

Research Paper

A 10-Year Epidemiological Study of Human Cystic Echinococcosis in Qazvin Province, Iran



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ABSTRACT

Background: Cystic echinococcosis (CE) is a parasitic infection with significant health problems and economic burden in most of developing countries. The current study aims to investigate the 10-year prevalence of human CE and the demographic and clinical characteristics of operated patients in Qazvin province, Iran.

Methods: In this cross-sectional study, the medical records of CE patients who had undergone surgery in two hospitals of Qazvin Province from 2009 to 2019 were studied.

Findings: There was 203 CE cases in ten years, corresponding to a surgical incidence rate of 1.49 in 100,000 people. Of these, 99(48.8%) were male, and 104 (51.2%) were female. Patients' age ranged from 8 to 86 years, with a mean age of 43±19.16 years. A significantly higher CE incidence was reported in patients aged 20-40 years. The majority of patients were housekeeper (48.8%), and the CE was diagnosed in almost all patients by imaging techniques. Involved organs were liver (n=119, 58.6%) and lungs (n=64, 31.5%), and their concurrent involvement was 4.5% (n=9). The highest CE incidence was recorded in 2015 (n=35, 17.2%). Affected organ and number of cysts had significant association with the CE recurrence in operated patients.

Conclusion: The CE has had a uneven incidence trend in Qazvin Province during 2009-2019. A further epidemiological study is recommended to evaluate more risk factors of CE for its control in this area.

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1. Introduction

Cystic echinococcosis (CE), resulting from infection with canid cestode *Echinococcus granulosus* (*E. granulosus*), is an important zoonotic disease. It is mainly characterized by the formation of unilocular cysts, most frequently in the liver or lungs [1]. On average, only about 10% of symptomatic CE cases have cysts in other organs like the heart, spleen, kidneys, bones, and brain [2]. The CE is a major public health concern with different geographic distribution worldwide. It is less prevalent in northern and central European countries, while is considerably endemic in the Mediterranean region, Middle East, Central Asia, southern and eastern Europe, and South Africa, and is highly endemic in South America and western China [3]. According to the World Health Organization report, more than 188,000 people are affected by CE each year and is responsible for a loss of 183,500 disability-adjusted life years [4]. The real global burden of CE is about 800 million dollars, of which about 760 million dollars is related to human burden and about 140 million dollars is related to animal burden [5]. To complete its life cycle, the *E. granulosus* requires two mammalian intermediate and definitive hosts. Its transmission cycle is mostly domestic, involving dogs and domestic ungulates (mainly sheep) which put human health at potential risk due to close physical contact between humans and dogs [6]. The socio-economic consequences of CE include medical costs (diagnostic and treatment costs) and non-medical costs such as monetary loss due to livestock CE and reduced productivity of asymptomatic individuals [7]. Despite considerable social and economic impacts on the affected population, CE remains one of the 17 neglected tropical diseases across the world [2].

The real burden of CE is likely to be underreported for three reasons; a large number of cases are asymptomatic for years before clinical signs become apparent. Besides, its diagnosis mostly relies on imaging methods that may not be available in the endemic areas. Moreover, the treatment of CE can be costly and challenging, frequently requiring surgical intervention [8]. CE is also a national public health concern in Iran. Due to its significant health and economic burden, there is a continuous need to perform the periodic investigation in medical centers to complete the epidemiologic profile of CE which can provide comprehensive information for public health experts and policymakers of the health system. Therefore, the present study aims to determine the epidemiologic characteristics of the CE with respect to the demographic characteristics of the patients.

2. Materials and Methods

In this cross-sectional study, we retrieved clinical records of patients with CE underwent surgery in hospitals of Qazvin Province, Iran from March 2009 to 2019. We included all operated patients with CE as the study samples. Inclusion criteria were residence in Qazvin Province and having operation due to CE. Exclusion criterion was incomplete clinical records. We collected data from the patients' medical records available in the hospital archives. It consists of clinical characteristics such as symptoms, surgical approach, affected organs (Liver, lungs, spleen, kidney & bones), number of cysts, diagnostic tests (abdominal sonography & Computerized Tomography (CT) scan, plain abdominal x-ray, and sonography), patients' complaints (abdominal pain, dyspnea, nausea & vomiting, fever, and weight loss & anorexia), and disease recurrence. We also collected the patients' demographic information such as gender, age, marital status, occupation, underlying diseases, and place of residence (urban or rural areas). We calculated the temporal distribution of CE in Qazvin Province between 2009 and 2019, and plotted the spatial distribution of CE among the cities of Qazvin Province (Qazvin, Buin Zahra, Alborz, Abyek, Takestan, Avaj) separately.

We used the statistics of Mean \pm SD, frequency, and percentage for describing quantitative and qualitative variables. The bivariate and multivariate logistic regression were used to determine the association between variables. The recurrence of CE was considered as the outcome variable in the regression models. Independent variables were age, gender, marital status, occupation, affected organ, number of cysts, and underlying diseases. We used Excel software version 2013 to calculate the annual trend in the incidence of CE. The distribution maps were prepared in ArcGIS v. 10.5 software and data were analyzed in STATA software v. 14.

3. Results

In total, 203 CE cases were admitted to hospitals in Qazvin Province during 2009–2019. Of these, 104 (51.2%) and 99 (48.8%) were female and male, respectively. The Mean \pm SD age of patients was 43 \pm 19.16 years, ranged from 8 to 86 years. The most CE infections occurred in those aged 20–40 years. The most of patients were housekeeper (48.8%) and married (76%); 105 (51.7%) patients were residing in rural areas and 98 (48.3%) in urban areas. The prevalence of underlying diseases in the patients was 15.8%.

Table 1. Clinical characteristics of the CE patients in Qazvin province from 2009 to 2019

Variables		No. (%)
Diagnostic test	Abdominal sonography & CT	86(42.4)
	Plain abdominal x-rays	59(29.1)
	Sonography	42(20.7)
Complaints	Abdominal pain	130 (64)
	Dyspnea	63(31)
	Nausea & vomiting	48(23.6)
	Fever	42(22)
	Weight loss & anorexia	33(16.3)
Affected organs	Liver	119(58.6)
	Lungs	64(31.5)
	Spleen	4(2)
	Kidney & bones	2(1)
Number of cysts	1	125(61.6)
	2	38(18.7)
	3	10(4.9)
Organ involved	Lung	64(31.5)
	Liver	119(58.6)
	Lung and liver	6(3)
	Other	14(10)
Recurrence of the disease	Yes	21(10)
	No	181(89.2)

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The patient's clinical characteristics, such as their complaints and anatomical sites of cyst distribution, diagnostic tests, number of cysts, affected organs, and recurrence of the disease are shown in Table 1. The mostly used diagnostic test was abdominal sonography & CT scan and the most prevalent complaint was abdominal pain. The liver (n=119, 58.6%) and lungs (n=64, 31.5%) were the most affected organs; in 4.5% of cases, both liver and lungs had been affected. More than 23% had more than one cyst in one or two organs, simultaneously. The recurrence of disease after the primary operation during follow-up was reported in 21 patients (Table 1).

There were fluctuations in the incidence of CE during the study years; it was increased during 2009-2015 (from 1.75 in 100,000 people to 2.78 in 100,000 people). In 2016, it was dropped to 0.32 in 100,000 people. From 2016 to 2019, there was a relatively upward trend from 0.32 in 100,000 people to 0.89 in 100,000 people. In 2015, there was the highest incidence rate of CE (17.2%). The lowest incidence rate (n=32, 15.8%) was reported in 2018 (n=3, 1.5%) (Figure 1). The mean 10-year incidence of the disease in each cities of Qazvin Province showed that Buin Zahra (2.1 in 100,000 people) and Alborz (0.95 in 100,000 people) ranked in first and last places, respectively, while the provincial incidence was 1.49 in 100,000 people (Figure 2).

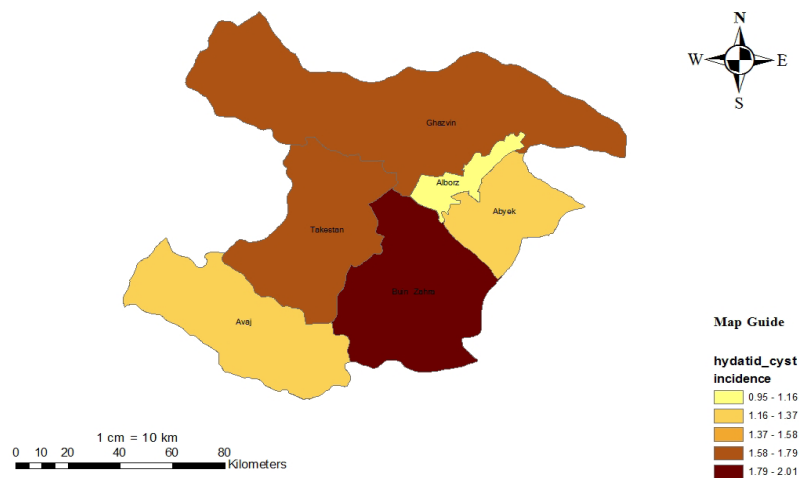


Figure 1. Annual trend in the CE incidence in Qazvin province from 2009 to 2019 (per 100,000 people)

We also tested the association between independent variables (age, gender, occupation, place of residence, underlying disease, affected organs, and number of cysts) and disease recurrence. The result showed that odds of disease recurrence had significant association with the affected organ (OR=0.67, 95% CI: 0.50-0.88) and number of cysts (OR=0.98, 95% CI: 0.97-0.99) (Table 2).

4. Discussion

This study evaluated the epidemiology and clinical features of CE among operated patients in Qazvin Province. To our knowledge, this is the first epidemiological

study of CE that has been conducted in Qazvin Province based on medical records. The data showed that CE infection was more prevalent among female patients aged >30 years, and housekeepers. The liver and lungs were the most affected organs. Moreover, imaging techniques were the main methods for the diagnosis of CE. Organ involvement and number of cysts had significant association with disease recurrence in CE patients.

This study indicates that the incidence of CE is about 1.49 in 100000 people in Qazvin Province that varied based on the residential status of patients. The incidence

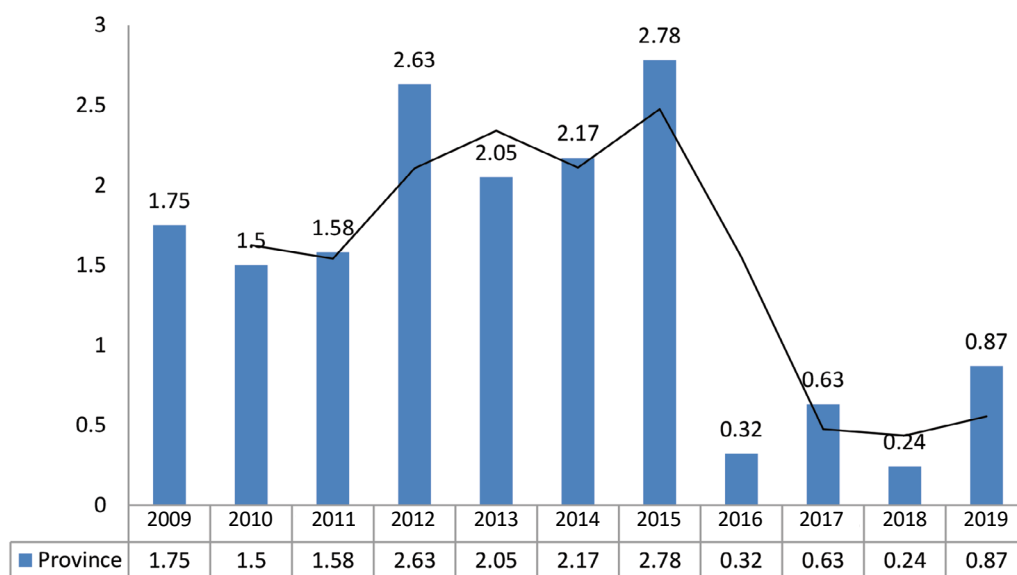


Figure 2. Distribution map of the CE incidence in different cities of Qazvin province during 2009-2019

Table 2. Logistic regression analysis results to determine the factors predicting CE recurrence

Variables	Crude OR	95% CI		P	Adjusted OR	95% CI		P
		Lower bound	Upper bound			Lower bound	Upper bound	
Age	1.00	0.97	1.02	0.92	1.01	0.96	1.01	0.34
Gender	0.67	0.27	1.69	0.40	0.71	0.24	2.15	0.55
Marital status	0.89	0.58	1.37	0.61	0.74	0.46	1.18	0.21
Occupation	0.95	0.80	1.14	0.64	1.01	0.81	1.25	0.94
Rural/urban areas	1.23	0.50	3.00	0.64	1.45	0.54	3.87	0.45
Affected organ	0.69	0.53	0.89	0.00	0.67	0.50	0.88	0.00
Number of cysts	0.98	0.97	1.00	0.04	0.98	0.97	0.99	0.03
History of underlying diseases	0.91	0.64	1.30	0.61	0.86	0.60	1.23	0.41

OR: Odds Ratio.

was higher in Buinzahra County and lower in Alborz County. The higher incidence of CE in Buin Zahra may be related to this fact that the disease is more prevalent in areas with sheep farms. Buin Zahra is the center of animal husbandry in Qazvin Province, and Shahsevan, as one of the most important pastoral nomads, resides in this area; their lifestyle could facilitate the CE infection in this region. The study results showed that the most of the patients were living in rural areas. The similar condition has been observed in other hospital-based studies in other provinces of Iran such as Hamadan and West and East Azerbaijan Provinces [9-11]. We predict that the number of CE cases in the rural areas is much higher, since many patients living in rural areas do not refer to the hospitals because of using a watch-and-wait strategy or prefer chemotherapy to the surgery when clinical symptoms are mild.

The annual trend of the CE incidence during 2009–2019 was evaluated in this study. There were fluctuations in the first third of this period. Then, it showed a rising trend in the middle third of the study period, resulting in an increase by above 2 in 100,000 people. Finally, there was a declining trend in the final third of the period. Almost more than half of the patients were female (female/male ratio: 51/49), which is not consistent with the result of other studies conducted in Ahwaz, Tehran, and Tabriz where the female/male ratio were 40/60, 42/58, and 48/52, respectively [12-14]. The subjects in these three studies were children, which may be the reason for this discrepancy. In other study, consistent with the present study, the infection rate was also higher in females [15]. Housewives have

been the most affected group in most studies conducted in Iran, especially in rural areas. The main reasons include more exposure to the infection sources like contaminated vegetables, geophagy, and more contact with dogs and animal breast pumping.

The age range of patients in our study (8-86 years) is consistent with that in previous studies in Iran. Studies by Rezaei et al. in Qom Province [16], Khalili et al. in Chaharmahal & Bakhtiari Province [17], Salehi et al. in North-Khorasan Province [18] also reported the same age range in patients. Moreover, the total number of patients (n=42, 20.7%) were at the age ≥ 80 years. The reason for this high number of patients in this age group might be the fact that CE is a disease with long incubation period and its clinical signs appear over time and with the increase of age.

In this study, as expected, hepatic cysts were more frequent than pulmonary cysts, with a ratio of 1.8:1. Almost all the studies carried out in Iran except one by Aslanabadi et al. in 2013 [19], have indicated that lungs are involved more, yielding similar results to ours. They have admitted that their result was unexpected. The prevalence of CE in uncommon anatomic sites (except in the liver and lungs) was 5.5% of all cysts, with spleen being the most frequent site of infection (2% of all cysts). The studies have reported higher prevalence ranging from 7% to 13% [20].

While this study provided valuable information, it had some limitations. One of the limitations was that in hospital discharge records, there was no information about

the history of contact with dogs or other definitive hosts (wolves or other canids). Since dogs play a key role in transmission of parasites, as one of the sources of infection, more studies are needed to evaluate its effect. Another limitation was the lack of information about the educational level of CE patients in patients' medical records. Moreover, there was no information about occupational exposures, cyst detection stages, serological or molecular nature of diagnostic tests.

5. Conclusion

Although there is uneven hospitalization rate due to CE during 2009-2019 in Qazvin Province and annual human CE incidence rates declined in the last four years, it remains a common infectious disease in the region. Therefore, it is necessary that the CE be considered as an urgent health problem by health policy-makers and develop methods for its surveillance. It is also necessary to update provincial data about the CE prevalence by conducting more studies in the future to estimate the disease and cost burden and evaluate the effectiveness of implemented control strategies.

Ethical Considerations

Compliance with ethical guidelines

The study was approved by the ethical committee of Qazvin University of Medical Sciences, Qazvin, Iran (Code: IR.QUMS.REC.1399.029).

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Authors' contributions

Conceptualization and project administration: Zahra Hosseinkhani and Amin Yazdani; Writing and editing: Zahra Hosseinkhani, Amin Yazdani, Bahas Sattarian; Methodology and data analysis: Zahra Hosseinkhani and Hadi Bagheri.

Conflict of interest

The authors declare no conflicts of interest

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