

# Clinical Features of Hydatid Disease in Guilan (the North Province of Iran): A Ten-Year Study

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**Background:** Hydatid cyst disease (HCD) or echinococcosis has a worldwide distribution and is endemic in many countries of Mediterranean region. In Iran, due to various climatic conditions, the rate of the disease is diverse in different parts. The most infected regions are the domains of Alborz and Zagros Mountains where there is a high rate of ruminants and the main carrier of people are animal husbandry. Due to the importance of this zoonotic helminth and the lack of information about hydatid cyst disease in Guilan province, we conducted a retrospective study from 2000 to 2010 to evaluate the HCD inpatients in this region.

**Objectives:** The aims of the study are to evaluate HCD inpatients and to review the clinical presentations of different organs affected by cysts and postoperative outcomes of hydatid cyst in Guilan province.

**Materials and Methods:** A retrospective descriptive investigation was performed by reviewing the medical records of patients with HCD, admitted to four private and governmental hospitals, in Rasht, capital of Guilan province from 2000 - 2010.

**Results:** Sixty-two patients (38 males, 24 females) with a mean age of  $46.38 \pm 18.36$  years (aged between 8 and 81 years old), with HCD were evaluated. In the 40 - 60 years old group, there was highest rate of HCD (40.3%). Housewives had the highest rate of infection (40.3%) followed by farmers (22.6%), pupil (8.1%), animal husbandries (6.5%) and others (22.6%). The most prevalent complaint of the patients with liver involvement were abdominal pain (75%) and nausea and vomiting (9.1%) and the most common findings of the inpatients with lung involvement the most common complaint was cough (40%). Seventy one percent of the patients had liver involvement and 16.1% had lung involvement. The most common diagnostic method was ultrasonography (48.14%). Computed tomography was diagnostic in 43.20% of cases and chest X-ray in 8.6% of cases. Overall, 96.8% of patients were cured and the mortality rate was 3.2%.

**Conclusions:** Echinococcosis is still an important health problem in Iran that needs further studies. Therefore, accurate information on the distribution of the disease is the first step to control and prevent it. In general, the situation of the hydatidosis in the human in Guilan shows somehow a resemblance with the other areas in Iran.

**Keywords:** *Echinococcus granulosus*; Echinococcosis; Iran

## 1. Background

Hydatid cyst disease (HCD) or echinococcosis, caused by the metacestode stage (larval form) of the dog tapeworm and it is usually endemic in raising countries, but also in most developed countries in Mediterranean region, such as South America and Africa (1-3). *Echinococcus granulosus* has a sustaining sheep-dog epizootic cycle which is difficult to break off and humans acquire the infection by incidental ingestion of eggs released by infected definitive hosts to the environment (4). It is enzootic in sheep which may serve as the reservoir and amplification vector and generate human cases (2). The most common routes of infection in people are ingestion of contaminated water or uncooked food, or direct hand-to-mouth fecal transmission (5, 6). Once ingested by an intermediate host, the eggs develop into oncospheres in the upper gastro-

intestinal tract. The oncospheres penetrate the intestinal wall, enter the portal vein and reach to the liver. A few oncospheres may pass the liver and become trapped in the lungs, or more rarely, in other organs and tissues (5, 7, 8). It is a unique parasitic disease which can growth and live almost anywhere in the body and demonstrates a spectrum of manifestations that vary according to growth stage, associated complications, and affected tissues (9).

The most common involved organ is liver (59 - 75%) and others include lung (27%), kidney (3%), bone (1 - 4%), and brain (1 - 2%) (10). Despite its distribution, the disease burden is generally underestimated and only recently WHO included echinococcosis as a part of a Neglected Zoonosis subgroup for its 2008 - 2015 control strategic plan (11, 12). Iran is an important endemic region of cystic hydatid disease (13). Given various climate conditions of Iran, the rate of the disease varies in various parts. The most

### Implication for health policy/practice/research/medical education:

Hydatid cyst is considered as an important health problem in Iran and since the agriculture and animal husbandry is common in Guilan, accurate information on the distribution of the disease is the first step towards controlling and preventing it.

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infected districts are the domains of Alborz and Zagros Mountains where there is a high rate of ruminants population and the main carrier of people are animal husbandry (14). Moreover, human hydatidosis is responsible for approximately 1% of admission to surgical wards and the rate of human infection is 0.6 - 1.2 /100000 (15). Hydatid cyst is a prevalent zoonotic helminth (16) and in Guilan province (North of Iran), induces various economic and health damages every year. Early diagnosis of this zoonosis can help to decrease the adverse outcomes.

## 2. Objectives

As symptoms and signs of this cystic infection is similar to the other cystic lesions and the information about this prevalent zoonosis is limited, we conducted a retrospective study from 2000 to 2010 to evaluate the HCD inpatients in Guilan province and review the clinical presentations, different organs affected by cysts, and postoperative outcomes of hydatid cyst.

## 3. Patients and Methods

### 3.1. Study Design

A retrospective descriptive investigation performed by reviewing the medical records of patients with HCD admitted to four private and governmental hospitals (Razi, Poursina, Golsar and Aria) in Rasht city, capital of Guilan province during the years 2000 to 2010.

### 3.2. Checklists

A checklist was designed to collect the necessary data out of the medical records. All patients' files were reviewed for: demographic data, presenting symptoms, anatomical locations of cysts, diagnostic methods, complications and the mortality rate.

### 3.3. Statistical Analysis

SPSS version 16 was used and the data were analyzed by using the descriptive statistic.

## 4. Results

From 2000 to 2010, sixty-two patients (38 males, 24 females) with a mean age of  $46.38 \pm 18.36$  years (aged 8 - 81 years old), with HCD were evaluated. In the 40 - 60 years old group, there was a higher rate of HCD (40.3%) compared with other groups. The frequencies of disease in urban and rural regions were 66.1% and 33.9%, respectively. Housewives showed the highest incidence of infection (40.3%) followed by farmers (22.6%), pupil (8.1%), animal husbandries (6.5%) and others (22.6%), respectively. Totally 93 complaints were reported by the patients, the most common one was abdominal pain which was presented

in 51.6% of cases. The symptoms at the time of presentation are shown in Table 1.

**Table 1.** Symptoms at the Time of Presentations in Patients With Human Hydatid Cysts

| Manifestations      | No. (%)   |
|---------------------|-----------|
| Cough               | 10 (10.8) |
| Sputum              | 8 (8.7)   |
| Dyspnea             | 5 (5.4)   |
| Chest pain          | 3 (3.2)   |
| Right flank pain    | 2 (2.1)   |
| Abdominal pain      | 48 (51.6) |
| Jaundice            | 3 (3.2)   |
| Pruritus            | 1 (1.1)   |
| Nausea and vomiting | 6 (6.4)   |
| Neck mass           | 1 (1.1)   |
| Earache             | 1 (1.1)   |
| Fever               | 3 (3.2)   |
| Weight loss         | 2 (2.1)   |

Inpatients with liver involvements had mostly complained about abdominal pain (75%), nausea and vomiting (9.1%). In patients with lung involvements the most common complaint was cough (40%). The anatomic locations of all cysts showed the liver was the most common infected organ, followed by the lung. Other organs were suprarenal area, kidney, mandible, supraclavicular area, pancreas and gallbladder one cyst in each of these organs were detected (Table 2).

**Table 2.** Organ Involvement of Hydatid Cysts

| Involved organs           | No. (%)   |
|---------------------------|-----------|
| Lung                      | 10 (16.1) |
| Liver                     | 44 (77)   |
| Lung and liver            | 2 (3.2)   |
| Other organs <sup>a</sup> | 6 (9.6)   |
| Total                     | 62 (100)  |

<sup>a</sup> There was one cyst in each: suprarenal area, kidney, mandible, supraclavicular area, pancreas and gallbladder

Among 12 patients with lung involvements, 8 patients (66.7%) showed right side cysts and 4 patients (33.4%) had left side involvements. Of 48 liver cysts identified, 69.6% were located in the right lobe and 21.7% in the left lobe. Three patients had both lobes infection. The infection in organs other than liver and lung, were seen in just 6 patients. In the present study, diagnosis was most commonly based on radiologic methods (75.3%). Ultrasonography (48.14%) was the most common radiologic method fol-

lowed by computed tomography (CT) (43.2%) and chest X-ray (8.6%), respectively. In 18.5% of patients, diagnosis was based on clinical manifestations, 3.7% based on pathology and 2.5% based on laboratory data. Out of the 62 patients, the surgery was conducted for 61 patients and one of them, a 78-year-old man with a history of deep venous thrombosis and Warfarin use was considered. Among 62 patients, 2 (3.2%) died, one of them died due to Spontaneous rupture of the liver Hydatid cyst into the peritoneum and another one due to bleeding and disseminated intravascular coagulation (DIC) after liver surgery. Follow-up was possible in 39 patients after discharge, 33 of them showed complete healing, but 2 patients had recurrent liver cysts, 3 patients had abdominal pain because of incisional hernia and one showed recurrence of lung symptoms.

## 5. Discussion

Hydatid disease remains a continuous public health problem in endemic countries and Iran has been announced as a hyperendemic region by World Health Organization (WHO). Our study presents a review on medical records of HCD patients in four referral hospitals in Guilan province during a 10-year period. In the present study, males proved to be more frequently affected than females. In the study of Rai et al., the male to female ratio of pulmonary Hydatid cyst was 2:1 (17). Ciobanca and Junie showed in their study a slightly higher incidence of hydatid cyst in males comparing to females (18). Furthermore, in the study of Mirshemirani et al. on children, hydatid cysts were more common in boys than girls (19), while Baharsefa et al. (20) and Rostami Nejad et al. (21) showed a higher rate of infection with hydatid cyst in females than males. In this study, the most of patients were in adult and older age groups, mostly were in 40 - 60 years old group. Arbabi et al. reported the highest rate of infection with hydatidosis in age group of 60 - 80 years in Hamadan province of Iran (22), while Hoseini et al. reported the most prevalence in the 20 - 40 years old patients in Kurdistan Province (23).

In the present study housewives showed the highest prevalence of hydatid disease. This result is comparable with other studies in Iran (24, 25) and with study in Taleghani Hospital of Tehran during 1994 - 2003 (26), in East - Azerbaijan province during 1995 - 2002 (27) and in Kurdistan Province during a five-year period (28) study. It seems that the high infection incidence among housewives is probably because of more involvement of rural women with infected soil and animals and more sprawls of infected dog stools in the environment of houses. Hydatid disease is generally considered as a rural disease because of the characteristics of the parasite life cycle, but in our study, more patients were from urban than from rural areas. This finding is in concordance with the study of Ok UZ et al. (29), Dopchiz et al. (30), Talaiezhadeh et al. (30) and (18) which demonstrated a predominance of hy-

datid disease in urban populations.

There may be geographical differences in the distribution of the involved organs that are related to some biological factors in the parasite or host (31). The liver and lungs are the most frequently involved organs of this disease. The incidence of simultaneous liver and lung involvement has been reported to be between 5.8 and 13.3% based on various reports (32-35). According to our study, the liver is the most common infected site of hydatid disease (71%), followed by the lungs (16.1%) and other organs (12.9%), being in accordance with other studies (36, 37). Probably more involvement of liver inpatients is because after entering the gastrointestinal tract, most cysts are entrapped by liver through portal system. Inpatients with lung involvement, the right lung were the more common site. It was comparable with Bagheri's and Aghajanzadeh's study (38, 39). This is to some extent because of better circulation of this site. Specifically, the right hepatic lobe is affected in 80% of cases and the left lobe in 20%. Less common sites are the lungs (15%) (1). In Langer's et al. study inpatients with liver involvement, cysts were found in the right lobe in 74.4%patients, the left lobe in 15.4%patients, and both lobes in 10.2%patients (40) which were similar to our findings.

In this study, the most common symptoms were abdominal pain (51.6%), cough (10.8%), sputum (8.7%) and others (29.9%). Langer et al. reported that the abdominal pain and tenderness were the most common complaints of inpatients with liver hydatid cyst (40). Mirshemirani's showed that coughing was the most common symptom inpatients with lung hydatid cysts (19). Moreover, Aghajanzadeh et al. reported cough, dyspnea, chest pain and sputum as the common chief complaints of the patients (39). In addition, Tor et al. in their study showed that out of 288 patients, 30 patients were asymptomatic, the rest (89%) were symptomatic, cough and chest pain were the most common symptoms (32). The diagnosis is based on the history of exposure in an endemic area, lab investigations, imaging techniques (X-ray, ultrasound, CT and MRI) and serological examinations (specific IgG, complement fixation, indirect fluorescent antibody (IFA), and enzyme-linked immunosorbent assay (ELISA) tests). The results of serological examinations always have to be related to the clinical diagnosis, imaging and other lab techniques. The severity of various serological tests used for hydatid disease varies from 64 to 87% (18, 41, 42). In our study, ultrasonography was the best and the most definitive technique of diagnosis. The Ultrasonography is the method of detection of both hepatic and extrahepatic echinococcal cysts; the sensitivity of this technique ranged from 93% to 98% (16). Farrokh in his study showed that ultrasonography had the sensitivity of 83% in diagnosing hydatid cysts and suggested that clinical history and examination with sonography were enough for hydatid cysts diagnosing (43).

Furthermore, in the study of Rahimnejhad, radiologic methods showed the sensitivity of 100% for lung cysts and 85.7% for liver cysts (44). Ultrasonography is considered the best diagnostic mean for providing essential information about the location, number, size, and type of cysts. Sonography has been recommended for diagnosis of hydatid cyst in endemic and hyperendemic regions. In the present study, 98.7% of patients underwent a surgery. Overall the management of both primary and recurrent hydatid disease is surgical, as antihelminthic chemotherapy alone has failed in many cases (45). In many parts of the world, including Iran, surgery remains the treatment of choice for most individuals suffering from CE (5). However, The World Health Organization recommendations state that medical therapy should be used for: patients with inoperable disease, patients with multiple cysts in two or more organs, patients with peritoneal cysts, patients following incomplete surgery or relapse and for prevention of secondary spread of echinococcal infection following spontaneous rupture or aspiration of cysts (45).

In the present study, two patients died, one of them died because of spontaneous rupture of the liver hydatid cyst and the other one, due to bleeding after liver surgery. The complications of hepatic hydatid cysts are generally rare and they include two main categories: rupture and secondary bacterial infection (1). Moreover, the major complications after surgical resection with hepatic disease include wound infection, biliary leak, and intraabdominal bleeding resulted in a hepatic lobectomy (16). In the present study, the mortality rate was 3.2%, in comparison with 1.72% in Mirshemirani study (19). In our study, recurrence rate was 7.69% (2 patients with liver cyst recurrence and 1 patient with lung cyst recurrence) while Mirshemirani reported 6.14% of recurrence rate in children hydatid cysts (19). As a whole the present study showed that hydatid cyst was mostly common in the age group of 40 - 60 years old and most of the patients were diagnosing living in urban areas. In general, the situation of the hydatidosis in the humans in Guilan province shows somehow a resemblance with the other areas in Iran.

Hydatid cyst disease is still an important health problem in Iran that needs further studies. Therefore, accurate information on the distribution of the disease is the first step toward the control and prevention of disease. Retrospective hospital survey data on human hydatid cysts cannot provide an accurate picture about the echinococcosis. A certain number of cases are not referred to the hospitals because the infection is asymptomatic or does not require surgical intervention or hospitalization, and some data were not recorded in their files.

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## Authors' Contribution

Study concept and design was performed by Mansour-Ghanaei and Joukar. Analysis and interpretation of data was carried out by Joukar. Data was collected by Javadi. Manuscript was written by Mansour-Ghanaei, Joukar and Souti.

## Financial Disclosure

The authors declare that they have no conflicts of interests.

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