		Lung CT findings (n= 6, %)	
Size** (Mean ± SD)	7.88±2.81	Size (Mean ± SD)	8.18±2.94	Size (Mean ± SD)	5.08±1.94
Solitary cyst	5 (55.56)	Solitary cyst	4 (44.44)	Solitary	5 (83.33)
Right lobe	7 (77.78)	Right Lobe	8 (88.89)	Right lung	3 (50)
Septations	6 (66.67)	Septations	5 (55.56)	Lower lobe	4 (66.67)
Well-defined margins	9 (100)	Well-Defined Margins	9 (100)	Well-Defined Margins	6 (100)
Hypoechoic	9 (100)	Thick wall	6 (66.67)	Consolidation	3 (50)
Vascularity	0 (0)	Floating lily sign	5 (55.56)	Calcifications	0 (0)
		Calcification	3 (33.33)	Cystic	6 (100)

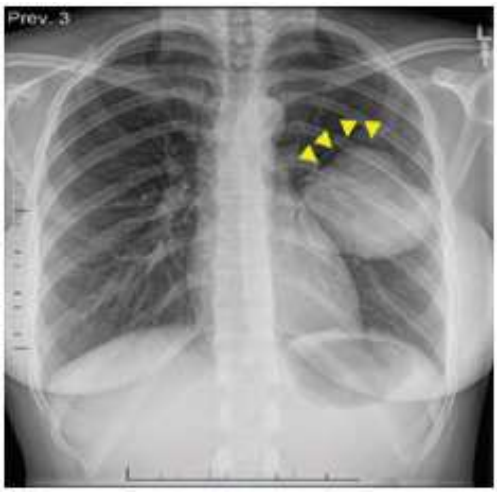
*n = number of subjects; CT, computed tomography; SD, standard deviation; **Size in centimetres.*



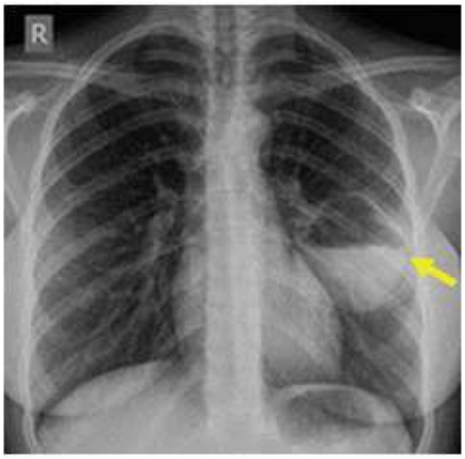
(A) An abdominal computed tomography section shows a large, thick, partially calcified wall cyst (yellow arrows) with internal calcification in the right hepatic lobe.



(B) A liver ultrasound section shows a well-defined hypoechoic lesion with peripheral curvilinear calcification (yellow arrows) in the right hepatic lobe.



(C) A chest radiograph shows a homogeneous irregularly rounded opacity in the left lung (yellow arrows).



(D) A chest radiograph shows a large thin-walled cavity with an air-fluid level (yellow arrow), which also displays irregular lines of the ruptured endocyst, also known as the "water-lily sign" due to a communication between the cyst and tracheobronchial tree.

Figure 1: Radiological features of hydatid cysts

Diagnoses and Treatment Modalities

Hepatic and lung hydatid cysts were found in 81.82% (n=9) and 54.55% (n=6) of the cases, respectively. Approximately half of the patients had both liver and lung involvements (n=5, 45.45%). While all patients with pulmonary CE were treated with a combination of medical and surgical treatments, three patients with hepatic CE were treated with either intervention alone (Medical treatment alone= 2 cases, surgical treatment = 1 case). Table 4 shows the diagnosis, treatment modalities and reoperation rates among participants. Due to partial regression of the cyst, reoperation was done for three subjects with hepatic involvement and one subject with pulmonary involvement.

Table 4: Diagnoses and treatment modalities

Diagnosis	n (%)
Hepatic CE* alone	4 (36.36)
Pulmonary CE alone	2 (18.18)
Hepatic and Pulmonary CE	5 (45.45)
Treatment	
Hepatic CE	**9 (81.82)**
Observation	0 (0)
Medical treatment alone	2 (22.22)
Surgical treatment alone	1 (11.11)
PAIR**	0
Open surgery	1 (11.11)
Medical and surgical treatments	6 (66.67)
Medication and PAIR	4 (44.44)
Medication and Open Surgery	2 (22.22)
Pulmonary CE	**6 (54.55)**
Observation	0
Medical treatment alone	0
Surgical treatment alone	0
Medical and Surgical Treatments	6 (54.55)
Reoperation	
Hepatic CE	3 (33.33)
PAIR after PAIR	2 (22.22)
PAIR after Open Surgery	1 (11.11)
Pulmonary CE	1 (16.67)
Open Surgery after Surgery	1 (16.67)

n = number of subjects; * Cystic Echinococcosis;

**PAIR: Puncture, Aspiration, Injection, and Re-aspiration

Discussion

The incidence of CE in this study is low compared to other regional studies, and most cases were travel related. Liver was the most affected organ by *E. granulosis* as per this study followed by the lungs.[4-5]

Hepatic CE

Clinical features of hepatic CE such as abdominal discomfort, palpable abdominal mass, nausea, and vomiting are mostly related to the pressure effect of the enlarging cysts. Less prevalent symptoms like obstructive jaundice might occur as the daughter cysts pass into the biliary tract. However, once the cyst ruptures, life-threatening presentations like peritonitis and anaphylactic shock can be elicited.[6-7]

Abdominal ultrasound remains the primary diagnostic tool for hepatic CE. Based on cyst activity and characteristics, there are five sonographic types of CE suggested by the revised Informal Working Groups on Echinococcosis (IWGE) classification in which CE1-CE2 are active forms and CE4-CE5 are inactive forms, as described in table (5).[7-9]

Compared to the abdominal US, an abdominal CT scan is more sensitive; features like border regularity, septations, and multivesicular rosette sign are more accurately seen in CT studies. Hepatic magnetic resonance imaging (MRI) is very useful, particularly if cysto-biliary communications are suspected.[9]

Table 5: WHO-IWGE 2001 and Gharbi 1981 sonographic classification of Cystic echinococcus

WHO-IWGE 2001*	Gharbi 1981	Description	Stage
CE1**	TYPE I	Unilocular anechoic cystic lesion with double line sign	Active
CE2	TYPE III	Multiseptated, 'rosette-like "honeycomb cyst"	Active
CE3 A	TYPE II	Cyst with detached membranes (water-lily-sign)	Transitional
CE3 B	TYPE III	Cyst with daughter cyst in a solid matrix	Transitional
CE4	TYPE IV	Cyst with heterogeneous hypoechoic/hyperechoic content. No daughter cysts	Inactive
CE5	TYPE V	Solid cyst with calcified wall	Inactive

*WHO-IWGE: World Health Organization-Informal Working Groups on Echinococcosis, ** CE: Cystic Echinococcus

Treatment of hepatic CE typically includes a combination of medical, PAIR (puncture, aspiration, injection, and re-aspiration), and or surgical via open or laparoscopic modalities.[7-9]

Surgical treatment is the mainstay of therapy and can be done endoscopically, under radiological guidance, or open surgery. It is indicated in large hydatid cysts with multiple daughter cysts, cysts with a high risk of rupture (superficially located single liver cysts), fistulizing, and infected cysts. It is contraindicated in dead, calcified, tiny cysts, in the presence of multiple cysts in multiple organs and whenever it is hard to approach the cysts.[7-9]

PAIR technique is used for cysts larger than 5 cm in diameter, CE1 and CE3a stages, deep liver cysts that are not amenable to drainage, recurrent cysts after surgery or chemotherapy, and if surgical intervention is needed contraindicated. As PAIR procedures carry a risk of spillage of cystic material into the peritoneum causing secondary Echinococcosis and anaphylaxis, it is contraindicated if the cysts have the potential to rupture, have biliary involvement or are inactive. When compared to surgery, PAIR is cheaper, requires a shorter hospital stay, and when combined with medical therapy; it is associated with considerable clinical and parasitological efficacy, fewer morbidities, lower rates of mortality, reoperation rate and disease re-occurrence.[8-11]

Medical treatment is used as adjunctive therapy before PAIR or surgical interventions. Medications will soften cysts' walls, reduce the risk of recurrence, and facilitate cysts' removal. Medical treatment with Mebendazole (40-50 mg/kg/day) or Albendazole (10-15 mg/kg/day) tablets should be initiated at least four days preoperatively and continued for 1-3 months after surgery. Medical treatment with no other interventions is mainly indicated for inoperable cysts due to underlying medical comorbidities of the patient, complex site of the cyst and small cysts. Medications should be extended to 3-6 months. This modality is contraindicated in large cysts that might rupture, inactive or calcified cysts, early pregnancy, and chronic liver diseases. Complete blood count and liver enzymes should be monitored every two weeks in the first three months, followed by every four weeks for 3-6 months.[8-9, 12]

Pulmonary CE

The pulmonary system is the second commonly affected system (17.2-22 % of cases). Due to the elasticity and compliance of the lungs, hydatid cysts typically grow faster in the lungs compared to other organs. Therefore, larger hydatid cysts are frequently seen in the lungs, where they can grow from a few millimetres to 10 centimetres in 1 year.[12] Similar to hepatic cysts, pulmonary cysts may remain asymptomatic for years, or they might cause cough, chest pain, and haemoptysis depending on their size, site, the pressure they create on the surrounding tissues and on whether or not they are ruptured. Most pulmonary hydatid cysts are simple; however, complicated cysts can occur due to pleural or bronchial cavity involvement.[12-13]

The diagnosis of pulmonary CE is based on imaging and serology. Blood tests may show eosinophilia, raised inflammatory markers and abnormal liver function tests. Indirect hemagglutination test and enzyme-linked immunosorbent assay (ELISA) are the most widely used serological methods for detecting CE but have high false-positive results. Meanwhile, ELISA is beneficial in following up with patients to detect recurrence.[13-14]

Imaging studies like plain chest x-ray and CT are necessary for the diagnosis, treatment, and follow-up. Non-complicated pulmonary hydatid cyst will most likely reveal homogenous rounded opacity with definite edges; however, in a ruptured cyst, communication might be found between the cyst and the tracheobronchial tree resulting in an air/fluid level.[13-16] When the cyst is completely collapsed, the endocysts can float over the cyst fluid, creating the water-lily sign (Figure 1).

Medical therapy of pulmonary hydatid cysts is indicated for smaller cysts, patients with contraindications to surgery, multiple cysts, recurrent cysts, and intraoperative spillage of hydatid fluid. Albendazole is favoured because it has a better bioavailability compared to Mebendazole. The recommended dosage of Albendazole is 10–15 mg/kg/day twice daily or Mebendazole 40–50 mg/kg/day three times daily for a minimum period of 3–6 months.[13, 15, 17]

Surgical intervention, the cornerstone treatment of pulmonary CE, aims to remove the entire hydatid cyst, preserve the lung parenchyma, and prevent intraoperative spillage of the larva. Different surgical techniques can be used, including enucleation, pericystectomy, cystotomy with capitonnage, open aspiration, and lung resection. This modality is indicated for cysts if they are extensive, superficial, at risk of rupture, infected, located in critical anatomical regions, and if there is a substantial mass effect. Postoperative follow-up includes monitoring liver functions and chest imaging monthly for the first three months, then every three months until the end of the first postoperative year.[15, 17-19]

The recurrence rate of pulmonary hydatid, which occurs mainly due to minor spillage of the hydatid cyst and/or inadequate treatment, can be as high as 22.0% and usually requires operative management. Nonetheless, successful resolution with medical therapy or PAIR was reported.[20-21]

This study has several strengths, including a comprehensive description of clinical and radiological characteristics of all CE cases over 5-year-duration with an assessment of their possible risk factors, the treatment modalities. According to the latest literature review, this is a pioneer study which delved into the epidemiology & clinical profile of hydatid cysts in Bahrain. However, this study is limited by its retrospective descriptive nature, and hence some relevant data like serological tests, original backgrounds, and treatment complications could not be obtained for some patients. The second limitation is that this study population was limited only to patients presenting to Salmaniya medical centre and did not include other hospitals in Bahrain, so the future prospective studies should be directed to include all patients with hydatid cysts at the national level to get a more accurate estimation of the disease incidence in Bahrain and to direct targeted preventive measures at the community level.

Conclusion

CE should be suspected in all patients who are found to have well-defined, hypoechoic cystic liver or lung lesions, mainly if there is a history of travel to endemic areas. A multi-speciality approach is usually required to manage symptomatic patients. Further studies are needed to determine the specific indications of PAIR and the exact role of serological tests in CE.

Conflict of Interests

Nil

Financial Support

Nil

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