

Analysis of hospital records of children with hydatid cyst in south of Iran

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Abstract The clinical manifestations of hydatidosis are various and related to anatomic location. Defining frequent symptoms and signs of the disease is imperative for early management of it. The aim of this report was to analyse the clinical features of infected children with hydatid cysts located in different organs. In this study, medical charts of 57 children between 3 and 16 years of age with hydatid cyst admitted to Pediatric Wards of Nemazee Hospital were evaluated over a 12 year period (from 2003 to 2014, prospectively). All the epidemiologic, clinical, paraclinical and therapeutic data were collected. The frequencies of hydatidosis in males and females were 42.1 and 56.1%, respectively. Hydatid cysts were found in the liver and lungs in 59.6 and 33.3% patients respectively and 2 patients had an asymptomatic cyst in the heart with concomitant liver and lung cysts. The right upper quadrant pain (100%) was the most common symptom in the liver cysts. Phlegm (78.9%), Dyspnea (57.9%), acute (47.4%) and chronic cough (47.4%) were mostly seen in lung hydatid cysts. Some symptoms such as fever (68.4%) and weakness (59.6%) were the most common presenting symptoms in both groups. All children were treated through surgical approaches plus medical treatment. In the present report, liver was the most common site of involvement in children. Liver hydatidosis should be considered in children with upper quadrant pain and

pulmonary hydatidosis in children complaining of phlegm and dyspnoea.

Keywords Hepatic echinococcosis · Pulmonary echinococcosis · Heart echinococcosis · Diagnosis · Child

Introduction

Hydatidosis is a parasitic disease in humans and animals caused by the larval form of *Echinococcus granulosus* tapeworm (McManus et al. 2003). Humans as intermediate hosts can be affected through ingesting tapeworm eggs, which are transmitted by contact with dogs and domestic animals or contaminated vegetable and water (Mandell and Dolin 2005; Polat et al. 2003; Turgut 2001).

Cystic echinococcosis, a public health problem is usually endemic in many areas of the world such as South America and Mediterranean area (Moro and Schantz 2006; Yang et al. 2006). Hydatid disease is endemic to Iran and leads to 1.0% of all surgical admissions (Ahmadi and Badi 2011). A review of the incidence of hydatidosis from 1996 to 2008 showed that the seroprevalence of human hydatidosis varies from 1.2 to 21.4% in different parts of Iran (Rokni 2009).

The Hydatid cyst usually grows in size slowly and is diagnosed in only 10–20% of patients younger than 16 years of age. Most of the adults have acquired it during childhood while symptoms and signs are absent (Kammerer and Schantz 1993; Todorov and Boeva 2000). Hydatid cyst involves different organs, but liver and lungs are typical regions (Bal et al. 2008). In the liver, pain in the right upper quadrant is the most common symptom and in complicated hepatic hydatidosis, symptoms such as fever,

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jaundice, or anaphylactic symptoms appear. In the lung, a nonproductive cough, bloody phlegm, fever, and chest pain are the most common symptoms, although in most cases, lung hydatid cysts are asymptomatic (Sayek et al. 2004). Rupture is the main complication leading to many conditions ranging from mild allergic manifestation to anaphylactic shock (Moro and Schantz 2009).

The presenting symptoms and signs of hydatidosis are often non-specific making the diagnosis difficult; therefore, clarification of the disease symptoms in children for better diagnostic and treatment approaches is important. In the present report, we report the clinical and para-clinical information of children admitted with hydatid cyst.

Patients and methods

This cross-sectional descriptive study was performed on children with the final diagnosis of hydatid cyst admitted to Nemazee Academic, Referral Hospital, affiliated with Shiraz University of Medical Sciences, Iran, from 2003 to 2014 prospectively.

All the epidemiologic, clinical, paraclinical and therapeutic data of 3–16 years old patients were collected from their medical charts. The patients over 16 years of age were excluded from the study. The study was approved by the ethical committee of Shiraz University of Medical Sciences, Shiraz, Iran.

The information including sex, age, demographic data, history of contact with a dog or domestic animals, hydatid cyst features (location, type and size), symptom distribution and duration, complications, clinical features, and the medical treatment were contained in the questionnaire and recorded by a pediatric resident physician.

Data were expressed as percentage. For cyst location, percentages were calculated out of the total number of patients and for symptoms it was calculated based on the frequency of each symptom relative to the number of patients with liver or lung cyst.

Results

In this report, we analysed the records of 57 children with the final diagnosis of hydatid cyst. The patients were between 3 and 16 years of age and included 24 (42.1%) males, 32 (56.1%) females, while gender of 1 case (1.75%) was not reported. The average age of children was 11.5 ± 6.1 years (ranged from 3 to 16 years). The patients residing in urban areas were 19 (33.3%) and in rural areas, 38 (66.6%). Among them, 39 (68.4%) had a history of contact with dogs or domestic animals.

Hydatid cysts were solely located in the liver and lungs of the patients. Out of 57 patients, 34 had liver (59.6%), 19 had lung (33.3%) cysts, 2 had more than one organ involvement (3.5%) and in 4 patients cyst location was not known. Also, two patients (3.5%) had cysts in their heart with concomitant cysts in the liver and lung. Table 1 shows the distribution of cysts in different organs. Right lobe of liver, left lobe of liver and upper lobe of the left lung was infected in 24 (42.1%), 10 (17.5%) and 8 (14%) patients, respectively.

Fever (68.4%) and weakness (59.6%) were the most common symptoms among all patients with hydatid cyst. In addition, out of 57 children, 30 (52.6%), 23 (40.3%) and 18 (31.6%) patients also complained of loss of appetite, nausea and vomiting and chills respectively. The right upper quadrant pain (100%) and phlegm (78.9%) were the most frequent symptoms in the liver and lung hydatidosis, respectively (Tables 2, 3). Among a wide range of complications associated with the hydatid cyst, the cholestasis and rupture occurred in 1 (1.75%) and 7 (12.3%) patients with hepatic cyst, respectively. Patients with heart hydatid cyst were asymptomatic in this study.

Through ultrasonography, three different types of hydatid cyst were identified in the present study; the hypoechoic cyst in 27 (47.4%), mixed cyst (solid–liquid) in 16 (28.1%) and solid cyst in 1 (1.75%) patient were the most common types, respectively. Also, the type of cysts in 13 children (22.8%) was not reported. The size of cysts varied from 10 to 185 mm in length (mean: 52.8 ± 36.8 mm) and 10 to 164 mm in width (mean: 46.1 ± 35.3 mm). The size of the heart hydatid cyst in one patient was 30×30 mm and the other one had two cysts in the heart with the size of 14.7×7.5 mm and 16.4×12 mm. Surgical intervention was used for the treatment of all (100%) children. In addition, anti-Echinococcus drugs including albendazole and mebendazole had been prescribed for them.

Discussion

Echinococcosis is one of the most important parasitic infections in Iran, with considerable impacts on economic and public health areas (Rokni 2009). Contrary to other reports, we observed that the most frequent site of hydatid cyst in children is the liver (59.6%) and lungs (33.3%). It has shown that the lung tissues have compressible nature, allowing the cyst to become larger in size before being symptomatic in children (Kilani and El Hammami 2002). Furthermore, many previous studies denoted that lungs are the most common region for hydatid cyst in children while the liver is the most frequent site in adults (Beggs 1985; Karaoglanoglu et al. 2001; Mandell and Dolin 2005). A

Table 1 Distribution of hydatid cyst in different organs

Location	Number of patients (%) N = 57	
Liver (N = 34)	Right lobe of the liver	24 (42.1)
	Left lobe of the liver	10 (17.5)
Lung (N = 19)	Lower lobe of the right lung	5 (8.8)
	Upper lobe of the right lung	5 (8.8)
	Lower lobe of the left lung	1 (1.7)
	Upper lobe of the left lung	8 (14)
Heart (N = 2)	Left ventricle	1 (1.7)
	Right ventricle	1 (1.7)
Data not available (N = 4)		4 (7)

Table 2 Symptoms and signs distribution in children with liver hydatid cyst

Symptom of liver hydatid cyst	Number of patients (%) N = 34
Symptoms	
Right upper quadrant pain	34 (100)
Flank pain	3 (8.8)
Abdominal pain	1 (2.9)
Signs	
Jaundice	7 (20.6)
Hepatomegaly	7 (20.6)

Table 3 Symptoms and signs distribution in children with lung hydatid cyst

Symptom of lung hydatid cyst	Number of patients (%) N = 19
Symptoms	
Phlegm	15 (78.9)
Dyspnea	11 (57.9)
Acute cough	9 (47.4)
Chronic cough	9 (47.4)
Hemoptysis	9 (47.4)
Chest pain	8 (42.1)
Signs	
Acute respiratory distress	4 (21.1)
Empyema and pleural effusion	4 (21.1)

study on hydatid cysts in Tunisian children revealed that hydatid cysts were primarily located in the lung (61.8%) and the liver was the second common site (34.85%) (M'rad et al. 2012) and in a similar study on children in the northwest of Iran, 67.8% of patients had the cyst in their lungs (Aslanabadi et al. 2013). However, some studies have yielded similar results to ours; in Serbia, 60.7% of children with echinococcosis, had liver hydatid cyst

(Djuricic et al. 2010). The present analysis showed that the Right lobe of the liver was more vulnerable to hydatid cysts, 42.1% patients had right lobe effected, which was in compliance with an earlier report (Mirshemirani et al. 2011). In the present report, the upper lobe of the left lung was more frequently affected (14%), while in other studies, lung cysts were most commonly located in the right lower lobe (Aslanabadi et al. 2013; Mirshemirani et al. 2009; Montazeri et al. 2007). Although the incidence of hydatid cyst in the heart is rare and seen in only 0.2–2% of all hydatid cases (Yekeler et al. 1993; Yilmaz et al. 2000), it has high mortality if left untreated. In our report, among patients with hydatid cyst 2 (3.5%) patients had cyst in the heart; located in right and left ventricle. Hydatid cyst of the heart is commonly observed in the left ventricle (50–60% of cases), interventricular septum (10–20%), right ventricle (5–15%), pericardium (10–15%), and right or left atrium (5–8%) (Kaplan et al. 2001).

The majority of patients (68.4%) in the present study, had a history of contact with animals and there was a greater frequency in patients living in rural areas (66.6%) versus urban areas (33.3%) which was in agreement with previous studies (Derfoufi et al. 2011).

Symptoms of hepatic hydatid disease are highly variable, associated with the location and type of the cysts and their complications (Romero-Torres and Campbell 1965) and the patients with small cysts may remain even asymptomatic (Santivanez and Garcia 2010). Symptomatic cysts may present with increased abdominal girth, hepatomegaly, palpable lesions, vomiting or abdominal pain, but, more serious complications may develop in critical areas such as bone marrow or reproductive system. Jaundice is a rare complication of hepatic cyst (Blanton 2011). Mirshemirani et al. studied hepatic hydatid cysts in under 14 years of age children. Abdominal mass (50%), jaundice and nausea (2%) had the highest and lowest prevalence (Mirshemirani et al. 2011). In our report, pain in the right upper quadrant was seen in 100% of the patients with liver cyst. Other symptoms and signs related to hepatic cyst, including jaundice, hepatomegaly and flank pain were seen in 20.6, 20.6 and 8.8% of the patients, respectively.

Chest pain, cough and hemoptysis are the most common symptoms which manifest due to hydatid cysts of the lung (Blanton 2011). In the present report, in lung involvement, phlegm (78.9%), dyspnea (57.9%), acute and chronic cough (47.4%) were the most frequent symptoms. A survey in Tabriz on children with hydatidosis reported that chest pain and cough are the main symptoms (Aslanabadi et al. 2013).

Different stages of cyst development cause variations in ultrasound imaging (Garcia et al. 1988). It was suggested that solid echo patterns are associated with mature cysts (Beggs 1983). We found three types of hydatid cyst in

ultrasonography imaging with solid cyst having the lowest frequency (1.75%).

Seven patients (12.3%) with cyst rupture were found in our study. Cyst rupture, which can be spontaneous or by trauma, may lead to anaphylactic reaction which may occur as a result of penetration of cyst contents into the peritoneum or blood circulation (Sanei et al. 2008).

Heart echinococcosis usually is asymptomatic and may present with nonspecific symptoms. However in later stages, in the case of cyst rupture into the heart, different symptoms appear depending upon the location of the cyst (Madariaga et al. 1984; Noaman et al. 2017; Ünlü et al. 2002). In the present report, patients with heart hydatid cyst were asymptomatic and symptoms such as dyspnea and chest pain that were seen in one patient could be explained by lung cyst. Patients were suspected of having heart hydatid cyst after detecting calcification in heart area, on chest x-ray.

All children (100%) in our study underwent surgery and the definitive diagnosis was made after operation. Although serological tests were also performed for patients, as hydatidosis is endemic in our region and due to false positive cases, positive results could not necessarily confirm the disease. Albendazole chemotherapy is the preferred treatment for small cysts (<5 cm in diameter) and ultrasound—or CT—guided PAIR is used for larger cysts in the early stage. Surgery is the main treatment for complicated cysts (ruptured cysts, cysts communicating with the biliary tract, large pulmonary cysts, or cysts of the central nervous system or bones (Kliegman et al. 2015). In a retrospective study in Tehran, conservative surgical treatment was performed in 90% of children with pulmonary hydatid cyst and no mortality and recurrence occurred (Mirshemirani et al. 2009). Also, Chautems et al. (2005) showed that surgical therapy leads to complete resolution of liver cyst in complicated cases.

Conclusion

In the present report, liver was the most involved organ in majority of children that was inconsistent with few earlier reports. The most common symptoms in the liver such as right upper quadrant pain (100%) and in lung hydatidosis such as, phlegm (78.9%) and dyspnea (57.9%) could be helpful in early diagnosis of the disease. All the children were surgically treated and were also prescribed anti-echinococcus drugs.

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Compliance with ethical standards

Conflict of interest The authors declared that they have no conflict of interest.

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