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Epidemiology and clinical characteristics in patients with hydatid cyst undergoing surgery at Besat Hospital of Hamadan, Iran

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Original Article

Abstract

BACKGROUND: Hydatid cyst is one of the most dangerous zoonoses caused by the larval stage of *Echinococcus granulosus*. Our aim was to determine the epidemiology of hydatidosis and the clinical characteristics of patients with hydatid cyst who had surgery at Besat Hospital of Hamadan, Iran, from 2006 to 2016.

METHODS: This cross-sectional study was conducted on all patients diagnosed with hydatid cyst who then had undergone a surgery at Besat Hospital of Hamadan, Iran, from 2006 to 2016. Demographic characteristics of the patients, the affected organs, their clinical features, drug history, and operation history were recorded. The collected data were analyzed in SPSS software.

RESULTS: The mean age in this study was 38.63 years. Among the participants, 138 (53.9%) were women and 118 (46.1%) were men. Among all patients, 100 (39.1%) were urban and 156 (60.9%) were rural residents. The rural population was significantly more affected by hydatid cyst than the urban population ($P < 0.001$). Female housewives (44.1%) were the most affected by this disease ($P < 0.001$). The liver was the most commonly involved organ (52.3%), followed by the lungs (31.2%).

CONCLUSION: Hydatid cyst has a global spread, causing massive human involvement and, if not treated properly, will result in death. Echinococcosis is a recurrent disease and a serious public health challenge.

KEYWORDS: Echinococcosis; Epidemiology; Surgery

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Introduction

Hydatid disease is caused by echinococcosis, which is a cystoid tapeworm parasite. Its granulosis and multiclacrisis types, the former causing hydatid cyst and the latter causing alveolar echinococcosis, are more common among humans. Hydatid cyst is one of the most dangerous zoonoses, which is caused by the larvae of Echinococcus granulosis. This parasite is a Cestoda and from the Echinococcus species which is considered to be the second most important parasitic worm in the world. This parasite has a global spread, but it is most commonly seen in Mediterranean beaches, southern Russia, Iran, Australia, and Bulgaria. The parasite has a global spread and the highest incidence of disease is in areas where livestock is prevalent.[1] Human infection to hydatid cysts is more common in areas where humans and dogs are in close contact. The definitive hosts of the parasite are carnivores, especially dogs and wolves. These

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animals are contaminated by eating mature Echinococcus and cysts that exist in the organs of intermediate hosts, such as sheep, deer, and goose. In the intestinal tract of the definitive host, the cyst turns into a parasite with 400-800 eggs. These eggs are excreted in the stool. Intermediate hosts, such as humans and sheep, are infected by eating vegetables, water, and grass infected with parasite eggs. In the intestine of intermediate hosts, like humans, the eggs are opened and the embryo is attached to the duodenum and jejunum, and then, enters the mesenteric veins and reaches the liver through the portal vein. Embryos may reach the lungs through the hepatic veins and lower inferior veins into the right atrium, and then, right ventricle. The parasite may enter the lymphatic system of the small intestine and through the thoracic duct into the right ventricle of the heart and enter the lungs without passing through the liver.[2] Humans become infected by accidentally eating water and vegetables contaminated with Echinococcus eggs. As the definitive hosts are dogs and other carnivores, humans and livestock are considered to be the intermediate hosts for this parasite. Infection with hydatid cyst is more commonly found in those who have close contact with dogs, such as ranchers, wool spinners, veterinarians, and those who live with livestock and in areas where dogs are kept,[3] The disease is clinically important, and parasitic cysts are often replaced in the liver and the lungs, which are vital organs, and can spread to other organs such as the brain, eyes, kidneys, spleen, heart, and reproductive organs, and even to the bones.[4] Therefore, surgery is required. 70% of lesions are in the liver, 25% in the lungs, and about 2% in the peritoneum. The brain can also be involved, which can be very dangerous, and the patient may suffer from seizures, symptoms of increased pressure (ICP), and other neurological symptoms. The hydatid cyst should be considered as one of the causes of stroke in young patients. The incubation period may last 5 to 20 years and varies according to the location, type, and severity of lesions.[5]

In Iran, the Middle East, and the areas where animal husbandry thrives, the disease is more prevalent.[6] A majority of third-world residents live in rural areas, and the majority of this population is exposed due to their involvement in farming and animal husbandry, and their long exposure to soil and contaminated materials. Hydatid cyst is still a health problem in endemic areas such as the Middle East and Mediterranean countries. Although echinococcosis is endemic in eastern Mediterranean countries, there is no coherent information and statistics on the disease in the region. The highest prevalence of human infection has been reported in the east and south of Europe, the Mediterranean coast, the Middle East, Latin America and Africa, and most often in rural population of these areas. It can be claimed that the disease has been eradicated through parasite control methods in Iceland, New Zealand, Tasmania, and southern Cyprus.[7]

Hydatid cyst is not a common disease in humans, but because of its dangerous nature and the difficulty of treatment, it is a health problem in many countries. Despite the advancements which human beings have made in every field of science, and have led to their superiority over nature, parasitic diseases continue to be the major health and economic problems of all human societies, and there is no area in the world in which these diseases cannot be found. The prevalence of parasitic diseases is heavily dependent on the economic, social, cultural, geographical, and climatic conditions of various regions. These diseases are more prevalent in poor and Third World countries, and although control and prevention plans have been effective in many areas, parasitic diseases have increased in some other parts of the world.[8] Drugs like albendazole and mebendazole cannot treat the

disease completely and surgery is usually required. Timely diagnosis can lead to significant improvements in the control and treatment of the disease. In most cases, the early stages are asymptomatic. Cystic echinococcosis can be asymptomatic in 65% of cases, and is often found after an examination for other diseases, so there is a need for easy and inexpensive methods to identify patients in endemic areas. The World Health Organization (WHO) recommends prophylaxis in areas with high prevalence.[9] Hydatid cyst or hydatidosis is one of the most common parasitic diseases among humans and animals which can be transmitted from canines to humans.[8] The importance of hydatid cyst is the involvement of vital organs of the body, especially the liver and lungs, and the significant loss of livestock. Despite the use of new therapies, surgical treatment is a selective treatment of the disease, which has an annually heavy economic and health burden for countries.

Considering the importance of hydatid cyst disease, the imposition of significant economic losses in livestock, and that there has not been a study on human hydatidosis in Hamedan, Iran, this study was conducted with the aim to determine the epidemiologic status of patients with surgical hydatid cyst in Besat Hospital of Hamedan over the past 10 years.

Methods

This study was a cross-sectional study. The study population consisted of all patients with hydatid cyst, hospitalized and operated in Besat hospital of Hamedan during the years 2006-2016. Sample size was all cases registered in the hospital medical records within a period of 10 years. The study was performed by referring to the medical records of the patients, the data were collected and recorded in a checklist by the researcher. The inclusion criteria was having all medical records of patients with hydatid cyst who underwent surgery during the years 2006-2016, and the exclusion criteria was incomplete file information.

The data included demographic characteristics, gender, age, occupation, residency (and issues related to hydatidiosis), the involved organ, clinical symptoms, drug history, history of previous surgeries, etc. The ethical standards of the study were considered prior to the study and were observed during the research. Data were analyzed in SPSS software (version 16; SPSS Inc., Chicago, IL, USA). Mean and standard deviation were used to describe the quantitative data and table, charts, and frequency ratios were used to describe qualitative data. In the analytical section, independent t-test and Mann-Whitney (based on the parametric results) were used for comparison of quantitative variables in two groups, and ANOVA and Kruskal-Wallis (based on the parametric results) were used for more than two groups. To compare the qualitative variables, chi-square test was used.

This study was proved by the Vice-chancellor for Research and Technology, Hamadan University of Medical Sciences (No. 9610126556). All participants received a questionnaire and written informed consent form in accordance with ethical committee requirements.

Results

During the period of 2006-2016, 256 patients with hydatid cyst underwent surgery in Besat Hospital of Hamadan. The frequency distribution of demographic characteristics is presented in table 1. The mean age of the cases was 38.63 years. The mean age in the female and male groups was 40.15 years and 36.85 years, respectively. Regarding the normal distribution of age, independent t-test was used to compare of the infection in these two groups. The age of the infection was not significantly different between men and women (P = 0.320). The minimum age for patients was 4 years and the maximum was 84 years. Nearly half of cases (48.5%) were

below 35 years of age. In chi-square test, women were more likely to be referred to undergo surgery than men, but this difference was not statistically significant (P = 0.210). Of the 256 cases, 100 (39.1%) were urban and 156 (60.9%) were rural residents. Those from rural areas were significantly more affected by hydatid cyst than the urban population (P < 0.001). The housewives encompass the highest rate of infection and were significantly more affected than others (P < 0.001) (Table 1).

Table 1. Frequency distribution of demographic information in patients

Variable	n (%)	P
Gender		0.210*
Female	138 (53.9)	
Male	118 (46.1)	
Residency		< 0.001**
Urban areas	100 (39.1)	
Rural areas	156 (60.9)	
Age		
0-15	33 (12.9)	
16-25	46 (18.0)	
26-35	45 (17.6)	
36-45	32 (12.5)	
46-55	41 (16.0)	
56-65	28 (10.9)	
66-75	26 (10.3)	
76-85	5 (2.0)	
Occupation		< 0.001***
Housekeeper	113 (44.1)	
Self-employment	48 (18.8)	
Student	36 (14.1)	
Child	8 (3.1)	
Farmer	13 (5.1)	
Employee	2 (0.8)	
Soldier	2 (0.8)	
Rancher	34 (13.3)	
History of anti-parasitic drugs use		
Yes	53 (20.7)	
No	203 (79.3)	
History of previous surgery		
Yes (secondary)	29 (11.3)	
No (primary)	277 (88.7)	

*k2 test, **nonparametric k2 test, ***nonparametric k2 test

Of the 256 cases, 226 (85.9%) had one affected organ and 36 (14.1%) had more than one affected organ. In more than half of the cases, the liver was the only affected organ (52%), 80 (31%) had only lung involvement, in 26 cases both liver and lungs were affected, and in 16 cases other organs were involved (Figure 1).

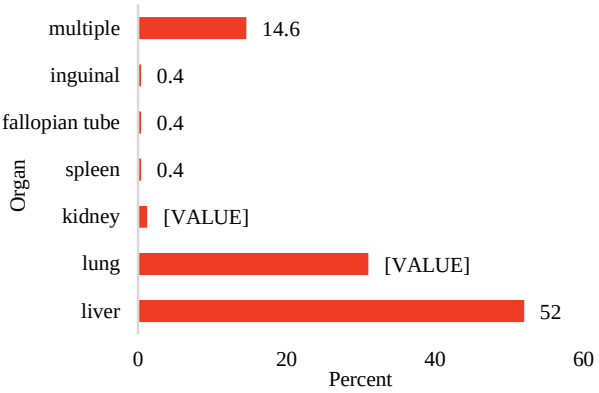

Figure 1. Frequency distribution of the cases in terms of the number of affected organs

The most common sign and symptoms were gastrointestinal symptoms, and among those, abdominal pain was the most frequent (176 cases including 110 cases of RUQ pain, 40 cases of epigastric pain, and 26 cases of abdominal pain). Respiratory complaints, nonspecific symptoms such as fever and chills (n = 70), nausea (n = 51) and vomiting (n = 49), early satiety (n = 46), weight loss (n = 15), and skin, urologic, and neurological symptoms were other signs and symptoms (Table 2).

Considering that the age variable had a normal distribution in the KS test, ANOVA was used to determine the relationship between age and the affected organs. According to this test, and with regard to P = 0.001, there was a significant relationship between age and the affected organs.

In young patients, lung involvement was more common than the elderly, as well as the involvement of other organs compared to the liver and lung involvement at older ages. Chi-square test was used to determine the relationship between gender and the affected organs. Based on the results of this test and

P = 0.040, there was a significant relationship between gender and the affected organs. In men, lung involvement was significantly more common than women; however, liver involvement was significantly more common in women (Table 3).

Table 2. Frequency distribution of cases, distinguished by signs and symptoms (nonparametric k2 test)

Signs and symptoms	Frequency	Percentage
RUQ pain	110	43
Epigastric pain	40	15.6
Diffuse abdominal pain	26	10.4
Dyspnea	74	29
Cough	67	26.2
Fever	41	16
Chill	29	11.3
Sweating	7	2.8
Chest pain	31	12.1
Vomiting	49	19.2
Nausea	51	20
Early satiety	46	18
Weakness and fatigue	3	1.2
Hematemesis	2	0.8
Hemoptysis	15	6
Weight loss	15	6
Abdominal distention	3	1.2
RUQ distention	10	4
Inguinal pain	1	0.4
Icterus	5	2
Orthopnea	3	1.2
Hematuria	2	0.8

To determine the relationship between the affected organs and occupation, due to the non-establishment of chi-square conditions, non-parametric chi-square test was used. Based on the results of this test and P = 0.038 there was a significant relationship between occupation and the affected organs. There is no meaningful relationship between the infected organs and the place of residency according to the result of the non-parametric chi-square test (P = 0.170).

Echinococcus infection is a recurring disease and a serious public health challenge. In the present study, people with hydatid cyst who underwent surgery in recent years have had an upward trend. However, the cause may be related to the improvement of the methods of detection, not controlling of the disease (Figure 2).

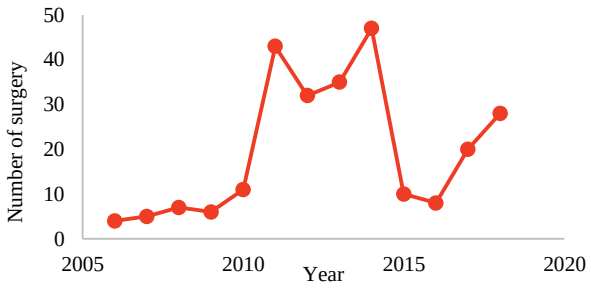

Figure 2. Number of surgeries performed per year

Table 3. The relation between gender and the affected organs

Affected organ	Female	Male	Total	P
liver	84 (60.9)	50 (42.4)	134	
Lungs	33 (23.9)	47 (39.8)	80	
Liver and lungs	13 (9.4)	13 (11)	26	
Kidney	2 (1.4)	1 (0.8)	3	
Kidney and liver	2 (1.4)	0 (0)	2	
Kidney and lungs	0 (0)	1 (0.8)	1	
Spleen	0 (0)	1 (0.8)	1	
Pericardium, liver and lungs	0 (0)	2 (1.7)	2	0.040
Brain, Liver and lungs	1 (0.7)	0 (0)	1	
Peritoneum and liver	1 (0.7)	2 (1.7)	3	
Peritoneum & urinary bladder	0 (0)	1 (0.8)	1	
Fallopian tubes	1 (0.7)	0 (0)	1	
Inguinal	1 (0.7)	0 (0)	1	
Total	138 (100)	118 (100)	256	

Discussion

Considering the importance of hydatid cyst disease, the imposition of significant economic losses in livestock, and that there has not been a study on human hydatidosis in Hamedan, this study was conducted with the aim to determine the epidemiologic status of patients with surgical hydatid cyst in Besat Hospital of Hamedan, Iran, Over the past 10 years. In much of Iran, cystic echinococcosis is a major problem in both the human and livestock, causing very significant economic losses. Dogs play an important role in transmitting *E. granulosus* in Iran, as in many other endemic areas.[9]

In our study, to determine the epidemiology of hydatidosis and the clinical characteristics of patients with surgical hydatid cyst in Besat Hospital of Hamedan during 2006-2016, it was shown that women (53.9%) are more likely to be infected by this disease than men (46.1%), which can be attributed to genetic differences, women's occupations, washing and eating raw vegetables, direct contact with pets and house cleaning. However, a study indicated that Echinococcus granulosis affects men and women equally. The most common occupation among cases was homemakers (44.1%), self-employed men (18.8%), students (14.1%), and ranchers (13.3%). Other studies in other parts of Iran,[10,11] and studies in other countries such as Yaghan et al. In Jordan,[12] Molan in Iraq,[13] also demonstrated that women are affected more than men. One of the reasons for this difference is that housewives are more likely to be in contact with sources of contamination, especially vegetables that are infected with parasite eggs. Another reason is that women are also involved in agricultural activities and animal husbandry. Another reason for this is career diversity among men, while women are mostly housewives, which makes this higher incidence to be more concentrated in a career group. The age pattern in this study was similar to that in other studies, including that by Kamali et al.[14] However, it did not match the age pattern in other studies such as those by Davoodabadi et al.,[15] and Noorian.[16] In total, about half of the cases (48.5%) in our study were under 35 years of age, which was a significant statistical finding.

Most cases were from rural areas, this finding was in agreement with that of the study by Moosazadeh et al.,[17] and Fallah et al.,[18] while in the study by Kamali et al., the urbanized population were more likely to have hydatid cyst, which may be because most of the population studied was urbanized.[15]

Moreover, 85.9% of patients had only one organ involvement, and 14.1% of patients had more than one affected organ. This result was in agreement with the findings of Islami et al.,[19] while in the study by Khazaei et al.[20] the involvement of multiple organs was significantly lower than that in our study (8.4%).

Consequentially, 52.2% of the patients had hepatic hydatid cysts and the most commonly involved organs were the liver and lungs (31.2%). These statistics are similar to the results of the study by Davoodabadi et al.,[15] and Noorian,[16] which reported that the most involved organs are liver (46.3%) and lung (28.1%), and the rate of liver involvement in women was higher than men.

Yad Yad et al.[21] reported that liver infection was 36.32% and total lung infection was 34.8%, although in our study liver involvement was significantly higher than lung involvement (52.3% vs. 31.2%). The most commonly reported symptoms were abdominal pain, nausea, vomiting, loss of appetite, and in those with pulmonary hydatid cyst, the most common symptoms were cough, dyspnea, fever, blood clots, and chest pain, which was in agreement with our study findings.

In the study by Khan et al.,[22] The rate of contamination of the liver was higher than that of other organs, although the rate of lung involvement was significantly higher in our study (31.2% vs. 18.6). The organs with the

subsequent highest levels of infection included the kidney (3.5%), spleen (3.5%), and uterus (2%). In the present study, after the liver and lung involvement, co-infection of the liver and lungs (10.2%), kidney involvement (1.2%), co-infection of the peritoneum and liver (1.2%) and co-infection of the liver and kidney (0.8%) were the most commonly affected organs. This finding was not in agreement with the result of the abovementioned study.

In the study by Parquahi et al.,[20] the main clinical complaints of patients included abdominal pain, nausea and vomiting, hepatomegaly, dyspnea, and cough. These findings were similar to that of the present study. However, in their study, the spleen was the second most affected organ which was not in agreement with our results.

Anti-parasitic drugs, including Albendazole or Mandendazole, were used for nearly three-fifths of patients. Appropriate treatment before and after surgery may reduce the risk of disease recurrence. In addition, in patients with several cysts, combination therapy with Albendazole and Pericamartol can be an alternative to surgical treatment.[23] In the present study, 20.7% of patients already had a history of anti-parasitic drug use, ultimately required surgery. In this study, 88.7% of the patients were infected with the disease for the first time (primary) and 11.3% of the rest had a history of this infection (secondary) and the surgical treatment for it.

The differences in results of various studies is related to demographic differences lifestyle, cultural, and social differences. It should also be noted that our study was conducted on patients in one hospital and patients with hydatid cyst who underwent surgery. This selection bias may restrict the generalization of the results to all patients with hydatid cyst. However, the present study has some limitations. First, the study cases were selected from a single center rather than multiple centers. Moreover, the study sample size was relatively small. Therefore, further studies, including large datasheets and multiple centers are needed to investigate the clinical significance of these parameters in hydatidosis patients.

Conclusion

Hydatid cyst has a global spread, causing massive human involvement and, if not treated properly, will result in death. Echinococcosis is a recurrent disease and a serious public health challenge. According to the WHO, Iran is one of the countries with moderate prevalence. Hydatidosis should be considered as a problem in Iran because it is endemic.

Conflict of Interests

Authors have no conflict of interests.

Acknowledgments

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