Liver Hydatid Cyst in Children (A 14-year Review)

Alireza Mirshemirani*, MD; Ahmad Khaleghnejad, MD; Jaefar Kouranloo, MD; Nasser Sadeghian, MD; Mohsen Rouzrokh, MD, and Shaghayegh Hasas-Yeganeh, MD

Pediatric Surgery Research Center, Mofid Children's Hospital, Shahid Beheshti University of Medical Sciences, Tehran, Iran

Received: Jul 11, 2010; Final Revision: Jan 12, 2011; Accepted: Mar 02, 2011

Abstract

Objective: Hydatid disease is still an important health hazard in the world. This disease is a parasitic infestation which is endemic in many sheep and cattle raising areas such as in Iran. The aim of this study was to evaluate the clinical appearance, diagnosis, and treatment of liver hydatid cyst in children.

Methods: This retrospective study evaluated 100 patients who were referred to Mofid Children's Hospital with liver hydatid cyst from March 1996 to March 2010. Medical records of 1 to 14 year old patients who had definitive liver hydatid cyst were included and analysis of variables such as age, gender, symptoms, diagnostic investigation, operative technique, hospital stay, mortality, morbidity and outcome of treatment were evaluated.

Findings: The patients consisted of 54 boys (54%) and 46 (46%) girls with an age range of 1-14 years (mean 11.8±4.6). The incidence rate increased by age. The patients had totally 110 cysts, right pulmonary lobe 81 (73%) cysts and left side had 29 (27%). Abdominal mass was the most common (50%) symptom. Abdominal sonography gave correct diagnosis in 94 (94%) patients. Conservative surgical treatment was carried out in 98 children. Two patients were treated medically as the cysts were small and calcified. The most common complication was wound infection in 3 cases. Mean length of hospitalization was 9 days. In 100% of our patients the type of parasite was *Echinoccocus granulosus*. The morbidity rate was 12% (prolonging external catheter drainage in 12 patients). There was only one (1%) mortality and 2 (2%) recurrences.

Conclusion: Due to the less invasive and high accuracy of liver sonography in diagnosis of hydatid cyst, we recommend it as the method of choice for the diagnosis in endemic regions. Surgery is the method of choice for treatment.

Iranian Journal of Pediatrics, Volume 21 (Number 3), September 2011, Pages: 385-389

Key Words: Liver Hydatid Cyst; Abdominal Mass; Diagnosis; Treatment; Children

^{*} Corresponding Author;

Introduction

Hydatid disease is a public health hazard in $Iran^{[1,2]}$. It is common in rural area where dogs and

cattle are kept. The primary hosts for the *Echinococcus granulosus* tapeworm are dogs and canines^[3]. They produce eggs in the intestine that pass in the stool. Eggs are ingested by intermediate hosts such as sheep and cows.

Human become accidental hosts by eating tapeworm eggs^[4]. In adults the liver is the most common organ infected by larval form of Echinococcus^[3-6]. But in children lungs are the most common organ infected by larval form of the *Echinococcus granulosus*^[3,7-10]. Diagnosis hvdatid cyst of the liver is made by history, physical, radiological and serological findings^{11,12}]. Symptoms are related with size, localization, rupture and infection of cysts. The most common symptom is abdominal mass. The cyst is identified by sonography and in some cases by CT scan. Immunodiagnosis makes the distinction of echinococcal cysts from benign cysts or malignant neoplasms possible^[11]. Ultrasonographic classification has provided a good, widely morphologic accepted description of the disease^[12]. Rupture of hydatid cysts occur infrequently $(3.2\%)^{[12,13]}$.

Subjects and Methods

This retrospective study was conducted in hospitalized patients operated for liver hydatid cysts in Mofid Children's Hospital in Tehran, Iran during March1996 to March 2010. A total of 100 patients were evaluated in this study based on pathological findings (Echinococcus granolosus was detected in 100% of our patients). Data were collected from medical records of patients in archives and analyzed in terms of age, gender, clinical features, diagnostic investigation (sonography, CT-scan imunoserology test), operative technique, post operative complication, mortality and duration of hospital stay. ELISA test was performed in all patients and IFA test for suspected ones. Cystotomy and partial pericystectomy were mostly used for patients

with unruptured hydatid cysts but in some cysts that were lager than 5 cm in diameter scolicidal agent (like hypertonic saline) was injected in cyst cavity after aspiration of cyst component and then resected. All patients were followed up for 2-5 years and recurrence of the cyst was evaluated. Descriptive analysis was performed by using frequency tabulations for categorical variables. No comorbidity was seen in our study group. All patient identifying information remained confidential.

Findings

The patients consisted of 54 (54%) boys and 46 (46%) girls (range 1-14 years) (Table 1). *Echinococcus granolosus* was detected in 100% of our patients by pathologic reports. Twenty five (25%) patients had associated lung hydatid cyst.

Incidence of hydatid cyst increased by age and 19 (19%) patients had coexisting lung cysts. Our patients had totally 110 cysts, right lobe had 81 (73%), and left side 29 (27%) cysts. 14 (13%) cysts were smaller than 4 cm in diameter, 74 (67%) cysts were 4-5 cm in diameter and 22 (20%) cysts were more than 5 cm in diameter.

Abdominal mass was the most common symptom (Table 2). Twenty five (25%) patients had associated lung hydatid cyst. Abdominal sonography was performed in all patients, which revealed hydatid liver cyst in 94 (94%) patients. CT scan confirmed the diagnosis in 6 (6%) suspicious patients.

Twelve patients (12%) had calcified cysts. Cystotomy and partial pericystectomy was done in

Table 1: Age distribution in 100 patients with liver cyst in Mofid Children's Hospital

Age (year)	Number (%)
0-2	2 (2)
3-6	11 (11)
7-9	16 (16)
10-12	27(27)
13-14	44 (44)
Total	100 (100)

Table 2: Symptom distribution in 100 patients with liver cyst in Mofid Children's Hospital

Symptoms	Number (%)
Abdominal mass	50 (50)
Fever	12 (12)
Hepatomegaly	12 (12)
Abdominal pain	10 (10)
Weight loss	8 (8)
Loss of appetite	4 (4)
Nausea	2 (2)
Jaundice	2 (2)
Total	100 (100)

98 patients but in patients with cysts larger than 5 cm in diameter, suction of cyst component after injection of scolicid agents, cystectomy was performed as an alternative method. Two patients in whom the cysts were small and calcified (confirmed by sonography guided percutaneous aspiration) were treated medically. All patients received oral Albendazol post-operatively for 3 to 6 months. Wound infection was the most frequent post operation complication (5 cases), other complications were rupture of cyst during operation (2 cases), spontaneous cyst rupture before operation (2 cases), and anaphylactic shock (2 cases). In cases with residual cavities, we performed drainage, and followed them by periodic ultrasonography. In our study the morbidity rate was 12% (prolonged external catheter drainage in 12 patients). Mortality was in one (1%) patient due to anaphylactic shock. The mean length of hospital stay was 9 days. On discharge, 94 (94%) patients were cured. All patients were followed up by periodic ultrasound examination for 2 to 5 years. Recurrences occurred in two (2%) cases which were treated medically.

Discussion

Hydatid disease is still a national problem in highly endemic countries and needs epidemiologic prevention for its eradication^[3].

The symptoms of hydatid disease depend on which organs are affected. Most patients with hydatid cysts are asymptomatic, and the diagnosis is usually made incidentally during clinical or radiological examination for unrelated reasons^[13]. The most commonly affected organ is the liver in adults, and lung in children. Surgery is the main form of treatment for hydatid disease.

A risk of surgery is that hydatid cyst ruptures and spreads tapeworm heads throughout the patient's body. To reduce this risk, we prescribe high doses of the drug albendazole in conjunction with surgery [14].

Of our 100 cases 54 (54%) were boys and 46 (46%) girls. Statistical analyses indicate that in children males are more likely to be infected with liver hydatid cyst [15,16], but in Bulent's report incident of hydatid cysts was equal in males and females [17,18]. Patients' age ranged from 1-14 years, and the youngest one was one year old, we believed that one year of age is the youngest age of this pathology but Prashant has reported hydatid disease in a six month old infant. Incidence rate of hydatid cyst increased by age in our patients. Seventy one (71%) cases were more than 9 years old.

Hydatid cysts in children involves more commonly lungs than liver but Schitogullri^[19] believes that liver cysts are more common in childhood and Talaizadeh^[16] found nearly equal incidences (41% -43%) of hydatid cyst in lung and liver in pediatric patients.

In our study cysts were seen most commonly in right liver lobe. Abdominal mass was the most common symptom in our patients similar to other studies [15,20,21]. Cysts may grow slower in the liver than in the lung due to less elasticity of the lung. This may explain the lower incidence of hydatid cyst in liver [15]. Liver sonography revealed a correct diagnosis in 94% near to the rate of 96.4% given by Koeseglu^[22]. Cystotomoy and pericystectomy were the preferred techniques of surgery similar to Kanat study [23], which are now the most used surgical techniques.

Intrabiliary rupture is the most common complication of hydatid cyst, with an incidence of 5% to 25% of patients^[13]. Significant risk factors for hydatid cyst perforation include younger age, cyst diameter of more than ten cm, and superficial cyst location^[13]. Rupture of hydatid cyst is very

rare and can occur spontaneously iatrogenically^[13]. We had spontaneous cyst rupture before operation in 2 cases, Wound infection was the most frequent postoperative complication in our study which was similar to others^[22,25]. In our study the morbidity rate was 12%, this rate was 14.5% in Balay's study[25]. There was one (1%) mortality case due to anaphylactic shock and 2 (2%) recurrences, in other series there were no recurrences and mortality cases^[19,22]. The mean length of hospital stay was 9 days similar to Balci report^[25]. Echinoccus granulosus was detected in all patients. As surgery is expensive and not available worldwide, therefore, during the last decades minimally invasive techniques have been introduced in the treatment of hydatid liver cyts. Recently some surgeons prefer percutaneousaspiration-injection and re-aspiration (PAIR) technique as a first line of therapy for hydatid liver cysts^[26]. Laparoscopic approach should be a valid surgical alternative choice expert surgeons^[26,27]. Although PAIR has been widely used in recent years, still some pediatric surgeons believe in surgical approach in complicated cases^[28]. Izadpanah et al^[29] suggest medical treatment for asymptomatic liver hydatid cysts.

Conclusion

In children the incidence of liver hydatid cyst increased by age. The most involved organ was lung and then liver. Due to the less invasive and high accuracy of abdominal sonography in diagnosis, we recommend it as the method of choice for the diagnosis in endemic regions. A Conservative surgical technique (cystotomy and partial pericystectomy) is sufficient in most cases. Mostly the postoperative results are favorable.

Acknowledgment

This study was financially supported by the office of the Vice chancellor for Mofid Children's Hospital Clinical Research Development Center (CRDC).

Conflict of Interest: None

References

- 1. Saidi F. Surgery of hydatid disease. Philadelphia: Saunders, 1976.
- Harandi MF, Hobbs RP, Adams PJ, et al. Molecular and morphological characterization of echinococcus granulosus of human and animal origin in Iran. *Parasitology* 2002; 125(4):367-73.
- 3. Tensaw IM. Hidatid cyst in children. *Ann Ped Surg* 2010;6(2):98-104.
- King CH. Cestodes (tapeworms). In: Mandell GL, Bennet JE, Dolin R (eds). *Principles and Practice* of Infectious Disease. 6th ed. New York: Churchill Livingstone. 2005; Pp. 3290-2.
- Ruiz-Rubelleo JF, Gomez-Alvarez M, Sanchez J, et al. Complications of exrahepatic echinococosis: Fistulization of an adrenal hydatid cyst into intestine. World J Gastroenterol 2008;14(9): 1467-9.
- 6. Celik M, Senol C, Keles M, et al. Surgical treatment of pulmonary hydatid disease in children: report of 122 cases. *J Pediatr Surg* 2000; 35(12):1710-3.
- 7. Beggs I. The radiology of Hydatid disease. *AJR Am J Roentgenol* 1985;145(3):639-48.
- 8. Karaoglanoglu N, Kurkcuoglu IC, Gorguner M, et al. Giant hydatid lung cysts. *Eur J Cardiothorac Surg* 2001;19(6):914-7.
- 9. Ahmad Khan R, Wahab Sh, Singh Chana R, et al. Isolated retropritoneal hydatid cyst in a child: a rare cause of acute scrotal swelling? *J Ped Sur* 2010;45(8):1717-9.
- 10. Mirshemirani A, Razavi S, Sadeghian N. Surgical treatment of pulmonary hydatid cyst in 72 children. *Tanaffos* 2009;8(1):56-61.
- 11. Moralioglu S, Ozen IO, Afsarlar C, et al. A rare cause of acute abdomen in children: an intrabilliary rupture of hydatid cyst. *Turk J Gastroenterol* 2010;21(3):331-2.
- 12. Anyfantakis D, Bievrakis E, Viachakis I. et al. Hepatopulmonary hydatidosis in a ten-year-old girl: a case report. *J Med Case Rep* 2010;4:205.
- 13. Sumer A, Caglayan K, Altinli E, Koksal N. Case report: Spontaneous liver hydatid cyst rupture in a child. *Israel J Emerg Med* 2009;9(I):13-6.
- 14. Tapeworms and hydatid disease. Better Channel. Available at: http://www.betterhealth.vic.gov.au/bhcv2/bhcarticles.nsf/pages/tapeworms_and_hydatid_disease. Access date: May 2009.

- 15. Montazeri V, Sokouti M, Rashidi HR. Comparison of pulmonary hydatid disease between children and adult, *Tanaffos* 2007; 6(1):13-8.
- Talaiezadeh A, Maraghi Sh. Hydatid disease in children: A different pattern than adults. Pakistan J Med Sci 2006;22(3):329-32.
- 17. Bulent K, Gultekin G, Serdar H, et al. Analysis of pulmonary hydatosis according to their segmentary location. *Clin Pulm Med* 2008; 15(1):8-12.
- 18. Congir AK, Salim E, Enon S, et al. Surgical treatment of pulmonary hydatid cyst in children. *J Pediatr Surg* 2001;36(6):917-30.
- 19. Sehitogullari A. Our results in surgical treatment in hydatid cyst of the lungs. *Eur J General Med* 2007;4(1):5-8.
- Rattan KN, Sharma A, Sharma Anita. Hydatid disease in children. *Indian J Chest Dis Allied Sci* 1998;40(1):73-7.
- Anadol D, Gocmen A, Kiper N, Ozcelik U. Hydatid disease in childhood; A retrospective analysis of 376 cases. *Pediatr Pulmonol* 1998;26(3):190-6.
- Koeseoglu B, Baken V, Onem O, et al. Conservative surgical treatment of pulmonary hydatid cysts in children: An analysis of 35 cases. Surg Today 2002;32(9):779-83.

- 23. Kanat F, Turk E, Aribas OK. Comparison of pulmonary hydatid cysts in children and adults. *ANZ J Surg* 2004;74(10):885-9.
- 24. Ulka R, Onen A, Onat S. Surgical treatment of pulmonary hydatid cysts in children: Report of 66 cases. *Eur J Pediatr Surg* 2004;14(4):255-9.
- Balcy AE, Ulka R, Eren & Thor;, Eren MN, cebeci E, Erdem K. Surgical treatment of pulmonary hydatid cysts in children. *Thoracic Cardiovasc* Surg 2002;Thema:Tuseday, February 19, 2002 special session-thoracic and vascular surgery 9730
- 26. Giorgio A, Disarno A, Giorgio S, et al. Percutaneous treatment of hydatid liver cyst. *Recent Patents Anti-Infective Drug Discov*2009; 4:29-36.
- 27. Giorgio A, Di Sarno A, De Stefano G, et al. Sonographic and clinical outcome of viable hydatid liver cysts treated with double percutaneous aspiration and ethanol injection as first line therapy: efficacy and long term follow-up. *AJR Am J Roentgenol* 2009;193(3): W186-92.
- 28. Farkas B, Budusan A, Ordeanu C. Management and results in liver hydatid disease in children. *AMT* 2009;11(1):161.
- 29. Izadpanah A, Saeedi F. Asymptomatic liver hydatid cysts: A preferred approach. *Iran J Med Sci* 2006; 31(3):1-4.