



Investigation of Prevalence and Risk Factors of Human Hydatid Cyst Disease in Jordan, Western Middle East: A Systematic Review (1990–2023)

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Abstract

Introduction: The primary objective of this systematic review is to enhance our understanding of the prevalence of human hydatid cyst disease through a new and comprehensive assessment.

Methods: All studies related to human hydatid cyst disease in Jordan from 1990 to January 2024 were searched in Google Scholar, Research Gate, and PubMed databases. Articles relevant to the prevalence, serology, surgery, review, and data were included in the systematic review.

Results: Only 8 studies met the inclusion criteria in the systematic review. According to these studies, the serological prevalence of hydatid cysts in Jordan was 5.2 %. During the surgical investigation, 801 cases of hydatid cysts were recorded, and the gender variable showed us that the amount of women (63.04%) who had cysts was more than men (36.95%). In terms of age variable, 82.62% of the people who underwent surgery were < 50 years old and 17.37% were > 50 years old.

Conclusions: The current study provides a better understanding of the prevalence and cases undergoing surgical intervention due to hydatid cyst disease in Jordan. The Middle East region is endemic for this disease, and Jordan is one of the countries situated in this endemic area. Therefore, investigating the incidence, familiarity with risk factors, and the lifecycle of this disease should be more important than ever before.

Keywords: *Echinococcus granulosus*, Hydatid disease, Cystic echinococcosis, Systematic Review, Jordan

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Introduction

Overview

Echinococcosis (hydatid disease) is a common parasitic disease between humans and animals that occurs due to the infection of the larval stages of cestodes of the *Taenidae* family and *Echinococcus* genus (1). In general, 3 types of hydatid cysts are observed in humans: cystic echinococcosis (CE) caused by *E. granulosus*, alveolar echinococcosis (AE) caused by *Echinococcus multilocularis*, and polycystic echinococcosis (PE) caused by *Echinococcus vogeli* and *Echinococcus oligarthrus*. Two types of these 3 cases are very important for humans in medicine, namely AE and CE, which can cause great damage to the medical community of countries (2,3). PE is the mildest type of hydatid cyst disease and is not important in the Middle East, most cases being reported in Central and South America (4,5). According to World Health Organization, hydatid cyst disease is one of the 17 neglected tropical diseases that causes 3 billion dollars in medical damages in the world annually (6,7). Depending

on the location of cysts and their growth rate, the clinical symptoms vary, among them can be mentioned the abdominal pain, lung and liver disorders. In rare cases, neurological disorders can be seen. Sometimes cyst rupture causes fever, anaphylaxis, lung wheezing and even the death of the patient (8,9). Hydatid cysts are diagnosed using medical examinations such as imaging and indirect immunofluorescence or ELISA tests (10).

The Situation in the Middle East

Echinococcus granulosus, the causative agent of hydatid disease, was first discovered in the Alaska region of North America (11). However, in terms of prevalence and distribution, this disease is found in almost all parts of the world, including: southern and central Russia, Central Asia, China, Australia, some areas of South America, northern and eastern Africa, and the Middle East (12). This disease has been reported in many countries in the Middle East, making the region endemic for this disease. However, accurate statistics on the prevalence of hydatid



cyst disease are lacking in many countries in the region. Most of the research on hydatid cysts worldwide has been conducted in three countries: Turkey, France, and China (13). Middle Eastern countries have diverse climates, but the predominant climate in this region is warm and arid, which contributes to the spread of this disease. Recent research in Middle Eastern countries indicates a seroprevalence of hydatidosis of 1.6% in Turkey and 5% in Iran (14,15). Surgical cases are also reported annually in countries from this region. Studies conducted in the western Middle East, in Lebanon, indicate that the liver is the most commonly affected organ. Also Qatar, one of the countries in the Persian Gulf region, has similar results, with 81.3% of patients having liver cysts (16,17). Jordan is considered as one of the countries where this disease is endemic in the region. Therefore, statistical surveys are very helpful in controlling and managing this disease better.

Life Cycle

The biological cycle of *E. granulosus* is of the heteroxenous type, occurring in two hosts; a definitive host, primarily the dog and other wild canids (wolf, jackal, coyote), and intermediate hosts, mainly sheep as well as other herbivores (cattle, goats, camels, horses), with humans incidentally involved in the cycle. The number of developed *Echinococcus* worms depends on the number of ingested protoscoleces. Dogs are typically infested with several hundred worms attached between the intestinal villi of the small intestine. The gravid segment, filled with eggs, detaches from the strobila (worm body) and is eliminated with feces into the external environment where it disintegrates and releases the eggs. Each egg or embryophore contains a hexacanth embryo or oncosphere. After ingestion by an intermediate host, the oncosphere is released from its shell under the action of digestive juices; it traverses the intestinal wall using hooks and its own secretions, measuring 20 to 25 μm in diameter, but its plasticity allows it to pass through all capillaries. It reaches the liver via the portal system, sometimes exceeding the liver through the suprahepatic veins and reaching the lungs. More rarely, localization can occur in any part of the body through the general circulation. Once in the organ, the embryo transforms through a process of “vesiculation” into a hydatid larva. The cycle is closed when the dog (definitive host) ingests the viscera (liver, lungs) carrying fertile cysts from parasitized animals (intermediate hosts). The ingested protoscoleces undergo the action of pepsin in the stomach and evaginate in the anterior part of the duodenum under the effect of bile and pH modification. They then develop into sexually mature worms. Each ingested protoscolex can give birth to an adult cestode within an average of six weeks after infestation. However, the duration of the prepatent period varies depending on the strains of the *E.*

granulosus species. Humans accidentally enter the parasite cycle, generally constituting a parasitic dead end. The hydatid, following asexual reproduction in the form of active polyembryony, contains several hundred thousand protoscoleces, infective elements for the definitive host. Hydatid development is very slow, and fertility (formation of protoscoleces) is only achieved after 15 to 18 months in sheep and cattle (18-21).

Material and Methods

Data Collection

This study is a systematic and retrospective study that investigated the prevalence of *hydatid cyst* disease based on serological data. Checklist search method (PRISMA) was collected from English language databases including: Google Scholar, ResearchGate, PubMed, Embase, Scopus using the following search terms: “Hydatid cyst”, “Serological prevalence”, “Cystic Echinococcosis”, “Hydatidosis”, “IgG antibody”, Hydatid cyst surgery “Epidemiology” and “Jordan”. The data included a wide range of literature, including articles, abstracts, national and international congresses. Only full-text articles between 1990 and 2023 were included in the statistical data of this systematic review.

Certain parameters regarding sex, age category and communion with dogs were statistically analyzed in the GraphPad Prism program. The *P* value was calculated using one-tailed Fisher’s exact test (Figure 1).

Study Area

The Middle East is a large region of 7.207.575 square kilometers in Southwest Asia and Northeast Africa, which has a warm and tropical climate in most areas. The country of Jordan, with an area of 89.350 square kilometers and a population of 10.193.000 people, is one of the countries in the Middle East region, with coordinates 29° and 34° N, 34° and 40° E. The Jordan intersects with Saudi Arabia in the south and east, Iraq in the northeast, Syria in the north and Palestine in the west. The climate is hot and dry. Most of the country’s regions are covered by dry deserts, which is the reason for the population dispersion in this country (22) (Figure 2).

Results

Surgical Cases Examination

A review of surgical cases in Jordan between 1990 and January 2024 showed that 801 patients underwent surgery in Jordanian hospitals (Table 1). Of this number, 296 (36.95%) were male and 505 (63.04%) were female, indicating that the incidence of the disease is higher in Jordanian women than in Jordanian men ($P < 0.0001$). Additionally, an analysis of the age variable showed that 82.62% were under 50 years old ($P < 0.0001$). The location of hydatid cysts is very important, so this important variable was also investigated. It was found that 65.56% of

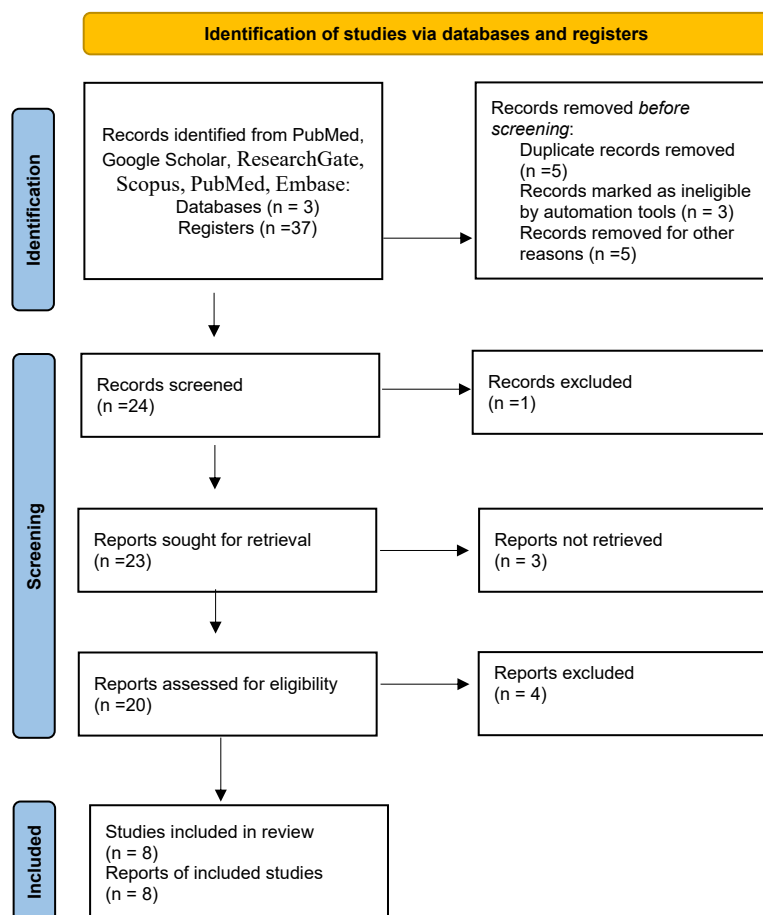


Figure 1. Flow chart of the study selection process showing inclusion and exclusion of studies identified

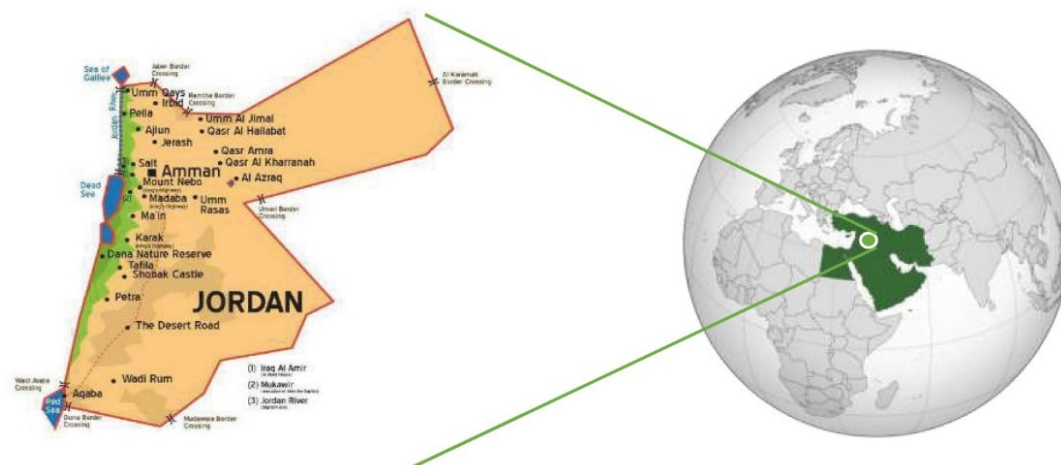


Figure 2. The Position of the Middle East in the world and the country of Jordan in the Middle East

Table 1. The status of CE Surgery Cases in Jordan (1990–2023)

Author Name (Reference)	Year of Study	Region	Population	No. of Samples (Prevalence %)
Amr et al, 1994 (23)	1976 -1986	Jordan – Jordan	Patients with CE	306 (100%)
Dowling et al, 2000 (24)	1990 - 1996	Jordan – Jordan	Patients with CE	44 (100%)
Yaghan et al, 2004 (25)	1994 - 2003	Jordan – Jordan	Patients with CE	65 (100%)
Daradkeh et al, 2007 (26)	1973 - 1999	Jordan – Amman	Patients with CE	169 (100%)
Al-Radaideh et al, 2017 (27)	2005 - 2015	Jordan – Jordan	Patients with CE	217 (100%)
Total	-	-	-	801 (100%)

the surgically treated cysts were in the liver, 17.74% in the lungs, 1.85% in the kidneys, 1.32% in the brain, 3.57% in the spleen, 6.88% in other parts of the body, and 3.4% in multiple locations throughout the body (Table 2).

Serological Examination

Nowadays, serological investigations play a crucial role in the control and rapid diagnosis of diseases. Serology is also highly important in the diagnosis of human hydatid cyst disease. Over a 33-year period in Jordan, three major studies were conducted using the ELISA test. In 1993, 1994, and 2003, a total of 4,570 individuals were included in these studies. Among the samples prelevated, 238 tests equivalent to 5.20% of all, were reported positive. In terms of gender distribution, 64.37% of the diagnosed samples were from females, while 38.62% were from males (Table 3).

Table 2. Subgroups of CE Surgery Cases Distribution in Jordan (1990–2023)

Variables	No. of Studies	Sample Size (Prevalence %)	P Value
Gender	5	-	0.0001
Men	-	296 (36.95%)	
Women	-	505 (63.04%)	
Total	-	801 (100%)	
Age category	2	-	0.0001
<50	-	233 (82.62%)	
>50	-	49 (17.37%)	
Total	2	282 (100%)	
Location of hydatid cyst	4	-	0.0001
Liver	-	495 (65.56%)	
Lung	-	134 (17.74%)	
Kidney	-	14 (1.85%)	
Brain	-	10 (1.32%)	
Spleen	-	27 (3.57%)	
Other	-	52 (6.88%)	
Mix	-	23 (3.04%)	
Total	4	755 (100%)	
Communion with dogs	2	-	
Yes	-	109 (31.14%)	
No	-	241 (68.85%)	
Total	2	350 (100%)	

Table 3. Serological Status of CE in Jordan (1990–2023)

Author Name (Reference)	Year of Study	Region	Diagnostic Method	No. of Samples	No. of Positive (Prevalence %)
Abo-Shehada, 1993 (28)	1993	Jordan – Jordan	ELISA	176	5(2.84%)
Moosa and Abdel-Hafez, 1994 (29)	-	Jordan – Jordan	ELISA	2,006	48(2.39%)
Qaqish et al, 2003 (30)	1998-2001	Jordan – Jordan	ELISA	2,388	185 (7.74%)
Total	-	-	-	4,570	238 (5.20%)

Discussion

Parasitic diseases are among the most significant health concerns worldwide, affecting millions of people annually (31). As hydatid cyst disease is considered a major parasitic disease of public health concern in both humans and animals, it can lead to various disruptions within communities (32). Therefore, we decided to conduct a systematic study of this disease in Jordan. From 1990 to January 2024, approximately 24 individuals with human hydatid cysts underwent surgery in hospitals in this country each year. In the present study, the occurrence rate of the disease in the Jordanian population was determined to be an average of 8 cases per 100,000 individuals, which is higher than in other countries in the Middle East. This rate is 0.74 in Fars province of Iran per 100,000 individuals (33), 6.6 in Turkey per 100,000 individuals (34), and 1.2 to 2.7 in Israel per 100,000 individuals (35). Therefore, effective strategies for disease control and better identification are needed in Jordan. Dogs are considered the definitive hosts of this disease, and many studies have shown a significant relationship between keeping dogs at home or direct contact with dogs and hydatid cyst disease (36,37). However, in this study, no significant correlation was observed between patient contacts with dogs (Table 2). Various variables in serological studies were very limited, only indicating that the prevalence of the disease is higher in women than in men (Table 4). However, overall, conducting serological studies greatly aids in faster and more accurate diagnosis, and health organizations in Jordan should pay more attention to this matter.

Conclusion

This comprehensive study has shown us that the rate of hydatid cyst surgeries per population in Jordan is higher compared to many other countries in the Middle East. Additionally, studies related to the serology of this disease during this 33-year period have been very limited, and specific variables have not been thoroughly investigated. Considering that Jordan is located in the Middle East, an endemic region for this disease, more extensive research should be conducted, and the precise prevalence rates should be documented in all provinces, considering the climatic conditions of each region. Many individuals may be infected with this disease without showing specific symptoms, making serological diagnostic tests essential for screening infected individuals. Public health is the most important issue in various countries; therefore, more

Table 4. Subgroups of CE Serology Distribution in Jordan (1990–2023)

Variables	No. of Studies	Sample Size	No. of Positive (Prevalence %)	P Value
Gender	2	-	-	0.00020
Men	-	1.908	83 (35.62%)	-
Women	-	2.486	150 (64.37%)	-
Total	2	4.394	233 (100%)	-

extensive and precise research on *hydatid cyst* disease in Jordan will contribute to better public health in the community.

Authors' Contribution

Conceptualization: Alireza Ghorbani.

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Formal analysis: Alireza Ghorbani, Yagoob Garedaghi.

Investigation: Alireza ghorbani, Khoulood Dekkiche.

Methodology: Alireza ghorbani, Yousef Gharedaghi.

Project administration: Alireza Ghorbani, Khoulood Dekkiche.

Resources: Fatemeh Hodaei, Yousef Gharedaghi.

Software: Alireza Ghorbani, Yousef Gharedaghi.

Supervision: Yagoob Garedaghi.

Validation: Alireza Ghorbani

Visualization: Alireza Ghorbani.

Writing—original draft: Alireza Ghorbani, Khoulood Dekkiche

Writing—review & editing: Alireza Ghorbani, Khoulood Dekkiche, Yagoob Garedaghi.

Competing Interests

None declared.

Ethical Approval

Not applicable.

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