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Systematic Review

Hydatid Cyst in the Pulmonary Artery: A Systematic Review

Dlsoz M. Hussein[1], Fahmi H. Kakamad[1,2,3*], Bnar J. Hama Amin[1,4], Dana H. Baqi[1,2], Soran H. Tahir[1,2], Abdulwahid M. Salih[1,2], Razhan K. Ali[4], Fattah H. Fattah[1,2], Gasha S. Ahmed[3], Berun A. Abdalla[1,3], Hussein M. Hamasalih[1,5], Hiwa O. Abdullah[1,3], Sabah J. Hassan[1,6]

1. *Smart Health Tower, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq*
2. *College of Medicine, University of Sulaimani, Madam Mitterrand Street, Sulaimani, Kurdistan, Iraq*
3. *Kscien Organization, Hamdi Str, Azadi Mall, Sulaimani, Kurdistan, Iraq*
4. *Shar Hospital, Sulaimani, Kurdistan, Iraq*
5. *College of Nursing, University of Sulaimani, Sulaimani, Kurdistan, Iraq*
6. *Ministry of Health, Sulaimani, Kurdistan, Iraq*

[*] ***Corresponding author***: *fahmi.hussein@univsul.edu.iq (F.H. Kakamad). Doctor City, Building 11, Apartment 50, Zip code: 46001, Sulaimani, Iraq*

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Abstract

Introduction

It is uncommon to have a hydatid cyst in the pulmonary artery. Typically, it results from an intracardiac cyst or a surgical cyst rupturing from a hepatic origin. The objective of this systematic review is to describe this rare disease.

Methods

The electronic search engine and database "Google Scholar" and "PubMed" were systematically searched for pertinent publications in the English language.

Results

A total of 45 papers were initially found by the systematic search; however, only 16 studies that comprised 16 patients were included. Eight (50%) of the patients were male and eight (50%) were female. Hemoptysis, dyspnea, bloody sputum, vomiting, and shortness of breath were the clinically significant symptoms. Twelve (75%) of the patients had a history of hepatic hydatid disease, followed by three patients (18.8%) with primary lung cystic lesions, and one (6.3%) patient with primary cardiac lesions. Severe pulmonary hypertension, septic shock, and massive hemoptysis all contributed to the death of three (18.8%) patients.

Conclusion

Localization of hydatid cysts inside the pulmonary artery is incredibly uncommon but life-threatening condition. Surgery may be the last remaining choice for some patients.

1. Introduction

Hydatidosis is a parasitic infection caused by *Echinococcus granulosus* larvae. The parasite enters the human body via the gastrointestinal route, most frequently, the liver and lungs are affected [1]. Even though the disease can spread to any organ system via systemic vessels, vital organs such as the heart and brain can occasionally be affected [2]. Hydatid cysts in the pulmonary arteries are extremely uncommon, they might develop as a result of cardiac hydatid cyst embolizing through the pericardium, interventricular septum, and cardiac chamber walls or by the passage of the hydatid cyst from the liver into the inferior vena cava, right heart chambers, and into the pulmonary artery and/or from lung hydatic lesions [3]. The various

symptoms of this disease can include anything from cough, dyspnea, and chest pain to severe hemoptysis. The disease must be managed as soon as possible, especially if they have severe hemoptysis or chest pain [4]. High mortality is associated with the cyst's location inside the pulmonary artery due to secondary dissemination, embolization, and anaphylactic shock after cyst rupture [5]. When pulmonary hypertension and cystic infiltrations of the lung occur in endemic regions, the presence of normal acute phase reactants should trigger an echocardiogram and a serology test for hydatid disease [6]. Ultimately, accurate evaluation of the unique radiologic signs of hydatid cyst embolization to the pulmonary arteries is essential for timely diagnosis and the implementation of suitable treatment [7]. The objective of the current study is to present a systematic review of studies that have been published regarding the hydatid cyst inside the pulmonary artery.

2. Methods

2.1. Study design

This study was a systematic review of the published studies on pulmonary artery hydatid cyst, which was conducted according to the Preferred Reporting Items for Meta-analyses (PRISMA) guideline.

2.2. Data sources and search strategy

The electronic search engine and database "Google Scholar" and "PubMed" were searched exhaustively for all pertinent publications in the English language, using the key terms "Pulmonary artery hydatid", "Pulmonary artery Echinococcus", "Pulmonary artery echinococcosis", "pulmonary hydatid" "Pulmonary arterial hydatid", "Arterial echinococcosis", "Vascular hydatid", "Vascular echinococcosis", "Pulmonary artery echinococcosis". To supplement data collection, references mentioned in the included papers were also checked.

2.3. Eligibility criteria

The study's selection criteria concentrated on all articles that discussed embolization, hydatid cysts in the pulmonary artery, and life-threatening consequences associated with arterial hydatid disease.

Articles were excluded due to 1) abstracts and titles that were unrelated, 2) language being non-English, 3) pre-print articles, and 4) being published in predatory journals.

2.4. Data extraction

Multiple data were collected from the included articles, including the year of publication, first author, country, age, clinical manifestations, treatments, and disease localization.

2.5. Data analysis and synthesis

The extracted data were calculated and thoroughly re-evaluated. They have been displayed as counts, proportions, and mean values

3. Results

The initial database searches turned up a total of 45 articles. The initial title and abstract screening resulted in the removal of three abstract papers, six duplicates, four non-English, and six irrelevant. The full-text screening of the remaining 26 articles resulted in the removal of seven unrelated studies and 3 papers being published in predatory journals. Sixteen papers were selected for the final analysis. All of the studies were case reports. The PRISMA chart is detailed in Figure 1. And, Figure 2 displays the geographical distribution of studies on hydatid cysts inside the pulmonary artery. The current systematic review included a total of 16 cases, eight cases (50%) were male and eight (50%) were female. The mean age was 42.62, ranging from 14 to 86 years. The included cases were associated with various manifestations: 8 (50%) cases had hemoptysis, 9 (56.2%) cases had dyspnea, 4 (25%) had bloody sputum, and 2 (12.5%) had vomiting and shortness of breath (Table 1 and 2).

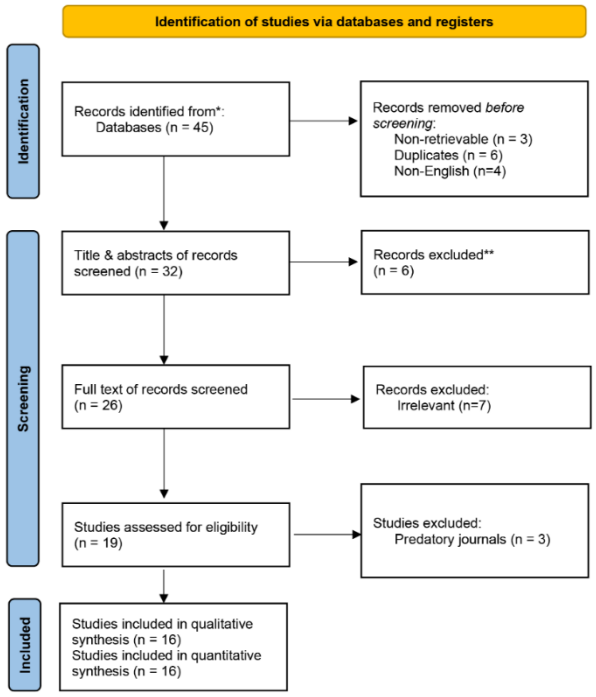

Figure 1. Study selection PRISMA flow chart.

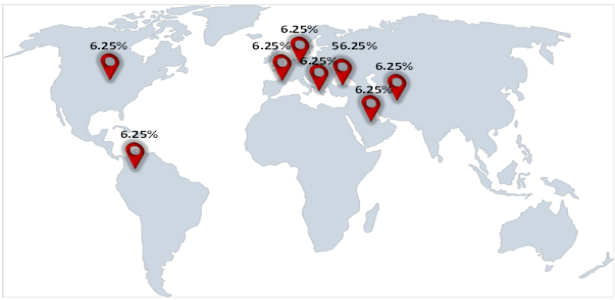

Figure 2. Geographical distribution of the studies on hydatid cysts inside the pulmonary artery.

Table 1. Characteristics of the included studies.

Author	Country	N. cases	Published date	Gender	Age	Clinical manifestations
Tercan et al. [1]	Turkey	1	2005	Male	39	Hemoptysis, dyspnea
Alper et al. [2]	Turkey	1	1995	Male	55	Dyspnea
Sirmali et al. [3]	Turkey	1	2006	Female	33	Fatigue, dyspnea, pain, swelling in the leg
Attaran et al. [4]	Iran	1	2019	Female	50	Dyspnea, hemoptysis, fever
Karantanas et al. [5]	Greece	1	2000	Male	67	Dyspnea, hemoptysis, bloody sputum
Çetin et al. [6]	Turkey	1	2017	Female	14	Fatigue, dyspnea
Namn et al. [7]	USA	1	2003	Male	32	Chest pain, vomiting, hemoptysis
Yagüe et al. [8]	Spain	1	1998	Male	44	Hemoptysis, vomiting
Tanju et al. [9]	Turkey	1	2005	Female	21	N/A
Koksal et al. [10]	Turkey	1	2006	Male	24	Cough with bloody stained sputum
Bulman et al. [11]	Columbia	1	2007	Male	37	Dyspnea, hemoptysis
Bayraktaroglu et al. [12]	Turkey	1	2009	Female	28	Dyspnea, hemoptysis
Demirpolat et al. [13]	Turkey	1	2014	Female	76	Weight loss, cough with bloody sputum, loss of appetite,
Schuuring et al. [14]	Netherland	1	2016	Female	44	Sudden thoracic pain, shortness of breath
Bakan et al. [15]	Turkey	1	2016	Male	32	Dyspnea, hemoptysis, bloody sputum
Almutairi et al. [16]	Saudi Arabia	1	2018	Female	86	Productive cough, mild shortness of breath

Table 2. Clinical characteristics of the involved patients.

References	Treatments	Lung hydatic lesion	Hepatic involvement	Cardiac involvement	Previous surgical cyst resection	Arterial cyst localization
Tercan et al. [1]	Surgical cyst resection	Yes	Yes	Yes	Yes	Dissemination from liver
Alper et al. [2]	Surgical cyst resection	Yes	Yes	Yes	Yes	Dissemination via IVC then through cardiac chamber
Sirmali et al. [3]	Surgical cyst resection	No	Yes	No	Yes	Dissemination via IVC
Attaran et al. [4]	Surgical cyst resection	No	Yes	Yes	Yes	Dissemination via IVC cyst rupture in cardiac chamber
Karantanas et al. [5]	On medication	Yes	No	No	No	dissemination from lung parenchyma
Çetin et al. [6]	On medication	No	No	Yes	No	Germinative membrane embolization from cardiac
Namn et al. [7]	On medication	No	Yes	No	No	Dissemination via IVC
Yagüe et al. [8]	Surgical cyst resection	No	Yes	No	Yes	Dissemination from liver
Tanju et al. [9]	Surgical cyst resection	Yes	No	No	Yes	Dissemination from lung
Koksal et al. [10]	Surgical cyst resection	Yes	Yes	No	Yes	Dissemination from liver
Bulman et al. [11]	Surgical cyst resection	Yes	Yes	No	No	Dissemination via IVC or cystotomy of lung hydatid cysts
Bayraktaroglu et al. [12]	On medication	Yes	Yes	Yes	Yes	Dissemination via IVC cyst rupture in cardiac chamber
Demirpolat et al. [13]	On medication	Yes	Yes	No	No	Dissemination via IVC
Schuuring et al. [14]	On medication	Yes	Yes	yes	No	Dissemination via IVC and lung parenchyma

Bakan et al. [15]	Surgical cyst resection	Yes	No	No	No	Lung hydatid cyst dissemination
Almutairi et al. [16]	On medication	Yes	Yes	No	Yes	Dissemination by cystotomy of lung and hepatic hydatid cysts

** IVC, Inferior Vena Cava*

The echinococcosis therapies used in the past may have contributed to hydatid cyst spread to the pulmonary artery. However, there was not a statistically significant association between them in this study (Table 3). Hemoptysis, a vascular involvement symptom, and a history of prior organ involvement are shown in Table 4 and their relationship was non-significant.

Table 3. Hydatid cyst dissemination to the pulmonary artery and its relation to the treatment methods.

Previous treatment methods	Dissemination of a hydatid cyst to the pulmonary artery from the infected organs					
	Liver	Lung and cardiac chamber	Liver and lung	Primary cardiac lesion	Total	*P-value*
Surgical cyst resection	6 (66.7%)	1 (11.1%)	2 (22.2%)	0 (0.0%)	9 (100.0%)	
Treatment on medication	4 (57.1%)	2 (28.5%)	0 (0.0%)	1 (14.3%)	7 (100.0%)	0.4
Total	10 (62.5%)	3 (18.7%)	2 (12.5%)	1 (6.25%)	16 (100%)	

Table 4. Hydatid cyst dissemination to the pulmonary artery and hemoptysis as a sign of vascular involvement.

Clinical sign of hemoptysis	Dissemination of a hydatid cyst to the pulmonary artery from the infected organs					
	Liver	Lung and cardiac chamber	Liver and lung	Primary cardiac lesion	Total	*P-value*
Yes	6 (75%)	1 (12.5%)	1 (12.5%)	0 (0.0%)	8 (100.0%)	
No	4 (50.0%)	2 (25.0%)	1 (12.5%)	1 (12.5%)	8 (100.0%)	0.2
Total	10 (62.5%)	3 (18.7%)	2 (25.0%)	1 (6.3%)	16 (100.0%)	

4. Discussion

Cystic echinococcosis is a zoonosis caused by the cestode *Echinococcus granulosus* larval stage. Carnivores, including dogs, are definitive hosts. However, sheep and other ruminants act as intermediate hosts. By consuming food or water that has been contaminated with dog feces that contain the parasite eggs, humans become infected. These embryos cross the intestinal wall to reach the hepatic portal system, where they encyst in the liver and other organs of the intermediate host [12]. The lungs filter 15%–25% of the embryos, the liver filters 60%–70% of them, and 10%–15% of them are transported by systemic circulation to other organs [1].

The parasite and the host both contribute to the hydatid cyst's wall structure. The fibrous tissue that makes up the pericyst, which is the outer layer, is created by the host in reaction to the infection. The protoscolices containing inner germinal layer known as the endocyst and the acellular outer laminated membrane known as the ectocyst combined make up the actual wall of the hydatid cyst [7]. Cyst rupture causes the protoscolices to spread, which can lead to the formation of secondary cysts in the infected tissues. Blood tests for hydatid infection can show anti-echinococcal antibodies and eosinophilia, but the latter is typically linked to cyst rupture [17].

Just a few ideas exist to explain the highly rare occurrence of a hydatid cyst inside the pulmonary artery [10]. The most common cause of localization is embolization from primary cardiac locations. Another possibility is that the embryos travel from the liver into the inferior vena cava and then travel to the pulmonary arteries via the right cardiac chambers [8]. Cysts in the lung parenchyma will result in a tiny break in the arteries' walls, allowing the parasites to pass through [5]. In the current analysis, hydatid cysts in 12 patients had either spread from the liver to the lung or had embolized through the inferior vena cava from the liver to the pulmonary arteries.

The heart can be afflicted in only 0.5% to 2% of all cases of hydatidosis. The interventricular septum, right or left atrium, right ventricle, pericardium, and left or right ventricle may also be damaged, though the left ventricle is most usually afflicted (50–60%) [1]. In the case described by Çetin et al., hydatid cyst in the bilateral pulmonary arteries was most likely caused by germinative membrane embolization with the spread of hydatid cysts from the right ventricle and the subsequent development of chronic thrombosis [6]. According to the case reported by Tercan et al., no cysts were found in the right heart chambers or walls, although a hepatic hydatid cyst had previously undergone surgical excision. Since the liver's scolices ruptured into the hepatic veins during surgery, it was suspected that the cysts in the pulmonary arteries were scolices emboli [1]. A total of 50% of pulmonary artery echinococcosis cases had a history of hepatic hydatid cysts being surgically removed in the present analysis.

Echinococcosis of the lungs might be asymptomatic or mimic thromboembolic disease. Common symptoms of pulmonary echinococcosis are chest pain, dyspnea, coughing up cyst

contents, hemoptysis, and/or an anaphylactic reaction [11]. Most intact lung cysts are found accidentally on chest radiography due to a primary infection in the lung parenchyma, which may go undetected for many years [16]. Clinical classifications of hydatid pulmonary embolism include acute fatal, sub-acute with pulmonary hypertension and mortality within a year, and chronic pulmonary hypertension [18]. The embryos located within an artery usually grow slowly, finally occluding it, and possibly asymptomatic at first. If the cysts continue to grow, symptoms may develop over time, and pulmonary perfusion may become inadequate when they compress a vital structure or obstruct blood flow, leading to anaphylactic shock [1]. The patient in the case study by Bakan et al., lived symptom-free for three years before having a cystotomy capitonnage for right-sided pulmonary hydatid cysts. It may be explained by the cysts' slow development rate as this might continue long enough for collaterals to develop [15].

When the cyst material is discharged from the cyst owing to trauma or surgery, anaphylactic shock may result in death [16]. Additionally, systemic cyst problems such as sepsis, respiratory failure as well as perioperative difficulties may be caused by spontaneous cyst rupture or by the dysfunction of the affected organ as a result of cyst growth [14]. Increasingly, hemoptysis in patients who are prone to have hydatid cysts could be a warning sign of vascular involvement, which requires close vital sign monitoring and reanimation [4]. In the current analysis, hemoptysis as a marker of vascular involvement and hydatid cyst dissemination to the pulmonary artery occurred in 50% of patients. And by this analysis, 4 patients have had pulmonary hypertension.

If a computed tomography scan shows spherical, fluid-attenuating intravascular masses that are hydatid cysts in the pulmonary arteries. These cysts may lead to artery hypertrophy. There has also been evidence of calcification around the edges of the cysts [7]. Furthermore, by exhibiting lesions that are cystic and highlighting homogenously hypointense lesions in T1 weighted images and hyperintense lesions in T2 weighted images, magnetic resonance imaging may help with diagnosis. The cyst walls have low signal margins that are visible on the T2-weighted images. Computed tomography and magnetic resonance imaging are helpful in follow-up to investigate in recurrence and formation of pseudoaneurysm. Hydatid cysts are also confirmed by pathological examinations [15]. Treatment for hydatid cysts is crucial and the fluid from a hydatid cyst is a highly effective anaphylactic agent. Because surgery has a high morbidity and mortality rate, it must be customized for each patient if can be complicated by artery or cyst rupture, disease propagation, anaphylactic shock, embolism, and the development of a pseudoaneurysm. As a result, this choice must be made with great care [14]. While in the right circumstances, a cyst should be surgically removed and additional medicine employing benzimidazole substances, such as albendazole and mebendazole, should be used as a therapy of choice [12]. The surgical methods used to treat hydatid disease of the lungs include enucleation of the lung, pericystectomy, cystotomy with or without capitonnage, and other surgical procedures [9]. Hydatid disease pre and postoperative medication are effective in eliminating spilled scolices in ruptured cysts. The surgical outcomes depend on the patient's previous diseases and the severity of their underlying diseases [4].

The findings of Namn and colleagues' investigation led to the advice of some prophylactic measures because they showed that surgical removal of hepatic echinococcal cysts proximal to hepatic venous structures also entails a risk of hydatid cyst embolization to the pulmonary arteries. These include clamping of the inferior vena cava, avoidance of traction on the liver, and cavocaval bypass if necessary [7]. In a case presentation with concurrent hydatid cysts inside the pulmonary artery and the lung parenchyma, according to Koksal et al., it is crucial to first remove the intra-arterial cyst followed by the care of the lung cyst with a parenchyma-saving procedure or lung resection is critical [10]. According to Çetin et al., it is increasingly becoming evident, that it is not feasible to completely remove the cyst due to the bilateral pulmonary arterial involvement. Bilateral pulmonary hydatidosis is treated surgically like that of chronic thromboembolic pulmonary hypertension, which is treated by pulmonary artery endarterectomy. However, special care should first be given to first removing the hydatid vesicles attached to the vascular endothelium before beginning an endarterectomy [6]. A hydatid cyst in the left pulmonary artery was not suspected in the first surgical intervention performed on the patient in the case reported by Yagüe et al., The second intervention was carried out due to anaphylactic shock caused by the rupture of the cyst in the right pulmonary artery.

Five cases of pulmonary echinococcosis in the current analysis were treated with medication, while the remaining cases were treated surgically. On the other hand, three patients with pulmonary echinococcosis all passed away from severe pulmonary hypertension, septic shock, and massive hemoptysis.

5. Conclusion

Hydatid disease of the pulmonary arteries is extremely unusual and may develop when cysts rupture into the hepatic veins, heart, or lung parenchyma either spontaneously or secondary to surgery. Within the pulmonary arteries, hydatid cysts have a particular appearance on both computed tomography and magnetic resonance imaging. For an early diagnosis and effective therapy, recognition of this illness is crucial.

Declarations

Conflicts of interest: The author(s) have no conflicts of interest to disclose.

Ethical approval: Not applicable, as systematic reviews do not require ethical approval.

Patient consent (participation and publication)**:** Not applicable.

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Authors' contributions: DMH, GHS and BAA participated in data collection. AMS designed the study; HOB, SJH performed the data analysis. FHK, SHT participated in preparing the

manuscript. BJHA, DHB, RKA, FHF and HMH critically revised the manuscript. FHK, BAA confirmed the authenticity of the data. All authors approved the final version of the manuscript.

Data availability statement: Note applicable.

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