Human hydatidosis in Tehran, Iran: A retrospective epidemiological study of surgical cases between 1999 and 2009 at two university medical centers

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Abstract. This study was conducted to evaluate retrospectively 203 patients diagnosed with hydatid cyst disease and treated surgically at two university medical centers between 1999 and 2009 in Tehran, the capital of Iran. Cystic echinococcosis (CE) affected more females 117 (57.6%) than males 86 (42.4%). A remarkable gender difference skewed towards females was observed, and the male/female ratio among CE cases ≤20 and ≥61 years old was 1.18 and 0.52, respectively. The age of the patients ranged from 8 to 82 years, and the age group 21-40 years (42.8%) was the most affected. A significantly higher number of hydatid cysts were recorded in the liver than in other sites (P<0.001), and more females had higher hepatic cysts (48.8%) than males (35.0%). The ratio of hepatic hydatidosis to pulmonary hydatidosis was about 11. There was a relative direct relationship between the ratio of liver to lung cases and age, and this ratio was significantly (P<0.01) higher in individuals >40 years of age (liver/lung ratio ≥43) than in those <40 years of age (liver/lung ratio 2.8–7.6). Unusual cyst locations in kidneys, brain and pelvic area, followed by spleen and spine was also observed. Single organ involvement was found in 95% of the patients, and was more common in females (55.2%) than in males (39.9%). Housewives had the highest rate of infection (53.5%) followed by labourers with 14.8%, which showed a significant difference (P<0.001). Similarly, urban dwellers was also over-represented among the cases (87% urban vs. 13% rural; P<0.001). In 69.5% of cases there was only one cyst, 16.3% had two cysts, 4.7% with three cysts, and 9.5% had four cysts or more. The results showed that further studies are needed to determine the prevalence, economic impact and risk factors of the disease in the area.

INTRODUCTION

Cystic echinococcosis (CE), also known as hydatidosis, is a zoonotic parasitic disease of human and mammalians caused by the larval stage of dog tapeworm *Echinococcus granulosus*. The parasite is found worldwide, including in the Middle East and North Africa from Morocco to Egypt (Eckert & Deplazes, 2004; Sadjjadi, 2006; Ahmadi & Hamidi, 2008). In Iran, CE is endemic and is maintained in three distinct cycles: a dog/livestock domestic cycle, a dog/camel desert

cycle, and a sylvatic cycle between wild carnivores and wild ruminants (Ahmadi & Hamidi, 2008). Dogs play an important role in transmitting the disease in Iran. In a series of studies that were carried out in Iran, the infection rate of *E. granulosus* in the definitive host dog was reported to range from 2% to 63% (Fallah *et al.*, 1995; Eslami & Hosseini, 1998; Maleky & Moradkhan, 2000; Dalimi *et al.*, 2006), while in the intermediate hosts such as sheep, goats, cattle and camels it ranged from 1.5% to 70% (Ahmadi, 2005; Ansari-Lari, 2005; Sadjjadi, 2006; Ahmadi &

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Meshkekar, 2011). In a recent study of echinococcosis in stray dogs, a very high prevalence of E. granulosus was reported from Tehran province (Maleky & Moradkhan, 2000). Human cases are regularly reported from different regions of the country (Nasseh & Khadivi, 1975; Ahmadi & Dalimi, 2006; Pezeshki et al., 2007; Ahmadi & Hamidi, 2008, 2010; Sarkari et al., 2010). It remains one of the serious health problems of the area and exerts heavy impact on the economy and social welfare of the people in Iran. Human hydatid disease is responsible for approximately 1% of admission to surgical wards, in Iran (Ahmadi & Hamidi, 2008; Sarkari et al., 2010). Careful study of retrospective hospital records provides a useful tool to evaluate the regional epidemiological situation of the disease (Pierangeli, et al., 2007; Ahmadi & Hamidi, 2008). However, from Tehran (capital of Iran), there is no figure available at international level concerning the human CE in university medical centers. Therefore, this retrospective study was undertaken to study epidemiological and some clinical characteristics of hydatidosis treated surgically at two university medical centers in Tehran over a 10-year period.

MATERIALS AND METHODS

A retrospective study was conducted by reviewing the records of patients that had been surgically treated for hydatid cysts between 1999 and 2009 at two large university hospitals in Tehran: Shohada-Tajrish and Imam-Hossein hospitals. People from different parts of the province are referred to these hospitals for surgery. Data such as age at the time of surgery, gender, occupation, place of residence, number of cyst and the anatomic location of cysts were recorded. The ratios between male and female cases and ratios between liver and lung involvement were calculated. The collected data were statistically analysed using SPSS software. Values of P < 0.05 were accepted as significant.

RESULTS

The total number of CE surgeries recorded in two university medical center registries over a 10-year period (1999-2009) in Tehran were 203 cases (Table 1).

Age and gender distribution pertaining to 203 confirmed cases of human CE and the male/female ratio by age groups are shown in Table 2. The youngest patient operated was 8 years old and the oldest was 82 years of age. Age group 21–40 years was the most affected and represented 42.8% of the total number of cases. An overall analysis of gender and age of the CE cases revealed that the number of females infected was higher than that of males with a 0.73 male to female ratio (Table 2), while the gender ratio in the general population is about 1.00.

Table 1. Number of surgical cases for cystic echinococcosis during 1999–2009 at two university medical centers in Tehran, Iran

Years	Number of surgical cases in two medical centres					
Tears	Shohada- Tajrish	Imam Hossein	Total (%)			
1999-2000	10	11	21 (10.3)			
2000-2001	10	7	17 (8.4)			
2001-2002	14	13	27 (13.3)			
2002-2003	11	14	25 (12.3)			
2003-2004	7	10	17 (8.4)			
2004-2005	8	4	12 (5.9)			
2005-2006	12	8	20 (9.9)			
2006-2007	7	6	13 (6.4)			
2007-2008	16	15	31 (15.3)			
2008-2009	11	9	20 (9.9)			
Total	106	97	203 (100.0)			

Table 2. Age and gender distribution of surgically confirmed cystic echinococcosis cases at two university medical centers in Tehran, Iran (1999-2009)

Age	Numb	Number and (%) of cases				
(years)	Male	Female	Total	female ratio ^a		
<u>≤</u> 20	13 (6.4)	11 (5.4)	24 (11.8)	1.18		
21 - 40	36 (17.7)	51 (25.1)	87 (42.8)	0.71		
41 – 60	21 (10.4)	24 (11.8)	45 (22.2)	0.87		
≥61	16 (7.9)	31 (15.3)	47 (23.2)	0.52		
Total	86 (42.4)	117 (57.6)	203 (100.0)	0.73		

^a The gender ratio in the general population was about 1.00.

Hydatid cyst distribution in various anatomical sites is shown in Table 3. The involvement of liver was the most frequent, and the second most frequent location was the lung (Table 3). The incidence of hepatic CE was significantly higher than that of CE in other organs (P<0.001), and more females had higher hepatic cysts (48.8%) than males (35.0%) (Table 4). Single organ involvement was found in 95% of the patients (Table 3), and was more common in females (55.2%) than in males (39.9%).

The age-group distribution of the patients with liver and/or lung cysts is shown in Table 4. The ratio of hepatic hydatidosis to pulmonary hydatidosis was about 11. There was a direct relationship between this ratio and age group, and this ratio in \leq 20 years old was 2.8, with \geq 43 in 41-60 years old or older

(Table 4). The findings indicated that the hepatic hydatid disease predominated in every age-group (Table 4).

In the present study 69.5% (132 cases) only had one cyst, and 9.5% (18 cases) had four cysts or more (Table 5). As observed in Fig. 1, a significantly (P< 0.001) higher CE was recorded in the housewives (53.5%) than other occupational groups. Similarly, urban dwellers were also over-represented among the cases (87% urban vs. 13% rural; P<0.001).

DISCUSSION

Hydatid disease poses an important socioeconomic and public health problem in many areas of the world. In Iran, CE is an endemic problem that is seen in both human (Nasseh

Table 3. Location of hydatid cyst with single and multiple organ involvement at two university medical centers in Tehran, Iran (1999-2009)

Cyst location	Male	Female	Total
	n (%)	n (%)	n (%)
Single- organ involvement			
Liver only	66 (32.51)	94 (46.30)	160 (78.81)
Lung only	7 (3.45)	6 (2.96)	13 (6.40)
Kidney	3 (1.48)	1 (0.49)	4 (1.97)
Brain	2 (0.99)	2 (0.99)	4 (1.97)
Ovary	_ ` ´	1 (0.49)	1 (0.49)
Supra-pubic	_	1 (0.49)	1 (0.49)
Leg	_	1 (0.49)	1 (0.49)
Buttock	_	1 (0.49)	1 (0.49)
Intraperitoneum	_	1 (0.49)	1 (0.49)
Epigastrium	_	1 (0.49)	1 (0.49)
Pelvis	2 (0.99)	1 (0.49)	3 (1.48)
Spine	1 (0.49)	1 (0.49)	2 (0.99)
Bilous canals	_	1 (0.49)	1 (0.49)
Total	81 (39.90)	112 (55.17)	193 (95.07)
Multiple- organ involvement			
Liver+lung	_	2 (0.99)	2 (0.99)
Liver+spleen	1 (0.49)	1 (0.49)	2 (0.99)
Liver+abdomen	1 (0.49)	_	1 (0.49)
Liver+ pelvis	1 (0.49)	1 (0.49)	2 (0.99)
Liver+ kidney	1 (0.49)	_	1 (0.49)
Liver+epigastrium	1 (0.49)	_	1 (0.49)
Liver+ bilous canals	_	1 (0.49)	1 (0.49)
Total	5 (2.46)	5 (2.46)	10 (4.93)
Overall	86 (42.36)	117 (57.64)	203 (100.0)

n: no. of cases

Table 4. Distribution of surgically confirmed cystic echinococcosis cases according to age groups and site of cysts in Tehran, Iran (1999 -2009)

	Liver			Lungs			C	Other organs		
Age Liver/Lung (years)	M n	F n	Total n (%)	M n	F n	Total n (%)	M n	F n	Total n (%)	ratio
<u>≤</u> 20	9	5	14 (6.9)	2	3	5 (2.5)	2	2	4 (1.9)	2.8
21-40	25	44	69 (34.0)	5	4	9 (4.4)	7	7	14 (6.9)	7.6
41-60	21	22	43 (21.2)	_	1	1 (0.5)	2	3	5 (2.5)	43
≥61	16	28	44 (21.6)	_	_	_	2	3	5 (2.5)	>43
Total ^a	71 (35.0)	99 (48.8)	170 (83.7)	7 (3.4)	8 (3.9)	15 (7.4)	13(6.4)	15(7.4)	28 (13.8)	11.3

M = Male; F = Female; n : no of cases

Table 5. The number of cysts among 190 cystic echinococcosis cases at two university medical centers in Tehran, Iran (1999-2009)

Male n (%)	Female n (%)	Total n (%)	
53 (27.9)	79 (41.6)	132 (69.5)	
18 (9.5)	13 (6.8)	31 (16.3)	
3 (1.6)	6 (3.2)	9 (4.7)	
7 (3.7)	11 (5.8)	18 (9.5)	
	n (%) 53 (27.9) 18 (9.5) 3 (1.6)	n (%) n (%) 53 (27.9) 79 (41.6) 18 (9.5) 13 (6.8) 3 (1.6) 6 (3.2)	

n:no. of cases

& Khadivi, 1975; Ahmadi & Dalimi, 2006; Pezeshki et al., 2007; Ahmadi & Hamidi, 2008; Sarkari et al., 2010) and livestock animals (Ahmadi, 2005; Ansari-Lari, 2005; Ahmadi & Dalimi, 2006; Ahmadi & Meshkekar, 2011). In many parts of Iran, the infection rate of E. granulosus in the definitive host (dog) was reported to be high, and in Tehran, almost one in every two (48%) of the stray dogs investigated by Maleky & Moradkhan (2000) was found infected.

In the absence of statistically sound epidemiologic data, retrospective analysis of hydatidosis based on medical records in regional hospitals may prove useful. Hence, our study presented 203 patients surgically treated for CE at two university medical centers in Tehran (capital of Iran), namely Shohada-Tajrish and Imam-Hossein hospitals with 106 cases (52.22%) and 97cases (47.78%), respectively. The highest number of CE cases (15.3%) was recorded in 2007-2008 and the lowest (5.9%) in 2004-2005 (Table 1).

In the current survey, most of the CE cases were middle-aged. Similar observations have been made elsewhere in Iran (Pezeshki et al., 2007; Ahmadi & Hamidi, 2008; Sarkari et al., 2010). In their recent study of the CE cases admitted to three university hospitals in Hamedan province, Ahmadi & Hamidi (2008) found that the age-group who presented with the largest number of cases was 20-39 years, followed by 40–59 years, and this is also true for CE at Tehran Milad Hospital in Iran (Pezeshki et al., 2007). In Argentina (Pierangeli et al., 2007) and Jordan (Al-Qaoud et al., 2003), however, young adult CE cases (aged 15-29 years in Argentina and 16–30 years in Jordan) were more common than in the younger or older age-groups. Cystic echinococcosis is a chronic disease and hydatid cyst grows very slowly in human when compared to animals. It takes years for a hydatid cyst to bring on the clinical disease in human host. This might explain the reason for the high rate of surgeries for CE in people who are in the age group 21-40 years old.

Women are the main sufferers of CE in our study with a male: female ratio of 0.73, while the gender ratio in the general population was about 1.00. A remarkable gender difference skewed towards females was observed in the age group \geq 61 years with male to female ratio being 0.52 (Table 2). These findings are in line with other studies by Saeed *et al.* (2000), Todorov & Boeva (2000), Al-Qaoud *et al.* (2003), Pezeshki *et al.* (2007) and Ahmadi & Hamidi (2008). The reason for this is not quite clear but handling

^a Each infected organ separately accounted in multiple organs involvement

and eating of raw vegetables (mainly contaminated with E. granulosus eggs) by women in the studied area might be accounted for this (Ahmadi & Hamidi, 2008). However, this finding contrasted with reports from east Iran (Nasseh & Khadivi, 1975) and Kyrgystan (Torgerson et al., 2003) where the number of surgical cases in adult men was higher than in women. Unlike adult cases, young males ≤20 years old showed considerable higher surgical incidence than females of comparable age at a ratio of 1.18 (Table 2). The variation in the male: female ratio with age group was not, however, statistically significant. A similar difference between the genders (in <20 years) was noted in Hamedan province of Iran (Ahmadi & Hamidi, 2008), Bulgaria (Todorov & Bovea, 2000) and Jordan (Al-Qaoud et al., 2003).

Our analysis of cyst locations showed that different organs were involved with CE but liver is the most commonly affected organ in both males and females. This observation is consistent with reports from several other studies, not only in Iran (Nasseh & Khadivi, 1975; Pezeshki et al., 2007; Ahmadi & Hamidi, 2008; Sarkari et al., 2010) but also in Iraq, Bulgaria, Jordan and Kyrgystan (Saeed et al., 2000; Todorov & Boeva, 2000; Al-Qaoud et al., 2003; Torgerson et al., 2003). Overall, incidence of hepatic CE observed in the present study was 11 times more than pulmonary CE (Table 4), indicating that this difference was statistically significant (P<0.001). The findings showed that the hepatic hydatid disease predominated in every age-group (Table 4), in accordance with studies by Ahmadi & Hamidi (2008) in Iran and Torgerson et al. (2003) in Kyrgystan. The present study showed that there was a relative direct relationship between ratio of liver to lung cases and age group. This ratio was significantly (P<0.01) higher in individuals >40 years of age (liver/lung ratio \geq 43) than in those <40 years of age (liver/lung ratio 2.8-7.6) (Table 4). Pathophysiological influences on the anatomic location of cysts are still unknown (Zahawi et al., 1999); however, it is likely the liver is more commonly infected because Echinococcus oncospheres after penetrating the intestinal wall are disseminated to the

liver via the portal vein. These oncospheres are first filtered through the liver capillaries before they can reach other organs. Majority of previous studies have reported that the lungs were more frequently invaded than the liver in children and adolescents (Todorov & Boeva, 2000; Al-Qaoud et al., 2003; Pierangeli et al., 2007), but for this work it was reversed. As was observed in this study, Ahmadi & Hamidi (2008) in Iran and Torgerson et al. (2003) in Kyrgystan, also reported that the hepatic hydatid disease was more commonly diagnosed and operated in children and adolescents.

In the present study, kidneys, brain and pelvic area, followed by spleen and spine were the organs where hydatid cyst was most frequently observed after the liver and lungs. About 13.8 % of confirmed cases of hydatid disease were diagnosed with localization other than in the liver and lungs (Table 4). Such results were reported in Iran (Ahmadi & Hamidi, 2010) and Turkey (Engin et al., 2000; Çöl et al., 2003). Unusual localizations of parasitosis range from 5% to 30% (Versaci et al. 2005; Ahmadi & Hamidi, 2010). These forms are of interest not only for epidemiological reasons but also because of the controversial pathogenesis and diagnostic problems that sometimes lead to an unclear clinical diagnosis.

In the studied hospitals, the vast majority (95%) of CE cases seeking surgical treatment showed only single-organ involvement. A similar percentage of the CE cases were investigated in Iran (Ahmadi & Hamidi, 2008; Sarkari et al., 2010) and Argentina (Pierangeli et al., 2007), but only 68.8% of the Iraqi cases studied by Molan (1993) showed single-organ involvement. Normally, most of the patients with primary CE have single organ involvement and harbour a solitary cyst (Eckert & Deplazes, 2004) as was observed in this study. Nearly 69.5% of the CE cases in our study presented with a single cyst followed by two, three and four or more with 16.3%, 4.7 and 9.5%, respectively (Table 5).

Overall incidence of CE was significantly higher in housewives (Fig. 1), and a significant difference in incidence was observed compared to other occupations (P<0.001). More contact with contaminated

soil, handling and eating contaminated raw vegetables imposed an important factor to cause more infection in housewives of the region. This result is similar to those reported in Iran (Pezeshki *et al.*, 2007; Ahmadi & Hamidi, 2008; Sarkari *et al.*, 2010), and Iraq (Saeed *et al.*, 2000).

The results showed that the prevalence of CE in the urban dwellers was significantly higher than the rural dwellers (87% urban vs. 13% rural; P<0.001). The majority of previous studies have reported that the rural dwellers were more frequently infected than the urban dwellers in Iran (Nasseh & Khadivi, 1975; Ahmadi & Hamidi, 2008), Iraq (Saeed et al., 2000), Jordan (Al-Qaoud et al., 2003) and Argentina (Pierangeli et al., 2007)), but for this work it was reversed.

In conclusion, CE remains an important health problem in many areas of Iran, including Tehran province. The findings of this study merit for more extensive epidemiological investigations of CE in human to determine the prevalence, economic impact and risk factors of the disease in the area. Also, effective programmes need to be implemented for controlling and reducing the disease in livestock, carnivores especially dogs and also humans in the region.

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