

Prevalence, Clinical Features, and Management of Hydatid Disease in Saudi Arabia: Systematic Review

Faten A. Al Braikan, MSc, PhD

Department of Clinical Microbiology and Immunology, Faculty of Medicine, King Abdulaziz University, Jeddah, Saudi Arabia

Correspondence

Dr. Faten A. Al Braikan

Department of Clinical Microbiology and Immunology, Faculty of Medicine
King Abdulaziz University
P.O. Box 80205, Jeddah 21589
Kingdom of Saudi Arabia
e-M: falbraikan@kau.edu.sa

Submission: 02 Jun. 2024

Accepted: 15 Nov. 2024

Citation

Al Braikan FA. Prevalence, clinical features, and management of hydatid disease in Saudi Arabia: Systematic review. JKAU Med Sci 2024; 31(2): 49-61. DOI: 10.4197/Med.31-2.5.

Copyright: ©The Author(s), YEAR. Publisher. The Journal of King Abdulaziz University - Medical Sciences is an Official Publication of "King Abdulaziz University". It is an open-access article distributed under the terms of the Creative Commons Attribution Non-Commercial License, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

Abstract

Hydatidosis, caused by *Echinococcus granulosus*, is an endemic parasitic disease worldwide. The most frequent anatomic locations are the liver and lung. This systematic review examined the prevalence, clinical features, diagnosis, and management of hydatid disease in Saudi Arabia. Six databases (PubMed, Medline, Ovid, Scopus, Web of Science, and Cochrane) were searched using the keywords "Hydatid and Saudi Arabia" in the title and abstract. All papers investigating the hydatid diseases in Saudi Arabia were included in the systematic review. Eight articles were considered suitable for the systematic review of 82 papers that were extracted through the database search. The studies enrolled 835 patients, of whom 440 cases were diagnosed with hydatid disease. The prevalence of hydatid disease among patients with liver diseases ranged between 3.9 and 5.6% during the period from 1978 to 2014. In addition, the liver was the most reported infected organ with hydatid diseases, followed by the lungs among patients. Some studies diagnosed hydatid disease among asymptomatic patients, while others reported gastrointestinal, respiratory, and musculoskeletal symptoms. The most commonly reported diagnostic methods were indirect hemagglutination (IHA), ultrasonography, and Computed Tomography (CT) scans for locating cysts. A combined approach of surgical interventions such as endocystectomy and cystopericystectomy with medical therapy has shown greater effectiveness despite some associated complications like anaphylaxis, mild hypernatremia, biliary leakage, and bronchopleural fistula. Improving treatment outcomes and minimizing complications requires a multidisciplinary approach that combines surgical expertise with medical therapy, increased public awareness, and early diagnosis promotion.

Keywords

Prevalence, Diagnosis, Hydatid, Echinococcosis, Saudi Arabia

INTRODUCTION

Hydatic disease or cystic echinococcosis is a parasitic infectious disease that can be transmitted from animals to humans caused by the larval stage of the tapeworm *Echinococcus*^[1]. Human infection with the parasite can occur through the consumption of food or water contaminated with its eggs or through direct contact with infected dog feces^[2]. Four species of *Echinococcus* may be associated with human infection: *Echinococcus granulosus*, *Echinococcus multilocularis*, *Echinococcus vogeli*, and *Echinococcus oligoarthritidis*. The most prevalent of these is tapeworm *E. granulosus*, which is responsible for cystic echinococcosis globally^[3,4]. Other species of *Echinococcus* cause uncommon medical conditions. For instance, *E. multilocularis*, the juvenile stage of the fox tapeworm, leads to alveolar *Echinococcus*^[5]. This condition generally appears as a lesion that occupies space in the liver and spreads to other organs. *E. vogeli* and *E. oligoarthritidis* are the species responsible for polycystic echinococcosis, which presents as multiple cysts on almost any organ^[6].

Hydatid disease is an international public health problem. It commonly affects developing countries, South America, West China, the Middle East, and North Africa (MENA), and other countries. The overall prevalence of the disease is underestimated due to low surveillance studies conducted in all prevalent regions. It was estimated that Hydatid disease has a global incidence rate of 1–200 per 100,000 yearly^[7]. Furthermore, the number of cases affected by hydatid disease in the MENA region in 1990 was 134,980 and increased to 207,368 in 2019^[8].

Hydatid disease tends to affect the liver (50%–77% of cases) due to its bowel venous drainage system. However, it can involve any visceral organ in the body, including the lung (15%–47%), spleen (0.5%–8%), kidney (2%–4%), and other organs^[9,10]. Despite the unusual involvement of organs other than the liver and lung, it can cause substantial morbidity and mortality^[11–13].

The disease could present with a range of symptoms that are not specific to the disease itself. The clinical manifestation of patients with hydatid disease depends on the organ affected and the size of the cyst, as well as the interaction between the cysts and the structure of the organ due to obstruction of blood/lymphatic flow, rupture, or subsequent bacterial

infection. The infection may remain asymptomatic in earlier stages when the cyst is small (<10 cm) and may remain for many years; however, serious complications can occur, including peritonitis, anaphylactic shock, and multiorgan failure. Accordingly, disease management is crucial to reduce the possible complications^[14,15]. In addition, secondary cyst infections are attributed to cystic-biliary communication. These infections are considered the most common risk factors for elevated intracystic pressure, erosion of adjacent structures by an enlarging cyst, and complications developing^[15–17].

Abnormal laboratory findings, including thrombocytopenia, leukopenia, and elevated liver function, are observed in hydatid disease; however, these are nonspecific and not diagnostic. Several approaches are used for diagnosing the disease, including serological tests, radiological evaluation, and histopathological and cytological examinations^[18,19]. The combination of imaging and serology investigations usually enhances diagnosis^[20]. Regarding radiological evaluation, ultrasound imaging is a highly sensitive and efficient way to detect liver lesions^[21]. Moreover, Computed Tomography (CT) scans and Magnetic resonance imaging (MRIs) are more effective in detecting and characterizing hydatid disease with greater sensitivity and specificity^[22]. CT scans can quickly and accurately diagnose cyst ruptures, providing the exact location and type and allowing prompt surgical intervention in emergencies^[23]. In terms of serological tests, Casoni skin and the indirect hemagglutination (IHA) methods are the most common approaches to detect the disease. The Casoni skin test involves injecting hydatid cyst fluid into the skin, while the IHA is a serum antibody test that is highly sensitive to the disease^[24].

Treatment of hydatid cysts is attributed to their size, location, clinical signs and symptoms, and patient characteristics. Management usually includes antiparasitic treatment, surgical resection of the cyst, or percutaneous puncture, aspiration, injection, and respiration (PAIR), depending on the World Health Organization (WHO) diagnostic classification^[25]. Smaller, uncomplicated liver cysts less than 5 cm in size can be effectively treated with albendazole alone or in combination with praziquantel. However, surgery is recommended for cysts larger than 10 cm and for cysts at risk of rupture or complications. In particular, the total cystopericystectomy technique is preferred as it has lower risks of postoperative complications^[26]. Additionally, Endocystectomy is used for hepatic cystic

echinococcosis management, which is the conservative and feasible surgical approach^[27].

Limited studies were conducted in Saudi Arabia to describe hydatid disease infection, its epidemiology, clinical presentation, diagnosis, and management to raise awareness of the problem and improve early diagnosis and management of the disease. Thus, we conducted this systematic review of the existing literature to provide an overview of the clinical features, management, and outcomes of hydatid disease in Saudi Arabia.

MATERIAL AND METHODS

This systematic review complied with established criteria (Preferred Reporting Items for Systematic Reviews and Meta-Analyses, PRISMA)^[28].

SEARCH STRATEGY

The systematic review was conducted through a literature search of PubMed, Medline, Ovid, Scopus, web of Science, and Cochrane databases using the keywords "Hydatid AND Saudi Arabia" in the abstract and title. The author screened studies examining hydatid disease in Saudi Arabia to select studies that matched the inclusion and exclusion criteria. Furthermore, key data points were retrieved from the final record of the included research.

INCLUSION AND EXCLUSION CRITERIA

The systematic review included all papers investigating hydatid diseases in Saudi Arabia: prevalence, involved organs, investigation, management, and disease outcome. Studies were selected based on the following criteria: published in English, involving clinical samples, and full-text availability. We excluded duplicated papers, published studies in languages other than English, narrative reviews, case reports, case series, studies with insufficient data or findings, studies with irrelevant findings, studies that did not include clinical samples and studies for which full text was unavailable.

SCREENING AND DATA EXTRACTION

A reference manager was used to check the output of the search technique for duplication. The titles and abstracts of the relevant studies were first screened. Afterward, relevant full-text papers were examined for inclusion criteria. An independent

author independently extracted data from a Microsoft Excel spreadsheet. The data included authors, year of publication, study design and period, objective, population characteristics, and study outcomes.

STRATEGY FOR DATA SYNTHESIS

A summary table was created using data from relevant studies to provide a qualitative interpretation of the findings and study components.

RISK OF BIAS ASSESSMENT

Out of 82 extracted studies, eight met our inclusion criteria and were assessed for risk of bias using the ROBINS-1 tool^[29] among non-randomized studies of interventions (NRSI). This tool was used to evaluate the quality of studies included, assessing aspects such as confounding, selection bias, measurement of outcomes, and intervention classification. The studies' risk of bias was categorized as low, moderate, serious, or critical based on predefined criteria. The overall risk of bias was reached using signaling questions. The risk of bias revealed the overall quality of the included studies.

RESULTS

OVERVIEW OF THE INCLUDED STUDIES

Eighty-two (82) papers were extracted from six databases (PubMed, Medline, Ovid, Scopus, Cochrane, and Web of Science) search. Of these, 28 were omitted as duplicates. Regarding the remaining 54 articles, 44 were excluded because they did not match the inclusion criteria: 3 were review articles, 19 did not include clinical samples, 16 were case reports, 2 were case series, and 4 were not full-access papers. Following screening and assessment, 2 articles were excluded because they did not match the study's objective. Eight articles were considered suitable for the systematic review (Figure 1).

The included papers were published in different hospital settings in Saudi Arabia, mainly in two cities (Riyadh and Jeddah). The studies enrolled 835 patients, of whom 440 cases were diagnosed with hydatid disease. The included studies were published between 1983 and 2016 (Table 1). The study design varied among the included studies; one of the papers utilized a case-control study design, six relied on retrospective analysis, and one was prospective studies. The studies

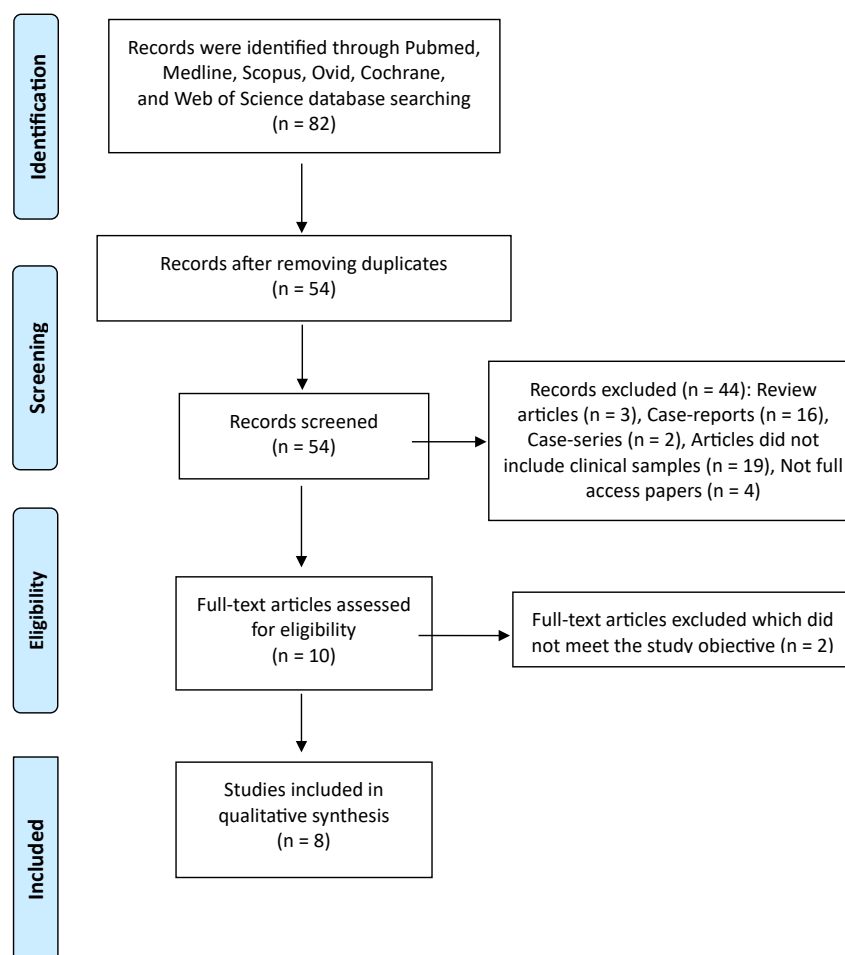


Figure 1. Flow diagram of study selection for the systematic review

Table 1. Characteristics of the included studies.

Study Reference Number	Study Design (Period)	Study Objective	Population Characteristic	Results
30	Retrospective (from March 1978 to September 1980)	To identify clinicopathological patients with liver disease	Number: 124 with liver disease. Out of 7 patients, they were diagnosed with hydatid disease. Gender: 4 males and 3 females with hydatid disease. Age: ranged from 17 to 50 years.	The prevalence of hydatid liver disease among patients with liver diseases who had liver biopsy performed was 5.6%. Laboratory findings: All patients had a normal level of bilirubin. SGOT levels ranged from 6 to 54 (i.u.), while SGPT levels ranged from 4 to 38 (i.u.). Alkaline phosphatase levels ranged from 18 to 94 (i.u.). Alpha-fetoprotein was tested among one patient (1/20000 ng ml ⁻¹). Eosinophilia was detected in four patients.

Table 1. Characteristics of the included studies.–Continued

Study Reference Number	Study Design (Period)	Study Objective	Population Characteristic	Results
31	Case-control study	Serodiagnosis of human hydatid disease	Number: Control group: 120 male blood donors + 114 patients (adult females and children) with no history of hydatid disease. Test group: 52 suspected hydatid patients.	Diagnosis: Out of 52 patients who were suspected to have human hydatid disease, 30 cases tested positive through serodiagnosis by IHA test. Among 20 cases of hydatid liver cysts, three were confirmed through ultrasonography, resulting in an 80% positivity rate. Among patients suspected of having hydatid disease (hepatomegaly, splenomegaly, or hepatosplenomegaly) and exhibiting symptoms, 50% had IHA titers of 1:128 or higher. The serological results of this control group showed that 70% had no hydatid IHA antibodies, while 25% had antibody titers of 1:4 to 1:8.
32	Retrospective study (September 1984 to August 1986)	To describe the management of liver hydatidosis.	Number: 68 patients with hydatid disease involving various organs (liver, lung, and others (brain, kidney, spleen, abdominal wall) at Riyadh Central Hospital. Gender: Male: 15 Females: 27 Age: from 8 to more than 60 years.	Diagnosis: 42 patients were diagnosed with liver cysts. The right lobe was the site with the most involvement (71%). Ultrasonography and Casoni's intradermal test were helpful in diagnosing cysts. CT was used to better localize cysts and plan suitable surgical procedures. Organ involved: •Liver: Liver alone (37 patients), liver and spleen (2 patients), liver and lung (2 patients), Liver, spleen, and kidney (1 patient). •Lung: Lung alone (20 patients), lung and spleen (1 patient), and lung and breast (1 patient). •Other organs: Brain (1 patient), kidney (1 patient), spleen (1 patient) and abdominal wall (1 patient). Symptoms: Abdominal pain (29 patients), abdominal mass (17 patients), jaundice (5 patients), and asymptomatic (4 patients). Laboratory findings: Leukocytosis (> 12,000/mm ³): 8 patients (17%). Eosinophilia (>8%): 14 patients (31.1%). Serum bilirubin (> 2 mg/dl): 5 (11.1%). Alkaline phosphatase (> 200 IU/dl): 10 patients (22.2%). Positive Casoni's test: 26 of 30 patients (87%). Positive IHA test: 20 of 28 patients (71%). Scan findings: -15 cases experienced plain abdominal and chest roentgenograms. •A raised dome of the diaphragm was detected in nine cases. •Soft tissue mass was detected in three cases. • Calcification was detected in three cases. - Ultrasound examination identified the cystic mass in all cases. -An isotope liver scan was performed among 15 patients, and all had the filling defect due to the cyst. -A CT scan was needed in 20 cases for better cyst localization.

Table 1. Characteristics of the included studies.–Continued

Study Reference Number	Study Design (Period)	Study Objective	Population Characteristic	Results
				<p>- An ERCP was performed on four patients with high serum bilirubin.</p> <p>- The remaining patient with high bilirubin had PTC.</p> <p>Management: Out of 33 patients underwent surgery. Endocystectomy with tube drainage was performed among 23 patients, and five patients underwent total cystoperiostomy. One case received medical treatment.</p> <p>Outcome: No complications were noted in these patients. One patient died of anaphylaxis following percutaneous transhepatic cholangiography. A 20% hypertonic saline solution was used as a scolicidal agent in this series, and mild hypernatremia was noted among some patients.</p>
33	Descriptive study (between 1988 and 1997)	To investigate the epidemiology of hydatid disease	<p>Number: 67 patients diagnosed with <i>E. granulosus</i> infection.</p> <p>Gender: Male: 53.7%, Females: 46.3%</p> <p>Age: mean: 39.2 (SD:18.2) years.</p>	<p>Diagnosis: All patients were diagnosed using serology, including IHA, Casoni skin test, radiography, ultrasound, and CT.</p> <p>Symptoms: Half of the patients had gastrointestinal symptoms, 49.3%. Respiratory (32.8%), genitourinary (11.9%), and musculoskeletal systems were also involved (3%). Approximately 3% of the patients were asymptomatic. Out of 40 patients had hepatic involvement, of which four were considered secondary, and the majority had cysts in the right lobe. One patient complained of back pain.</p> <p>Cyst location: The cyst was found in the spleen, the tail of the pancreas, and the extensor muscles of the back and the lower limbs.</p> <p>Treatment: 56.7% of patients received surgical treatment, 20.9% had surgical and medical therapy, and 22.4% had albendazole.</p> <p>Outcome: Out of 97% of the patients survived, but 20% had a disease recurrence. Two patients died with a case fatality rate of 3%. One patient (a 66-year-old female) died two weeks after the operation for hydatid disease of the liver and bronchus. The other patient was an 85-year-old who had had surgery five years earlier and died of unknown causes.</p>
34	Retrospective study (from 1983-1996)	To assess the medical records of patients with histopathologic diagnosis of hepatic granuloma	<p>Number: 116 patients</p> <p>Gender: Male: 77. Females: 39</p> <p>Age: from 18 to 65 years.</p>	<p>The prevalence of hydatid disease among patients who had hepatic granulomas identified by liver biopsy was 4.3%.</p> <p>Symptoms: Anorexia and abdominal pain.</p>

Table 1. Characteristics of the included studies.–Continued

Study Reference Number	Study Design (Period)	Study Objective	Population Characteristic	Results
35	Observational study (December 1999 to December 2004)	To describe the presentation, diagnosis, and management of patients suffering from Echinococcosis.	Number: 117 patients with echinococcosis. Gender: Male: 63.24%, Females: 36.76% Age: mean: 40.9 (SD: 20.7) years.	<p>Clinical presentation: Out of 29 patients (24.8%), complications related to Echinococcosis were present.</p> <p>Symptoms: The most typical symptoms were pain in the right upper quadrant (51 cases, 43.6%), followed by cough ± sputum (22 cases, 18.8%). Pain in the upper abdomen was detected in 21 cases, or 17.9%. Nine patients were presented with jaundice ± cholangitis. A few cases were presented with fever and Lump L Thigh (3 and 1 case, respectively).</p> <p>Diagnosis: Laboratory findings: The IHA test gave a sensitivity of 78.6. The initial results of the hematological profile indicated a mean hemoglobin level of 12.7 (SD:17.4) g/dl and a mean white blood cell count of 9.3 (SD:4.9) /cc.</p> <p>Scans: All cysts were visualized using ultrasound scans (cases with extrapulmonary Echinococcosis), chest X-rays, and CT scans (used in selected cases).</p> <p>Management: Endocystectomy was the most frequent procedure in both the liver and lung. Albendazole was the most frequently used drug among 78 cases (68.4%) at a dose of 15 mg/kg in two divided doses. Mebendazole was used among the rest of the patients at a 50 mg/kg dose in three divided doses. Both these drugs were efficacious. However, Albendazole was more efficacious than Mebendazole.</p> <p>Outcome: Eight patients experienced biliary leakage, while three suffered from bronchopleural fistula. Most patients with these conditions could recover with conservative treatment, with only two of the biliary leakage cases and one of the bronchopleural fistula cases requiring surgery. Additionally, 15 patients experienced complications related to infections. No recurrences were recorded during the follow-up period; all patients who underwent surgery also received medical treatment. However, multiple factors prevented 25 patients (21.4%) from undergoing operative treatment and were treated with medical care. The mean hospital stay was 14.4 days (2–187 days). Patients receiving surgical treatment were discharged earlier, with a mean stay of 11.9 days, while those receiving medical treatment alone were generally sicker and tended to stay longer, with a mean of 23.6 days.</p>
36	Retrospective study (2006-2014)	To measure morbidity and	Number: 77 patients underwent hepatectomy	The prevalence of hydatid cysts among patients undergoing liver resection was 3.9% as a benign indication.

Table 1. Characteristics of the included studies.–Continued

Study Reference Number	Study Design (Period)	Study Objective	Population Characteristic	Results
		mortality and assess predictors of outcome after hepatectomy.	Gender: Male: 53.3%, Females: 46.8% Age: mean: 49.1 years.	
37	Prospective study, (1995 to 2005)	To identify risk factors associated with endobronchial rupture among patients with hydatid cysts.	Number: 32 patients undergoing thoracotomies for pulmonary hydatid cysts Gender: Male: 21 Females: 11 Age: mean: 32 (SD: 15) years.	Diagnosis: Chest radiography and CT were used to identify the cysts' precise location, size, and integrity. Outcome: 53.1% of the patients (17 in total) had ruptured cysts (group 1), while 15 patients had intact cysts (group 2). Most cysts (68.7%) were found in the right lung. Out of 21 (65.6%) cysts were identified in the lower lobes, 6 (18.7%) in the upper lobes, and 5 (15.6%) in the lingual and middle lobes. The fistula diameter was the only significant risk factor associated with cyst rupture.

IHA = Indirect hemagglutination; CT = Computerized tomography; ERCP = Endoscopic retrograde cholangiopancreatography; PTC = per-cutaneous transhepatic cholangiography; SD = Standard deviation.

included diverse patient populations within the age range of six months to 95 years, and most of them were patients with liver disease.

RISK OF BIAS ASSESSMENT OF THE INCLUDED STUDIES

Regarding the risk of bias assessment, Table 2 summarizes the studies that have been included in the hydatid disease pattern. Based on our assessments, the overall quality was between moderate and serious. The probable sources of bias were the presence of confounding, measurement of reported results, and classification of interventions that were mostly reported in an unclear way for a conclusive judgment.

DISCUSSION

Hydatid disease is an endemic chronic disease in certain areas that causes devastating acute complications^[38,39]. Few clinical studies on hydatid diseases were carried out in Saudi Arabia. Therefore, this systematic review aims to investigate the prevalence, clinical features, diagnosis, management, and prognosis of hydatid disease in Saudi Arabia.

The prevalence of hydatid disease was reported only among patients with liver diseases. In the

current systematic review, three studies discussed the prevalence of hydatid diseases among patients with liver diseases only. The prevalence of hydatid disease was 5.6% from 1978 to 1980 in Jeddah. However, in Riyadh, the rate decreased to 4.3% from all liver biopsy specimens obtained over 14 years from 1983 to 1996. During the period from 2006 to 2014, the rate was 3.9%^[30,34,36]. These findings suggest a positive development in the management and control of hydatid diseases among patients with liver conditions. The decreasing prevalence may be attributed to various factors, such as advancements in medical technology, improved diagnostic techniques, and enhanced public health measures. WHO has estimated that the rate of human infection exceeds 50 per 100,000 individuals each year in areas where echinococcosis is endemic. High prevalence is noted in various regions of Argentina, Central Asia, and East Africa^[40]. In our study, it is difficult to estimate the prevalence of hydatid disease as these data were extracted from symptomatic patients who underwent liver biopsy or resection. Hence, this underestimated the total number of hydatid diseases in Saudi Arabia. Future studies should strive to encompass a broader range of patient demographics and disease presentations in recent years to gain a more comprehensive understanding of the true prevalence of the disease and avoid prevalence influenced by changes in public health across years.

Table 2. Robvis traffic light plot figure

Study ID	D1	D2	D3	D4	D5	D6	D7	Overall
Kassimi et al., 1983 ^[30]	-	+	-	-	+	+	-	-
Hossain et al., 1985 ^[31]	-	+	-	+	X	+	X	X
Al-Kraida et al., 1988 ^[32]	-	+	-	+	+	-	-	-
Alam, 1999 ^[33]	-	+	-	+	+	+	X	X
Al Mofleh et al., 2000 ^[34]	-	-	+	-	-	+	+	-
Fahim, and Al Salamah, 2007 ^[35]	-	+	+	+	+	+	-	-
Al-Alem et al., 2016 ^[36]	-	+	-	+	+	+	-	-
Ashour et al., 2016 ^[37]	-	-	+	-	+	-	-	-
Domains: D1: Bias due to confounding D2: Bias due to the selection of participants D3: Bias in the classification of interventions D4: Bias due to deviation from intended interventions D5: Bias due to missing data D6: Bias in measurements of outcomes D7: Bias in measurement of reported results	Judgment Low Moderate Serious							

Moreover, future research should strive to encompass underrepresented regions, such as the southern and northern parts of Saudi Arabia.

In line with the literature, the organ most reported infected with hydatid diseases in our review was the liver, followed by the lungs. However, hydatid cysts could be seen in various other parts of the body. According to the included studies, the liver was the most reported infected site for hydatid diseases^[30-32,34-36], whereas the right lobe was the most involved site among patients

with hydatid liver disease^[32-34]. In addition, the lung was the second affected organ, which was reported in three studies^[32,33,35,37], where one study reported that most cysts were found in the right lung^[37]. Rarely, cysts were also seen in other sites alone or with the liver and lung, such as the spleen, kidney, brain, breast, abdominal wall, the tail of the pancreas, peritoneal, and the extensor muscles of the back and the lower limbs^[32,33].

There are variations in the clinical features of hydatid disease, which can make a presumptive

diagnosis difficult. From the included studies, four studies reported the symptoms of hydatid disease^[32-35]. Hydatid disease can cause symptoms in various body systems, including the liver, gastrointestinal, respiratory, genito-urinary, and musculoskeletal. The reported signs and symptoms involve back and abdominal pain, even in the right upper quadrant of the abdomen and upper abdomen, anorexia, and hepatomegaly^[32-35]. Other symptoms based on the site, such as cough, anorexia, fever, jaundice, incidental urticaria, anaphylactic reaction, and disease recurrence, may also occur^[32,33,35]. Additionally, a lump on the left thigh and complications such as cholangitis, intrabiliary and intrabronchial rupture of the hydatid cyst, compression, and infection can be associated with hydatid disease^[35,37]. However, three studies identified some asymptomatic patients with hydatid disease, and patients with primary pulmonary hydatidosis had concomitant asymptomatic hepatic cysts^[32,33,35]. These symptoms are nonspecific, which may lead to ignorance, delaying the diagnosis and worsening the prognosis. The review outlines various symptoms of hydatid disease and emphasizes the need for a detailed analysis to differentiate these symptoms from other prevalent diseases in Saudi Arabia, like tuberculosis, to avoid misdiagnoses or delays in diagnosis. Future studies should focus on distinguishing these overlapping symptoms in clinical practice.

The laboratory findings discussed in the included studies revealed a wide range of variations in the biochemical parameters among patients with hydatid disease. For instance, bilirubin levels varied among the included studies. One study reported normal levels among patients with hydatid liver disease^[30], and another study reported high levels among 11.1% of patients with the disease among different organs^[32]. Moreover, one study reported the variation level of SGOT and SGPT; where the SGOT levels ranged from 6 to 54 i.u, SGPT levels ranged from 4 to 38 i.u^[30]. Two studies reported alkaline phosphate levels, one of which reported variation from 18 to 94 i.u^[30], and the other found that 22.2% had abnormal levels^[32]. One study tested alpha-fetoprotein levels in one patient with hydatid disease, reporting a result of 1/20,000 ng/mL-1^[30]. Moreover, leukocytosis was detected in 17% of patients with hydatid disease^[32]. Another study detected a high mean (Standard deviation [SD]) of leukocytes (9.3 (4.9))/cc^[35]. Additionally, two studies indicated eosinophilia among patients with hydatid disease, with prevalence rates of 5.8% and 31.1%^[30,32]. Another study among patients with echinococcosis

reported a mean (SD) hemoglobin level of 12.7 (17.4) g/dl^[35]. The variations in the findings could be attributed to factors such as the stage of the disease, the involvement of different organs, and the individual patient's response to the infection.

Regarding serodiagnosis, four studies used the IHA test, which was practical for diagnosis because of its high sensitivity (50%, 71%, 78.6, and 100%) in diagnosing the hydatid disease^[31-33,35]. One study reported that 50% of suspected hydatid disease patients had IHA titers of 1:128 or higher^[31]. The increased IHA antibody production against parasite-associated antigens can lead to immunopathological lesions^[41]. Additionally, two studies used Casoni's intradermal test (16.4% and 87%). Despite it concluded that it was helpful in diagnosis^[32,33]. In accordance with previous studies, Casoni's test showed a varied accuracy (50 to 90%)^[42-44]. Performing both the Casoni's and IHA tests in suspected hydatidosis patients enhances the diagnostic accuracy of the disease. In this review, the sensitivity and specificity of diagnostic methods for hydatid disease in various healthcare settings in Saudi Arabia have not been critically analyzed. Therefore, it's important to assess the accessibility of these diagnostic tools in all healthcare facilities and compare the techniques used to global standards for improved accuracy.

Regarding scan findings, four studies used ultrasonography, which effectively diagnosed hydatid diseases^[31-33,35]. In addition, four studies used CT^[32,33,35,37], and three used chest radiography^[33,35,37] to identify the cysts' precise locations, sizes, and integrities. Moreover, a study among patients with pulmonary disease used chest X-rays as a screening tool for the disease^[35]. These findings underscore the significance of utilizing a combination of serodiagnostic tests and imaging techniques for the diagnosis of hydatid disease.

Surgery is currently the most effective treatment for removing *E. granulosus* cysts and achieving a complete cure, with a success rate of up to 90% in patients^[45]. However, it may not be practical for some patients whose cysts are located in risky positions, multiple sites, or different organs, or who are at high risk or have inadequate surgical facilities^[46]. In the present systematic review, four studies used surgery techniques to manage hydatid disease, such as endocystectomy^[32,35] and cystoperiocystectomy^[32], and two studies did not mention the surgery type used^[33,37]. However, post-surgery complications were reported in the studies, such as anaphylaxis, hypernatremia, biliary

leakage, and bronchopleural fistula, which can occur in patients with hydatid disease^[32,33,35,37].

Additionally, the included studies reported hydatid management through surgical and/or medical therapy such as mebendazole^[35] or medical therapy only as albendazole^[33,35] or antihelminthic^[35], which effectively managed hydatid diseases. One study revealed that Albendazole was more effective than mebendazole^[35]. On the other hand, it was found that patients who underwent surgical treatment tended to have shorter hospital stays than those who received only medical treatment^[35]. The review covers surgical and medical management techniques but lacks a critical evaluation of their effectiveness and cost-benefit analysis. Future research should further explore the advantages of surgery over medical treatments, such as albendazole, in different clinical contexts. Additionally, a more comprehensive examination of complications and strategies for prevention would be valuable. Analyzing the long-term outcomes for patients with hydatid disease, including recurrence rates after treatment and potential chronic complications, would significantly enhance the review, as well as understanding the long-term socioeconomic impact of the disease, particularly for those in rural communities or with limited access to healthcare.

Considering that hydatid disease is zoonotic, it would be helpful to enhance the existing public health measures in Saudi Arabia aimed at controlling the disease. These measures could involve deworming livestock, enhancing sanitation, and educating at-risk populations about preventive practices. Furthermore, it would be beneficial to investigate the potential impact of veterinary and agricultural policies in reducing transmission, such as enforcing stricter hygiene standards in slaughterhouses and promoting responsible pet ownership. This could provide more practical insights^[45]. Therefore, collaboration between veterinarians and public health workers is crucial for effectively controlling hydatidosis^[47]. Moreover, increasing public awareness will improve treatment outcomes and minimize complications of hydatid disease in Saudi Arabia.

One limitation of this study is that the included studies were limited to a specific geographical area. The majority were conducted in Riyadh, and only one study was carried out in Jeddah. Therefore, the findings may not represent the true information about hydatid diseases in Saudi Arabia.

CONCLUSION

The systematic review highlights the presence of hydatid disease in Saudi Arabia. However, the prevalence cannot be truly estimated. The symptoms of hydatid disease were mainly in the gastrointestinal, respiratory, and musculoskeletal systems based on the site and size. The most common diagnostics tests were serological tests, such as IHA and Casoni's test, and imaging, such as ultrasonography and CT, for cyst locations. Endocystectomy and cystopericystectomy were effectively used to manage hydatid disease, where surgeries combined with medical therapy could be more effective. However, complications such as anaphylaxis, mild hypernatremia, biliary leakage, and bronchopleural fistula were reported. Increasing public awareness, promoting early diagnosis, and adopting a multidisciplinary approach combining surgical expertise with medical therapy are crucial to improving treatment outcomes and minimizing complications of hydatid disease in Saudi Arabia.

CONFLICT OF INTEREST

The author declared that there is no conflict of interest that is related to this study and this article.

DISCLOSURE

The author did not receive any form of commercial support, including compensation or financial assistance, for this case report. Additionally, the author has no financial interest in any of the products, devices, or drugs mentioned in this article.

ETHICAL APPROVAL

Not applicable.

REFERENCES CITED

- [1] Eckert J, and Thompson RC. Historical Aspects of Echinococcosis. *Adv Parasitol.* 2017;95:1-64. doi:10.1016/bs.apar.2016.07.003
- [2] McManus DP, Zhang W, Li J, and Bartley PB. Echinococcosis. *Lancet.* 2003;362(9392):1295-1304. doi:10.1016/S0140-6736(03)14573-4
- [3] Polat P, Kantarci M, Alper F, Suma S, Koruyucu MB, and Okur A. Hydatid disease from head to toe. *Radiographics.* 2003;23(2):475-537. doi:10.1148/rg.232025704
- [4] Torgerson PR, and Budke CM. Echinococcosis--an international public health challenge. *Res Vet Sci.* 2003;74(3):191-202. doi:10.1016/s0034-5288(03)00006-7

- [5] Kern P, Wen H, Sato N, et al. WHO classification of alveolar echinococcosis: principles and application. *Parasitol Int.* 2006;55 Suppl:S283-S287. doi:10.1016/j.parint.2005.11.041
- [6] Meneghelli UG, Martinelli AL, Llorach Velludo MA, Bellucci AD, Magro JE, and Barbo ML. Polycystic hydatid disease (*Echinococcus vogeli*). Clinical, laboratory and morphological findings in nine Brazilian patients. *J Hepatol.* 1992;14(2-3):203-210. doi:10.1016/0168-8278(92)90159-m
- [7] Nunnari G, Pinzone MR, Gruttadauria S, et al. Hepatic echinococcosis: clinical and therapeutic aspects. *World J Gastroenterol.* 2012;18(13):1448-1458. doi:10.3748/wjg.v18.i13.1448
- [8] Rahim F, Qasim NH, Zhumagaliuly A, and Dzhusupov K. Human Cystic Echinococcosis in The Populations of MENA Countries, With A Focus on The United Arab Emirates, From 1990 To 2019: From Genetic Epidemiology of Rare Disease to Systematic Analysis for the Global Burden of Disease Study 2019. 2023 doi:10.21203/rs.3.rs-3190738/v1
- [9] Akbulut S. Parietal complication of the hydatid disease: Comprehensive literature review. *Medicine (Baltimore).* 2018;97(21):e10671. doi:10.1097/MD.00000000000010671
- [10] Fadel SA, Asmar K, Faraj W, Khalife M, Haddad M, and El-Merhi F. Clinical review of liver hydatid disease and its unusual presentations in developing countries. *Abdom Radiol (NY).* 2019;44(4):1331-1339. doi:10.1007/s00261-018-1794-7
- [11] Ben-Hamda K, Maatouk F, Ben-Farhat M, et al. Eighteen-year experience with echinococcosis of the heart: clinical and echocardiographic features in 14 patients. *Int J Cardiol.* 2003;91(2-3):145-151. doi:10.1016/s0167-5273(03)00032-9
- [12] Göğüş C, Safak M, Baltacı S, and Türkölmez K. Isolated renal hydatidosis: experience with 20 cases. *J Urol.* 2003;169(1):186-189. doi:10.1016/S0022-5347(05)64064-5
- [13] Nourbakhsh A, Vannemreddy P, Minagar A, Toledo EG, Palacios E, and Nanda A. Hydatid disease of the central nervous system: a review of literature with an emphasis on Latin American countries. *Neurol Res.* 2010;32(3):245-251. doi:10.1179/016164110X12644252260673
- [14] Dadoukis J, Gamvros O, and Aletras H. Intrahepatic rupture of the hydatid cyst of the liver. *World J Surg.* 1984;8(5):786-790. doi:10.1007/BF01655782
- [15] Eckert J, Deplazes P, Craig PS, Gemmell MA, Gottstein B, Heath D, Jenkins DJ, Kamiya M, and Lightowler M. Echinococcosis in animals: clinical aspects, diagnosis and treatment. WHO/OIE Manual on echinococcosis in humans and animals: a public health problem of global concern. 2001:72-99. doi: 10.5555/20013100617
- [16] Mandolkar S, B R, PL A, GT S. Cystocutaneous fistula of the left lobe of liver: An extremely rare presentation of hydatid liver cyst. *International Surgery Journal.* 2015;2(1):109. doi:10.5455/2349-2902.isj20150224
- [17] Kjossev KT, and Teodosiev IL. Cutaneous fistula of liver echinococcal cyst previously misdiagnosed as fistulized rib osteomyelitis. *Trop Parasitol.* 2013;3(2):161-165. doi:10.4103/2229-5070.122150
- [18] Kammerer WS, and Schantz PM. Echinococcal disease. *Infect Dis Clin North Am.* 1993;7(3):605-618.
- [19] Bektas S, Erdogan NY, Sahin G, Kir G, and Adas G. Clinicopathological findings of hydatid cyst disease: a retrospective analysis. *Ann Clin Pathol.* 2016;4(3):1071. doi: 10.47739/2373-9282/1071
- [20] BEKÇI TT. Diagnosis and treatment of human hydatid disease. *Eur. J. Gen. Med.* 2012;9(12):15-20. doi:10.29333/ejgm/82498
- [21] Safioleas M, Misiakos E, Manti C, Katsikas D, and Skalkeas G. Diagnostic evaluation and surgical management of hydatid disease of the liver. *World J Surg.* 1994;18(6):859-865. doi:10.1007/BF00299087
- [22] Tüzün M, Altınörs N, Arda IS, and Hekimoğlu B. Cerebral hydatid disease CT and MR findings. *Clin Imaging.* 2002;26(5):353-357. doi:10.1016/s0899-7071(02)00449-7
- [23] Alexiou K, Mitsos S, Fotopoulos A, et al. Complications of Hydatid Cysts of the Liver: Spiral Computed Tomography Findings. *Gastroenterology Res.* 2012;5(4):139-143. doi:10.4021/gr460e
- [24] Tuazon AO, and Pasterkamp H. Hydatid disease of the lung (pulmonary hydatidosis). In: Chernick V, Boat TF, Kendig EL, editors. *Kendig's disorders of the respiratory tract in children.* Philadelphia (PA): WB Saunders. 1998;1050-7
- [25] Brunetti E, Kern P, and Vuitton DA; Writing Panel for the WHO-IWGE. Expert consensus for the diagnosis and treatment of cystic and alveolar echinococcosis in humans. *Acta Trop.* 2010;114(1):1-16. doi:10.1016/j.actatropica.2009.11.001
- [26] Ferrer Inaebnit E, Molina Romero FX, Segura Sampedro JJ, González Argenté X, and Morón Canis JM. A review of the diagnosis and management of liver hydatid cyst. *Rev Esp Enferm Dig.* 2022;114(1):35-41. doi:10.17235/reed.2021.7896/2021
- [27] Al-Saeedi M, Ramouz A, Khajeh E, et al. Endocystectomy as a conservative surgical treatment for hepatic cystic echinococcosis: A systematic review with single-arm meta-analysis. *PLoS Negl Trop Dis.* 2021;15(5):e0009365. Published 2021 May 12. doi:10.1371/journal.pntd.0009365
- [28] Page MJ, McKenzie JE, Bossuyt PM, et al. The PRISMA 2020 statement: An updated guideline for reporting systematic reviews. *Int J Surg.* 2021;88:105906. doi:10.1016/j.ijisu.2021.105906
- [29] Sterne JA, Hernán MA, Reeves BC, et al. ROBINS-I: a tool for assessing risk of bias in non-randomised studies of interventions. *BMJ.* 2016;355:i4919. Published 2016 Oct 12. doi:10.1136/bmj.i4919
- [30] Kassimi MA, Ali M, Zimmo SK, Khan MA, and Anees AM. Pattern of liver disease in the western region of Saudi Arabia. *Ann Trop Med Parasitol.* 1983;77(2):179-186. doi:10.1080/00034983.1983.11811695

- [31] Hossain A, Bolbol AS, and Chowdhury MN. Serodiagnosis of human hydatid disease in Riyadh, Saudi Arabia. *Ann Trop Med Parasitol*. 1985;79(4):439-442. doi:10.1080/00034983.1985.11811942
- [32] Al-Kraida A, Alam MK, Qazi S, Al-Qasabi QO, and Bashier AM. Hydatid disease of the liver in Riyadh. *Ann. Saudi Med*. 1988 Mar;8(2):117-21.
- [33] Alam AA. Epidemiology of hydatid disease in Riyadh: A hospital-based study. *Ann Saudi Med*. 1999;19(5):450-452. doi:10.5144/0256-4947.1999.450
- [34] Al Mofleh IA, Al Rashed RS, Ayoola EA, et al. Hepatic granulomas in an arab population: a retrospective study from a teaching hospital in Riyadh. *Saudi J Gastroenterol*. 2000;6(1):41-46.
- [35] Fahim F, and Al Salamah SM. Cystic echinococcosis in Central Saudi Arabia: a 5-year experience. *Turk J Gastroenterol*. 2007;18(1):22-27.
- [36] Al-Alem F, Mattar RE, Fadl OA, Alsharabi A, Al-Saif F, and Hassanain M. Morbidity and mortality and predictors of outcome following hepatectomy at a Saudi tertiary care center. *Ann Saudi Med*. 2016;36(6):414-421. doi:10.5144/0256-4947.2016.414
- [37] Ashour MH, Hajjar WM, Ishaq M, et al. Pulmonary hydatid cysts: the naturally occurring models for rupture. *Asian Cardiovasc Thorac Ann*. 2016;24(7):670-675. doi:10.1177/0218492316658374
- [38] Laajam MA, and Nouh MS. Hydatidosis: clinical significance and morbidity patterns in Saudi Arabia. *East Afr Med J*. 1991;68(1):57-63.
- [39] Halezeroglu S, Celik M, Uysal A, Senol C, Keles M, Arman B. Giant hydatid cysts of the lung. *J Thorac Cardiovasc Surg*. 1997;113(4):712-717. doi:10.1016/S0022-5223(97)70228-9
- [40] Lodhia J, Chugulu S, Sadiq A, Msuya D, and Mremi A. Giant isolated hydatid lung cyst: two case reports. *J Med Case Rep*. 2020 Oct 24;14(1):200. doi: 10.1186/s13256-020-02524-4.
- [41] Warren KS. A functional classification of granulomatous inflammation. *Ann N Y Acad Sci*. 1976;278:7-18. doi:10.1111/j.1749-6632.1976.tb47011.x
- [42] Kattan YB. Intrahepatic rupture of hydatid cyst of the liver. *Br J Surg*. 1975;62(11):885-890. doi:10.1002/bjs.1800621108
- [43] Malaika SS, Attayeb A, Sulaimani S, and Reddy JJ (1981). Human echinococcosis in Saudi Arabia. *Saudi Med J*. 1981;2:77-84. doi:10.5555/19832901052
- [44] Langer JC, Rose DB, Keystone JS, Taylor BR, and Langer B. Diagnosis and management of hydatid disease of the liver. A 15-year North American experience. *Ann Surg*. 1984;199(4):412-417. doi:10.1097/0000658-198404000-00007
- [45] Eckert J, Gemmell MA, Meslin FX, Pawlowski ZS. WHO/OIE manual on echinococcosis in humans and animals: a public health problem of global concern. Paris: World Organisation for Animal Health. 2002 Jan:20-72. Accessed 03 March 2024. <https://www.who.int/publications/item/929044522X>
- [46] WHO Informal Working Group. Guidelines for the treatment of cystic and alveolar echinococcosis in humans. *Bull WHO*. 1996;74:231-42. Accessed 03 March 2024. <https://cir.nii.ac.jp/crid/1573387451068892672>
- [47] Gessese AT. Review on Epidemiology and Public Health Significance of Hydatidosis. *Vet Med Int*. 2020 Dec 3;2020:8859116. doi: 10.1155/2020/8859116