Epidemiological, Laboratory and Clinical Features of Childhood Hydatid Disease

Çocukluk Çağında Kist Hidatik Hastalığının Epidemiyolojik, Laboratuvar ve Klinik Özellikleri

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Abstract

Objective: Hydatid disease is a parasitic infestation by a tapeworm of the genus Echinococcus. It is an important cause of morbidity in endemic areas and can be life threatening. In our country it can occur from childhood onwards and usually requires the prolonged use of medications.

Material and Methods: Twelve paediatric cases admitted to the Ege University Children's Hospital with various symptoms and diagnosed with hydatid disease between 2009 and 2011 were included in the study.

Results: During a 3-year period 12 patients (5 female, 7 male; mean age 11±2 years; range 6 to 14 years) were admitted to the hospital because of fatigue, nausea, fever and cough, abdominal pain, or back pain, or were diagnosed incidentally. Complete blood count tests revealed mild eosinophilia (mean=490±158/mm^3); two cases admitted with fever (16%) had elevated C reactive protein levels and leucocytosis. All patients were treated with Albendazole (10 mg/kg/d), seven were treated surgically, four (33%) were treated with percutanous drainage (PAIR: puncture, aspiration, injection, re-aspiration) and two (16%) did not need invasive procedures and remain in follow up.

Conclusion: This study presents the course of hydatid disease and emphasizes the diagnostic challenges in follow up. *(J Pediatr Inf 2013; 7: 53-6)*

Key words: *Echinococcus* spp, childhood, hydatid disease

Özet

Amaç: Kist hidatik hastalığı Echinococcus ailesine ait barsak solucanının etken olduğu parazitik bir enfestasyondur. Endemik bölgelerde halen önemli morbidite nedenidir ve hayatı tehdit edebilmektedir. Ülkemizde çocukluk çağında da sıklıkla görülebilen bu hastalık nedeniyle uzun süreli ilaç kullanımı gerekli olabilmektedir.

Gereç ve Yöntemler: Ege Üniversitesi Çocuk Hastanesi'ne çeşitli yakınmalar ile başvuru sonucu kist hidatik tanısı alan olgular 2009-2011 yılları arasında çalışmaya dahil edilmiştir.

Bulgular: Üç yıllık izlem sürecinde toplam 12 olgu (yaşları 6 ile 14 arasında değişen (11±2); 5 kız, 7 erkek olgu) halsizlik, bulantı, kusma, ateş, öksürük, karın ağrısı, sırt ağrısı nedeniyle başvurmuş ya da tesadüfen tanı almıştır. Tam kan sayımında ılımlı eozinofili (490±158/mm^3), ateş yakınması ile başvuran iki olguda (%16) C reaktif protein düzeyinde artış ve lökositoz saptanmıştır. Tüm hastalar Albendazol tedavisi almış olup (10 mg/kg/gün), yedi olgu opere edilmiş, dört olguya (%33) perkutan drenaj (PAIR: puncture, aspiration, injection, re-aspiration) tedavisi uygulanmıştır. Geri kalan iki olgu (%16) için herhangi bir girişim gerekmemiş olup halen izlemleri sürmektedir.

Sonuç: Bu çalışma çocukluk çağında kist hidatik hastalık sürecini göstermekte ve tanısal zorluklara dikkat çekmektedir. *(J Pediatr Inf 2013; 7: 53-6)*

Anahtar kelimeler: *Echinocccoccus* spp, çocukluk çağı, kist hidatik

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Introduction

Hydatid disease is a parasitic disease that was recognized centuries ago (1). This zoonotic infection is caused by adult or larval (metacestode) stages of cestodes belonging to the genus *Echinococcus* and the family *Taeniidae*. Four species of *Echinococcus* have been identified, namely *E. granulosus*, *E. multilocularis*, *E. oligarthrus* and *E. vogeli.* Humans are intermediate hosts in which the infective metacestode stage develops after peroral infection with eggs.

Metacestodes may also develop in humans, causing various forms of echinococcosis. Their cysts can develop in all anatomic sites following oral ingestion of eggs but the liver and lungs are the most frequently affected organs. *E. granulosus* is the most common strain causing hydatid disease in our country and the Middle East, and causes hydatid disease on all continents and in tropical zones around the world (2, 3). Current control programmes are predominantly based on the control of dog populations, regular dosing of dogs with praziquantel to eliminate *E. granulosus*, improved control of animal slaughter, and health education (4). However, the disease remains an important clinical problem in some areas and also affects children and adolescents. We analysed the epidemiological, laboratory and clinical characteristics of human cystic echinococcosis to determine the course of the disease in a paediatric population.

Material and Methods

Twelve paediatric cases who were admitted to Ege University Children's Hospital with various symptoms and who were diagnosed with hydatid disease between 2009 and 2011 were included in the study. Demographic data, causes of admissions, white blood cell and eosinophil counts, type of organ involvement, levels of specific antibody titres for *E. granulosus* and selected treatment options were recorded.

Statistical analysis

Continuous variables are presented with mean±standard deviation and the categorical variables as frequency and related percentage.

Results

Over a 3-year period 12 patients (5 female, 7 male; mean age 11±2 years; range 6 to 14 years) were admitted to the hospital due to fatigue, nausea, fever and cough, abdominal pain or back pain or were diagnosed incidentally. Eight of these patients (67%) had isolated lung, liver or vertebral involvement. The other patients (33%) were diagnosed with concurrent liver and lung, liver and omentum or liver and spleen involvement. Children diagnosed with hydatid disease in our hospital mostly live in rural areas; only a small proportion (20%) live in urban areas. Complete blood count tests revealed mild eosinophilia (mean=490±158/mm^3); two cases admitted with fever had elevated C reactive protein levels (5 mg/dL and 8 mg/dL with a normal value of < 0.5 mg/dL) and leucocytosis (15700/mm^3 and 17000/mm^3 with normal values of 4000-7000/mm^3). All patients were treated with Albendazole (10 mg/kg/d, twice daily at mealtimes), seven were treated surgically and four patients were treated with percutanous drainage; two patients required no invasive procedures and are still in follow up. Complete blood count and ELISA (Enzyme-linked Immunosorbent Assay) tests were performed at the time of diagnosis and during the third, sixth and twelfth months of therapy. ELISA titres of the patients in follow up are shown in Table 1. One patient (Case 3) had spontanous rupture of the pulmonary cyst (Figure 1 and 2) and was hospitalized in the intensive care unit. Another patient (Case 9) developed toxic hepatitis during the standard-dose regimen of Albendazole treatment (10 mg/kg/d, twice daily at mealtimes). Elevated levels of transaminases dropped to normal values 3 days after discontinuation of therapy.

Table 1. ELISA titres of children with hydatid disease in follow up

Case	ELISA (At time of diagnosis)	Treatment	3 months	6 months	12 months
1(L+Li)	1/40000	PAIR	1/40000	2.PAIR	1/20000
2(Li+O)	1/20000	PAIR+Operation	1/20000	1/20000	1/20000
3(L)	1/20000	Rupture+Operation	1/160	1/160	1/160
4(L)	1/20000	Medical	1/10000	1/5000	1/5000
5(Li)	1/640	PAIR	1/160	1/160	1/160
6(Li)	1/1280	PAIR	1/320	1/320	1/320
7(Li)	1/20000	Medical	1/20000	1/10000	1/20000
8(V)	1/40000	Operation	1/1280	1/320	1/80
9(L)	1/20000	Operation	1/20000	1/1280	1/20000
10(Li+S)	1/20000	Operation	1/1280	1/1280	-
11(Li)	1/40000	Operation	1/160	1/1280	1/160
12(L+Li)	1/20000	Operation	1/1280	1/1280	1/10000

L: Lung, Li: Liver, O: omentum, PAIR: puncture, aspiration, injection, re-aspiration, S: spleen, V: vertebra

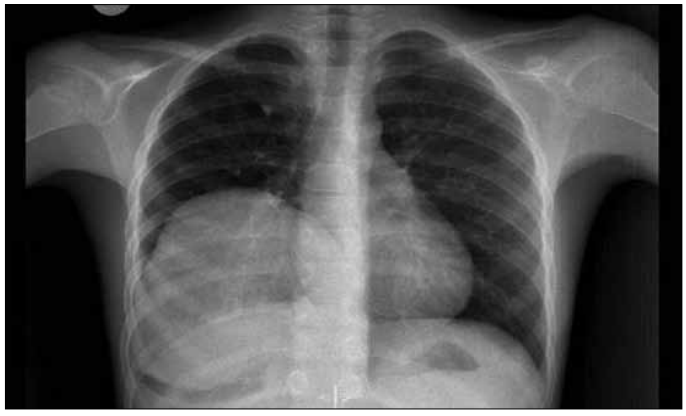

Figure 1. Pulmonary hydatid cyst (Case 3) before spontaneous rupture

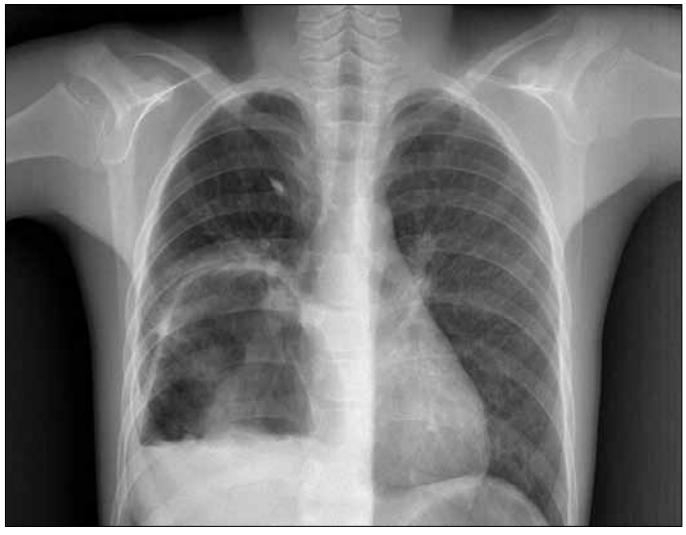

Figure 2. Ruptured cyst (Case 3)

Discussion

In our country hydatid disease remains a public health problem related to suboptimal control of the disease in animals and insufficient environmental hygiene. Children diagnosed with hydatid disease in our hospital mostly live in rural areas where dogs, the definitive hosts, are straggling. In a province-based study recently performed in Turkey the prevalence of the disease was found to be 0.15% in children (5). Autopsy studies indicate a burden of disease in all age groups (6). Many cases have been reported with the presence of cysts in the lungs, liver, orbita and nervous system (7-11). In our study group, causes of admissions varied according to the location of the involved body sites. Children with abdominal pain were diagnosed with liver disease according to the WHO classification based on ultrasound imaging (12). All patients took Albendazole either alone or as pre-postprocedure adjuvant therapy. Patients with only liver involvement were treated successfully with percutanous drainage or surgery even they have high titers of antibodies in follow up. The observation of elevated antibody titres in some patients with multiple organ involvement (Cases 1 and 2) brings the significance of serological tests into question, although it is not possible to make any firm conclusions given the small patient population. Interestingly the patient with spontaneous rupture of the pulmonary cyst who needed intensive care because of respiratory distress had low titres in follow up. This observation questions whether minimally invasive procedures such as percutanous drainage or medical therapy alone increase the risk of recurrence given the intact membrane of *Ecinococcus spp.*, which has a germinative layer and is thereby hard to invade. In some cases surgery alone leads to low levels of antibodies and provides complete cure. Other cases appear cured due to the absence of cysts in whole body imaging, but the persistence of high levels of antibodies confuses caregivers in follow up. For example in our case series one patient (Case 2) had concurrent liver and omentum cysts. Initially she was treated with PAIR for liver disease but surgery was necessary due to continued high serological titres and the presence of a unilocular cyst, which was classified as a CE3 transitional cyst according to the WHO classfication, and which may have contained daughter cysts. After surgery serological titres remained high despite complete cure of the liver cysts. We hypothesize that the presence of an omental cyst that was inoperable but regressed with medical therapy was the cause of high ELISA titres for *E. granulosus*.

Another challenge concerns the shortage of medical options despite studies conducted to find new therapies and build a consensus (13). Because even toxic effects of Benzimidazoles are usually reversible like in our patient (Case 9); it relapses and impedes prolonged use of therapy.

Conclusion

In this study we wish to emphasize the need for reliable diagnostic tests and therapeutic choices for follow up in such patients.

Conflict of Interest
No conflict of interest was declared by the authors.

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Author Contributions
Concept - B.Y.,F.V. ; Design - B.Y., F.V.; Supervision - F.V.; Data Collection and/or Processing - B.Y., S.Ş.,

Z.S.B. ; Analysis and/or Interpretation - B.Y.; Literature Review - B.Y.; Writing - B.Y.; Critical Review - D.D.E., M.K., F.V.

Çıkar Çatışması
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Yazar Katkıları

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