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Demographics of Patients with Surgical and Nonsurgical Cystic Echinococcosis in East Azerbaijan from 2001 to 2012

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Abstract: Echinococcosis is very frequent helminthic infection in human being. The disease is endemic in Iran and particularly in its northwestern region, East Azerbaijan. The objective of this study was to study demographics of patients with cystic echinococcosis in East Azerbaijan; as well as to report the sites of involvements. In this cross-sectional study, 318 patients with surgical and nonsurgical cystic echinococcosis were recruited from three referral centers in Tabriz within a 10-year period of time. The patients' demographics (age and gender) and the site of infection were investigated. Among the patients, females were predominant (57.5 vs. 42.5%). The mean age of patients was 32.59±18.47 (range: <1-87) years and the most frequently age group was 20-30 years (23.6%), followed by the age groups of 30-40 years (21.7%), 10-20 years (14.2%), 40-50 years (11.9%), 50-60 years (11.3%), <1 year (6%), 60-70 years (4.7%), 1-10 years (4.1%) and finally, >70 years (2.5%). The lung was the most frequently infected organ (48.1%), followed by the liver (28.9%), gallbladder (3.5%), spleen and kidney (each one in 2.2%), abdomen, intestine, pleura, bronchus and mediastinum (each one in 1.6%), chest wall and heart (each one in 1.3%), brain, pericardium, facial sinus, pancreas and esophagus (each one in 0.6%) and the ovary, uterus, axillary region, pelvis and femur (each one in 0.3%). In conclusion, this study showed that the patients with hydatid cysts in East Azerbaijan Province are mainly females in the age group of 20-40 years. The lung and liver are the major sites of infection in these patients.

Key words: Hydatid cysts, echinococcus granulosus, demographics, epidemiology infection

INTRODUCTION

One of the most prevalent helminthic infections all over the world is Echinococcus (hydatidosis) which occurs from the Middle East, Asia, Africa and Europe, to the American continents (Altintas, 2003; Moro and Schantz, 2006; Magambo *et al.*, 2006; Akhtar *et al.*, 1998). One of the endemic areas in this regard is Iran (Shahnazi *et al.*, 2011) in which, previous studies from different parts have reported a seropositivity rate of 3-5.9% (Sadjjadi, 2006; Pazoki *et al.*, 2006). The incidence of the surgical disease has increased from 5 cases per 100,000 to 27 cases per 100,000 in the past decade. Many of these patients have been reported to be children (Torgerson *et al.*, 2006). Due to its high prevalence and consequent morbidity, cystic echinococcosis imposes increasing public health and economic concern (Dakkak, 2010). Echinococcus is caused by *Echinococcus granulosus*, a larval cestode which is mainly transmitted between dogs and domestic livestock. However, human being could also be infected in this cycle (Craig and Larrieu, 2006; Al-Qudah *et al.*, 2008; Ghazani *et al.*, 2008; Mirani *et al.*, 2000; Sabry and Lotfy, 2009; Haroun *et al.*, 2008; Mellau *et al.*, 2010;

Azlaf and Dakkak, 2006; Bardonnat *et al.*, 2003). In this parasitic disease of mammals, metacestodes play a pivotal role. Families of dogs or cats host the mature worms in their intestines. Many species of herbivorous or omnivorous mammals are regarded as intermediate hosts which contain metacestodes in their tissues. These metacestodes can be found as large cysts (cystic echinococcosis or hydatid disease), as structures containing vesicles (alveolar echinococcosis), or as an intermediate form forms (polycystic echinococcosis). When human beings ingest the cestode eggs, they become infected (Romig *et al.*, 2011; Sabry, 2007). It is reported that near 70-80% of hydatid cysts in human cystic echinococcosis are found in the liver, 10-20% in the lungs and almost 5% in other sites (Eckert *et al.*, 2001; Torgerson and Budke, 2003). Although, it is not definitely proved, previous reports have shown that various risk factors may be connected with human cystic echinococcosis including livestock raising, pastoral life, occupation in rangeland, female gender, ethnicity, dog ownership, poor hygiene and low socioeconomic status (Dowling *et al.*, 2000; Larrieu *et al.*, 2002). This study aimed to investigate the demographics of patients with cystic echinococcosis in East Azerbaijan, Northwestern

Iran; as well as to determine the sites of involvements in these patients. Although the disease is very frequent in this region, there is no similar study available in the literature.

MATERIALS AND METHODS

Study design and patients: In this descriptive, cross-sectional study, 318 patients with definite diagnosis of surgical and nonsurgical cystic echinococcosis were recruited from three main referral teaching centers (Imam Khomeini, Imam Reza, Sina) in Tabriz, Northwest of Iran; within a 10 year period from 2001-2012. This study was approved by the ethics committee of Tabriz University of Medical Sciences.

Variables: Patients’ age and gender, as well as the location of infection were documented in all cases with definite diagnosis of human cystic echinococcosis. The sites of involvement were also reported stratified by the patients’ age and gender.

Statistical analysis: Data were analyzed using the SPSS statistical software package (version 18.0; SPSS Inc, Chicago). Variables are shown as Mean±Standard deviation (range), or frequency (percentage).

RESULTS

Three hundred eighteen patients with hydatid cysts, including 183 females (57.5%) and 135 (42.5%) with a male to female ratio (M/F) of 0.74 were studied (Fig. 1).

The mean age of the patients was 32.59±18.47 (range: <1-87) years, including the age groups of under 1 year (n = 19), 1-10 years (n = 13), 10-20 years (n = 45), 20-30 years (n = 75), 30-40 years (n = 69), 40-50 years (n = 38), 50-60 years (n = 36), 60-70 years (n = 15) and over 70 years (n = 8) (Fig. 2).

The hydatid cysts were present in the lung in 153 patients, in the liver in 92 patients, in the gallbladder in 11 patients, in the kidney and spleen each one in 7 patients, in the abdominal cavity, intestine, pleura, bronchus and mediastinum each one in 5 patients, in the chest wall and heart each one in 4 patients, in the brain, esophagus, pericardium, facial sinuses and pancreas each one in 2 patients and in the ovary, uterus, axillary cavity, pelvis and femur each one in 1 patient (Fig. 3).

Stratified by the patients’ gender, involvement of the spleen, abdomen, intestine, facial sinus, esophagus, axillary region and pelvis was predominant in the male patients while involvement of the lung, liver, gallbladder, kidney, pleura, bronchus, mediastinum, chest wall, heart,

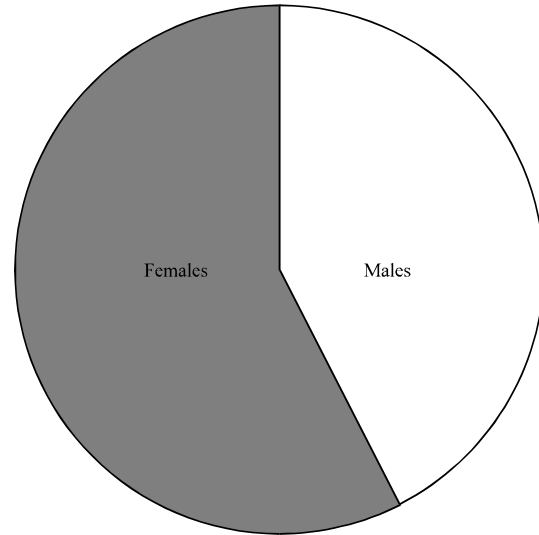


Fig. 1: Gender of the studied patients with hydatid cysts

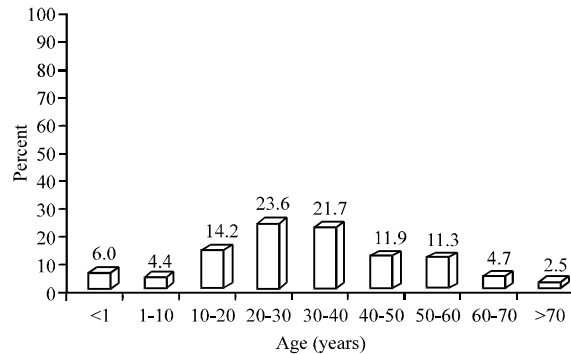


Fig. 2: Age groups of the studied patients with hydatid cysts

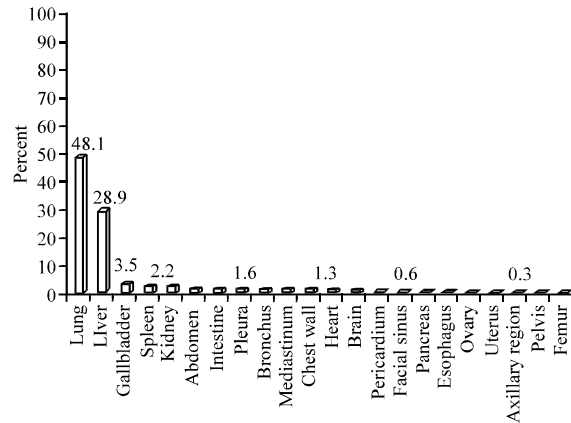


Fig. 3: Locations of hydatid cyst in the studied patients

Table 1: Locations of the hydatid cyst in the studied patients stratified by their gender

Location	Gender	
	Male	Female
Lung	76 (49.7)	77 (50.3)
Liver	29 (31.5)	63 (68.5)
Gallbladder	3 (27.3)	8 (72.7)
Spleen	4 (57.1)	3 (42.9)
Kidney	1 (14.3)	6 (85.7)
Abdomen	3 (60.0)	2 (40.0)
Intestine	3 (60.0)	2 (40.0)
Pleura	2 (40.0)	3 (60.0)
Bronchus	1 (20.0)	4 (80.0)
Mediastinum	2 (40.0)	3 (60.0)
Chest wall	1 (25.0)	3 (75.0)
Heart	1 (25.0)	3 (75.0)
Brain	1 (50.0)	1 (50.0)
Pericardium	1 (50.0)	1 (50.0)
Facial sinus	2 (100.0)	0 (0.0)
Pancreas	1 (50.0)	1 (50.0)
Esophagus	2 (100.0)	0 (0.0)
Ovary	0 (0.0)	1 (100.0)
Uterus	0 (0.0)	1 (100.0)
Axillary region	1 (100.0)	0 (0.0)
Pelvis	1 (100.0)	0 (0.0)
Femur	0 (0.0)	1 (100.0)

Data presented as frequency and values in parenthesis represents the percentage values

ovary, uterus and femur was more frequent in the female patients. Involvement of the brain, pericardium and pancreas was equal in both the male and female patients (Table 1).

Considering the patients' age groups, involvement of the lung, liver, pleura and ovary was seen most frequently among the age group of 20-30 years. Involvement of the spleen, kidney, mediastinum, uterus, pelvis and femur was more frequent among the age group of 30-40 years. Hydatid cysts of the abdomen and intestine were more common in the age group of 50-60 years. Involvement of the bronchus and heart was more frequent among the age group of 40-50 years. Involvement of the gallbladder was more common in the age group of 30-50 years. Involvement of the chest was more frequent in the patients aged 10-50 years while the pericardium was involved frequently among the patients aged 30-40 years and 50-60 years. The facial sinus was involved in the patients aged under 1 year, as well as in those aged 30-40 years. The brain was involved only in the patients aged 10-20 years and the axillary region only in patients under 1 year. The pancreas and esophagus were involved in the patients aged 20-40 years and over 60 years, respectively.

DISCUSSION

In the present study, demographics of 318 patients with surgical and nonsurgical cystic echinococcosis, as well as the frequency of infected locations were evaluated in the Northwestern Iranian Province of East Azerbaijan.

This is the first epidemiological survey in this area which is considered as an endemic region for the disease according to the official reports.

Based on the findings, females were predominant (57.5 vs. 42.5%; a M/F ratio = 0.74) and the mean age was 32.59±18.47 (range: <1-87) years. The most frequently age group was 20-30 years (23.6%), followed by the age groups of 30-40 years (21.7%), 10-20 years (14.2%), 40-50 years (11.9%), 50-60 years (11.3%), <1 year (6%), 60-70 years (4.7%), 1-10 years (4.1%) and finally, >70 years (2.5%). The lung was the most commonly involved organ (48.1%), followed by the liver (28.9%), gallbladder (3.5%), spleen and kidney (each one in 2.2%), abdomen, intestine, pleura, bronchus and mediastinum (each one in 1.6%), chest wall and heart (each one in 1.3%), brain, pericardium, facial sinus, pancreas and esophagus (each one in 0.6%) and the ovary, uterus, axillary region, pelvis and femur (each one in 0.3%). Available data in the literature are very heterogeneous in this regard and vary greatly based on the region of investigation.

In a pioneer study by Amr *et al.* (1994) 306 subjects with hydatid cysts were studied in Jordan. Among these patients, 60% were females and 40% were males. The liver was the most commonly infected organ (57.8%), followed by the lung (26.4%). Other infrequently involved sites were the kidneys, spleen, brain, soft tissues of neck, appendix and mesentery. Over 35.8% the cases were aged <21 years.

In line with this study, majority of our patients were females, either. Although the liver and lung are also two frequently involved organs in Amr *et al.* (1994) study, in contrast to this finding the lung preceded the liver in term of the most commonly infected site in our series. The other mentioned involved sites were also found in our patients.

In a Chinese study by Xu *et al.* (2002), 560 patients with cystic echinococcosis were evaluated. In this study, over 36% of the patients were under the age of 14 years. This is in contrast with our finding and also this common idea that the children are main victims of hydatid cysts (Romig *et al.*, 2011). These conflicting results most probably originate from the selected studied communities. In the present study, the patients were recruited from three main referral centers in Tabriz, the capital of the East Azerbaijan Province. It's noteworthy that there is also another referral centre in this city which is dedicated to the children and neonates. As this children hospital was out of the sampling source in the present study, lower rate of pediatric patients is estimated.

In another study in Kazakhstan, Torgerson *et al.* (2002) evaluated 1405 patients with cystic echinococcosis among them 7.8% were under 6 years old and 23.4% aged between 7 and 16 years of age. In adults 53.4% were females and 46.6% males.

Although, gender prevalence of this study is in accordance with our finding, conflicting data regarding the age groups can be justified again by the mentioned sampling method in the preset survey.

In Turkey, Miman *et al.* (2010) studied 91 patients with cystic echinococcosis. The male patients consisted 59% of the cases vs. 41% for the females; with a mean age of 36.2 years. Lung and liver were the most common sites of involvement (totally 73.6%).

Although, in contrast with our population male patients were predominant in this study, the mean age of the patients and the most prevalent involved sites are similar.

In an Iranian series by Ahmadi and Badi (2011) in Tehran, 203 patients diagnosed with hydatid cyst disease were evaluated. The disease affected more females (57.6%) than males (42.4%). The age group 21-40 years (42.8%) was the most affected one. Other unusual locations were the kidneys, brain and pelvic area, followed by spleen and spine. In this study, the authors concluded that the gender and age are two major determinants of the site of involvement in patients with hydatid cyst disease.

Finding of this study in term of the gender and age of patients with hydatid cyst disease, as well as the sites of involvement are in line with ours. Interestingly, we also found that the age group of 20-40 years was the most commonly involved one in these patients.

This age and gender-dependency of the disease is also emphasized in another study by Oudni-M'Rad *et al.* (2007). They evaluated 121 Tunisian children with hydatid cysts and found that the lung was the most commonly involved site (59%) followed by the liver (35%). Infection was more common among the females (F/M ratio = 1.96) and the greatest number of cases was seen in the 4-9 year age groups.

Except for the most frequently reported age group, the other data including gender ratio and common sites of infection are similar to our findings.

In another Iranian study by Sarkari *et al.* (2010) in the southwestern Yasuj district, 36 cases with cystic echinococcosis were examined. Among these patients 49.6% were females and 50.4% were males and the highest rate of infection was in the cases aged between 30 and 39 years. Hepatic cyst was recorded in 81% of the cases and nephrotic cysts were the second most prevalent ones.

In comparison with our finding, they found an almost equal involvement in both males and females and older age of patients was reported. In addition, they reported a higher rate of the liver involvement in comparison with the lung. Low number of the cases is a great limitation in this series.

In an Italian study, Conchedda *et al.* (2010) studied 540 patients with cystic echinococcosis. In this survey, the male-to-female ratio was 1.36. They concluded that this ratio indicates that the traditional male occupations are associated with the disease. For both ages, incidence of the disease increased by age. The liver was the most common localization (72%) in this survey.

This idea that the higher rate of male patients is due to their occupation makes sense. However, it should be noted that this tradition varies greatly in different geographical areas. For example, in contrast to the Western country, females still have a prominent role in livestock raising in developing countries; particularly in rural areas.

In Tunisia, Bellil *et al.* (2009) investigated the epidemiological features of extrapulmonary hydatid cysts in 265 patients. The M/F sex ratio was 0.61 and the mean age of 38.7 years. In this series, the hydatid cysts involved the kidney (24.1%), the central nervous system (22.6%), the liver (19.6%) and the spleen (11.3%). Present results are in line with this report regarding the patients' gender and age, as well as the sites of involvement.

In another study in Egypt, Ibrahim *et al.* (2007) evaluated 41 human cases of proved hydatidosis granulosis, including 22 females (53.7%) and 19 males (46.3%). The liver was the most commonly involved organ, followed by the lung, spleen, brain, eye, pelvic and bones of upper and lower limbs. We also reported similar results in present study.

Salem *et al.* (2011) studied 63 human hydatid cysts in Mauritania. In this study, the lung was the most common site of involvement (52%), followed by the liver (33%), spleen (4%), brain (3%), heart (2%), breasts (2%), kidney (2%), diaphragm (1%) and peritoneum (1%).

We also showed that the lung was the most frequently involved organ in these patients. In a 13-year (1990-2003) retrospective study by Ernest *et al.* (2010) in Tanzania, a total of 171 hydatidosis patients were evaluated. The disease affected more females (59.1%) than males (40.9%) and a higher number of infections were recorded in individuals <30 years of age (22.8-30.4%) than in those >30 years of age (2.3-9.4%). Liver was the most commonly affected organ (81.5%). Female gender and younger age were concluded to be the main risk factors of hydatidosis in this study.

Except for the most commonly infected site, other results of the mentioned study are almost in line with ours. It should be noted that we stratified the sites of involvement by the age and gender of the patients (Table 1, 2).

Table 2: Locations of the hydatid cyst in the studied patients stratified by their age groups

Location	Age group (years)								
	<1	1-10	10-20	20-30	30-40	40-50	50-60	60-70	>70
Lung	6 (3.9)	11 (7.2)	31 (20.3)	39 (25.5)	29 (19.0)	12 (7.8)	16 (10.5)	4 (2.6)	5 (3.3)
Liver	(6.5)	1 (1.1)	9 (9.8)	25 (27.2)	20 (21.7)	13 (14.1)	10 (10.9)	7 (7.6)	1 (1.1)
Gallbladder	1 (9.1)	1 (9.1)	0 (0.0)	2 (18.2)	3 (27.3)	3 (27.3)	1 (9.1)	0 (0.0)	0 (0.0)
Spleen	0 (0.0)	0 (0.0)	0 (0.0)	1 (14.3)	2 (28.6)	1 (14.3)	1 (14.3)	1 (14.3)	1 (14.3)
Kidney	1 (14.3)	0 (0.0)	0 (0.0)	0 (0.0)	3 (42.9)	2 (28.6)	1 (14.3)	0 (0.0)	0 (0.0)
Abdomen	1 (20)	0 (0.0)	1 (20.0)	0 (0.0)	1 (20.0)	0 (0.0)	2 (40.0)	0 (0.0)	0 (0.0)
Intestine	1 (20)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0(0.0)	3 (60.0)	1 (20.0)	0 (0.0)
Pleura	0 (0.0)	0 (0.0)	0 (0.0)	2 (40.0)	1 (20.0)	1 (20.0)	0 (0.0)	1 (20.0)	0 (0.0)
Bronchus	1 (20)	0 (0.0)	1 (20.0)	1 (20.0)	0 (0.0)	2 (40.0)	0 (0.0)	0 (0.0)	0 (0.0)
Mediastinum	0 (0.0)	0 (0.0)	0 (0.0)	1 (20.0)	3 (60.0)	1 (20.0)	0 (0.0)	0 (0.0)	0 (0.0)
Chest wall	0 (0.0)	0 (0.0)	1 (25.0)	1 (25.0)	1 (25.0)	1 (25.0)	0 (0.0)	0 (0.0)	0 (0.0)
Heart	0 (0.0)	0 (0.0)	0 (0.0)	1 (25.0)	0 (0.0)	2 (50.0)	1 (25.0)	0 (0.0)	0 (0.0)
Brain	0 (0.0)	0 (0.0)	2 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Pericardium	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (50.0)	0 (0.0)	1 (50.0)	0 (0.0)	0 (0.0)
Facial sinus	1 (50)	0 (0.0)	0 (0.0)	0 (0.0)	1 (50.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Pancreas	0 (0.0)	0 (0.0)	0 (0.0)	1 (50.0)	1 (50.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Esophagus	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (50.0)	1 (50.0)
Ovary	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Uterus	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Axillary region	1 (100)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Pelvis	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)
Femur	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)	1 (100.0)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0.0)

Data presented as frequency and values in parenthesis represents percentages

As mentioned earlier and according to the reported studies, the heterogeneity of results is clear in this regard. It is previously shown that different genotypes of *Echinococcus granulosus* isolates from various animal sources may contribute to clinically or histologically heterogeneous diseases. For example, Ahmadi and Dalimi (2006) showed that the parasites isolated from sheep and human pertained to the same genotype while camel isolates were different. This justification, along with the ethnical differences and socioeconomic variations may contribute to this great inhomogeneity of available data in the literature.

CONCLUSION

Patients with hydatid cysts in East Azerbaijan Province are mainly females in the age group of 20-40 years. The lung and liver are the major sites of infection in these patients.

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