

East African Medical Journal Vol. 80, No 8 August 2003

ECHINOCOCCOSIS MULTILOCULARIS IN SOUTH-EASTERN ANATOLIA, TURKEY

A. K. Uzunlar, MD, F. Yilmaz, MD, Department of Pathology, Medical Faculty, Dicle University Diyarbakir, Turkey and M. Bitiren, MD, Department of Pathology, Medical Faculty, Harran University University, S. Urfa, Turkey

Request or reprints to: Dr. A.K Uzunlar, Department of Pathology Medical Faculty, Dicle University, Diyarbakir, Turkey

## ECHINOCOCCOSIS MULTILOCULARIS IN SOUTH-EASTERN ANATOLIA, TURKEY

A. K. UZUNLAR, F. YILMAZ and M. BITIREN

### ABSTRACT

**Objective:** To quantify the human prevalence, elucidate possible risk factors and present other epidemiological parameters of human alveolar echinococcosis in the south-eastern region of Turkey.

**Design:** A retrospective study.

**Settings:** South-eastern region (Anatolia) of Turkey by two medical centres: Dicle University Medical Faculty in Diyarbakir and Harran University Medical Faculty in Urfa.

**Subjects:** Forty seven cases of human alveolar echinococcosis were presented between the period of 1980 and 2000.

**Methods:** For the majority of our patients, personal anamnestic, clinical data and addresses were available from their files, and we visited their villages to investigate the risk factors for human alveolar echinococcosis. Epidemiological parameters were evaluated.

**Results:** The 47 cases consisted of 18 (38.3%) males and 29 (61.7%) females. The average age was 34.6 years. In all cases, lesions occurred in the liver. No village or district had more than one case. The majority of patients (73.8%) were farmers and 33 had a history of dog ownership.

**Conclusion:** This region has a higher incidence than Turkey as a whole, and domestic dogs and free-ranging dogs seem to be the main source of infection.

### INTRODUCTION

Human alveolar echinococcosis (AE) is one of the most lethal of the larval cestode infections (1). It is characterised by hepatic disease resulting from infection by the larval (metacestode) stage of echinococcosis multicularis, a small tapeworm of foxes, *Vulpes spp.* and *Alopex lagopus*. Completion of the cycle is ensured by the predator relationship existing between foxes, dogs (the definitive host) and small rodent (the intermediate host) (2). Humans are infected by the accidental ingestion of eggs, which are voided in the faeces of infected definitive hosts, either by direct contact with infected canids or indirectly from the contaminated environment. More than 98% of primary infections of humans occur in the liver, typically with long asymptomatic periods (5-15 years). A possible secondary metastatic focus within the liver or adjacent organs and or lungs and brain is a serious complication (3).

Alveolar echinococcosis is confined to the northern hemisphere and is endemic in Russia, Ukraine, Moldova, Turkey, some central European countries and central Asia, including China (1, 4, 5). Although AE is found throughout Turkey, it is predominantly seen in the eastern Anatolia region of Turkey, which borders Iran and Armenia, and includes the provinces of Elazig, Agri, Erzurum and Van; and in the south-eastern Anatolia region of Turkey, which borders Syria and Iraq and includes the province of Diyarbakir, Urfa, Antep, Siirt, Batman, Sirnak and Mardin. Anatolia is the Asian part of Turkey, extending from the Bosphorus and Aegean coast eastward to the borders of Georgia, America, Iran and Syria.

The current study was undertaken in south-eastern Anatolia, Turkey, by two medical centers, with the aim of quantifying the human prevalence, elucidating possible risk factors and presenting other epidemiological parameters.

### MATERIALS AND METHODS

Between the period of 1980 to 2000, a total of 47 cases of human AE (42 histologically confirmed, three clinically confirmed and two radiologically plus serologically confirmed) were reported by two medical faculties (Dicle University Medical Faculty in Diyarbakir and Harran University Medical Faculty in Urfa) in the south-eastern Anatolia region of Turkey.

For the majority of our patients, personal anamnestic, clinical data and addresses were available from their files. Nineteen patients were still alive and contactable and we visited their villages to investigate the risk factors for EA. Ten patients had moved; separatist terrorism in the region has forced people to migrate to other regions or big cities in Turkey. Fourteen patients had died and we did not have the full addresses of four patients. An investigation was performed on the definitive and intermediate host of EA in the villages of the 19 patients.

Epidemiological parameters such as incidence and prevalence, age, sex, occupation and possible risk factors as well as the site of infection in AE patients were evaluated.

### RESULTS

A total of 47 cases of human AE were documented between 1980 and 2000, representing an average of 2.2 cases/year. As far as we know, 19 AE patients are presently living in the south-eastern Anatolia of Turkey. The prevalence is 0.4 cases/100 000 (Table 1).

The distribution of cases according to province reveals that the majority of patients have been or continue to be residents of the eastern provinces of the region, Diyarbakir (n=15, 31.9%), Mardin (n=9, 19.1%) and Batman (n=7, 14.8%) (Figure 1)

The 47 patients consisted of 18 (38.3) males and 29(61.7%) females. The average age was 34.6 years. The average age of men and women was 35.4 and 34.1 years, respectively; the ages of AE patients ranged from 7 to 70 years (males, 7-53 years; females, 9-70 years). Twenty(42.6%) of the 47 patients were between 30 and 40 years old.

**Table 1**

*Prevalence of human alveolar echinococcosis in the south-eastern Anatolia region of Turkey*

| Province   | Inhabitants* | Cases | Cases/ 100000 |
|------------|--------------|-------|---------------|
| Diyarbakir | 1371145      | 8     | 0.6           |
| Urfa       | 1459213      | 3     | 0.2           |
| Siirt      | 270865       | 2     | 0.7           |
| Batman     | 427012       | 3     | 0.7           |
| Mardin     | 683356       | 2     | 0.3           |
| Sirnak     | 345654       | 1     | 0.3           |
| Total      | 4557245      | 19    | 0.4           |

\* 2000 census

**Table 2**

*Incidence of human alveolar echinococcosis in some countries*

| Country                           | Period    | New cases | Av. Per year | Incidence* |
|-----------------------------------|-----------|-----------|--------------|------------|
| Austria<br>(whole country 14)     | 1983-1990 | 14        | 1.8          | 0.02       |
| Germany<br>(Bavaria15)            | 1985-1990 | 50        | 10           | 0.09       |
| France<br>Doubs(16)               | 1960-1992 | 85        | 1.7          | 1.40       |
| Switzerland<br>(whole country 17) | 1984-1992 | 65        | 7.2          | 0.10       |
| Turkey (13)                       |           |           |              |            |
| Whole Country                     | 1980-1990 | 76        | 6.9          | 0.13       |
| Whole Country                     | 1991-1997 | 122       | 17.2         | 0.19       |
| Present study                     | 1980-1990 | 18        | 1.6          | 0.49       |
|                                   | 1991-2000 | 29        | 2.9          | 0.63       |

\*Examples of incidences per year per 100 000 inhabitants.

In 47 patients, lesions occurred in the liver, and in 11 patients, other organs were also involved (lung in four cases, omentum in three, kidney in one, spleen in one and breast in one). In all, 76% of patients had lesions in only one organ; in 24% of cases, more than one organ was involved.

All patients were resident in different villages. We did not establish the existence of any village or district with more than one case. In other words, we did not find any endemic focus. In addition, familiar clustering of AE was not identified.

Data on the occupation was available for 42 AE patients. In all, 31(73.8%) were farmers, three (7.1%) workmen, two (4.3%) unskilled workers and two (4.3%) hunters, and four (9.5%) had other occupations (drivers, teachers). The occupational distribution of all cases showed that the majority of female and male patients were farmers. Out of the 47 patients, 43 were from rural areas; nearly all females in rural areas take part in arable farming and animal husbandry. Of the 47 patients, 33 had a history of dog ownership and 18 had a domestic cat.

## DISCUSSION

Human AE, thought to be a rare helminthic zoonosis, usually has a fatal outcome if untreated and therefore can have a serious public health impact. By the end of the 1980's, endemic areas of AE were known to exist in four countries in central Europe(Austria, France, Germany and Switzerland). Since 1989, AE has been identified in red foxes in six other countries: the Netherlands, Belgium, Luxemburg, Liechtenstein, Poland, and the Czech Republic. AE is also endemic in Turkey, Russia, Ukraine, Moldova, and other states of the former USSR (5).

In Turkey, the first human AE case was described in 1939 (6). Alveolar echinococcosis is a serious health problem in Turkey and is most prevalent in the eastern and south-eastern regions of Turkey (7). Although no detailed published data is available on the development of this infection in animals in Turkey, foxes, rats, wolfs, wild rodents and domestic animals may be responsible (8). There has only been a single report of AE in a wild animal in Turkey, in a fox in the north-west (9).

In the South-Eastern region of Turkey, the life cycle of AE is predominantly sylvatic, involving domestic dogs or free-ranging dogs as definitive hosts and various species of rodents as intermediate hosts. Humans acquire the infection from this cycle by the ingestion of parasite eggs released in the faeces of infected dogs. The contribution of dogs to environmental contamination is very high because there is a high proportion of dog ownership in the region. This aspect of the problem is different from the other endemic countries and the other regions of Turkey.

Most people in this region live in rural areas , and work as arable farmers or in animal husbandry. Seventy percent of our AE patients have been or continue to be farmers. Farmers and their families are exposed to AE to a much higher degree than are people from other professions. The Eastern provinces of this region are very poor, and hygiene and sanitation standards are low. These factors, together with the involvement of the domestic dog as a definitive host, would contribute to the greater transmission opportunities in this region. Some high-risk activities such as hunting or collecting wild herbs were not significant in the region, because only two patients had a history of hunting.

The region is not forested . Some patient's villages are situated in large scrub areas. As a result, the areas are suitable hunting areas for dogs. Craig *et al.* (1) reported

that human AE is not only related to dog ownership or contact, but also the presence of a higher proportion of landscape characterised by scrub/grassland. Free-ranging dogs, closely associated with the human population, disseminate embryophores of the cestode inside or outside the houses(10). In the 19 villages investigated, there were many free-ranging dogs. It is thought that this risk is very important in the region.

The ability of the house mouse to serve as an intermediate host for some European strains of AE has been pointed out (8). Petavy *et al.* (10) noted that the house mouse is not a favourable host because of the severe tissue response around the cyst. We did not find any evidence concerning the house mouse. Domestic cats are clearly less suitable than dogs as final hosts for AE. However, they may contribute to the dispersal of embryophores in rural areas (11). In this study, 18 (38.3%) of the 47 patients had domestic cats.

Information on 157 cases recognised by Uysal *et al.* (12) in Turkey in 1986 revealed that the patients were aged 4-70 years, with 76% between 30 and 50 years. Among the patients whose gender was recorded, 74 were females and 80 were males. The female to male ratio was 0:9. Most of the female patients described themselves as housewives and most of the males considered themselves farmers. The original place of residence was recorded for 146 of the 157 AE patients discussed by Uysal *et al.* (12) and the vast majority (133 cases) came from eastern Anatolia, central Anatolia or the shores of the Black Sea.

On the other hand, 159 cases of AE were reported by Altintas *et al.* (13), in Turkey between 1980 and 1998, excluding south-eastern Anatolia. Of these cases, only 89 were reported from eastern Anatolia. Together with our 47 cases, a total of 206 cases have been recorded in Turkey since 1980.

This study revealed that 29 (61.7%) of the AE patients were females and 18 (38.3%) were males and the female to male sex ratio was 1:6. Altintas *et al.* (13) reported that the female to male ratio was 1:4 in their study, whereas Uysal *et al.* (12) noted that the female to male sex ratio was 0:9. In south-eastern region, women are at higher risk for AE than men. There are two possible explanations for this situation; Females in rural areas in this region take part in farming and animal husbandry more than males, because male farmers work in the western provinces of Turkey as seasonal workers for a few months of nearly every year. As a result, female farmers are exposed to the possible risk factors for AE more than males and women have more contact with dogs than men do, because they prepare all household food.

From 1980 to 1998 in eastern Anatolia (13) and from 1980 to 2000 in south-eastern Anatolia, the incidence was 4.6 cases/year and 2.2 cases/year, respectively. According to these results, the south-eastern Anatolia region is the second most prevalent area in Turkey. The region has a higher incidence than both Turkey as a whole and some European countries. Some incidences (14-17) are presented in Table 2.

We did not find any endemic focus in the south-eastern region of Turkey. This study shows that the region has a higher incidence than Turkey as a whole, and domestic dogs and free-ranging dogs seem to be the main source of the infection.

## REFERENCES

1. Craig, P.S., Liu, D., Marcpherson, C.N.L., *et al.* A Large focus of alveolar echinococcosis in central China. *Lancet.* 1992; **340**:826-831.
2. Raush, R.L. Life cycle patterns and geographic distribution of Echinococcus species. In: Thompson, R.C.A., Lymbery, A.J. (Eds). Echinococcus and Hydatid Disease. CAB International, Wallingford. 1995; 89-134.
3. Ammann, R. and Eckert, J. Clinical diagnosis and treatment of echinococcosis in humans. In: Thompson, R.C.A., Lymbery, A.J. (Eds). Echinococcus and Hydatid Disease. CAB International, Wallingford. 1995; 411-463.
4. Craig, P.S., Giraudoux, P., Shi, D., *et al.* An epidemiological and ecological study of human alveolar echinococcosis transmission in south Gansu, China. *Acta. Trop.* 2000; **77**:167-177.
5. Eckert, J. and Deplazes, P. Alveolar echinococcosis in humans: The current situation in central Europe and the need for counter measures. *Parasit. Today.* 1999; **15**:315-319.
6. Altintas, N. Cystic and alveolar echinococcosis in Turkey. *Annals Trop. Med. and Paras.* 1998; **92**:637-642.
7. Akinoglu, A., Demiryurek, H. and Guzel, C. Alveolar hydatid disease of the liver: A report on thirty-nine surgical cases in eastern Anatolia, Turkey. *Am. J. Trop. Med. Hyg.* 1991; **45**:182-189.
8. Martynenko, V.B., Shubin, A.G., Mordosov, *et al.* The possibility of the occurrence of village foci of alveolar hydatidosis in Yakutiya. *Meditsinskaya Parazitologiya I Parazitarnye Bolezni, Moskva.* 1984; **62**:25-27.
9. Keles, M. and Palancl, A. Ekinokokkus alveolaris (Erzururn ve cevresinde gorulen 54 vakamn butun yonleriyle incelenmesi). *Doguda Saglik Tlp Dergisi.* 1985; **1**:19-23.
10. Petavy, A.F., Deblock, S. and Walbaum, S. Life cycles of Echinococcus multilocularis in relation to human infection. *J. Parasitol.* 1991; **77**:133-137.
11. Petavy, A.F., Prost, C., Gevrey, J., Gilot, B. and Debluck, S. Infestation naturelle du chat domestique (*Felis catus* L.) par Echinococcus multilocularis Leuckart, 1863 (Cestoda): Premier cas en France decele en zone peri-urbaine. *Comptes Rendus de l'Acade'mie des Sciences, Paris.* 1988; **307**:553-556.
12. Uysal, V. and Paksoy, N. Echinococcosis multilocularis in Turkey. *J. Trop. Med. Hyg.* 1986; **89**:249-255.
13. Altintas, N., Yazar, S., Yolasigmaz, *et al.* Turkiye'de 1980-1998 yillari arasinda saptanan alveolar echinococcosis olgulari. *T. Parazitoloji Derg.* 1999; **23**:133-136.
14. Auer, H. and Aspöck, H. Incidence, prevalence and geographic distribution of human alveolar echinococcosis in Austria from 1854 to 1990. *Parasitol. Res.* 1991; **77**:430-436.
15. Nothdurft, H.D., Jelinek, T., Mai, A., Sigl, B., Sonnenburg, F. and Loscher, T. Epidemiology of alveolar echinococcosis in southern Germany (Bavaria). *Infection.* 1995; **23**:85-88.
16. Bresson-Hadni, S., Laplante, J. J., Lenys, D., *et al.* Seroepidemiologic screening of Echinococcus multilocularis infection in a European area endemic for alveolar echinococcosis. *Am. J. Trop. Med. Hyg.* 1994; **51**:837-846.
17. Ecker, J., Jacquier, P., Baumann, D. and Raeber, P.A. Human echinococcosis in Switzerland. 1984-1992. *Schweiz. Med. Wochenschr.* 1995; **125**:1989-1998.