Hepatic hydatid disease in children and adults living in different areas in Turkey

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ABSTRACT

Objective: To compare the clinical features of the hepatic hydatid disease in the operated children and adults living in the east and west part of Turkey.

Methods: Between January 2001 and May 2005, 105 patients were operated with the diagnosis of hepatic hydatid cyst in Trakya and Yuzuncu Yil University Hospitals, Turkey. The patients (n=105) were retrospectively evaluated in 4 groups; Edirne Ch: (18 children under 18 year-old) and Edirne Ad: (20 adults) were from Edirne, Van Ch: (22 children under 18 year-old) and Van Ad: (44 adults) from Van. The patients in each group were analyzed according to their clinical and radiological findings.

Results: The frequency of hepatic hydatid cysts in children was significantly higher in boys in Edirne Ch group and in girls in Van Ch group (p<0.05). In adults, the disease was also seen significantly higher in males in Edirne Ad group and females in Van Ad group (p<0.05). There were no difference symptoms of the disease, concomitant extra hepatic cysts and total cyst number in children and adults in the same region (p>0.05). The number of huge hepatic cysts and history of contact with animal were more common in children and adults living in Van.

Conclusion: While the course of hepatic hydatid disease has the similar clinical features among the children and adults in the same region, remarkable regional differences have been found on it.

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Cystic echinococcosis is a parasitic disease caused by "Echinococcus granulosus". Lung and liver are the organ where the larval form of the agent most commonly settles.[1] The disease is often seen in the areas where sheep breeding is common such as China, Mediterranean and Balkan countries, South America, the Middle East.[1-4] Although, the disease is considered to be eradicated from New Zealand and nearly from Australia, it is still a public health problem in endemic areas with the incidence rate of cystic echinococcosis ranging from 5-20 per 100,000 population.[2,3] On the other hand, it is an important matter leading to a great number of medical visits and generating health care costs as a result of the diagnostic procedures involved and associated medical problems. Patients with hydatid disease often require multiple surgical interventions. Extensive secondary hydatid disease often becomes inoperable and involvement of the bones usually requires amputation. Economic losses to affected families include surgical and hospital expenses as well as loss of income.[3,4] Thus, knowledge about the specific characteristics of the age groups of the patients and regional differences of the course of the disease may provide to plan of feasible health politics in the endemic areas. Echinococcal cysts in children frequently settle in lungs; however, in adults they are common in the liver.[2-5] The pathophysiology of this difference in predilection of parasite for lungs and liver in children and adults still remains unclear.[6] Variability of the disease in age groups led us to examine in pediatric and adult population of hepatic hydatid disease with regional differences. We hypothesized that regarding the fact that hydatid cyst is a zoonosis, the clinical features of the disease among the children and adults living in Van region where stock breeding

is very intensive may be different from the other regions in Turkey. Van region is located in the east Anatolia and has border with Iran. It is a passage between Caucasia and the Middle East with dense rural population. As a sample of another areas located in the west part of Turkey, Edirne where living is mainly depended on industry and agriculture has border with Bulgaria and Greece. Moreover, it is a bridge between Anatolia and Europe (**Figure 1**). In the area, the urban population is dense just opposite to Van region. Hydatid disease is endemic in both Edirne and Van regions. In this study, we aimed to compare differences in clinical presentation and in types of cysts located in the liver in children and adults operated for cystic echinococcosis in 2 different regions in Turkey.

Figure 1 - The map of Turkey. Van is located in the east Anatolia and Edirne is located in the north-west of Turkey.

Methods. Between January 2001 and May 2005, 105 patients were operated with the diagnosis of hepatic hydatid cyst in Trakya University Hospital, in Edirne and Yuzuncu Yil University Hospital, in Van. The patients have been analyzed retrospectively in 4 groups. Group Edirne Ch was formed from 18 children under 18 years old of age and they were living in Edirne region. Group Edirne Ad was formed from 22 adults living in Edirne region. Group Van Ch was formed from 21 children under 18 years old and they were living in Van region and Group Van Ad was formed from 44 adults living in Van region. To evaluate the differences in 2 regions, children and adults were analyzed according to their age, gender, symptoms, history of contact with animals, presence of huge cysts, ruptured cyst and number of hepatic cysts. The parameters were collected for the study from the medical records of the operated patients in both regions. A special form was created to record the patients' data and the form was filled by the authors after the medical records of the patients were investigated. The patients were evaluated with detailed history and careful physical examination in the clinics. For the diagnosis of hydatid cyst, all patients had undergone radiologic evaluation of posteroanterior pulmonary x-ray and at least one or several different radiological methods including abdominal ultrasonography and computed tomography have been used. Indirect hemaglutination test had also been used in some children to identify the suspicious masses. In patients with hepatic cysts, the final diagnosis was confirmed by surgery and it was not encountered with any case of alveolar echinococcosis among our patients. The bile duct ruptures were managed by T-tube drainage during the operation. Acute ruptured hydatid cysts were treated with emergency surgery as mentioned by Derici et al.[7] The other patients with hydatid disease were applied 3 courses of albendazole (10 mg/kg per day) in the period of 3-4 months which was begun 2 weeks before the operation and it has been continued 13-15 weeks after the operation as mentioned by Demirbilek et al[1] and Horton.[8] While the numeric results were expressed as mean±SD, categorical results were expressed as percentage. Obtained data were evaluated with the aid of x^2 and Kruskal-Wallis variant analysis method. The difference was considered significant as $p<0.05$. The study protocol was approved by the Local Ethics Committee, Trakya University Faculty of Medicine.

Results. In Edirne Ch group, male patients (n=11 [61.1%]) were more than females (n=7 [38.9%]) ($p<0.05$) and female patients (n=13 [61.9%]) were more than males (n=8 [38.1%]) in Van Ch group ($p<0.05$) (**Table 1**). The mean age of the children was significantly higher in Van Ch group than Edirne Ch group ($p<0.05$). In Edirne Ad group, male patients were more than females ($p<0.05$) and female patients were more than males in Van Ad group ($p<0.05$) (**Table 1**). The mean age of the patients was significantly higher in Edirne Ad group than Van Ad group ($p<0.05$). In children, there was statistically significant regional difference in the history of contact with animal (n=17 [80.9%] versus n=9 [50%]) and number of huge cyst (≥10 cm in diameter) located in the liver (n=13 [65%] versus n=5 [27.8%]) between Van Ch and Edirne Ch groups. However, the parameters of most frequent symptoms (hepatomegaly, abdominal pain and fever), mean cyst number, solitary hepatic cyst, multiple hepatic cysts and ruptured hepatic cysts were not statistically different in the children groups (**Table 2**). In adults, there was statistically significant regional difference in the history of contact with animal, multiple hepatic cysts, ruptured hepatic cysts, solitary hepatic cysts and number of huge cysts respectively between Van Ad and Edirne Ad groups ($p<0.05$) (**Table 2**). In our study, the clinical features of hydatid cyst in children and adults living in the same region were also investigated. Statistically, no significant difference was found between Edirne Ch and Edirne

Ad groups with respect to the parameters of gender, most common clinical symptoms, history of contact with animal, number of solitary hepatic cyst, multiple hepatic cysts, huge cyst, ruptured hepatic cysts, hospital stay and recurrence (p>0.05). No significant difference was also found when Van Ch and Van Ad groups were compared regarding to the same criteria (p>0.05).

Discussion. Epidemiologic studies show that the hydatid disease is often seen in developing countries and affect mostly young adults. In children, there are many studies showing either male or female dominance.[4] In our study, the number of male patients in Edirne and the number of female patients in Van region were higher in all age groups. In other words, while there was no difference with respect to gender distribution between children and adults living in the same region, the gender difference between the 2 regions was striking. In addition, in Van region, the disease is seen in females at their second or third decade. Closer contact with dogs and an immunosuppressive role of pregnancies have been hypothesized as causes of these findings. In Edirne, the disease was more common amongst the young children and old adults. The higher incidence of hydatid cysts in boys and male adults in Edirne should be studied more in details; in fact, in most endemic countries, cystic echinococcosis is more frequent in females, but usually only after puberty. Behavioral and genetic factors could play a role in Edirne to favor male infection. Hydatid cyst may be latent in children and adults for a long time but it may also rupture through biliary tract and also cause inflammation.[3,5,9] Onset symptoms of hydatid cysts are nearly always hepatomegaly and abdominal palpable mass.[9] In our study, hepatomegaly, abdominal pain and fever were most common symptoms and no regional and age differences were found on these parameters in children and adults. Cystic echinococcosis is one of the major specific public heath problem in our country.[1-3,5,10-12] In endemic regions as Turkey, hydatid cysts must be the first to be suspected in differential diagnosis of all cystic masses seen in whole of the body without considering that they are symptomatic or asymptomatic.[1-4,9-11] With

Table 1 - Gender and age distribution of hepatic hydatid cyst seen in children and adults.

Characteristics	Edirne Ch (n=18)	Edirne Ad (n=22)	Van Ch (n=21)	Van Ad (n=44)
Male	11 (61.1)*	14 (63.6)*	8 (38.1)	16 (36.4)
Female	7 (38.9)	8 (36.4)*	13 (61.9)	28 (63.6)*
Mean age (±SD)	12.1±3.3§	53.6±14.8§	16.3±1.8	37±13

Data are expressed as number and (%).
*Significant difference between the groups of Edirne and Van according to sex (p<0.05)
§Significant difference between the groups of Edirne and Van according to age (p<0.05)

Table 2 - The differences in clinical presentation and in types of cysts located in the liver.

Clinical presentation	Edirne Ch n=18	Edirne Ad n=22	Van Ch n=21	Van Ad n=44
Symptoms (hepatomegaly, abdominal pain, fever)	18 (100.0)	21 (95.5)	21 (100.0)	42 (95.5)
Contact with animal (%)	9 (50.0)*	5 (22.7)*	17 (80.9)	33 (75.0)
Other cases in the family (%)	0 (0)	0(0)	3 (14.2)	4 (9.9)
Mean cyst number (±SD)	1.6 ± 0.9	1.3±0.7*	2.2±2.5	1.75±0.9
Solitary hepatic cyst (%)	11 (61.0)	16 (72.0)*	11 (52.0)	19 (43.0)
Multiple hepatic cysts (%)	7 (39.0)	6 (28.0)*	10 (48.0)	25 (57.0)
Huge cyst (≥10 cm) (%)	5 (27.8)*	8 (38.4)*	13 (65.0)*	27 (61.4)
Ruptured hepatic cysts (%)	3 (16.6)	1 (4.5)*	8 (38.0)	11 (25.0)

Data are expressed as number and (%).
*Significant difference between the groups of Edirne and Van region according to being child or adult (p<0.05)

the advent of visualizing systems such as ultrasound and computerized tomography the disease became more easily diagnosable. Radiodiagnostic procedures give more detailed about localization and dimension of the cyst.[13] In our study, it was found that the features of the cysts diagnosed in the patients were different in 2 endemic regions. These differences were clear in adult population. The period of time that elapsed between Echinococcus granulosus egg ingestion and the development of clinical symptoms is an important variable that might be accounted for by a number of parameters including the genetic characteristics of the population and perhaps of the parasite too, immune suppression due to malnutrition, but also access to medical care. This time period could be one of the main reasons for the differences in cyst size.

Briefly, hydatid disease shows similar clinical features between children and adults living in the same region, but when the clinical presentation and anatomical characteristics of the cysts located in the liver were considered, remarkable regional differences were found in different endemic regions, even in the same country. The destruction of the parasite's living circle between dog and sheep is the most important step in preventing the disease. Regional characteristics of the course of the disease and risk groups should be considered when the health organizations are planning in the endemic countries. In addition, public education on hygienic rules, controlling the offal consuming and improvement of veterinary services may be the important points in prevention of the disease.

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