



Hydatid disease among adults and children: it is time to worry!

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ABSTRACT

Background: Hydatid disease is a public health issue as it continues to produce substantial morbidity and mortality. The most common affected organ among adults is the liver and among children, is the lungs. Its treatment includes medical therapy, surgical intervention or percutaneous drainage. We aimed to identify the clinical, therapeutic and evolutionary features of hydatid cyst among adults and children.

Method: We conducted a retrospective study including all patients hospitalized in Pediatric and Infectious disease department for hydatid cyst between 1990 and 2018.

Results: Overall, we enrolled 71 patients, among whom 37 (52.1%) were male. The median age was 28 years [3-87years]. The revealing symptoms were fever in 19 cases (26.7%) and cough in 18 cases (25.3%). Hepatic hydatid cyst was the main site, noted in 45 cases (63.4%), among which adults represented 37 cases (52.1%). Hydatid serology was positive in 87.2% of the cases. The treatment was surgical and followed by medical treatment based on albendazole in 43 cases (60.5%). The median duration of treatment was 4 months [1-60 months]. The most common adverse reactions of albendazole were elevated hepatic enzymes and reversible alopecia noted in 7 (12.7%) and 5 cases (9%). The disease evolution was marked by the occurrence of recovery in 53 cases (74.7%).

Conclusion: Our study highlighted the burden of hydatid disease in our region, which remains endemic. It might be misdiagnosed since not only clinical symptoms are misleading, but also, imaging results can be concealing.

Keywords: adults, albendazole, children, echinococcosis, treatment

INTRODUCTION

Hydatid disease is a public health issue as it continues to produce substantial morbidity and mortality and it represents a burden from an economic point of view. The disease is spread worldwide with an annual incidence rate ranges from 1 to 200 per 100000 person (1). It is endemic to the Mediterranean countries, central Asia, northern and eastern Africa, Australia and southern America (1, 2) where the incidence rate can exceed 50 per 100000 person-years (3).

The most common affected organ, among adults, is the liver in 60-75% of the cases, while the lungs are affected in 15-30% of the cases. Less commonly, others sites are involved. However, the most common affected site in children is the lungs (2, 4). The hydatid cyst (HC) is asymptomatic and as it grows, it might cause compression of adjacent organ and lead to complications which explains clinical symptoms. Less frequently, the HC is accidentally discovered when imaging is made for other purpose. As soon as the diagnosis is established, the treatment is indicated. It includes medical, surgical or endoscopic interventional treatment depending on the size, the location of the cyst and possible complication associated.

In the light of the lack of recent and exhaustive data of HC in our region, we aimed to identify the clinical, therapeutic and evolutionary features of HC among both adults and children.

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MATERIALS AND METHODS

Study Design

We conducted a retrospective study including all patients hospitalized in Pediatric and Infectious disease department for HC over a 29-year period between 1990 and 2018.

Data Collection and Case Definition

Data were collected from the medical records of patients on pre-established sheets. We included all cases of HC, all sites combined. Patients who were lost to follow up were excluded from the study at enrolment. The diagnosis of HC was based on imaging result associated to a positive hydatid serology. In default, the diagnosis was confirmed after surgery. A negative hydatid serology didn't exclude the diagnosis.

Multiple organ involvement of HC was defined as concurrent hydatid involvement of 3 or more non-contiguous organs.

Recovery was defined as the absence of recurrence during the follow-up period.

The disease evolution was rated stable when the size of the HC was reduced or stabilized.

Progression of hydatid lesions was defined as the appearance of new HC or the increase in size of the HC.

Statistical Analysis

Statistical analysis was performed using SPSS 20. Qualitative variables were presented as numbers and percentages. Quantitative variables were carried out by means and standard deviation if they were normally distributed. Otherwise, medians and interquartile ranges (IQR) were performed.

RESULTS

During the study period, we encountered 71 patients with HC, among whom 37 patients (52.1%) were male. The median age was 28 years [3-87years]. We identified 48 adults (67.6%) and 23 children (32.4%) aged less than 18 years.

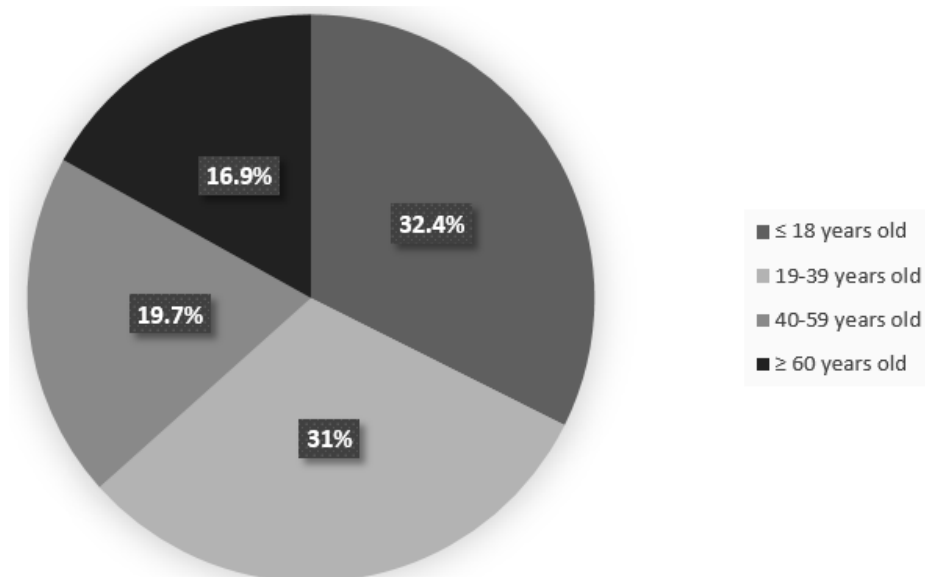


Figure 1: Distribution of cases of hydatid cyst by age groups

The incidence was 2.4 cases per year. According to residency, 59 patients (83%) came from rural areas. Forty-seven patients (66.2%) had a close contact with dogs and 4 patients (5.6%) had a family history of HC.

The revealing symptoms were fever in 19 cases (26.7%), cough and chest pain in 18 (25.3%) and 15 cases (21.1%), respectively. Nine patients (12.7%) were asymptomatic and imaging made for other reason revealed the diagnosis (**Table 1**). Hepatic HC was the main site, noted in 45 cases (63.4%), among which adults represented 37 cases (52.1%). It was followed by pulmonary HC in 33 cases (46.5%), among which children represented 20 cases (28.1%) (**Table 2**). Seventeen patients (23.9%) had simultaneous infection of the liver and the lungs. Unifocal HC was noted in 38 cases (53.5%) and multiple organ involvement in 13 cases (18.3%) (**Table 1**). The mean size of the cyst was 8 ± 3 centimeters.

Table 1: Clinical symptoms, laboratory results, treatment and evolutionary features of patients with hydatid cyst

Variables	Number	Percentage (%)	
Total	71	100	
Clinical symptoms	Fever	19	26.7
	Cough	18	25.3
	Abdominal pain	18	25.3
	Chest pain	15	21.1
	Dyspnea	8	11.2
	Hemoptysis	8	11.2
	Asymptomatic	9	12.7
Hydatid cyst presentation	Unifocal	38	53.5
	Bifocal	20	28.2
	Multiple organ involvement (≥ 3 organs)	13	18.3
Laboratory results	Leukocytosis	33	46.5
	Increased C-reactive protein	33	46.5
	Increased erythrocyte sedimentation rate	24	33.8
	hypereosinophilia	7	9.8
	Hepatic cytolysis	11	15.4
Treatment modalities	Surgical and medical treatment	43	60.5
	Surgical treatment	16	22.5
	Medical treatment	12	17
Disease evolution	Recovery	53	74.7
	Stable	13	18.3
	Progression of hydatid lesions	5	7

Table 2: Distribution of hydatid cyst site among adults and children

Hydatid cyst site	Adults		Children		Total	
	N	%	N	%	N	%
Total	48	67.6	23	32.4	71	100
Liver	37	52.1	8	11.2	45	63.4
Lungs	13	18.3	20	28.1	33	46.5
Kidney	10	14	1	1.4	11	15.4
Heart	4	5.6	1	1.4	5	7
Spleen	5	7	-	-	5	7
Soft tissue	4	5.6	-	-	4	5.6
Bone	2	2.8	-	-	2	2.8
Brain	-	-	2	2.8	2	2.8
Pancreas	2	2.8	-	-	2	2.8

N: Number; %: Percentage

Regarding laboratory investigations, increased levels of inflammatory markers, such as increased C-reactive protein and leukocytosis, were noted in 33 cases (46.5%). Hypereosinophilia was noted in 7 cases (9.8%). Hydatid serology was positive in 87.2% of the cases.

The HC treatment was surgical and followed by medical treatment based on albendazole in 43 cases (60.5%). The use of albendazole alone was indicated in 12 cases (17%) in front of multiple HC that were difficult to access (**Table 1**). The median duration of treatment was 4 months [1-60 months]. Among 55 cases (77.4%) who received albendazole, adverse reactions were noted in 12 cases (21.8%). Elevated hepatic enzymes and reversible alopecia, noted in 7 (12.7%) and 5 cases (9%), appeared after a mean duration of treatment of 2 ± 1.5 months and 7 ± 8 months, respectively. The disease evolution was marked by the occurrence of recovery in 53 cases (74.7%). Thirteen patients had a stable course of the disease (18.3%) (**Table1**).

DISCUSSION

Despite preventive measures, the hydatid disease remained relatively high in our region and continued to be endemic. It's a multisystem disease that affects commonly the liver in adults and the lungs in children (2, 4). Concomitant involvement of the liver and the lungs were reported in 16-23.1% of the cases (2), which was similar to our results. Other sites can be affected as well, such as the brain. Cerebral HC, a rare disease, doesn't exceed 2 % of the cases and is more frequent in children and young adults, representing 50 to 70% of the cases. However, all ages can be affected (5, 6). We noted 2 cases of cerebral HC, which were both among children. Renal HC is extremely rare, occurring in patients aged

between 30 and 50 years. The renal cyst is usually single, unilateral and lies in the cortex of the kidney (7, 8). In our study, it represented 15.4% of the cases. Cardiac HC was reported in about 0.02- 2% of the cases (9, 10). The most common location in the heart is the left ventricle, noted in 60% of the cases (10). The scarcity of these sites is explained by the passage of the parasite by two filters: the liver then the lungs, before reaching other parts of the body.

When the HC get enlarged, clinical symptoms begin. Cough, dyspnea, chest pain and hemoptysis are common presentation of pulmonary HC (4, 11). Hepatic HC remains asymptomatic for a while, then, as it grows, it can cause abdominal pain and discomfort. But, it can also cause obstruction of bile ducts, which manifest as jaundice and pruritus (11, 12). In our study, 25.3% of the patients had abdominal pain and cough.

Previous studies reported a predominance of males infected with pulmonary HC among both children and adults, with a percentage of boys up to 74% in children (2). This might be explained by the fact that boys are more active outdoors, which increases the odds of infection, whether after playing with an infected dog or consuming contaminated food. In our study, we noted a predominance of males as well.

Imaging results cannot always differentiate HC from other lesions. That's why, when combined to a positive hydatid serology, they may confirm the diagnosis of HC. Otherwise, the clinician is confronted by a dilemma and the diagnosis remains challenging. Previous studies reported a positive serology in 57% of the patients with pulmonary HC (13). In our study, it was positive in 87.2% of the cases. Cyst rupture or fissuration affects *Echinococcus* IgG positivity, which was very low in intact cysts. Also, the cyst size, location and vitality of larval cysts interfered in the results (12). Actually, pulmonary, cerebral and splenic HC cause less antibody response compared to hepatic and bone HC (13).

As for HC treatment, radical surgery was the most effective therapy (14), while conservative surgery was associated with significantly higher recurrence and morbidity rates (15). In fact, using Albendazole increased the incidence of spontaneous rupture of HC, but continuing medical treatment can result in complete recovery and eliminate the need for surgery (16, 17). Therefore, effective treatment includes both chemotherapy and surgery. Otherwise, medical therapy is the only choice in inoperative forms or multifocal hydatidosis (6).

Besides treatment, preventive measures are the key tool to control the hydatid disease and eventually eradicate it, that's why, policymakers should prioritize a control program and health education system as soon as possible.

Conflict of interest

The authors declare no conflicts of interest related to this report.

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